

ANGLIA RUSKIN UNIVERSITY

FACULTY OF HEALTH EDUCATION, MEDICINE AND SOCIAL CARE

**CO-DEVELOPING A HEALTH LITERACY FRAMEWORK TO INTEGRATE NUTRITION
INTO STANDARD CARE IN SICKLE CELL DISEASE**

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**A thesis in partial fulfilment of the requirements of Anglia Ruskin University for the
degree of Professional Doctorate**

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University**

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(ii); Abstract

ANGLIA RUSKIN UNIVERSITY

ABTRACT

FACULTY OF HEALTH EDUCATION, MEDICINE AND SOCIAL CARE

PROFESSIONAL DOCTORATE

**CO-DEVELOPING A HEALTH LITERACY FRAMEWORK TO INTEGRATE NUTRITION
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Nutrition in Sickle Cell Disease (SCD) is a neglected part of standard care impacting patient outcomes, despite it being widely researched. Moreover, the clinical features of SCD, a marginalised genetically inherited blood disorder, are responsible for growth and nutritional deficiencies that require nutrition service provision. Thus, a need exists to identify the influencing factors affecting the lack of nutrition integration in SCD. Presently, a paucity of research exists on how to integrate nutrition into standard care in SCD using health literacy in a novel way to support policy and practice development.

The study adopted a four phased sequential participatory Learning Alliance Methodology, involving sickle cell service users and carers (n=11) and service providers (n=7), between March to December, 2020, to co-develop a health literacy framework to support nutrition integration in SCD. Independent focus groups (phase one), network meetings (phase two, three and four) and an evaluation questionnaire, was used to collect the data.

Thematic analysis of the focus groups outcomes revealed four common themes namely; (1) Invisibility of SCD, (2) Under-recognised importance of nutrition, (3) Lack of priority to nutrition and (4) Multi-level factors affecting nutrition and service provision that together reflect key influencing factors identified as knowledge and care gaps, essential to tailor policy and practice in nutrition in SCD. Following consensus development and validation through network meetings, the evaluation of the health literacy framework (phase four), found the framework to be a valuable educational, communication and policy tool.

Overall the findings confirm the complexity, invisibility and neglect of nutrition service provision as part of standard care in SCD, a health inequality impacting patient experience, access and health outcomes, explained by the marginalisation of SCD. Hence, the influencing factors identified in the study require a whole systems policy and practice strategy to integrate nutrition into standard care in SCD.

Keywords: Sickle Cell Disease, Nutrition integration, Influencing factors, Health literacy framework, Learning Alliance Methodology, Policy and practice

(iii); Table of Contents

Acknowledgements-----	i
Abstract-----	ii
Contents-----	iii
List of Figures/Diagrams-----	iv
List of Tables-----	v
List of Abbreviations-----	vi
Research Terms and Definitions-----	vii

Chapter 1:

Introduction-----	15
1.1 Thesis Statement-----	15
1.2 Overarching aim and objectives-----	16 -17
1.3 Gap in knowledge-----	17-19
1.4 Rationale for the study-----	19-25
1.5 Thesis structure-----	25-26
Summary-----	27

Chapter 2:

Literature Review-----	28
Exploring the influencing factors affecting nutrition integration in SCD-----	28
2.1.1 Literature review strategy-----	28
(i) Search Design-----	28
(ii) Inclusion and exclusion criteria-----	29
(iii) Systematic literature review-----	30-31
(vi) Critical evaluation of the literature-----	32-34
2.1.2 Medical management in SCD-----	35
(i) Health factors in SCD-----	35-37
(ii) Social factors in SCD-----	37-43
2.1.3 Role of nutrition in SCD-----	43

(i) Clinical features affecting nutrition in SCD-----	43-47
(ii) Main nutritional problems-----	48-50
2.1.4 Nutritional management in SCD-----	51
(i) Global context-----	52-57
(ii) UK context-----	57-58
2.1.5 Influencing factors in nutrition in SCD-----	58
(i) Socio-ecological factors -----	58-60
(ii) Multi-factorial factors-----	60-62
2.1.6 Policy and practice development in nutrition integration in SCD-----	62-63
(i) Health literacy in policy and practice development -----	63-65
Summary-----	66

Chapter 2.2

Theoretical and Professional Perspectives-----	67
2.2.1 Freire's theoretical framework-----	67
(i) Rationale-----	67-69
(ii) Freire's theoretical concepts-----	69-74
2.2.2 Professional policy and practice perspectives-----	75
(i) Policies for nutrition management in SCD-----	75-76
(ii) Dietetic clinical practice-----	77-79
Summary-----	79

Chapter 3:

Methodology -----	80
3.1 Justification of the research methodology-----	80
3.1.1 Research paradigm and research strategy-----	80-85
3.1.2 The Learning Alliance Methodology-----	85-88
3.2 Sampling and Recruitment-----	89
3.2.1 Sampling method-----	89-90
3.2.2 Inclusion and exclusion criteria-----	91
3.2.3 Recruitment of participants-----	92-96

3.3 Research data collection methods and processes-----	97-98
3.3.1 Phase one: Conducting independent focus groups-----	100-103
3.3.2 Analysis of focus group data-----	104-112
3.3.3 Phase two: First Learning Alliance (LA) Network meeting-----	112
(i) Formation of the LA Network-----	112- 113
(ii) Consensus development-----	113-114
3.3.4 Phase three: Second LA network meeting-----	114
(i) Co-development of the health literacy framework-----	114-115
(ii) Co-development process-----	115
3.3.5 Phase four: Third LA network meeting-----	116
(i) Evaluation of the health literacy framework-----	116-120
(ii) Dissemination of initial research findings-----	120
3.4 Ethical perspectives-----	121-124
Summary-----	124

Chapter 4:

Results-----	125
Part one: Findings of the thematic analysis of the focus groups-----	125
4.1 Background of the research project participants-----	125-128
4.2 Focus group findings and themes-----	130
4.2.1 Medical management – Findings-----	132-136
4.2.2 Nutritional management – Findings-----	136-148
4.2.3 Nutrition service provision – Findings-----	148-156
4.2.4 Socio-ecological facilitators-----	157-158
4.3 Synthesis of SCSU-C and SCSP views-----	158
4.3.1 SCSU-C views-----	158 -159
4.3.2. SCSP views-----	160-161
4.3.3 Shared participant perspectives-----	162-163
4.4 Reflections on the process of the LA network meetings-----	164
4.4.1 First LA network meeting reflection (12/10/20) -----	164
4.4.2 Second LA network meeting reflection (22/10.20) -----	165
4.4.3 Third LA network meeting reflection (12/12/20) -----	166

Part two: Findings of the thematic analysis of the evaluation questionnaire-----	167
4.5 Evaluation questionnaire findings and themes-----	167-168
4.5.1 Theme 1: Improve knowledge and understanding of nutrition-----	169-170
4.5.2 Theme 2: Raise awareness of influencing factors affecting nutrition---	170-171
4.5.3 Theme 3: Valuable tool for communication and training-----	171
4.5.4 Theme 4: Improve policy and service provision outcomes-----	171-172
4.6 Conceptualisation of health literacy as an expansive and integrative concept---	172
4.6.1 Health literacy thematic concepts-----	172-173
4.6.2 Health literacy Nutrition Integration Conceptual Framework-----	174
4.6.3 Health literacy – Nutrition Integration Framework (HL-NIF) Blueprint—	176-177
Summary-----	179

Chapter 5:

Discussion-----	180
5.1 Marginalisation of people living with SCD-----	180-187
5.2 Influencing factors affecting nutrition in SCD-----	187
5.2.1 Medical management-----	188-190
5.2.2 Nutritional management-----	190-198
5.3 Socio-ecological factors-----	198-200
5.3.1 Micro level factors-----	200-203
5.3.2 Meso level factors-----	203-206
5.3.3 Macro level factors-----	207-210
5.3.4 Socio-ecological Facilitators-----	210-212
5.4 Clinical and nutritional policy and practice development in SCD-----	212
5.4.1 Evaluation of the health literacy framework-----	212-216
5.4.2 The LA methodology in healthcare intervention development -----	216-218
5.4.3 Health literacy in policy and practice in nutrition in SCD-----	219-222
5.5. Nutrition integration in SCD-----	223
5.5.1 Evaluation of policy and practice documents in SCD-----	223-227
5.5.2 Policy and practice recommendations to integrate nutrition-----	227-238
5.5.3 Reflexive perspectives on my personal and professional learning and development-----	238-242

5.6 Limitations and strengths-----	242
5.6.1 Limitations-----	242-244
5.6.2 Strengths-----	244-246
Summary-----	246

Chapter 6:

Conclusions and Recommendations-----	247
6.1 Conclusions-----	247-255
6.2 Contribution to knowledge-----	255-256
6.3 Recommendations for policy and practice change in nutrition in SCD-----	256-258
6.4 Future Directions-----	258-259
Summary-----	259-260

(iv); Figures/Diagrams

Chapter 2

Figure 2.1 PRISMA Framework: Identification and Screening (Page 31)

Chapter 3

Figure 3.1 The four phase research process (Page 99)

Chapter 4

Figure 4.1 Thematic mapping of the analysis of the focus group (Page 131)

Figure 4.2 SCSU-Cs concerns about the lack of nutrition service provision available to them as part of their standard care in SCD (Page 148)

Figure 4.3 A summary of the shared perspectives between SCSU-C and SCSPs about nutritional management in SCD (Page 163)

Figure 4.4 Health Literacy - Nutrition Integration Conceptual Framework (Page 175)

Figure 4.5 Health Literacy – Nutrition Integration Framework (Blueprint) (Page 178)

(v); List of Tables

Chapter 2

Table 2.1: Systematic literature review search strategy (Page 30)

Table 2.2 Search terms, aims and strategy for the critical evaluation of the literature (Page 33)

Chapter 3

Table 3.1: Characteristics of sickle cell research participants (Page 96)

Table 3.2: Characteristics of the Learning Alliance (LA) Network (Page 97)

Table 3.3: Summary of the four themes of the focus groups in phase one (Page 108)

Table 3.4: Summary of the four themes of the evaluation questionnaire in phase four (Page 119)

Chapter 4

Table 4.1 Background information and non-identifiable codes of the SCSU-C and SCSP participants (Page 129)

Table 4.2 Perceptions of optimum nutrition held by SCSU-Cs and SCSPs (Page 145)

Table 4.3 Summary table to show views of SCSU-Cs versus SCSPs about the nutritional management in SCD (Page 161)

Table 4.4 Summary of the findings and themes of the evaluation questionnaire (Page 168)

Table 4.5: Espousing health literacy thematic concepts from the main themes of phase one (Page 174)

(h) References-----Page 261-283

(h) Appendices-----Page 284-327

Appendix 1 Participant Information Sheet

Appendix 2 Original ethics approval

Appendix 3 First ethics amendment approval

Appendix 4 Second ethics amendment approval

Appendix 5 Third ethics amendment approval

Appendix 6 Research project advert

Appendix 7 Participant consent form

Appendix 8 Phase one question schedule

Appendix 9 Extract from the digital copy and summary of phase one data and developing themes

Appendix 10 APPG – SCAT Nutrition in SCD supporting letter

Appendix 11 Initial version of health literacy framework co-developed by the LA Network

Appendix 12 Phase four evaluation questionnaire

Appendix 13 Chronological timelines for key threshold concepts and pivoting moments in the research project

Appendix 14 Overlapping conceptual perspectives of the research project

Appendix 15 Additional points raised by the LA Network

Appendix 16 Agenda for the focus groups and network meetings

Appendix 17 Extracts of developing themes (phase one) from Excel Spreadsheet

Appendix 18 Thematic mapping of the evaluation questionnaire (phase four)

Appendix 19 Research output (Examples one to six)

Appendix 20 List of articles screened in the systematic literature review

(vi); List of Abbreviations

ACS	Acute Chest Syndrome
ADL	Activity of Daily Living
APPG-SCAT	All Party Parliamentary Group for Sickle Cell and Thalassaemia
ASCAT	Annual Sickle Cell and Thalassemia Conference
ASH	American Society of Haematology
BAPEN	British Association of Parenteral and Enteral Nutrition
BDA	British Dietetic Association
BEM	Black and Ethnic Minority
BMI	Body Mass Index
BSH	British Society of Haematology
BSNA	British Specialist Nutrition Association
CEO	Chief Executive Officer
DoH	Department of Health
DHSC	Department of Health and Social Care
DRM	Disease- Related Malnutrition
Hb	Haemoglobin

HbS	Sickle Cell Haemoglobin
HCP	Health Care Professional
HL	Health Literacy
HL-NIF	Health Literacy – Nutrition Integration Framework
ICB	Integrated Care Board
ICS	Integrated Care System
IT	Information Technology
LA	Learning Alliance
LAN	Learning Alliance Network
LAM	Learning Alliance Methodology
LOS	Length of Stay
LTC	Long Term Condition
MUST	Malnutrition Universal Screening Tool
NHS	National Health Service
NICE	The National Institute for Health and Care Excellence
PAG	Patient Advisory Group
PCN	Primary Care Network
PHE	Public Health England
PPI	Patient Public Involvement
PHOF	Public Health Outcomes Framework
SCD	Sickle Cell Disease
SCSU	Sickle Cell Service User
SCSU-C	Sickle Cell Service User – Carer
SCSP	Sickle Cell Service Provider
SCS	Sickle Cell Society
SEM	Socio-Ecological Model
QOL	Quality of Life
VOC	Vaso -Occlusive Crisis
UK	United Kingdom
US	United States
WHO	World health Organisation

(vii); Research Terms and Definitions

Medical Management

The medical management of SCD, refers to the identification, assessment, diagnosis, disease modifying treatment and pharmaceutical management of the clinical features associated with the pathophysiology of people living with SCD.

Nutritional Management

The nutritional management of SCD, refers to the identification, assessment, diagnosis and treatment of the nutritional needs, risks, challenges and health outcomes associated with the clinical features of the pathophysiology of people living with SCD.

Nutrition in SCD

Nutrition in SCD, refers to the nutritional needs, risks, challenges and health outcomes of people living with SCD as a consequence of the clinical features associated with the pathophysiology of the condition.

The role of nutrition in SCD

The role of nutrition in SCD relates to the consequences of the clinical features associated with the pathophysiology of the condition as it impacts on the nutritional needs, risks, challenges and health outcomes of people living with SCD.

Nutrition landscape in SCD

The nutrition landscape in SCD provides a comprehensive view to understand nutrition service provision in the condition as it relates to the nutrition needs, risks, challenges, resources, education and training, service users and service providers, health planners, multiple stakeholders in SCD care provision, policy and practice guidelines, nutrition standards and service quality standards.

Nutrition as a management option in SCD

Nutrition as a management option in SCD refers to how the knowledge of nutrition of expertly trained healthcare professionals, can be applied to address the nutrition needs, risks, challenges and health outcomes associated with the clinical features of the pathophysiology of the condition, through the process of identification, assessment, diagnosis, treatment and monitoring.

Integrating nutrition into standard care in SCD

Integrating nutrition into standard care in SCD refers to the inclusion of nutrition as a management option by improving the access, experience and outcomes of people living with SCD and whereby nutrition knowledge is used by expertly trained healthcare professionals, supported by policy and practice, to address the nutrition needs, risks, challenges and health outcomes associated with the clinical features of the pathophysiology of the condition, through the process of identification, assessment, diagnosis, treatment and monitoring.

Health literacy in nutrition in SCD

Health literacy as used in the context of this research project, aims to take a more comprehensive view to understanding the nutrition landscape in SCD. Health literacy is described in the literature as a range of outcomes to health education. Since health education is a key problem associated with poor nutrition knowledge in SCD (Matthews, 2016g) and health education is directed towards improving health literacy (Nutbeam, 2000), in the context of this research project, health literacy will be used instead of health education, in relation to policy and practice development to support nutrition service provision in SCD.

Definitions of health literacy

To contextualise the comprehensive view adopted by the researcher, health literacy is proposed as an expansive and integrative concept to both understand the nutrition landscape in SCD, and moreover in the development of policy and practice to support nutrition service provision, based on the following integrated definition of health literacy;

- “Health literacy is linked to literacy and entails people’s knowledge, motivation, and competencies to access, understand, appraise and apply health information, in order to make judgements and take decisions in everyday life concerning healthcare, disease prevention and health promotion – to maintain or improve quality of life during the life course” (Sorenson et al., 2012, p3).

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Chapter 1

(g) Introduction

This chapter provides an overview of the research problem, the lack of nutrition service provision as part of standard care in Sickle Cell Disease (SCD). The research problem is explored in light of SCD being a marginalised condition and how this has influenced the choice of research project approach, paradigm and methodology towards promoting policy and practice guidance in nutrition in SCD. A summary of the originality and contribution to knowledge is provided in relation to the overarching aim and objectives of the research project, with the co-development of a health literacy framework aiming to integrate nutrition into SCD management.

1.1 Thesis Statement

The study takes a novel approach to explore the integration of nutrition into standard care in SCD, using health literacy as an expansive and integrative concept to contribute to policy and practice development. Freire's (1970) seminal work, *'Pedagogy of the Oppressed'*, informs the theoretical underpinning of the study. As such, the study is a four phased qualitative enquiry using a sequential participatory Learning Alliance (LA) methodology, an emerging methodology in Health and Social Care (Moreno-Meguizamo, 2017), to identify the myriad of influencing factors affecting nutrition integration in SCD care. The purpose being to co-develop a health literacy framework involving Sickle Cell Service User/Carer (SCSU-C) s and Sickle Cell Service Provider (SCSP)s (the study participants) to address the knowledge and care gaps defining the nutrition landscape and management in SCD.

1.2 Overarching Aim and Objectives

Aim: *‘To support the integration of nutrition into standard care in SCD using a health literacy framework’.*

Nutritional management in SCD is not currently part of standard care provision available to people living with SCD, internationally and in the UK. A confounding factor pertains to the lack of policy and practice guidance to support nutrition service provision in SCD. Nutrition integration is a term used to express the need for nutrition to be considered as an adjunct management option alongside the existing medical management in SCD. To date there is a paucity of research focussed on how to integrate nutrition into standard care in SCD, since very little is known about the current nutrition landscape in SCD. As a result, more information is needed to understand the existing nutritional management of people living with SCD. More specifically, there is need to identify and understand the myriad of influencing factors affecting nutrition integration that may be contributing to the lack of nutrition service provision in SCD. It is hoped that the findings of this research project would inform the development of future policy and practice guidance and change in nutrition service provision, that is tailored to the unique needs of the SCD patient population, providing a more patient centred and personalised care provision.

Objective 1: To identify and understand the SCSU-C and SCSP knowledge, views and experience of the medical and nutritional management of SCD as well as the influencing socio-ecological factors and facilitators affecting nutrition and service provision to people living with SCD, to answer research question (1): *What are the influencing factors affecting nutrition integration in SCD?*

Objective 2: To form a Learning Alliance Network (LAN) between the SCSU-C and SCSP groups, then use the data collected in Objective 1, to co-develop and evaluate a health literacy framework to contribute to policy and practice guidance and change in support of

nutrition integration in SCD, to answer research question (2): *How can a health literacy framework support the integration of nutrition in SCD?*

Objective 1 (as it relates to phase one) and Objective 2 (as it relates to phase two, three and four), seeks to meet the overarching aim of this four phased research project, to support the integration of nutrition in SCD using a health literacy framework. Objective 1 draws on the principles of the participatory LA methodology (Moreno-Meguilzamo et al., 2015) and the Learning Alliance Palliative Care and End of Life (LAPCEL) study (Smith and Moreno-Meguilzamo, 2017;2019) using a multi-stakeholder approach to knowledge co –construction and sharing.

Objective 2, builds on the knowledge of co-construction and collaborative and partnership working of the participatory LA methodology, to promote the formation of a LAN between the SCSU-Cs and SCSPs. The purpose of the LAN is to reach a consensus viewpoint about the data collected in phase one of the research project towards the co-development of the health literacy framework. The partnership and collaborative working, by the members of the LAN in turn, gives them the opportunity to consider suitable suggestions to inform policy and practice guidance and change that can improve nutrition service provision, tailored to the unique needs and challenges of people living with SCD.

1.3 Gap in Knowledge

To date, no framework exists to support the integration of nutrition in SCD, confounded by the overarching gap in knowledge that pertains to the lack of research on how to integrate nutrition into standard care in SCD, using health literacy as an expansive and integrated concept to contribute to policy and practice development.

Originality:

Key original elements of the research project is firstly, acknowledgment of the SCD patient population as a marginalised group, and secondly: the need to identify and understand the factors that are driving the lack of nutrition service provision available to people living with SCD. As previously mentioned, the study uses Freire's (1970) framework as its theoretical underpinning to understand the knowledge, views and experiences of the study participants of the medical and nutritional management in SCD. To the best of my knowledge, this is the first time that Freire's framework has been used in studies relating to nutrition in SCD. Of note, there are parallels between Freire's principles and the chosen research methodology, the LA methodology (see Appendix 14). Both Freire's principles and the LA methodology principles, acknowledge the importance of working with marginalised patient groups, underpinned by critical social theory and critical theory, respectively. Their methodology promotes collaborative and partnership working to facilitate emancipation and empowerment amongst the marginalised groups involved in the study.

Contribution to knowledge:

The novel use of health literacy as an expansive and integrative concept to contribute to policy and practice supports the main output of the research project, the co-development of the health literacy framework. The subsequent development of the health literacy conceptual framework and Health Literacy –Nutrition Integration Framework (HL-NIF) (blue print) (Chapter 4, section 4.6), adds to the original contribution to knowledge. Adopting a socio-ecological lens to the research project - a first in nutrition in SCD - contributes to understanding the multiple influencing factors that result in the lack of, and explains the complexity of, nutrition and service provision through a whole systems lens. This whole systems lens resulted in the reframed conceptualisation of optimum nutrition in SCD, providing an expansive understanding of the multiple factors influencing the nutrition needs

of people living with SCD, thus adding to the theoretical knowledge contribution of the study, an important factor underpinning the rationale for the study.

1.4 Rationale for the study

Positionality of the researcher:

The rationale for the study and its choice of research topic, approach, paradigm and design, is influenced by my personal experiences of marginalisation, as a result of racism and oppression. Being exposed to the effects of racism growing up in Apartheid South Africa has framed my worldview for social justice, advocacy and equality. Living in a society fraught with marginalisation, that determined where I could or could not live, work or play, was evidence of the social injustice which has informed my lens of the world I live in.

Experiencing personal invisibility, oppression and disempowerment, all the more driving a deep set need to advocate on behalf of the voiceless and marginalised in my community.

My own experience of empowerment, through education as a means of emancipation, propelled the value I now place on both education and empowerment, particularly in those communities who were not able to advocate for themselves. As a consequence, my decision to undertake doctoral level research, was strongly influenced by my own journey of emancipation propelled by education and personal empowerment. A number of articles (Matthews, 2015c, 2016d, 2018h) and my debut book (Matthews, 2019i), narrate the positive impact of my own emancipation and empowerment. It is believed that personal empowerment, motivation and autonomy (Nutbeam, 2008) are key concepts, which when coupled with education (Freire, 1970) can support emancipation in marginalised groups. These personal experiences would prepare me for my encounter with SCD.

What is SCD?

SCD is a marginalised genetically inherited red blood cell disorder associated with high levels of morbidity, mortality, disability and poor Quality of Life (QOL) outcomes (Fitzburgh et al., 2005; Lucas and Mason, 2008; Aljuburi et al., 2012; Piel et al, 2021). SCD is considered a global public health problem according to the World Health Organisation (WHO, 2006) and United Nations (UN, 2008). SCD originates in the tropical regions of the world, because of its advantage against malaria (Sergeant, 2001). The gene is primarily found in African and non-African people in equatorial Africa, southern Italy, northern Greece, southern Turkey, the eastern province of Saudi Arabia and India (Asnani, 2010; Serjeant, 2013; Hyacinth, Adekeye and Yilgwan, 2013).

Moreover, SCD is a growing health burden, evidenced in the growing numbers of individuals predicted to be affected with the haemoglobinopathy (Piel et al., 2017). Overall, SCD has a higher prevalence than any other genetic condition in the UK, with an estimated 15 000 people, National Institute for Health and Care Excellence (NICE 2021). The estimated SCD patient population in the UK is based on the data collected by Public Health England (PHE, 19/20) in England, that shows that 1 in 2,517 babies screened for SCD, were found to be screen positive for significant conditions, and 1 in 78 were carriers of the sickle cell gene. However, with the influx of populations known to have a high risk of SCD into the UK, there is a greater need to keep accurate records of the number of people affected with the disorder. The increased number of carriers and those with the condition, has public health implications.

Predominantly, SCD affects people from a Black and Minority Ethnic (BME) background (Green et al, 2012; SCS, 2018; NICE, 2021). Although the research topic is focussed on nutrition in SCD, it is prudent to acknowledge the marginalisation of SCD as a patient population. The impact of the marginalisation of the SCD patient population has been emphasised recently, in a watershed report by the All Party Parliamentary Group for Sickle

Cell and Thalassaemia (APPG-SCAT, 2021). This report was commissioned by the Sickle Cell Society (SCS), in response to the coroner's report about the avoidable death of a 21-year-old sickle cell patient, (Evan Smith, 2019) whilst seeking care in the National Health Service (NHS), in the UK. A number of failings in the quality of care provision provided to people living with SCD, in relation to their experience, access and outcomes of care, were highlighted in this report. The report further highlighted the role of racism being an underlying reason for the poor quality of care provided to people living with SCD (House of Commons briefing, 2021), evidence of the invisibility and health inequalities in the medical management in SCD.

In addition to the effects of the invisibility and related health inequalities in SCD management, high hospital readmission rates to treat the clinical symptomology of people living with SCD add to the already high cost and health burden associated with this chronic condition (Aljuburi et al., 2012; Aljuburi, 2015). The healthcare burden in SCD, is not limited to the clinical symptomology, as a number of social and wider determinants of health (Matthews, 2015b; Berghs et al., 2020) was found to impact on the QOL and health and wellbeing outcomes of this patient group – as I myself witnessed while working as a social liaison officer in a SCD service in East London. Augmenting these existing confounding factors, is the under recognition of the nutritional needs and nutritional management available to people living with SCD – and yet, the role of nutrition is currently overlooked as a management option in SCD.

Nutrition in SCD:

The under recognition of the importance of nutrition in SCD, is a significant confounding factor in the nutritional management of people living with SCD. The absence of nutrition as a topic to be addressed in SCD management, whilst working as a social liaison officer in a SCD service, provided overwhelming evidence of the invisibility of the importance of nutrition in SCD (Matthews, 2014/15a). In this role, I observed that the medical management of SCD

was given the highest priority. Over time, a growing body of scientific evidence confirmed my own observation, that nutrition was not part of standard care in SCD (Prasad, 1997; Hyacinth, Gee and Hibbert, 2010; Matthews, 2015/16e; Matthews, 2019j; Khan et al., 2016; Ohemeng and Boadu, 2018; Umeakunne and Hibbert, 2019; Kamal et al., 2021). This observation underscored the main problem of the research project.

The lack of nutrition management in SCD, relates directly to the fact that nutrition is not currently integrated or embedded into existing care provision. Accordingly, people living with SCD are left to self-diagnose, self-manage and treat their often complex nutritional problems, which would normally be done by expertly trained healthcare professionals such as Dietitians. Without adequate nutrition service provision available to people living with SCD, their nutritional needs will remain invisible, overlooked and unmanaged, negatively impacting the health outcomes of the SCD patient population. Despite the role of nutrition being researched for over 60 years as highlighted in the following review articles (Reed, Redding – Lallinger and Orringer, 1987; Prasad, 1997; Hyacinth, Gee and Hibbert, 2010), there remains a disparity between the development of the medical management and the nutritional management in SCD (Hyacinth, Gee and Hibbert, 2010).

Importantly, this latter review article makes a causal link between the clinical features of SCD and nutrition where the clinical features are a direct contributor to the nutritional needs and problems experienced by people living with SCD, to varying degrees. To improve the nutritional health outcomes of people living with SCD, there is need for adequate nutrition service provision to be available to them, as part of their standard care provision. For nutrition to be integrated into standard care in SCD, the invisibility of nutrition in standard care provision in SCD needs to be reviewed and addressed to improve patient outcomes. Thus in 2014, an article written by a Dietitian, (Couch, 2013), about acute trauma as an emerging speciality in Dietetics, proved to be pivotal. The article demonstrated how the role of nutrition was beneficial to improve clinical outcomes in acute trauma patients, in relation to their Length of Stay (LOS), recovery time, and improved clinical outcomes. Since high

readmission rates is a problem in SCD, it seemed plausible to consider the role of nutrition in SCD management, in this way linking nutrition research to clinical practice, a gap identified in SCD (Nartey et al., 2021).

Need for service provision:

Given the paucity of knowledge on the state of nutritional management in SCD, I conducted a cross sectional survey in 2015 to scope out the existing nutrition service provision for people living with SCD in the UK, including assessing the level of involvement and the knowledge of Dietitians about SCD. The findings of the survey confirmed knowledge and care gaps in nutrition service provision in SCD (Matthews, 2016g), yet 96 percent (%) of the Dietitians reported they have a role to play in the nutritional management of people living with SCD. The findings of the survey were strategic in prompting me to undertake this doctoral research project, an attempt to contribute to the evidence base in support of the role of nutrition in SCD and to improve nutrition service provision in SCD. The lack of nutrition service provision in SCD is currently becoming a health inequality affecting the experience, access and health outcomes in people living with SCD.

However, in order to provide adequate nutrition service provision in SCD, it is important to address the nutritional needs of people living with SCD at a population level and not only in certain cohorts of patients, as evident in the existing scientific literature, where research is mostly conducted in the Haemoglobin (Hb) SS patient cohort (Heyman et al., 1985; Hyacinth, Gee and Hibbert, 2010; Hyacinth, Adekeye and Yilgwan, 2013), the most common and severe genotype of SCD. Given the complexity of SCD, and the nutritional needs of people affected by SCD over the life course, a multi-level, whole systems approach to nutrition service provision is indicated. In this way, the research project will adopt a population level approach and include all the patient groups, such as paediatric, adolescent, adults, pregnant and the elderly. In the study, the data will reflect the knowledge, views and experiences of the recruited SCSU-Cs and SCSPs, with limitations accounted for in any

under-represented patient groups. This approach will provide valuable insights in support of the co-development of the main research output to inform policy and practice development.

Need for policy and practice:

Adequate policy and practice development is required to support nutrition service provision in SCD. Numerous factors have been identified that influence the medical and nutritional management of people living with SCD. These factors include: the effects of the clinical symptomology of SCD, the impact of the social and wider determinants of health (Matthews, 2015b; Berghs et al., 2020), and the under recognition of the role of nutrition in SCD (Matthews, 2016f). Cumulatively, these factors sustain the complexity of the management of SCD and the service provision required to improve patient experience, access and outcomes. From these factors, it is possible to infer, that the nutritional management of SCD is complex. Therefore, a more comprehensive approach is needed to understand the nutrition landscape in SCD and the nutritional needs of people living with SCD in order to tailor policy and practice guidance and service provision.

The existing research identifies under nutrition and the macro and micro nutrient deficiency, as the most common nutritional problems in Hb (SS) patients, which is subsequently, linked to poor health outcomes (Heyman, et al., 1985, Hyacinth, Gee and Hibbert, 2010) and health status of people living with SCD. However, this is a narrow view to understand the role of nutrition in SCD. Ultimately, a wider approach to understanding the myriad of influencing factors affecting the role of nutrition in this complex disease was needed. A pivotal point in the research project in 2021 (see Appendix 13) identified the value of adopting a health literacy perspective to understanding the current nutrition landscape in SCD. However, instead of using health literacy in the traditional sense - as pertaining to health education and literacy only - I wanted to use it in a novel way and based on three key characteristics of health literacy:

1. Health literacy is linked to health outcomes and could be supported by Freire's (1970) principles (Dearfield, Barnum and Pugh-Yi, 2017)
2. Health literacy is a determinant of health (Nutbeam, 2008; Rudd, 2015)
3. Its role as identified by Sorenson et al., (2012) in three health domains: healthcare management, public health and health promotion, all three relevant domains to explore the lack of nutrition in SCD.

In this way, the project seeks to use health literacy as an expansive and integrated concept to contribute to policy and practice development in nutrition in SCD, as evidenced in the thesis structure below.

1.5 Thesis structure

The thesis structure includes a total of six chapters, to address the identified gap in knowledge. Below is an overview of these chapters.

The current chapter, Chapter 1 serves to introduce the research project to support the justification of the research project and its related overarching aim and objectives.

Chapter 2 consists of two parts, the first reflecting the lack of research on the nutritional management of SCD when using the PRISMA framework, warranting instead, a critical evaluation of the literature to identify the influencing factors affecting nutrition and service provision in SCD. The critical evaluation focussed on the following key topics: the medical management in SCD, the role of nutrition in SCD, the nutritional management in SCD, influencing factors affecting nutrition in SCD and nutrition integration in SCD. Part 2 (Chapter 2.2), provides an overview of the main theoretical principles underpinning and guiding the research project aim and objectives and key professional practice perspectives reflecting the lack of policy and practice guidance to support nutrition service provision in SCD.

Chapter 3 critically considers the evidence base in support of the philosophical and methodological choices which underpin the research project design, ethics, recruitment of participants, methods and processes and the thematic analysis of the data, in order to achieve the overall aim and objectives of the research project.

Chapter 4 is divided into two parts. Firstly, part one provides background information of the research participants and reports the findings of the thematic analysis of the focus groups conducted in phase one of the research project, including reflections of the LAN meetings in phase two, three and four. Secondly, part two reports the findings of the evaluation questionnaire conducted in phase four. Collectively, the findings of part one and part two aim to answer research question 1 and objective 1 and research question 2 and objective 2 (see Chapter 1, section 1.2), respectively. An overview of the health literacy thematic concepts, derived from the four main themes of the focus group analysis of phase one, is provided, which is used to develop the Health Literacy -Nutrition Integration Framework (NIF) blueprint.

Chapter 5 critically evaluates and discusses the findings of the research project, as reported in Chapter 4, the main influencing factors affecting nutrition integration – reflected in the core knowledge and care gaps in relation to existing literature. The four main points to be discussed include: the marginalisation of people living with Sickle Cell Disease (SCD), the influencing factors affecting nutrition in SCD, socio-ecological factors impacting the integration of nutrition management in SCD and a discussion on the gaps in the current practice guidance influencing the study recommendations to develop future policy and practice guidance in supporting the integration of nutrition in SCD. A discussion of the value of the LA methodology in healthcare intervention development as well as the main limitations and strengths of the research project is also included.

Chapter 6 provides an overview of the key conceptual conclusions based on the core knowledge and care gaps reflecting the multiple influencing factors underpinning the medical and nutritional management of people living with SCD, as reported and discussed in Chapter

4 and 5, respectively. As well as providing a summary of the contribution to knowledge, this chapter highlights the key policy and practice recommendations towards achieving the integration of nutrition in SCD, including the next steps to extend the impact of the research project, aimed at integrating nutrition into standard care in SCD.

Summary:

The marginalisation of SCD, drives the invisibility and health inequalities surrounding the medical and nutritional management in SCD, impacting on the experience, access and outcomes of people living with SCD. As such, nutrition service provision needs to be tailored to the unique nutritional needs and challenges of people living with SCD. The nutritional management of SCD is in itself multi-factorial and complex.

Without adequate nutrition service provision, the nutrition needs of people living with SCD, will not be addressed and potentially add to the high levels of morbidity, mortality and disability which are associated with the clinical symptomology of SCD.

Therefore, a multi-level, collaborative and comprehensive lens to understand the full extent of the role of nutrition in SCD and the development of tailored policy and practice guidance to support the integration of nutrition in SCD is needed. To achieve this, health literacy is considered in an expansive and integrated way to contribute to policy and practice development to address the many influencing factors reflected in the knowledge and care gaps in nutrition in SCD.

The next chapter will critically review the existing literature on the role of nutrition in SCD, in the context of identifying and understanding the current nutrition landscape in SCD and the influencing factors affecting nutrition integration into standard care in SCD.

Chapter 2

Literature Review

Exploring the influencing factors affecting nutrition integration in SCD:

This literature review is divided into two parts. Part one aims to identify and critically analyse academic and grey literature that relate to the influencing factors affecting nutrition integration into standard care in Sickle Cell Disease (SCD). The review will also explore the role of health literacy in policy and practice development to support nutrition service provision in SCD. Exploring the literature in this way will help to identify what already exists on the topic, in support of future policy and practice development and serve as grounds for future research in nutrition management in SCD. The literature review has tracked the following objectives;

- The nutritional management of people living with SCD
- The influencing factors affecting nutrition and service provision in SCD
- How health literacy is able to support nutrition integration in SCD

2.1.1 Literature review strategy

(i) Search Design:

The aim of conducting the review is to identify and critically analyse studies that relate to the influencing factors affecting the lack of nutritional management of people living with SCD as part of their standard care provision. Existing studies on the role of nutrition in SCD explore and present the nutritional problems of patients living with SCD, linked to the clinical features of the condition. However, this review is focused on exploring the lack of nutritional management of patients living with SCD and the influencing factors affecting nutrition integration in standard care in SCD to inform future policy and practice guidance tailored to the needs of the people living with SCD.

(ii) Inclusion and exclusion criteria:

As this research project was the first known attempt at integrating nutrition into standard care in SCD at policy and practice level, the overarching aim was to understand the current nutrition landscape, by identifying the influencing factors affecting nutritional management and integration of nutrition into standard care in SCD. Since the first recorded research on the role of nutrition is from the 1960's, this time filter was applied to the systematic literature search strategy to identify as many sources as possible. Hence the literature search sought to include as many relevant peer reviewed literature sources as well as grey literature sources and information on the topic. These points thus informed the boundaries of the inclusion and exclusion criteria applied to the literature review as listed below.

The inclusion criteria included the following;

- All studies that had nutrition and SCD, regardless of methods, location, including all patient cohorts
- Including influencing factors in relation to nutrition management and service provision in SCD
- Peer reviewed journals in English and translated into English, full text and abstracts
- Latest and most up to date nutrition and health and social care related policies, guidelines and reports in relation to nutrition and SCD

The exclusion criteria included the following;

- Animal studies
- Non-English studies
- Studies not related to the research question
- Studies mentioning influencing factors but not in the context of nutrition management in SCD

(iii) Systematic literature review

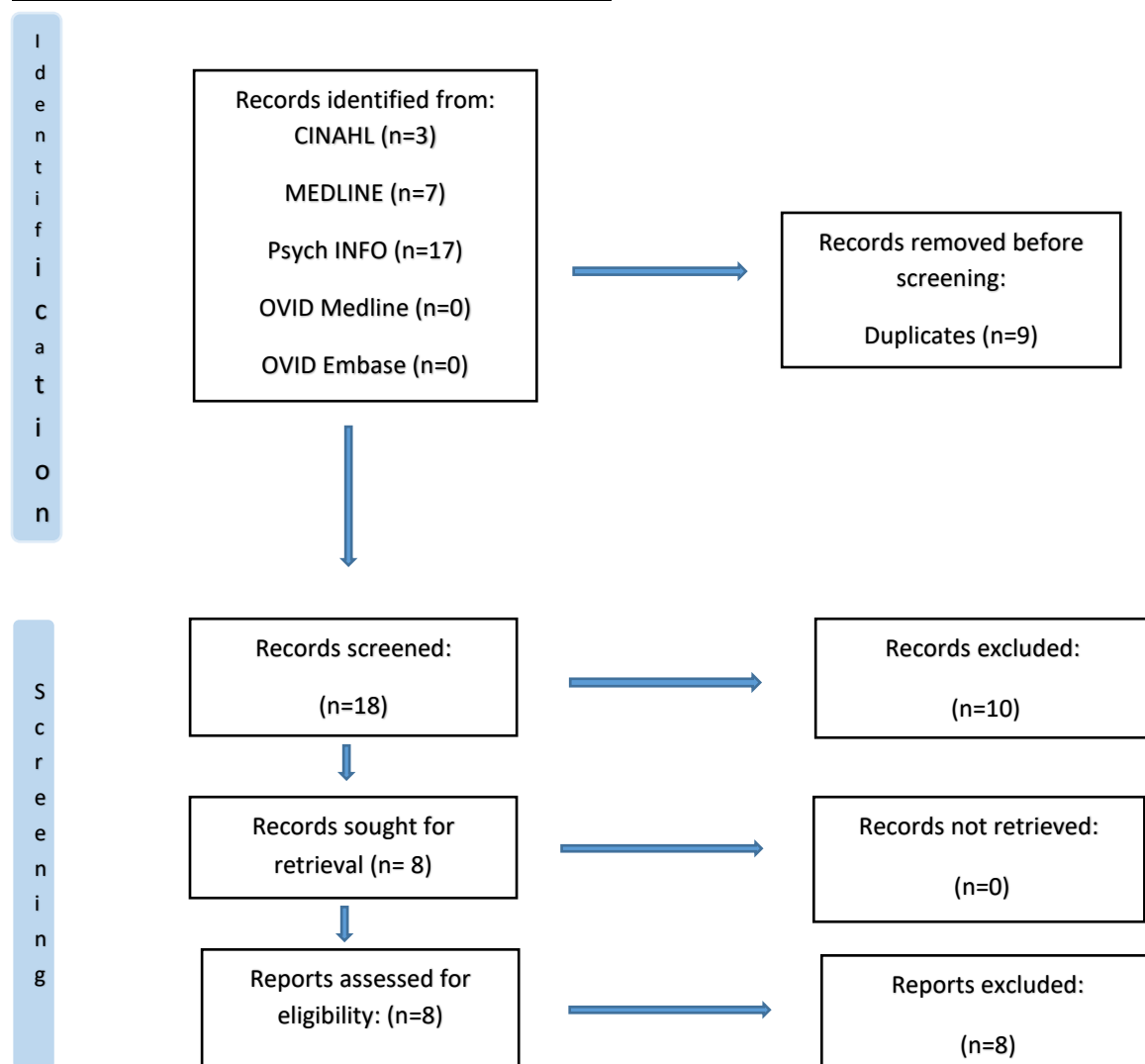
Initially, a systematic literature search strategy was conducted using OPEN Athens including the following health databases: CINAHL, MEDLINE, Psych INFO, OVID Embase, and OVID Medline, commonly used in healthcare research (Hicks, 2008), that consisted of journal articles, peer reviewed articles and abstracts. Based on the research question and aim of the review, the following keywords and phrases were identified to conduct the systematic literature review: 'sickle cell disease', 'sickle cell patients', 'nutritional management', 'integration', 'influencing factors' and 'healthcare'. Table 1 below, illustrates the main search terms, alternative terms and data bases used to conduct the literature search. Boolean operators 'OR' and 'AND' were applied as relevant to connect the main search terms and phrases and alternative terms eg ('sickle cell disease' OR 'disorder' OR 'anaemia') and 'AND' to combine all the individual search terms and phrases eg ('sickle cell disease' OR 'disorder' OR 'anaemia') AND eg ('nutri* management' OR 'care' OR 'service provision') to define the relationship between the search terms. Advanced searches included the use of truncation (* or?) to include all spellings and associated terms relevant to each keyword and phrase, whilst the Critical Appraisal Skills Program (CASP) framework (2018) was used to critically assess the quality of the relevant research papers identified in the review.

Table 2.1: Systematic literature review search strategy:

Main Search Terms	Alternative search terms	Data bases searched
'Sickle cell disease'	'anaemia' 'disorder'	CINAHL MEDLINE Psych INFO OVID Embase OVID Medline
'Sickle cell patients'	'service users' 'clients'	
'Nutritional management'	'care' 'service provision'	
'Influencing factors'	'barriers' 'challenges' 'risks' 'needs'	
'Integration'	'inclusion' 'incorporation' 'embedding'	
'Healthcare management'	'service provision' 'treatment'	

A research protocol was considered to ensure the quality of the literature review. The Preferred Reporting Items for Systematic and Meta-Analysis (PRISMA) guidance (2020) was used as it links both quantitative and qualitative checklists for research in health-related topics, see Figure 2.1 below. Although a total of (n=27) reports were identified, none of these reports were eligible for inclusion in the review, based on the inclusion and exclusion criteria. A total of (n=18) reports remained for screening after the removal of (n=9) duplicates, whilst (n=10) reports were excluded for screening as they were not conducted in a SCD patient cohort. After screening, the remaining (n=8) reports listed in (Appendix 20) were not eligible for inclusion in the review and therefore excluded as, although they contained some of the inclusion criteria such as influencing factors, it was not in the context of nutritional management in SCD and therefore did not meet the study aims and objectives.

Figure 2.1: PRISMA: Identification, Screening:



(vi) Critical evaluation of the literature:

Rationale:

Since the systematic literature review identified a paucity of studies, investigating the influencing factors affecting nutrition integration in SCD care provision, as illustrated in Figure 2.1 (PRISMA framework) above, an alternative literature search strategy was warranted. Alternatively, a critical evaluation of the literature was adopted, building on the articles that were identified from the systematic literature review (Appendix 20), after revising the inclusion and exclusion criteria in section 2.1.1 (ii) to include for example studies that considered influencing factors in SCD, even if it didn't include nutrition. For this reason, a wider search strategy drawing on research in other disciplines was necessary. In this way, the critical evaluation not only considered peer reviewed papers in SCD and nutrition, but in a wider context of health, education, psychology and social care, including grey literature sources and information to scope out the influencing factors affecting nutrition management and nutrition integration in SCD. Hence, the critical evaluation included research, policies, practice guidance and reports that inform the existing nutrition landscape in SCD and how the nutritional needs of people living with SCD are being managed. It can therefore be inferred that the lack of literature eligible for the inclusion in the systematic review points to a gap in studies exploring the nutritional management of people living with SCD, which the research project will aim to highlight through the critical evaluation of the existing literature.

Search strategy for the critical evaluation:

The search strategy for the critical evaluation was complex as there were many search variables to consider to undertake the review of the literature to identify the influencing factors affecting nutrition management and integration in SCD care. Since this study has not previously been undertaken, it was important to cover as much literature, both peer reviewed and grey literature (web sources, reports and guidance, white papers, professional articles

and policies and guidance) not only related to nutrition in SCD but the wider health, nutrition, education and social care context, as it relates to SCD.

Consequently, a series of targeted searches using a wide range of search engines including Google scholar, Google, PubMed, Anglia Ruskin University (ARU) library search, books, contemporary and professional sources were included to conduct the critical evaluation literature search. A total of five searches were undertaken for this part of the review, see Table 2.2 below. Table 2.2 illustrates the search terms, aims and phrases, timeframes and search strategy used to conduct a range of targeted searches to meet the aim of each targeted search in line with the overall aim and objectives of the literature search and research project.

Table 2.2: Search terms, aims and strategy for the critical evaluation of the literature review

Search terms and phrases/Timeframes	Aim of the search	Search Strategy
1.'Medical management in SCD' (health and social care factors)	Causal links between the medical and nutritional implications in SCD	○ Targeted Google scholar searches ○ Books ○ Contemporary and professional guidance and policies ○ National health and social care policies and guidance ○ Health and social care reports
2.'Nutritional management in SCD' – 1960's to present day	Role of nutrition in SCD – Nutritional consequences	
3.'Influencing factors affecting nutrition and service provision' – 1960's to present day	Socio-ecological and multifactorial factors in nutrition in SCD	
4.'Nutrition integration in SCD – 1960's to present day'	Understand global and UK context of nutrition in SCD	
5.'Health literacy in policy and practice – 1980's to present day'	Health literacy as an expansive and integrative concept in nutrition in SCD	

To ensure that the most up to date sources (particularly policies and guidance as it relates to health and social care) were included in this critical evaluation, the searches were periodically updated.

Appraisal of the literature sources identified in the critical evaluation:

To ascertain the trustworthiness and level of bias of the studies included in the critical evaluation, (both peer reviewed and grey literature), the CASP framework (2018) was used to critically appraise the sources. Each of the peer reviewed studies and papers used in the literature review were checked for validity, results and its relevancy to the study aims (Burls, 2009). Similarly, the grey literature was checked in terms of currency, relevancy, authority and the intended purpose of the source, as it relates to the research project aims. For example, a range of government sources, the Kings fund and British Dietetic Association (BDA) policy reports and guidance have been included in the literature review and checked in relation to the references used and those included in the documents. The journal and peer reviewed articles and papers were checked to establish clear aims, appropriate methodology and the transferability of the results to meet the study aims (Mhaskar et al, 2009), so testing the trustworthiness of the sources to enhance the rigor of the critical evaluation.

Focus of the critical evaluation:

In this way, the critical evaluation was important to ensure that the most relevant, valid and trustworthy literature sources were identified and included to meet the aim of the literature search, to explore the influencing factors affecting nutrition management and integration in standard care in SCD. Thus, the critical evaluation was focussed on the following key topics that will be explored below:

- The medical management in SCD (health and social factors)
- The role of nutrition in SCD (causal links between the clinical features and nutrition)
- The nutritional management in SCD (global, UK and clinical practice context)
- Influencing factors affecting nutrition in SCD (socio-ecological and multifactorial factors)
- Nutrition integration in SCD (health literacy in policy and practice development).

2.1.2 Medical management in SCD

(i) Health factors in SCD:

The medical management in SCD is complex and needs to be considered in terms of its complex pathophysiology. The clinical presentation is influenced by the changes that happen to the Sick Haemoglobin (HbS) molecule. The often-devastating pathophysiological consequences of the condition is in response to the cellular, molecular and structural changes which the HbS molecule undergoes, under hypoxic conditions (Chakrovorty and Williams, 2015). It is these structural changes that causes the haemoglobin molecule to become rod-like and stiff, as the proteins clump together (Asnani, 2010; Chakravorty and Williams, 2015). Despite SCD being the most common genetically inherited condition, warranting ongoing medical, nutritional and pharmaceutical interventions, it remains to be considered as a chronic long term condition. Long term conditions are considered by (Nolte and McKee, 2008), as conditions requiring ongoing interventions.

A host of clinical features are commonly seen in SCD (Kato et al., 2006; Ataga et al., 2008; Rees, Williams and Gladwin, 2010). These clinical features have an affinity to increase levels of morbidity and mortality affecting the disability and QOL of people living with SCD; these clinical features and related symptomology occurring over the life course of this population. The main clinical features resulting from the pathophysiology of SCD include the following: haemolysis (the rapid breakdown of red blood cells resulting in high red cell turnover, chronic anaemia and fatigue, increased oxidative stress causing chronic inflammation; vaso-occlusive crisis (blockage of small and large blood vessels) and impaired immunity, due to a poorly functioning spleen, thereby increasing the infection risk of people living with SCD (Chakrovorty and Williams, 2015). Not only do these clinical features result in the complications associated with this condition, these same clinical features are considered as causal links between SCD and the nutritional implications in SCD (Hyacinth,

Gee and Hibbert, 2010). However, this causal link is overlooked in terms of the role of nutrition as part of standard care in SCD.

Thus the complications associated with SCD are attributed to the clinical features mentioned above. The hallmark feature of SCD is vaso-occlusive crisis (commonly known as sickle cell crisis) (Ballas, 2011), and this is also one of the complications that dominates the treatment options in SCD medical management and research. After vaso-occlusion, Acute Chest Syndrome (ACS), which is a kind of lung injury, is the second most common reason for hospital admission in people living with SCD (Chakravorty and Williams, 2015; Serjeant, 2013). In addition, pulmonary complications are noted to be a major contributing factor to acute morbidity and mortality in SCD (Platt et al., 1994, Serjeant, 2013). Pulmonary hypertension, a chronic complication of SCD, contributes a significant risk to the wellbeing and life expectancy of people living with SCD (Machado and Gladwin, 2005). Other factors that impact the wellbeing of people living with SCD over their lifespan are avascular necrosis, leg ulcers and stroke (Serjeant, 2013). Priapism (Stuart and Nagel, 2004), sickle nephropathy (Ataga and Orringer, 2000) and cholelithiasis (Meshikes and al-Fraj, 1998) are further complications of SCD contributing to the morbidity associated with this disease (Fitzburgh et al., 2005; Lucas and Mason, 2008; Aljuburi et al., 2012; Piel et al, 2021)) which includes cerebrovascular events.

In an attempt to reduce the overall morbidity and mortality associated with the clinical symptomology in SCD, a range of disease-modifying treatment options are used. Most commonly, blood transfusion has been used as a standard management option in SCD for the management of both acute and chronic complications of the disease (Rees, Williams and Gladwin, 2010). Other treatment options include Hydroxyurea, found to significantly reduce the incidence of vaso-occlusive crisis, hospitalisations and mortality in high income countries, and has a high safety profile (Charache et al., 1995). The only curative treatment in SCD to date is Haematopoietic Cell Transplantation (HCT), (Bhatia and Walters, 2008; Kato et al., 2018), and in recent years, studies investigating the benefits of gene therapy are

also gaining momentum (Chakravorty and Williams, 2015) as a management option in SCD. However, most recently a new drug Crizanlizumab (NICE, 2021c) - (used in the prevention of adhesion of platelets to red blood cells), is the first new pharmaceutical therapy in the last 20 years to be approved for use in the medical management of SCD. In addition, Gardner (2018) highlights other treatment options including L-Glutamine (aimed at preventing oxidative stress), and Voxelotor (focussed on preventing polymerisation of the sickle red blood cells) as additional treatment options in the management of SCD, to address the healthcare burden in SCD.

In order for a more complete evaluation of the healthcare burden associated with SCD, the nutritional needs of people living with SCD needs to be brought to the forefront of the standard management of SCD. Hyacinth, Adekeye, and Yilgwan (2013), identify nutrition as a cost effective management option in SCD, particularly in developing countries. However, since the nutritional implications in SCD is correlated to the clinical features as it is experienced by people living with SCD, there is grounds to believe, that the current way nutrition is considered in standard SCD management needs to be challenged. Moreover, in SCD, high readmission rates are a huge cost factor. The significance of high readmission rates in SCD, is closely tied to the high health care utilisation and health burden costs associated with the complexity of the medical management of this chronic condition (Aljuburi, 2015; Aljuburi et al., 2012) in the UK and globally. The estimated cost to the NHS, now nearly nine years ago to treat sickle cell crisis, the primary cause for hospitalisation (Aljuburi et al., 2012, Aljuburi et al., 2015; Pizza et al., 2014; Nice, 2014) was approximately £19 million in (2010/11). A more recent study conducted by Jobanputra et al., (2021), as a 10-year cohort analysis study, revealed an increase in the overall treatment cost to the NHS of £76.4 million, up from £41.9 million, at the start of the study, however the social factors may also be a contributory factor to the health and wellbeing of people living with SCD.

(ii) Social Factors in SCD:

A number of social structural factors are relevant in understanding the health and healthcare experiences of people living with SCD. Notwithstanding, sociology is about society, and for this reason, texts like Craig et al., (2012) and Chatoo et al., (2019), consider multiple minority communities such as refugees, asylum seekers and economic migrants, as well as the historically ignored, Gypsies, Roma and Travellers, when they discuss issues of race and ethnicity and the need for its acknowledgement and inclusion in welfare policy and practice. Similarly, this research project seeks to influence policy and practice change, by drawing attention to how these social factors influence SCD care provision, and how in turn, these same social factors impact on the nutritional management of people living with SCD, a key focus of the research project not currently explored in existing literature in nutrition in SCD.

Race and racism:

It is a belief over the years that race and ethnicity are key social structural factors that influence patterns of health and illness (Taylor and Blakemore, 2007, Karlsen, 2007; Walch and Tait, 2014; Craig et al., 2012; Chatto et al., 2019). Race is defined by Field and Blakemore (2007, pg69) as related to the physical differences in people, more specifically that race is about the social distinctions (e.g. Apartheid) that people make and have about others, including the beliefs and myths they hold about different races such as their intelligence and skills. Race is argued to be a socially constructed distinction and in sociological terms argued to be a concept used to understand its use in the domination and oppression of non-white people by white Western colonisers (Walsch and Tait, 2014).

In the twenty years from the publication of the book the '*Politics of Sickle Cell*' (Anionwu and Atkin, 2001), racism continues to be a factor affecting the SCD patient population in the UK and around the world. More specifically, institutional racism underpinning the level of

healthcare provision in SCD as a minority ethnic group continues to be a problem (Anionwu and Atkin, 2001; Redhead, 2020; APPG –SCAT, 2021). Institutional racism relates to the limiting experiences and difficulties faced by ethnic minority groups, when seeking health and social care service provision (Anionwu and Atkin, 2001) described as; ‘The collective failure of an organisation to provide an appropriate and professional service to people because of the colour, culture, or ethnic origin’, (Macpherson report, 1999, p28). Institutional racism, therefore, has a significant impact on the level and quality of service provision for minority ethnic groups.

According to Redhead (2021), different types of racism are prevalent in the management of people living with SCD in post-colonial Britain, whilst Power – Hays and McGann (2020) highlight a range of racial discriminations in relation to SCD care provision in the US. In the article by Redhead (2021, p197), the assumption of nurses and doctors about people living with SCD being ‘addicted to drugs’, provides evidence of the ‘wider state and societal stereotyping of black communities’, of which people living with SCD form a part of. These views and perceptions, provide evidence about people’s attitudes and behaviours which amount to discrimination through unwitting prejudice, ignorance, and thoughtlessness (Macpherson, 1999 p 28); a major disadvantage to minority ethnic groups.

Moreover, racism and its effects has known psychological effects such as anxiety, mental and physical effects impacting the health experiences of ethnic minority groups, resulting in ethnic inequalities in health (Nazroo, 2003). Thus, the health of ethnic minority groups is believed to be undermined by racial discrimination, and needs to be challenged. Institutional level discrimination is believed to be found in all the major institutions in contemporary British society including the NHS (Field and Blakemore, 2007).

Ethnicity:

Ethnicity refers to the ways in which social groups can be distinguished from one another.

The term is defined by Dyson (2005, p 20) as “a socially constructed difference used to refer

to people who see themselves as having a common ancestry, often linked to a geographical territory, and perhaps sharing a language, religion and other social customs". Furthermore, ethnicity is believed to represent a set of structural influences that affects the standard of living and material circumstances affecting the lives of individuals and groups (Nazroo, 2001). In the UK, the non-white ethnic minority groups have increased to a total of 18% of the total UK population, according to the 2021, Census data, a significant increase from 8.7 % in 2001, and 14% in 2011 (ONS, 2011). Despite minority ethnic groups having equal entitlement to effective and appropriate health and social care, sociologists are concerned that minority ethnic groups still experience social inequality, less than favourable treatment thereby decreasing their life chances compared to the white population (Walsch and Tait, 2014).

SCD is the most common genetic condition in the UK (SCS, 2008; 2018), however, SCD has been repeatedly subjected to low levels of awareness, under resourcing and inequalities (Anionwu and Atkin, 2001, Redhead, 2021, APPG -SCAT, 2021). More recently, the inclusion of SCD as a high-risk patient population group during the COVID 19 pandemic, resulted in SCD being brought under the radar of healthcare providers in the UK, highlighting several inequalities in care (APPG -SCAT, 2020; Berghs, Cronin-Chavez, and Ebenso, 2020). However, this unexpected attention has succeeded to some degree, to address the longstanding invisibility of the condition, raising a critical consciousness and awareness of this long term condition, on the national health agenda. Increasing the awareness of the infection risk of people living with SCD, has in turn, helped to raise awareness of the health patterns and inequalities in SCD care provision.

Health inequalities:

The ongoing commitment of the government to reduce health inequalities is evident in a number of policies and white papers including for example, '*Our Health, Our Care, Our Say*' (Department of Health -DOH, 2006) and more recently amongst others, '*Levelling up the*

United Kingdom' (DOH, 2022) and reform suggested by The Kings Fund (2022a) – exploring new ways to tackle health inequalities, involving endurance, partnership working and disruption of the normal patterns of working. The Health and Social Care Act, according to the Department of Health and Social Care (DHSC, 2012-) and the New NHS Plan (2019), continue to place the reduction of health inequalities high on the agenda, and has been embedded in the both the NHS and Public Health Outcomes Frameworks (2021/22) and the new GP Outcomes Framework (NHSE, 2019), the Core20PLUS5 (NHSE, 2022), being an example of an initiative aimed at facilitating action at local and national levels towards reducing healthcare inequalities in marginalised adults in the UK.

In SCD, evidence of the marginalisation of SCD, is emphasised in the APPG – SCAT (2021) report, '*No one is listening*', a topic previously identified to impact on the medical management and outcomes of people living with SCD (Power-Hays and McGann, 2020; Redhead, 2021; Dyson, 2019; Anionwu and Atkin, 2001). Thus, building on the work of the government through the Office for Health Improvement and disparities, the NHS Race and Health Observatory (2022a) was established with the aim to drive policy change, shape research, influence leadership, engage public and create practical resources and tools to reduce the health inequalities experienced by patient groups from minority ethnic groups. In this way, the work of the NHS Race and Health Observatory is focussed on addressing the longstanding health inequalities many marginalised population groups are subjected to.

Furthermore, the NHS Race and Health observatory, commissioned The Kings Fund to independently review the scope of the health inequalities in NHS service provision to people from minority ethnic groups (Robertson et al., 2021). Health inequalities are described by the Kings Fund (2022b) as the avoidable, unfair and systematic difference's in health between groups of people, implying some population groups can have significantly worse health outcomes than others (The Kings Fund, 2021a). More than one type of health inequality is identified (Kings Fund, 2022b), and specific to nutrition in SCD, pertains to the lack of access and available nutrition service provision that impact on the patient's experience and

ultimately health outcomes, because of poor nutrition service provision and resources available to people living with SCD. Hence, ongoing efforts are required to adequately address the health inequalities and disparities in care provision, as actioned by the WHO commission on social determinants of health, through the report '*Closing the gap in a generation*' (Friel et al., 2008), and SCD is no exception, as often times the health inequalities are exacerbated by the social inequalities in the condition.

Social Inequalities:

Poor socio-economic status has been linked to health consequences by the Marmot Review (2010) evident in occupation banding of minority groups, lower incomes, stressful working conditions and poorer job security. This supports the view held by sociologists that the patterns of health and ill-health in members of minority ethnic groups are influenced by social factors. For this reason, a wider perspective is needed to consider the breadth of factors at play, which includes social divisions such as social class. Social class, is a term used to identify groups of people in relation to their occupation, income and wealth, considered to play a role in the life chances and social mobility of the people in these groups (Walsch and Tait, 2014). In this way, and since people living with SCD are predominantly known to be from an ethnic minority background, social class is an important factor to consider in this patient population.

Many people living with SCD are reported to be affected by low education attainment (APPG – SCAT, 2009), they may have a high incidence of sickness absence from school (Schwartz, Radcliffe and Barakat, 2009; Salih, 2019) and consequentially, employment opportunities and experiences (Bergs, et al. 2021; Berghs and Dyson, 2022; Pirez et al., 2022), has been found to be an important factor affecting their socio-economic status, and increasing their risk of social and material inequalities. People living with SCD live in poorer social conditions (Purdy, 2010, Aljuburi et al., 2013), a contributory factor to the wider determinants of health (Dahlgren and Whitehead, 1991). Furthermore, the health outcomes of people living with

SCD, may be impacted by the social determinants of health (Berghs et al., 2020; Sonenklar et al., 2022; Khan et al., 2022; WHO, 2004) potentially driving the health and social inequalities in the condition. Confounding the impact of school and work absences in SCD, and the overall clinical symptomology and features in SCD, the condition is associated with a shorter life expectancy (about 20 years), than those without SCD (Lubeck et al., 2019).

Past studies have shown that people living with SCD with Hemoglobin (Hb) SS, have a reduced life expectancy; 42years for males and 48 years for females (Bonds, 2005; Platt et al., 1994). However, people living with SCD are living longer with improvements in medical advancements (Aljuburi et al., 2012) and long term follow up (Gardner et al., 2016).

Following the review of haemoglobinopathy services in the UK, NHS England (2020a) have commissioned Haemoglobinopathy Co-ordinating Centres (HCC) to lead on co-ordinating, supporting and promoting a system -wide network approach to delivering haemoglobinopathy services in the UK, a positive step in improving healthcare provision in SCD. In this way, the HCCs serves to reduce the impact of potential institutional racism, health inequalities and resulting poor health outcomes in the healthcare management of people living with SCD. However, the link between nutrition and SCD remains under recognised and thus needs greater attention in SCD management.

2.1.3 The role of nutrition in SCD

The role of nutrition in SCD is an important consideration in the wider context of standard care in SCD management. In this way, failing to acknowledge the direct causal links which exist between the clinical features of SCD and nutrition is a significant oversight in relation to the complexity of the symptomology and complications as experienced by people living with SCD. For this reason, it is necessary to consider how the clinical features in SCD and the nutritional implications in SCD are linked, and the nutritional consequences linked to this causal association.

(i) Clinical features affecting nutrition in SCD:

In a global context, nutritional anaemia caused by Iron (Fe), Folate and Vitamin B12 deficiency, is one of the top ten contributors to the global burden of disease (WHO, 2008). However, anaemia associated with haemolytic anaemia in SCD, is not considered in the same way. Chronic haemolysis is identified as one of the main clinical features in SCD and the resultant pathophysiology holds significant implications for both the medical and nutritional management of the condition. Chronic haemolysis, resulting from the polymerisation of HbS (Kato et al., 2018), refers to the rapid breakdown of red blood cells with a significantly reduced half-life of the red blood cells ranging between 10 and 30 days (Bensinger and Gillette, 1974), a reduction of 75% (Alayash, 2017; Quinn et al., 2016) when compared to the standard half-life of 120 days for normal red blood cells. Oxidative stress, recognised as both a cause and effect of haemolysis (Kato et al., 2018), is a significant consequence of chronic haemolysis and believed to worsen the symptoms associated with chronic haemolysis (Fibach et al., 2008).

Oxidative status is believed to be assessed by the balance between pro-oxidants and anti-oxidants, where pro-oxidants referred to as Reactive Oxidant Species (ROS) are classified as radicals and non-radicals. When cells experience oxidative stress, ROS are generated in excess oxidising proteins, lipids and DNA, resulting in cell death and organ damage (Fibach et al., 2008) caused by tissue ischaemia and infarction (Asnani, 2010; Chakravorty and Williams, 2015). Over time, the organ damage can contribute to the increased morbidity and mortality associated with the condition ((Fitzburgh et al., 2005; Lucas and Mason, 2008; Aljuburi et al., 2012; Piel et al, 2021). Oxidative stress is a significant clinical problem in people living with SCD and particularly in pregnancy, it is believed to be an underlying cause for several complications including preeclampsia, preterm birth, intrauterine growth restriction and premature membrane rupturing (Roberts, 1990; Kingdom et al., 2000).

Accordingly, consideration for the role antioxidants such as Vitamin C and E may play in protecting against oxidative stress in pregnant women (Mousa et al., 2019) is advised.

Chronic haemolysis leads to increased energy demands on the body, increasing the cardiac demand and utilisation of energy (Asnani, 2010 Salaman et al., 1996; Hibbert, et al., 2006; Hyacinth, Gee and Hibbert et al., 2010). Moreover, chronic haemolysis is responsible for the chronic anaemia and fatigue commonly experienced by people living with SCD. Increased total energy requirement by the body as catabolism exceeds anabolism and nutrient substrates are used for energy and therefore not available for growth and repair leading to the risk of developing malnutrition. Moreover, the increased myocardial energy demand results in a long-term subclinical state of inflammation in people living with SCD (Akohouse et al., 2007; Hibbert et al., 2005; Hibbert et al., 2006). On the other hand, malnutrition may also be caused by reduced caloric intake (Fung et al., 2001, Mandese et al., 2016; Prasad, 1997; Osei et al., 2016) adding to the nutritional problems associated with SCD. Children and adolescence in particular, are impacted by the effects of chronic haemolytic anaemia, vascular injury and organ dysfunction (Heyman et al., 1985; Reed, Redding –Lallinger and Oringer, 1987; Prasad, 1997; Mandese et al., 2016). In this way, the resulting reduced energy levels experienced by people living with SCD may in turn affect their QOL, productivity and overall experiences of poor health and wellbeing outcomes (Hyacinth, Gee and Hibbert et al., 2010). However more can be done to raise awareness of the implications of haemolytic anaemias, as experienced in SCD, and its role in the pathophysiology of this chronic condition.

Vaso-occlusion is recognised as the hallmark feature of SCD (Ballas, 2011) and the landmark complication of SCD (Kato et al., 2018). Vaso – occlusion is the blockage of small and large blood vessels with sickle shaped red blood cells due to ongoing deoxygenation of the haemoglobin molecules and polymerisation of the HbS molecule (Kato et al., 2018). The blockage of the red blood cells is what contributes to the tissue ischaemia and infarction that can lead to tissue and organ damage (Kato et al., 2018), which is exacerbated by the

ischemia associated with chronic haemolysis. The organ and tissue damage can lead to a number of the multi-organ complications associated with SCD which adds to the high risk of morbidity and mortality in the condition ((Fitzburgh et al., 2005; Lucas and Mason, 2008; Aljuburi et al., 2012; Piel et al, 2021). The nature of the vaso-occlusion episodes, commonly known as sickle cell crisis, can happen and reoccur unpredictably over a lifetime in people living with SCD (Smith et al., 2005). Sickle cell crisis in children, known as dactylitis, can start as early as 6 months of age. In children, dactylitis can lead to shortened and deformed bones (Serjeant et al., 1994; Serjeant. 2013).

Importantly, repeated painful crisis episodes are recognised as the main reason for emergency hospital admission in SCD (Ballas and Lusardi, 2005; Aljuburi et al., 2012). Furthermore, the significance of the hallmark sickle cell crisis is linked to it being considered a measure of severity of the disease and as an early mortality predictor (Steinberg, 2005) in people living with SCD. Equally important, is the fact that SCD, is a blood disorder and therefore it affects all the systems of the body. The resulting tissue and organ damage from repeated episodes of vaso-occlusion, can affect all the main organs and systems of the body including, the spleen, skeletal, genito-urinary, gastrointestinal, hepato-biliary, cardiopulmonary and central nervous system (Meshikes and al-Fraj, 1998). The multi-organ damage in SCD, underscores the multiple health risks and complications facing people living with SCD, thereby increasing the level of risk in relation to the morbidity and mortality of vulnerable people living with SCD.

The immune system of people living with SCD is identified as having high susceptibility for abnormalities. In SCD, immuno-deficiency is due to early loss and poor functioning of the spleen (Asnani, 2010). The immune-deficiency is impacted by the level of white cells in people living with SCD and importantly to note, is used as an indicator of the severity of the condition. The risk of infection in people living with SCD, may be used as an indication of disease severity (Okocha et al., 2016), and is particularly influenced by serum levels of micronutrients, especially those involved in the antioxidant system of the body including zinc,

copper, selenium and magnesium. One study (Okocha et al., 2016), aimed to establish the link between serum micronutrient levels and disease severity in people living with SCD, as previous studies used blood counts and markers of inflammation (Anyaegebu et al., 1998; Okocha et al., 2014) rather than micronutrients to indicate disease severity. Out of the four nutrients, serum copper was found to be significantly correlated with disease severity score, $p=0.001$ with no significant correlation observed between disease severity scores and the other micronutrients.

The level of white cells plays a role in the vascular -occlusion episodes experienced by people living with SCD. The vascular -occlusion episodes are because of the adherence of the white cells to the endothelium of the red blood cells (Okpala, 2004). A serious life-threatening complication of the immune system in SCD is acute splenic sequestration in particularly patients with HbSS disease, however it is rare in adults (Solanki et al., 1986). For this reason, it is important to consider the risks to infants and young children under the age of eight who are more susceptible, with children under the age of five at a 30% greater risk of splenic sequestration (Edmond et al., 1985). Children under three years of age have an increased susceptibility to encapsulated organisms such as *Strep Pneumoniae* and *Salmonella SPP* that can lead to pneumococcal sepsis (Asnani, 2010). As a consequence of the immune – deficiency and susceptibility to encapsulated organisms, people living with SCD are at an increased risk of bacterial infections, and are known to have subnormal immune status and function (Ashaba et al., 2015) with infection seen as a complication of the condition (Hyacinth, Adekeye, Yilgwan, 2013). For this reason, they have a higher risk of contracting infections especially in the presence of higher oxidative stress at tissue level and therefore there is a need for the antioxidant system to be kept intact and well maintained (Okocha et a., 2016).

Infections are linked to malnutrition, as patients who are malnourished have an increased risk of infection (Hyacinth, Gee and Hibbert, 2010; Hyacinth, Adekeye, Yilgwan, 2013, Cox et al., 2013). The malnutrition risk of people living with SCD, place an increased demand on the

body to provide adequate amounts of nutrients to maintain health and wellbeing over the life course of these patients. Particular to people living with SCD, many have been found to have low levels of Zinc (Zn), (Prasad, 1997). Zn, is known to strengthen the immune system and reduce the risk of infections; in this way more consideration needs to be given to the nutritional implications in SCD, and the main problems observed in this patient population.

(ii) Main nutritional problems in SCD:

Under nutrition:

Malnutrition, relates to both under and over nutrition. In SCD, under nutrition is the main reported problem, although, over nutrition (overweight and obesity) is on the increase, in predominantly, HbSC patients (Hall et al, 2018; Ibemere, 2023). Malnutrition is defined as “*a deficiency of energy, protein and other nutrients that causes adverse effects on the body (shape, size and composition), the way it functions and clinical outcomes*”, (BAPEN, 2003), as well as “*including social and psychological and clinical outcomes*” (NICE, 2012).

Disease Related Malnutrition (DRM), is identified as a term not routinely used to describe the under nutrition in SCD (Matthews, 2019j). This term however, reflects one of the main causes of malnutrition (disease related) as identified by BAPEN (2009), connecting the cause of the malnutrition to the disease or condition itself. Relevant to SCD, social-related (e.g., loneliness, poverty, isolation), psychological factors (e.g., anxiety, depression) and physical factors (e.g., impaired taste, appetite, pain), may also be relevant, (Kruizenga et al., 2017, p8), and reflect the range of factors that may affect the broader malnutrition risk in people living with SCD, not explicitly explored in existing literature in SCD, adding to the limited understanding of the factors affecting nutrition in SCD.

Understanding the consequences of both DRM and malnutrition in the context of under nutrition in SCD is relevant. The consequences of both DRM and malnutrition is similar for

example, both conditions result in a range of adverse clinical outcomes such as impacting the morbidity and mortality of those affected and the fact that it has a detrimental effect on the physical and psychosocial health of patients (Coreira and Waitzberg, 2003; Amaral et al., 2007; Freijer et al., 2013). In addition, frailty, unintentional weight loss, weakness, immobility, sarcopenia and poor endurance are further consequences of malnutrition (Leach et al., 2013) emphasising the costly, clinical and financial impact of DRM, if left untreated, that may be adding to the cost and health burden in SCD patients affected by DRM or malnutrition.

Significantly, malnutrition affects the function and recovery of every organ system (Saunders, Smith and Stoud, 2010)- this is critical in SCD which is a blood disorder, with verified multi-organ/system involvement (Meshikhes and Al-Faraj, 1998; Alexandre and Yahya, 2015). The effects of DRM and malnutrition increases the use of healthcare services and the costs associated with it (Stratton, Green and Elia, 2003, Amaral et al., 2007; Norman et al., 2007; Feldblum et al., 2009), necessitating consideration given to the problems linked to under nutrition in SCD. In the context of the high hospital readmission rates observed in SCD, increased hospitalisation is known to increase a patient's risk for malnutrition (Elia, Zellipor and Stratton, 2005).

Importantly, under nutrition, is identified as a 'significant complication of SCD' (Hyacinth, Gee and Hibbert, 2010, p 58). Growth retardation, delayed sexual maturity and a weak immune system, are some of the many complications which could be considered partly due to nutritional deficiencies in SCD (Reed, Redding-Lallinger, and Orringer, 1987; Prasad, 1997; Hyacinth et al., 2010; Hyacinth, et al., 2013; Serjeant, 2001), as a result of the under nutrition in the condition, exacerbated by the factors mentioned above. In addition, pica (often correlated to lower haemoglobin levels) is a relatively unknown problem in SCD, however it is known to have an unusually high prevalence in people living with SCD (Ivascu et al., 2001). Similarly, not often recognised, people living with SCD may also be at high risk of osteopenia and osteoporosis (Esiokack, Yilmaz and Ilhan, 2022).

Moreover, under nutrition in SCD is linked to a range of micronutrient deficiencies including the following; Vitamin D (Almeida and Roberts, 2005; Buison et al., 2004), B2, B6, B12, A, C and E (Natta et al., 1992), folic acid, iron (Mohanty et al., 2008), calcium, magnesium (Oladipo et al., 2005; Zehtabchi et al., 2004), and zinc (Reed, Redding-Lallinger and Orringer, 1987; Zemel et al., 2002; Prasad, 2009; Vanderjacht et al., 2008), omega 3 fatty acids (Tomer et al., 2010; Daak et al., 2013) in many people living with SCD, as well as nutrients like Zn, glutamine (Williams et al., 2004), L-arginine and antioxidants (Vanderjacht et al., 2008).

Growth failure is a common problem in children with SCD and a serious consequence of the malnutrition associated with the condition (Heyman, et al., 1985; Prasad, 1997, Reed, Redding-Lillenger and Oringer, 1987). Wasting is recognised as a type of malnutrition. Wasting relates to poor weight in relation to height which is an indication of moderate to severe malnutrition. In US studies involving low weight children living with SCD (Warrior et al., 1994; Henderson et al, 1994), 41% and 25% of low weight children living with SCD were identified with moderate and severe under nutrition respectively and 11% with wasting. Similarly, stunting is recognised as a type of malnutrition. It has been found that children and adolescents living with SCD in comparison to their healthy peers, are leaner and shorter (Henderson et al, 1994; Silva and Viana, 2005). The presence of delayed skeletal maturation in both sexes' and menarche in girls with SCD (Adewoyin, 2015; Zemel et al., 2007.) were also identified.

Over nutrition:

Observations of increasing numbers of people living with SCD who are overweight or obese is rising, in the UK. A recent study conducted by Hall et al., (2018), assessing the high body mass index in children as a retrospective single-centre audit, revealed that 64 (17%) of the 385 patient records included in the audit were overweight or obese, of which a significantly higher proportion of the children with HbSC being obese or overweight compared with those with HbSS. Interestingly, high BMI did not correlate with disease severity, the highest

incidence of obesity was females with HbSC. Similarly, in a study by Ibemere (2023) characterising the prevalence of overweight and obese status among adults with SCD, there were higher odds of overweight (BMI >25kg/m²) in individuals with HbSC or HbSB + thalassemia regardless of treatment modifiers such as Hydroxycarbimide, when compared to those with HbS or HbSB0 – thalassemia, without any treatment modifiers such as Hydroxycarbimide. The author concludes that early identification, prevention and intervention for increasing BMI is necessary to reduce the incidence of obesity-related complications, and the need to consider how best these nutritional problems can be prevented and managed.

2.1.4 Nutritional management in SCD

Over the last 60 years, extensive research has been conducted on the role of nutrition in SCD. The nutritional management in SCD has thus been explored in existing scientific literature in the context of identifying the main nutritional problems associated with the condition, with possible nutritional specific interventions to address these nutritional problems. However, what has not been explicitly explored in the existing scientific literature is how the nutritional management of SCD can be integrated into standard care in SCD, despite calls for nutrition in SCD to be considered in this way (Prasad, 1997; Hyacinth, Gee and Hibbert, 2010; Matthews, 2015/16e; Matthews, 2019j; Khan et al., 2016; Ohemeng and Boadu 2018; Umeakunne and Hibbert, 2019; Kamal et al., 2021).

Furthermore, despite the extensive research conducted on the role of nutrition in SCD (over 60 years), the translation of the findings has not resulted in policy and practice change to support nutrition service provision, as highlighted by Nartey, et al., (2021) albeit in an African context. The link between research and clinical practice has been neglected. As a consequence, the nutritional management of people living with SCD has followed a narrow course and a narrow perspective in how nutrition in SCD is understood, as most of the

studies have been cohort based, addressing the under nutrition and related micro and macro nutrient deficiencies, through for example, educational interventions and programs.

Below the nutritional management in SCD will be evaluated from a global and UK perspective based on a number of review articles, primary research and policies and guidance, to understand the current nutrition landscape in SCD, and the existing clinical practices to manage the nutritional needs of people living with SCD.

(i) Global context:

Prasad's (1997) editorial article, identifies nutrition to be a fairly common occurrence in SCD. In the article, the author discusses a number of clinical problems related to the malnutrition risks of people living with SCD, and nutritional deficiencies that increase the malnutrition risk in these people. However, despite the malnutrition risks identified in people living with SCD, not much is being done to address these nutritional risks further exacerbating the poor health outcomes of people living with SCD. As a result, Prasad (1997, p 424) concludes that SCD has both a molecular and genetic basis, however, 'practically nothing has been done to correct the obvious nutritional problems' in the condition. Moreover, the author encourages physicians in SCD management to address the nutritional issues in SCD, as the correction of the nutritional deficiencies in SCD, may improve clinical outcomes in people living with SCD.

The review article by Hyacinth, Gee and Hibbert (2010), draws attention to the fact that attempts have been made to develop clinical care provision in SCD, since 1910. However, it is only since the 1980's that under nutrition in SCD has been considered as a 'serious complication of the condition' (de Franceschi, et al., 2000; Heyman et al., 1985; Tomer et al., 2001) that should be treated as 'part of required clinical care'. In the same review, under nutrition was identified as a 'critical feature of the condition' (Al-Saqladi et al., 2008; Badoloo, Jackson and Jahoor, 1989; Barden et al., 2000; Khan and Dinko, 2009; Natta and Reynolds,

1984; Soliman et al., 1999), however, this remains unaddressed at an empirical level, despite the availability of existing review articles such as (Reed, Redding-Lallinger and Orringer, 1987) and (Prasad, 1997). The authors of the review, therefore, call on the development of novel nutritional approaches to underpin the clinical management of SCD. Although, not explicitly expressing the need for the integration or embedding of nutrition into SCD care provision, the authors do recognise under nutrition in SCD as a serious complication of the condition that should be treated as part of clinical care. However, the role of the Dietitian in managing the nutritional needs of people living with SCD, is not recognised in existing literature, and this oversight can play a significant part in maintaining the status quo of the need for nutritional service provision in SCD.

Similarly, Hyacinth, Adekye and Yilgwan (2013), and Hyacinth, Gee and Hibbert (2010), recognise the nutritional problems in SCD, and consider the adoption of nutritional approaches in SCD to address the nutritional problems of people living with SCD. Although this is essential, the problem of the neglect of nutrition in standard care is not explicitly addressed, where Dietitians, trained in SCD, can provide a more holistic nutritional assessment and management to effectively address the nutritional problems and needs of people living with SCD. To date, existing research on the role of nutrition in SCD, reflect a limited involvement of Dietitians with an interest in SCD, further purporting the *status quo* of the lack of translation of research findings to support policy and practice development in nutrition in SCD.

Khan et al., (2016), calls for nutrition to be considered to combat the 'perils' of SCD and argue that care provided to people living with SCD, needs to include information about nutritious meals (low-income meals) to enhance protein and calorie consumption. Moreover, in regard to education about nutrition in SCD, emphasis should be placed on the role nutritional deficiencies play in SCD complications, and herein the Dietitian who is trained in SCD, can play a pivotal role not only in educating patients and families but can lead on the treatment and nutritional management of the nutritional needs of people living with SCD. The

overwhelming similarity with the preceding articles is the fact that although much has been reported about nutritional supplements and its benefits as a management option, not much has changed in clinical practice, meaning the nutritional needs of people living with SCD continue to be an overlooked management option. Hence, more work is needed to embed or integrate nutrition into standard care provision and to link research to clinical practice (Prasad, 1997; Nartey et al., 2021).

Nutrition has been identified as one of the most important but often forgotten aspects of care of people living with SCD (Ohemeng and Boadu, 2018). Similarly, Umeakunne and Hibbert (2019), add to this notion of the importance of nutrition, as the authors believe, nutritional interventions should be included as an 'adjunct' treatment in conjunction with standard practice. In this way, these authors, acknowledge the importance of the medical management of SCD, but also argues in favour of the role of nutrition to augment the medical and clinical management of people living with SCD. Umeakunne and Hibbert (2019), plainly state that the inclusion of nutrition is not 'either or' but should be provided 'alongside standard care provision', in response to the ongoing existence of nutritional deficiencies which exist and contribute to the slowed growth, development and reduced quality of life observed in people living with SCD. Moreover, they acknowledge the fact that the management of SCD is complex and multi-factorial (pg 15). Similarly, a number of contributing factors are at play affecting the nutritional assessment of people living with SCD (Matthews, 2015b). Despite over 100 years of the existence of SCD, nutrition is yet to be recognised as an 'adjunct therapy' for addressing multiple -diet – related chronic disorders in SCD (Umeakunne and Hibbert, 2019, p15).

On the other hand, Kamal et. al., (2021) attempts to address the determinants of the severity of SCD in adults in Saudi-Arabia. The authors conducted a prospective longitudinal study with 62 adult patients living with SCD over 93 months, to investigate the impact of nutrition on the outcome of SCD and Health related Quality of Life (HRQoL). Patients were required to provide medical and dietary history, a physical exam, anthropometry, protein and energy

intake, measurement of micronutrient levels, estimated sickle cell severity score and determinants of HRQoL (SF-26v2). In the study, protein energy and micronutrient deficiency were found to be independent predictors of SCD severity and morbidity. In addition, patients living with SCD were found to have various degrees of macro and micro nutrient deficiencies contributing to SCD severity, hospitalisation and poor HRQoL. The authors Kamal et al., (2021, p12) conclude that “integrating and correcting the macro and micronutrient deficiencies in SCD into the standard management of SCD is critical to improve clinical outcomes and QOL” in people living with SCD.

The review article by Dekker et al., (2012), sums up the possible association between SCD morbidity and micronutrient deficiency but recognises the paucity of research in this area. On the other hand, the findings of the Kamal et al., (2021) study, responds to calls from the study by Cox et al., (2011) to determine the relationship between malnutrition and severity of SCD outcomes, which would benefit from a longitudinal study. Thus, their study provides evidence in support of the link between malnutrition in SCD and severity of SCD outcomes. In addition, Kamal et al., (2021), identifies nutrition as an independent determinant of disease severity and health outcomes in SCD, thus supporting the recommendations by Cox et al., (2011) that nutritional interventions may play a role in enhancing healthy survival rates in people living with SCD, as nutrition is an independent determinant of poor severity and poor health outcomes in people living with SCD. In the same way, Mandese et al., (2016), found that inadequate nutritional intake, weight and BMI, have a significant impact on SCD severity indices in children living with SCD. However, Mandese et al., (2016), also highlight the fact that current disease treatment guidance makes no reference to the importance of maintaining an adequate complete nutritional intake. Although earlier studies (Cox et al., 2011; Dekker et al., 2012; Mandese et al., 2016) call for specific nutritional interventions to be ‘appropriately tested’ before any recommendations can be made, the longitudinal study by Kamal et al., (2021) provide compelling evidence in support of nutrition as an independent determinant of disease severity and HRQoL outcomes.

Similarly, there is a need for more qualitative studies to be conducted to gauge the perspective of people living with SCD, caregivers and healthcare providers about nutrition in SCD. The qualitative study by Levers- Landis et al., (2001), a situational analysis of problematic situations conducted with 37 caregivers of children living with SCD aged 5 to 13 years, is an example. Of the number of domains discussed in the interviews, that included medication adherence, nutrition, social problems, academic difficulties and minimising and coping with pain episodes, it is reported that 35/37 caregivers reported experiencing problems with their children's nutrition, followed by reports of the children's experiences of pain and how they cope with having SCD and pain. Notably, the reports of problems with nutrition were amongst the younger aged children and 93 nutrition-related problems were coded as part of the research project, compared to a total of 56 pain related problems, for which many caregivers encouraged their children to rest, keep well hydrated and avoid extreme temperatures. Yet, despite the high volume of nutrition related problems identified, very little has been noted about the practices of care givers to manage the nutrition related problems of their children, highlighting an ongoing need for the role of nutrition as a management option in SCD that can be addressed if nutrition is integrated into standard care provision in SCD; currently more emphasis is placed on pain prevention and management in SCD care.

In this study, more emphasis was placed on caregivers needing support with psychology or social work, even though these problems scored lower on the problem lists, whilst guidelines to support the nutrition needs of the children were not addressed by the researchers. In addition, no mention is made to offer support to caregivers who have identified having experienced problems with the nutrition of their children, even though suggestions were made to address the pain management and psychological needs of those affected.

Likewise, the study by Mitchell et al., (2004), highlight the fact nutrition counselling is not available as standard care as it is in other paediatric chronic illnesses such as cystic fibrosis - where nutrition is integrated into standard care provision (Spielberg and Clancy, 2016) and

diabetes, further evidence of the disparities in care provision for patients living with SCD and the value of nutrition care provision. The goal of the study by Mitchell et al, (2004) was to conduct focus groups of parents and caregivers of children living with SCD to assess how parents perceive eating patterns, growth and nutritional status in their children with SCD. Interestingly, nutrition was identified as a key self-management strategy in SCD according to Tanabe et al., (2010). Therefore, these findings, provide compelling evidence for nutrition to be considered as an important management option in SCD not merely as a perfunctory activity, signalling the need for policy and practice to support nutrition integration in SCD.

(ii) UK context:

Building on the research that has already been undertaken globally, a primary scoping cross sectional survey was conducted in 2015 in the UK (Matthews, 2016g), the first of its kind to assess the existing nutritional management in SCD. The study aimed at exploring the involvement, knowledge and attitudes of Dietitians about nutrition in SCD. Albeit a low response rate, the findings provided insight into how the nutritional needs of people living with SCD were managed. A total of 50 Dietitians responded out of a possible >7000; the survey was emailed to all British Dietetic Association (BDA) registered Dietitians at the time. Significantly, 89% (n=44) of the respondents were from London, known to have the highest prevalence of people living with SCD in England and the UK (Aljuburi et al., 2012; Lucas and Mason, 2008). Despite the low response rate, the data was able to provide some useful insights into the practices of Dietitians involved in SCD management. There was an overwhelming consensus amongst the respondents, that Dietitians have a role to play in managing the nutritional needs of people living with SCD, despite the lack of existing nutrition service provision in the condition.

Out of 33 Dietitian responses, 61% (n=20) reported to have some knowledge of SCD, while 18% (n=6) reported to have no knowledge of SCD. Furthermore, the findings revealed that

out of 45 Dietitian responses, 96% (n=43) of the Dietitians agreed that they have a role to play in managing the nutritional needs of patients living with SCD, while 87% (n=39) agreed that the condition warrants regular dietetic input. Moreover, 87% (n=39) of Dietitians agreed that people living with SCD are not readily referred for dietetic input, warranting more research to explore the factors influencing SCD patient engagement with the Dietitian. Overall, the lack of involvement in the nutritional management in SCD, was attributed to a lack of policy and nutritional guidance and a lack of understanding of the nutritional implications of SCD.

Overall, the survey provided valuable primary data to identify key gaps in nutrition knowledge of Dietitians, and the lack of nutritional resources and management guidelines and standards of care to provide tailored treatment and management options to people living with SCD. Moreover, the gaps explain the lack of nutrition service provision available to people living with SCD, and the poor referral rates of these patients to the Dietitian, further evidence to support the lack of nutritional management in SCD care provision and the need to understand the myriad of influencing factors that may affect nutrition and service provision in SCD management.

2.1.5 Influencing factors in nutrition in SCD

(i) Socio-ecological factors in nutrition in SCD

The Socio- Ecological Model (SEM) perspectives are quite extensively used in health promotion interventions, to help healthcare professionals to understand the multiple layers influencing health problems (National Cancer Institute, (NCI, 2005). The adoption of the SEM was first introduced conceptually to explain human development in the 1970's (Bronfenbrenner, 1977), before being accepted as a theory in the 1980's. However, the SEM, developed by McLeroy et al., (1988), identifies 5 layers of influence to explain health-related

behaviours and conditions, which I have adopted in the research project, namely, intrapersonal or individual factors, interpersonal factors, institutional or organisational factors, community and policy factors. These multiple layers of influence, relate to individual behaviour and are shaped by the social environment in which people interact (NCI, 2005).

Although a socio-ecological framework can be applied as methodology to frame a research project, this has not been the approach used in this research project, but rather the concepts which make up the layers of influence (McLeroy et al., 1988) have been used to structure part of the data collection in the focus groups, in phase one of this research project (see Chapter 3, section 3.4.1). In this way, a socio-ecological lens was used to identify and understand the myriad of influencing factors affecting the nutritional landscape in SCD and the lack of nutritional management in this chronic condition.

More benefits of adopting a socio-ecological lens to the research project is supported by the views of Greenhalgh (2009), in that this approach provides the opportunity to explore the health needs of vulnerable populations across a multi-level system, which is more than just the medical management allowing a more holistic evaluation of their health needs.

Furthermore, Kumar and Preetha (2012) identify the benefit of this approach to help tailor interventions to the specific needs of the patient populations, which may be a more cost-effective approach to promoting the health and wellbeing outcomes of those at risk.

In addition, Henderson and Baffour (2015) states that using a socio-ecological framework can be used as a practical theory in framing a problem, but this requires the use of qualitative inquiry and informs design and analysis. Moreover, it is found to be very relevant to research conducted in marginalised groups. Adopting a socio-ecological lens to the research project helps to frame the research problem, to provide clarity to the way the problem of nutrition in SCD can be understood. The adoption of the components of the SEM system developed by McLeroy et al., (1988), thus enables the research enquiry to consider the research problem at the personal, interpersonal, institutional, community/policy level.

Thus Early (2016) calls on healthcare systems to take a broader (theoretical) approach to health and healthcare, calling on the use of ecological frameworks, as it goes beyond just focussing on the person but considers interaction between and interdependence of the individual, interpersonal, organisational, community and society/policy factors.

Consequently, Davis (2015) identifies the need for collaborative, comprehensive, cross sector approaches to health but included other sectors such social services, public health, education, housing, to address the social determinants of health in underserved communities, relevant to the SCD population.

Moreover, a social ecological approach has been used in mental health and wellbeing with good effect (Reupert, 2017). The SEM system, recognises the dynamic inter relatedness among personal and environmental factors which extend to family, school, community and mental health agencies as they recognise the need for collaborative and coordinated service provision in this patient population, which is very relevant to people living with SCD.

Therefore, there is a need to acknowledge the multiple intersecting levels and corresponding influencing factors between individuals, families, organisations and the community including improving research opportunities for communities from minority ethnic backgrounds, relevant in taking a wider perspective to understanding nutrition in SCD.

(ii) Multi-factorial factors in nutrition in SCD

A review of the literature on the role of nutrition in SCD, predominantly focusses on under nutrition and nutrient deficiencies as experienced by people living with SCD. However, taking this approach may limit our understanding of the possible multi-factorial factors affecting the nutritional needs of people living with SCD. Of these multifactorial factors, limited access to food (Agamah, 2019) and food insecurity in low-income individuals (Cahill et al., 2020), were identified as barriers to nutrition, relevant to marginalised groups as found in Sonenklar et al., (2022) and Khan et al., (2022), linked to the impact of the social determinants of health in

people living with SCD. Food insecurity is identified as a key predictor of wellbeing (Coleman-Jensen, 2020). Furthermore, disabilities are identified as a strong factor for food-insecurity (Coleman-Jensen, 2020). In this way, people living with SCD, are at an increased risk of food insecurity, owing to the multiple complications leading to increasing disability, morbidity and mortality (Fitzburgh et al., 2005; Lucas and Mason, 2008; Aljuburi et al., 2012; Piel et al, 2021). Upon closer inspection, the underlying factors influencing the poor access to food and high rates of food insecurity are what needs more attention in future research studies in nutrition in SCD.

However, without the availability of nutrition service provision, the wider context of nutrition in SCD will be overlooked. The lack of formal nutrition service provision in SCD, in which the role of nutrition as an adjunct treatment to the medical management in SCD (Umeakunne and Hibbert, 2019) is overlooked, the *status quo* of poor access to and high rates of food insecurity will remain. For these reasons, it is imperative that nutrition in SCD is more than just food and a breadth of factors need to be considered when planning nutrition service provision for people living with SCD.

Employment is an important factor in accessing food and food insecurity in SCD. Berghs, Cronin-Chavez, and Ebenso, 2020; Berghs et al., 2021; Bergs and Dyson, 2022), discuss the impact of employment in a SCD context, making education (access to knowledge) equally essential to the affordability and food insecurity narrative in SCD. Equally, knowledge of both healthcare professionals in SCD, (Dennis-Antwi, Dyson and Ohene-Frempong, 2008) and the people living with SCD however, needs to be factored in to ensure a more holistic approach to the problem of food insecurity and nutrition in SCD as a whole.

Nutrition has a multifactorial aetiology in SCD (Matthews, 2015b; Umeakunne and Hibbert, 2019). Nutrition in SCD, is described as having a number of factors affecting its overall intake, assessment and management. In this way, consideration needs to be given to the psychosocial predisposition of people living with SCD, with the most common psychological presentations for SCD being depression and anxiety (Asnani, 2004). In addition, the wider

determinants of health such as housing, employment, socio-economic factors, is recognised by Matthews (2015b) to influence the nutritional needs of people living with SCD. For this reason, nutrition in SCD is more than simply an environmental factor, as reported in the article by Rees, Williams and Gladwin (2010), as its wider role in the management of SCD need to be reconsidered. The findings of the Sickle Cell Health Awareness, Perspectives and Experiences (SHAPE) survey (2022), are noted to confirm, the significance of the effect of health inequalities on people living with SCD in the UK, in line with the findings of the studies by (Sonnenklar et al., 2022) and (Khan et al., 2022) linking the impact of the social determinants of health to the health outcomes and inequalities in SCD.

Equally in the SHAPE survey (2022), it was found that most patients and caregivers, believed that SCD was not recognised as being as important as other conditions, with fatigue identified as the most common physical symptom experienced by 90% of the participants. Moreover, Berghs, Cronin-Chavez, and Ebenso, (2020), in their article on the disproportionate impact of COVID 19 on people living with SCD, include nutrition as an area of impact. However, the nutritional management of these people is not currently part of their standard care provision, further exacerbating the impact nutrition may have on the health and wellbeing outcomes of people living with SCD, due to COVID 19. Although nutrition is recognised in this context, the nutritional needs and management of people living with SCD remain a neglected management option in standard care in SCD that need to be addressed. All these factors cumulatively, attest to the complexity of nutrition in SCD, and the need to provide a more tailored nutrition service provision, where nutrition is recognised and integrated into standard care in SCD.

2.1.6 Policy and practice development in nutrition integration in SCD

The possibility of adopting health literacy to support policy and practice development in nutrition in SCD, comes as health literacy is considered in a different perspective from the

traditional understanding to be about literacy and health education. The article by (Dearfield, Barnam and Pugh- Yi (2017), that considers health literacy as linked to Freire's principles in health interventions, creates an opportunity to achieve this transforming perspective, that has also become a pivoting point in the research project in 2021 (see Appendix 13). This transformed perspective of health literacy in the context of policy and practice development in nutrition in SCD, is demonstrated in Appendix 14, illustrating the overlapping conceptual perspectives of the research project. In this way, health literacy is considered as an expansive and integrated concept, being an evolving and composite term, linked to health outcomes (Nutbeam, 2008; Rudd, 2015; Passche and Wolf, 2007), and considered a determinant of health, to be reviewed below and applied to policy and practice development in nutrition in SCD.

(i) Health literacy in policy and practice development:

Historically, the term health literacy was first used in 1974 (Simonds, 1974), and included in discussions about health education as a policy issue affecting the health system (Ratzan, 2001, pg 21). Berkman, Davis and MacCormack, (2010, p17) considers health literacy to be dynamic, and in particular, health literacy is seen as an evolving term that will continue to evolve (Nutbeam, 2008; Rudd, 2015) in response to advancing science and technology, the delivery of healthcare and public health services, thus increasing expectations and responsibility on individuals and groups in order to understand and act on information.

Furthermore, Paasche- Orlow and Wolf (2007), proposes a logic model, whereby health literacy is linked to health outcomes. In this model, health literacy is proposed to influence health outcomes at three critical points namely, healthcare, interaction between patients and their healthcare professionals and self-care. These authors, thus place health literacy as a risk factor, that should be identified and appropriately managed in clinical care. On the other hand, health literacy is also seen as an asset (Nutbeam, 2008) to be built as an outcome of

health education and communication that supports greater empowerment in health decision – making. In this way, Dearfield, Barnum and Pugh-Yi (2017), have considered Freire's (1970) framework to community interventions to support this process.

Building on the value of promoting health –decision making, a summary outcome model for health promotion has been developed by Nutbeam (1996). In addition, Nutbeam (2000) states that health literacy is also a result of health promotion actions such as education, social mobilisation and advocacy. Moreover, health literacy is identified as a process, linked to more established health promotion models and theories, that can be seen as part of an individual's, development towards improved quality of life (Sorenson et al., 2012). Thus, health promotion is not done 'on' people or 'to' people but rather 'by' or 'with' people (Nutbeam, 2000, p265).

In the Ottawa charter (1986), health promotion has come to be understood as 'public health action', directed towards improving peoples control over modifiable determinants of health such as personal, public policy and living and working conditions. The implicit health outcomes associated with high levels of health literacy includes improved knowledge and understanding of the health determinants, changed attitudes and motivation to adopt health behaviours and choices, and improved self-efficacy (Nutbeam, 2008). In this way, health literacy progressively allows for greater autonomy, motivation and personal empowerment (Nutbeam, 2008), essential to support decision making in individuals toward improving their health outcomes.

Understanding these characteristics of health literacy, and its role in health promotion and improving health outcomes, is beneficial in the context of developing nutrition service provision in SCD. Developing nutrition service provision in SCD is complex and adopting a health literacy lens enables the research project to explore the role of nutrition in SCD in a comprehensive way. Going by the definition of health literacy by the Institute of Medicine

(IOM, 2004) that “health literacy is a concern for everyone in health promotion, disease prevention and early screening, healthcare and maintenance and policy making”, this assumption of the concept, is feasible.

Similarly, health literacy is considered in a more integrated way, by Sorenson et al., (2012). In this study, health literacy is considered more than health education, but encompasses 12 dimensions, in their integrated conceptual model of health literacy. The 12 dimensions included the following: knowledge, motivation and competencies of accessing, understanding, appraising, applying health research information within a healthcare environment to support disease prevention and health promotion. In these 12 dimensions, health literacy is identified to cover the three health domains of healthcare management, public health and health promotion, that supports an expansive and integrative approach to health promotion in the context of policy development in SCD.

Thus, in the context of the research project, health literacy in nutrition in SCD relates to the three domains of health. The original aim of health literacy as defined by Nutbeam (2000) is, ‘the cognitive and social skills that determine the motivation and ability of individuals to gain access to understand, and use information in ways that promote and maintain good health’. as “concerning the capacity of people to meet the complex demands of health in a modern society” to ‘manage their health outcomes’. In other words, enabling people to take responsibility for their own health. Without first developing nutrition service provision, the nutritional needs of people living with SCD cannot be met, moreover, the health literacy (knowledge of nutrition in SCD) of the sickle cell service users and providers will not be possible and continue to negatively impact the health outcomes of people living with SCD.

In this way, using a health literacy lens, drawing on the links of health literacy to health outcomes, being a determinant of health and the relevance of health literacy in the three

health domains, these characteristics of health literacy provides a more holistic and comprehensive lens through which nutrition service provision in SCD can be considered.

Summary:

SCD is a complex condition and this complexity need to be acknowledged in the nutritional implications linked to the condition. Importantly, the causal links which exist between the clinical features of the condition and the nutritional problems associated with SCD, needs more recognition in the management of SCD. The critical evaluation of the literature has revealed the following gaps to consider:

- Nutrition in SCD is viewed through a narrow lens, reflecting mainly the under and over nutrition and the nutrient deficiencies as experienced by people living with SCD
- The oversight of the multiple influencing factors related to the complexity of SCD (health and social factors), that may impact on the nutritional needs of people living with SCD
- Most research conducted has been cohort based
- There is a paucity of policy and practice to support nutrition service provision
- Nutrition is not currently part of standard care provision in SCD
- Limited Dietetic input, in research and clinical practice – evidence of poor knowledge, lack of resources and limited involvement
- Currently, no framework exists to support the integration of nutrition into standard care in SCD

All in all, these gaps, require a comprehensive approach to support the development of nutrition service provision available to people living with SCD, as part of their standard care. The next part of this chapter will review the literature in regards to the theoretical and professional practice perspectives informing the research project, to address the identified gaps in knowledge, relating to the research project aim and objectives.

Chapter 2.2

Theoretical and Professional Practice Perspectives:

Part 2 of the literature review chapter provides an overview of the main theoretical (see Appendix 14) and professional practice perspectives underpinning the research project and its contribution to policy and practice change. First, it will explore Freire's (1970) seminal work, '*Pedagogy of the Oppressed*' as the main theoretical underpinning of the research project. Secondly, it will explore the role of the Dietitian and Dietetic practice, the development of policy to support nutrition service provision, and promote the value of integrating nutrition into standard care in SCD, the main aim of the research project.

2.2.1 Freire 's theoretical framework

(i) Rationale:

The theoretical approach of the research project is underpinned by Freire's (1970) seminal work, '*Pedagogy of the oppressed*'. There are a number of factors that make adopting a Freirean lens to this research project necessary. Firstly, the gaps in knowledge and care provision in SCD, resulting in the neglect of nutrition as part of standard care in SCD, speaks to the marginalisation established in SCD, evidenced in the APPG-SCAT (2021) report. Thus the ongoing marginalisation of the SCD population, purports the potential for injustice and inequality that needs to be exposed in nutrition in SCD, starting with the research context.

The paucity of studies investigating the nutritional management of people living with SCD (See Figure 2.1), reveals a number of shortcomings that need to be addressed in nutrition in SCD. These shortcomings, in particular caused by the lack of translation of existing scientific literature on the role of nutrition in SCD, have a direct bearing on the current clinical and nutritional policy and practice in SCD, and therefore the level and quality of care available to

SCD patients. Hence, a light needs to be shone on these shortcomings in nutrition in SCD, and Freire's concepts, provide the lens to do so.

Freire's lens is poignant to demonstrate the link between education, knowledge and critical consciousness. Without the translation of the existing scientific literature about the role of nutrition in SCD, what is known about nutrition in SCD is diminished and contained and available only to a handful of people who have access to scientific journals and publications. This oversight further monopolises what can be known about nutrition in SCD, especially relevant to the people who would benefit the most and closely reliant on healthcare professionals to provide the range of care they may need. In this way, the knowledge/power ratio is maintained in this relationship reinforcing the lack of critical consciousness about the value of nutrition in SCD management.

This lack of critical consciousness, a key concept of Freire's pedagogy of the oppressed, defined as the *'awakening to the expression of social discontents seen as precisely the real components of an oppressive situation'* (Freire, 1970, p10) signifies that people are not in a position to see the problems, its negative effects and therefore lack the impetus to take action to change the status quo. In so doing, the status quo of nutrition in SCD is maintained and so too the effects of the marginalisation, which by and large defines the SCD population.

Dyson (2019) calls on scientists in SCD to consider the benefits of research to improving the patient outcomes and not simply conducting research for research sake. For this reason, how researchers view the SCD population, and their role in society and their value and worth as human beings deserving nutritional management as part of their standard care, needs consideration. Central to Freire's framework, is the need to consider people as human subjects that have value and worth and not for people to be seen as mere objects, reliant on others to meet their needs.

This also speaks to the need to consider the holistic lived experiences of people living with SCD, which includes the whole person and not reduced to simply their medical management

but should be extended to include the nutritional implications, based on the plethora of existing scientific evidence. Therefore, adopting Freire's lens to discussing the existing scientific literature on the role of nutrition in SCD, not only serves to increase critical consciousness on the subject, but also supports the development of praxis (critical reflection to take action) that eventually leads to empowerment, the ability to take more control and make decisions towards changing the current status quo to improve nutrition service provision in SCD, inherent in the aim of this research project.

There has however been a number of criticisms about Freire's framework. In particular, the framework has been criticised in connection with the language used being described as difficult, pompous, convoluted, to mention only a few (Schugurensky, 1998). The use of sexist language by Freire was criticised by feminists, as well as the absence of references to race and gender issues, with seemingly more emphasis placed on class as an oppressive structural problem (Shugurensky, 1998). Nevertheless, Freire's (1970) seminal framework remains the chosen theoretical framework for the research project to be explored below.

(ii) Freire's theoretical concepts:

The key conceptual principles of Education, Empowerment, Critical consciousness and Praxis, apply to the observed knowledge and care gaps, lack of nutrition service provision and lack of policy and practice to support a change in nutrition service provision, to people living with SCD, as a marginalised patient group. These concepts will be evaluated below as it relates to the research problem.

Education

Withholding knowledge is a form of oppression and disempowerment as discussed in Freire's (1970) seminal work. The power/knowledge struggle is recognised by Freire (1970, p 81) to be everywhere and can be used by both the oppressors and the oppressed, and is a

key critical conceptualisation of the work of Foucault (1976). The conceptualisation of the relationship between power and knowledge, therefore underpins the essence of Freire's book, demonstrating how knowledge has been used to exercise power over people to keep them in a state of powerlessness and oppression.

To circumvent this power imbalance, Freire recommends education to be provided in a dialogical way, and this form of communication is believed to neutralise the power/knowledge struggle. In this way, engaging in dialogical communication in SCD, between sickle cell service users and service providers, there is more opportunity to address the power/knowledge struggle currently perpetuated by the lack of nutrition service provision in SCD; this mutual dialogue acknowledges the key aspect of Freire's Pedagogical framework, relating to the need for those who are oppressed to be seen as more human, thus the oppressed seen as subjects and not merely as objects. In this way, the effectiveness of dialogue in action, (Igor and Freire, 1987), reflects the value of humanisation, linking praxis, dialoguing and conscientization (developing and changing consciousness), to collectively promote emancipation and empowerment.

Hence, poor knowledge of nutrition in SCD as a consequence of the lack of nutrition service provision, holds serious risks to the people living with SCD in terms of poor health outcomes. Therefore, health education in particular, is recognised as a factor to increase health literacy (education/knowledge), believed to increase the decision making of individuals and their ability to take control of their health outcomes (Nutbeam, 2000). In this way, both education and health literacy in this context, are important when considering nutrition service provision in SCD, as health literacy (education/knowledge) is also considered a shared function of social and individual factors (IOM, 2004). In this way, Freire's conceptualisation of the importance of education is relevant in nutrition in SCD, as currently there are low levels of knowledge, education and service provision in SCD (Matthews, 2016g), that needs to be addressed to empower the people living with SCD to improve their QOL outcomes.

Empowerment

Building on the dialogical approach to education (Freire, 1970), education can be seen as a process of empowerment. Freire (1970, p 50) describes empowerment as living from the point of being rather than having or doing. In other words, empowerment is not dependent on what you have (possessions or knowledge) or the position that you hold, rather it is how you perceive yourself and what you are able to do. Self – depreciation (Freire, 1970, p 37) is a term used to describe how individuals who are oppressed see themselves, for example as ‘lacking self-worth’, being convinced that they are unfit and seeing themselves as being ignorant and others ‘knowing more than they do’. This may be true of the people living with SCD, with studies demonstrating drug seeking and attention seeking behaviours (Maxwell, Streetly and Bevan, 1999). As such, people living with SCD may be referred to as being ‘sick, lazy and unproductive’. When individuals see themselves through a self -depreciating or disempowered lens, ignorance and stagnation is the consequence and disempowerment the outcome. For this reason, Dyson (2019) promotes a new perspective to reframe the disability of SCD to be more about what people living with SCD ‘can do’, as opposed to the traditional assumption that because of disability people living with SCD ‘can’t do’ and achieve more than is expected.

Relevant to nutrition in SCD, empowerment and health are portrayed as multi-level concepts (Balcazar, Keys and Vryhof, 2019). As such, empowerment is about helping those that lack their own power to understand their own situation and to empower them to take more control over their own situations (Balcazar, Keys and Vryhof, 2019). To achieve this power transfer in people, it is necessary that empowerment factors in people’s needs, their rights and their choices (Dearfield, Barnum and Pugh-Yi, 2017). Empowerment as a process enables people to gain a greater influence and control over what is most important to them and their own lives and their environments (Balcazar, Keys and Vryhof, 2019). In other words, empowerment is about enabling and equipping people to take control over their life, similar to health literacy, which is deemed critical to empowerment (Nutbeam, 2008). Therefore,

both powerlessness and empowerment are about control, the difference being, empowerment seeks to enable people to gain greater control over decisions affecting their lives, health and important aspect of health. In the same way, health literacy seeks to enable individuals to exert greater control over their life and situations. Thus, empowerment in health is believed to be able to help individuals to achieve these health benefits owing to the fact that the process enables people to gain greater control over the decisions affecting their lives and health (Sadan, 1997; Nutbeam, 1998). Therefore, empowerment is an important concept when considering nutrition and service provision in people living with SCD.

Critical consciousness

Critical consciousness is elicited through education according to Freire (1970). Freire (1970, p10), describes the concept of critical consciousness as an 'awakening to the expression of social discontents seen as precisely the real components of an oppressive situation'. In SCD, without critical consciousness the expression of the reality of both people living with SCD and providers remains an unconscious problem. A lack of critical consciousness for both the people living with SCD and providers drives the propensity to continue with the *status quo*, maintaining a reality of unconscious oppressive behaviour that is accepted as the norm. The belief is that critical consciousness stirs up awareness of the social discontent within individuals to challenge the social norms and accepted realities, an important concept in changing the *status quo* pertaining to current nutrition landscape in SCD.

However, Freire's concepts of critical consciousness and praxis were criticised in how it was portrayed as resulting in automatic transformation, in this way believed to be an oversimplification of the range of factors that is known to be required to promote social change (Schugurensky, 1998; McLaren and Leonard, 2004). In this way, critical consciousness may need other factors to be in place, not only causing the stirring up of the social discontents, but needs education, empowerment and reflection and action to facilitate the change expected. If not, a lack of critical consciousness can potentially keep individuals

contained in an oppressive environment or in a SCD context, satisfied with a service provision that may not be meeting their full health needs.

Nutrition in SCD is an example of an unmet health need in SCD management/ treatment, nutrition being the white elephant in the room. However, Giroux (1993), emphasises the value of Freire's work as providing – a 'language of possibility' – aimed at propelling and motivating people (the oppressed) to take an active role in shaping their communities to address those factors that keep them oppressed. Hence, critical consciousness requires time for critical reflection towards taking action to change the current *status quo*, nutrition in SCD a prime opportunity for praxis and change.

Praxis

Education according to Freire (1970) not only elicits critical consciousness, but drives reflection and action. In his book, Freire highlights praxis, which he describes as 'reflection and action'. Praxis is an important element of education and is used to describe the enablement, embodiment and realisation of skills. Praxis in this context, relates to autonomy and responsibility, key concepts in health literacy relevant to facilitate behaviour change and transformation. However, Freire (1970, p 60) says that there can be no transformation without action. Moreover, Freire points out that action should not be emphasised to the detriment of reflection. In other words, once critical consciousness is reached amongst people living with SCD and providers, both groups should in the first instance spend time to reflect on the social discontents surrounding the provision of healthcare services. The ensuing learning, following the reflection, should then be channelled into focussed action to propel transformation. However, failing to take the time to reflect on the social discontents, can render the action less meaningful, as it may not address the true problems, such as the lack of nutrition service provision in SCD. Action may however require individuals, such as people living with SCD and providers to display empowerment rather than disempowered

behaviours that is critical to the co-development of the health literacy framework, the main output planned for the research project.

Qualitative approach

According to Freire (1970), a dialogic approach to education supports a qualitative research approach. A qualitative approach to research, promotes joint responsibility, co-operation and elicits critical consciousness about the social discontents surrounding the problem to be researched (Freire, 1970, p 53). A qualitative approach can reduce isolation of the oppressed or individualism and promotes fellowship and solidarity. An example of a qualitative approach is adopting a problem -posing approach to education, whereby both people living with SCD and providers become co-investigators or engage in mutual conversation and this in turn facilitates feelings of empowerment. As previously mentioned, education is the practice of freedom (Freire, 1970) and by allowing a qualitative approach to research, people feel less disempowered and oppressed when they are included in the process and given the opportunity to contribute to the discussions. Furthermore, qualitative research promotes 'peoples thinking' (Freire, 1970, p 81) which occurs in and among people together seeking out a reality. Freire says; "I cannot think for other people or without others, nor can others think for me" (Freire, 1970, p 81) and qualitative research creates the opportunity for people to develop their thinking on the subject being researched. A participatory based research approach such as the Learning Alliance Methodology (LA) Methodology (Moreno-Leguizamon et al., 2015) is an example of a qualitative research approach that embodies dialogic theory and promotes empowerment, partnership working, mutual communication and collaborative working. Moreover, adopting a participatory methodological approach adds more opportunity for collective learning and transformation.

2.2.2 Professional policy and practice perspectives

(i) Policies for nutrition management in SCD:

As already established, nutrition is a neglected part of standard care in SCD, despite the more than 60 years of existing scientific literature on the role of nutrition in SCD. Moreover, the growth and nutritional deficiencies caused by the clinical features of SCD, require nutrition service provision, yet to date there is a paucity of policy and practice guidance to support nutrition service provision in SCD. In an attempt to break this cycle of lack of policy and practice to inform clinical practice in the nutritional management of SCD, the inaugural national nutrition standards were commissioned by the Sickle Cell Society, in the UK, and were included in the national clinical standards for the management of adult SCD patients (SCS, 2018, pg 75-77). These nutritional standards were developed following the findings of the primary survey conducted by Matthews (2016g), reflecting the low levels of Dietetic involvement in SCD, due to a lack of nutrition guidelines, policies and standards. To date, these national nutrition standards are the only policy document in the UK directly relating to nutritional management in SCD. The development of the nutrition standards, was based on best practice (NICE Guidelines, 2006) about the nutrition support of adult patients, a literature review of existing scientific literature on the role of nutrition in SCD and my clinical knowledge, skills and experience working as a Dietitian and social liaison officer in a SCD service. Currently these nutrition standards are not implemented in any existing SCD related policies, reflected in the lack of nutrition services available to people living with SCD.

The development of the national nutrition standards was aimed at raising awareness and increasing the knowledge of the role of nutrition as a viable management option in SCD, and the following three nutrition standards were accepted and included in the national clinical standards for adult patients living with SCD (SCS, 2018, p75-77);

- Nutrition Standard 1: Dietitians to be part of the SCD MDT
- Nutrition Standard 2: All patients with SCD to be screened for malnutrition by appropriately trained healthcare professionals (NICE, 2006)
- Nutrition Standard 3: All patients with SCD found to be at risk of malnutrition need to be treated by appropriately trained healthcare professionals (NICE, 2006).

Four further recommendations were included in addition to the nutrition standards, calling on increased involvement of the BDA and all other relevant stakeholders and professionals to be aware of the role of nutrition in SCD and the need for more research to be conducted in nutrition in SCD. In addition to these nutrition standards and recommendations, the only other reference to nutrition management guidance is the following; a Chapter on the nutritional management of SCD (Stopler, 2008), a Canadian consensus paper, and the Annual Review Proforma (makes some reference regarding checking the weight and height of patients), whilst the UK Forum on Haemoglobin Disorders Quality Standards (2021) report, mentions the word 'Dietetics'. However, no specific measures to assess the quality of the nutrition services available to the patients are included, and no reference is made to the National Nutrition Standards (SCS, 2018, p 75-77) despite it being part of the official National Clinical Standards for adults living with SCD. This lack of policy and practice in nutrition in SCD, leaves a huge gap in the nutritional management and related policy and practice guidance to support nutrition service provision in SCD, in the NHS. In this way, there is a critical need to link existing and future research on the role of nutrition to clinical practice (Nartey et al., 2021), and for the development of policy and practice guidance to address the neglect of nutrition service provision as part of standard in SCD, to improve patient outcomes in nutrition in SCD.

(ii) Dietetic clinical practice:

Role of the Dietitian in nutritional management in SCD:

The reality is, that without policy and practice guidance to address the neglect of nutrition as part of standard care in SCD, the development of nutrition service provision to manage the growth and nutritional deficiencies caused by the clinical features of SCD, will continue to impact the health outcomes of the SCD patient population. However, in addition to policy and practice change in nutrition in SCD, there is need for a well trained workforce with the necessary knowledge and skills of nutrition in SCD. Importantly, Dietitians are recognised as diet and nutrition experts, and with quality, tailored education and training in nutrition in SCD, are well suited to work as part of the SCD MDT, called for in the national nutrition standards (SCS, 2018, pg 75). In addition, as part of standard practice, a core skill of Dietitians is to translate research into clinical practice (BDA, 2020). An example of linking research to clinical practice in Dietetics is observed in the nutritional advancement of COPD management (Kominek et al, 2017). A lack of linking research to clinical practice in nutrition in SCD, has been highlighted by Prasad (1997) and Nartey et al., (2021).

In an attempt to link research to clinical practice, the findings of the study by Matthews (2016g) has led to the inclusion of the national nutrition standards in the standards for the clinical management of adults living with SCD (SCSs, 2018, pg 75-77), however, very little has been done to incorporate these standards into existing policy and practice in the standard management of people living with SCD, (as previously mentioned) maintaining the current *status quo* of the neglect of nutrition as part of standard care, meaning people living with SCD do not have access to nutrition service provision as part of their standard care provision impacting patient outcomes in nutrition in SCD. Therefore, Dietitians trained in SCD, when included in the SCD MDT (Nutrition Standard 1 – (SCS, 2018, p 75), are in a prime position to screen, identify and assess the nutritional needs, risks and challenges of

SCD patients as part of their standard care provision and Dietitians are well placed to support the nutritional management of SCD patients in primary care.

Role of the Dietitian in clinical practice in primary care:

The role of the Dietitian is strongly emphasised by the BDA and the British Specialist Nutrition Association (BSNA) (2013). In this document, the authors underpin the importance of nutritional care provision by Dietitians to help the government in the UK achieve their objectives for people living with long terms conditions, in particular improving the QOL outcomes (Domain 2) and experience of healthcare provision (Domain 4) of the NHS Outcomes Framework (2021/22). It is believed that Dietitians can make a difference in managing the nutritional needs of people living with long term conditions, which includes people living with SCD, in primary care (BDA, 2015; Matthews, 2021k).

Dietitians in primary care can therefore play a key role in the nutrition management of many chronic long term conditions, including SCD, in this way the BDA has undertaken a programme of work called Future Dietitian 2025; its purpose, 'to ensure that the profession remains relevant in this changing world and that Dietitians and the wider dietetic and nutrition workforce are fit for the future' (Hicks, 2017). For this reason, Dietitians have been included in the Additional Roles Reimbursement Scheme (ARRS), as part of the latest GP contract agreement (2020/21-2023/24) (NHSE, 2020), in recognition of the close connection between the impacts of nutrition interventions on health outcomes, to ensure optimal health (Hicks, 2017).

To improve the health and cost burden goals in primary care service provision, a personalised care approach has been adopted. Personalised care (NHSE, 2019) is an all-age whole systems approach to integrating services and community support around the person giving people more choice and control over their health and wellbeing to live as independently as they wish and be more involved in the decision-making process of their care provision. In this way, individuals living with SCD, can be supported and enabled to talk

about their nutrition needs in addition to their medical needs, in order for them to better achieve their health and wellbeing outcomes. This will however, benefit from considering the myriad of influencing factors that may impact not only on the level and quality of nutritional service provision in SCD, but on the myriad of influencing factors that may directly affect the nutritional needs of people living with SCD and their overall health and wellbeing outcomes.

Summary:

Part one of the literature review chapter focussed on outlining the main gaps in the literature and the influencing factors affecting nutrition integration in SCD, in the context of how the marginalisation of SCD impacted on both the medical and nutritional management of SCD.

In this way, part two of the literature review chapter provided an overview and rationale for the choice of Freire's theoretical framework, in acknowledging the marginalisation of SCD, and the need to address the lack of critical consciousness and praxis of the role of nutrition in SCD management, to improve patient's outcomes in nutrition in SCD.

Furthermore, part two reflected on the role of the Dietitian and the required policy and practice change that is needed to support the nutritional management of people living with SCD, to address the neglect of nutrition as part of standard care in SCD.

The next chapter will focus on the chosen philosophical and methodological decisions underpinning the research project to achieve the research project aim and objectives, in acknowledgement of the marginalisation of the SCD patient population.

Chapter 3

Methodology

This chapter critically considers the evidence base in support of the philosophical and methodological choices which guide and underpin the research project design, ethics, recruitment of participants, methods and processes and the thematic analysis of the data, in order to achieve its overall aim and objectives.

3.1 Justification of the Research Methodology

3.1.1 Research Paradigm and Research Strategy:

This research project's design, population, choice of methods and data collection were conducted to reflect the multifaceted and complex lens through which key methodological, philosophical and theoretical elements were considered based on my own biographical experiences. In this way, my positionality as a researcher is considered an important part of the choice of methodology (Sikes, 2004) and since research is subjective, the beliefs, values, ontology and epistemology of the researcher informs the philosophical assumptions of the research project (Jackson, 2013, pg 50). The choice of research methodology is therefore not a one-size-fits-all exercise (Hood, 2006), nor does the research paradigm and research strategy fit neatly into a box, as highlighted by Blaikie (2010). Both these authors (Hood, 2006; Blaikie, 2010) acknowledge the range of factors influencing the decision-making processes required to conduct a research project including its research strategy.

Research strategy:

Traditionally, 'what' questions would be answered using an inductive research strategy, which aims to provide a description of a social phenomenon and help to understand the problem from the perspective of the participant's experience of everyday life (Robson, 2011;

Blaikie, 2010, 2007). While a phenomenological approach to the research enquiry (which relates to a paradigm of personal knowledge and subjectivity), as set out by Objective 1 (chapter 1, section 1.2) is relevant, i.e. placing an emphasis on personal perspectives and interpretation (Lester, 1999), it is important to note that within this interpretivism approach, the researcher forms a key part of the research (Ritchie et al., 2013; Brown, 2015).

Although, there is a need to understand the phenomenon of the factors affecting nutrition and service provision, based on the responses of the SCSU-C and SCSP groups, failing to recognise that the SCD patient population is predominantly from a marginalised background (SCS, 2018; NICE, 2021a) would only give a partial view to interpreting the full context of the perspectives of the participants. It has been established that people living with SCD are impacted by racial discrimination and marginalisation (APPG -SCAT, 2021; Redhead, 2021; Dyson, 2019; Anionwu and Atkin, 2001), which may therefore influence the views and perspectives of the participants. While racial discrimination was not explicitly explored as part of data collection, in answering the aim and objectives of the research project (chapter 1, section 1.2), it is necessary to acknowledge and consider ethnicity and the marginalisation of SCD when interpreting the results. The impact of race and inequality was allowed to emerge naturally from the comments, views and the experiences of the participants, not to detract from the main purpose of the study. This is particularly pertinent in relation to interpreting the data collected, and required the adoption of an appropriate critical lens and paradigm to fully understand the context of the influencing factors in nutritional management in SCD.

Social constructionism:

A constructionist approach, building on the findings of the first part of the research project (Objective 1, section 1.2) was adopted to meet Objective 2 (section 1.2). The action of co-constructing the health literacy framework would draw on a social constructionist paradigm. Social constructionism refers to the construction of social properties as a consequence of

the interactions between people and knowledge is actively constructed (Ritchie et al., 2013), meaning social properties do not have a separate existence, and do not exist in their own right (Robson, 2011). According to Schwandt (2007), social constructionism is concerned with the way people experience the world as they interact with others in social situations, which is the *modus operandi* for this research project, that includes the co-development and evaluation of the health literacy framework. The social interaction between the SCSU -Cs and SCSPs, using a constructionist paradigm produces as many realities as there are number of participants, and includes me, as the researcher (Robson, 2011 p 24).

In this way, constructionist epistemology refers to 'truth' or 'meaning' that emerges in and out of people's engagement with what already exists in the world (Moon and Blackman, 2017). Furthermore, these authors state that constructionist research is helpful in generating contextual understandings of a defined topic or problem, relevant to the aim of the research problem, to understand the knowledge, views and experiences of SCSU-Cs and SCSPs of nutrition and service provision in SCD. Equally, the values of the researcher and the participants are considered as part of the data collection process giving the participants a central and integral role in the research project to co-construct the health literacy framework. As such, social constructionism, unlike positivism, believes there is more than one single view or truth, implying that there can be a range of views, thereby allowing consideration for different voices, reflecting all the participants, and that there is no neutral, objective perspective (Alderson, 1998, p1009). Furthermore, in social constructionist research, the expectations, values, background and roles of the participants are considered, albeit implicit in the comments provided (Alderson, 1998) and since the target audience (SCD population) is from a marginalised background, it is feasible to consider a critical realist constructionist lens to the research project.

Critical realist constructionism:

The critical realist paradigm is a relatively new paradigm, Baskhar (1975) and is a brand of philosophy that distinguishes between the real world and the observable world. This means that unobservable structures in the world (Eg faith, race, ethnicity, health) cause observable events, such as change in beliefs, or the effects of racial discrimination or disempowerment. Therefore, it is believed that critical realism helps people to understand the world by understanding the unobservable structures in society that generates events. Hence, it can be known that race and ethnicity is real because of the observable effects (change in beliefs, values, inequality etc.). Pertinent to the SCD patient population, racism and resulting oppression have become key factors driving the marginalisation of SCD, as showcased in the APPG–SCAT (2021) report titled, '*No one is listening*'. The report highlighted how racism has impacted on the quality and levels of healthcare provision in the medical management of people living with SCD seeking care in the NHS. Therefore, in light of the marginalisation of the SCD patient population, adopting a critical realist lens to the research project is feasible.

Furthermore, the critical realist paradigm (Robson, 2011) aligns well with the philosophical and conceptual principles which underpins Freire's (1970) seminal work, as an emancipatory objective is relevant within a critical realist research agenda (Haigh et al., 2019). Both the critical realist paradigm and Freire's principles support emancipation and empowerment of marginalised and oppressed groups. In particular, Freire's (1970) '*Pedagogy of the oppressed*' uses education and empowerment, critical consciousness and praxis as tools to promote emancipation through mutual dialogue. The emancipatory function of critical realism relates to the collaborative style of the research approach, whereby there is active participation of the oppressed or marginalised groups in the research project. In this way, a critical realist constructionist lens also has an affinity to emancipatory styles (Robson, 2011). Therefore, a critical realist constructionist lens is most appropriate to the research enquiry and not only supports the emancipatory and empowerment function of the research project, but would benefit from a participatory methodology with a dual aim of knowledge sharing and

co-production to facilitate the co-development of the study intervention, the health literacy framework to support the integration of nutrition into standard care in SCD.

Participatory Action Research:

Paulo Freire's – '*Pedagogy of the oppressed*' (1970) lays a solid foundation to consider the role of Participatory Action Research (PAR) as a critical methodological approach to the research project, in conjunction with the LA methodology. Participatory research approaches offer many benefits to maximise its value as a research methodology. Participatory health research, (International Collaboration PHR), is considered to be an umbrella paradigm, and not one with narrow epistemological principles. Furthermore, PAR seeks to add value to the participants. According to Reason and Bradbury (2001, p1) PAR is defined as "a participatory, democratic process concerned with developing practical knowing in the pursuit of worthwhile human purposes". As such, the participatory approach offers the opportunity to really engage the participants in the research process, thereby promoting empowerment, similar to the principles of the LA methodology.

Cornwall and Gaventá (2001), emphasise the benefit of using research methods that promote consciousness-raising and reflection, to empower the capacity of participants to challenge unfair and unjust structures of power. Paulo Freire (1970), says empowerment is about enabling people to live from a point of being, not based on their possessions or what they have, but more about how they see themselves and what they can do. As such, people from marginal backgrounds, who lack the power to effect change in their own environments, such as the poor and disenfranchised, are the focus of action researchers (Somekh, 2006) to facilitate emancipation and empowerment through collaboration and participation.

A key characteristic of action research is for all participants in the research projects to be involved in the decision-making process. This is critical in SCD in order to promote inclusion and empowerment within each participant. In addition, researchers are deemed to be participants of the research, and work together with the participants to ensure that no one

person or group have more power or influence over the other (Ahmed, 2009; Brien, 1998).

Action research in the healthcare field is recognised as having the potential to improve people's health and decrease health disparities. This makes action research a desirable and appropriate research methodology for the research project in nutrition in SCD, key principles on which the LA methodology draws on. It is understood that action research is able to bridge the gap between research and practice, in so doing, able to address social justice, thereby facilitating conditions enabling people to have greater control over the determinants of health (Cargo and Mercer, 2008).

Minkler and Wallerstein (2008) point out that participatory research uses a wide range of terms to describe the research methodology, used widely in countries including the UK, Australia and New Zealand. More importantly, action research shares core principles and characteristics with the Learning Alliance (LA) methodology, drawing on collaboration and partnership working, undergirding its participatory nature, a key characteristic of the LA Methodology. However, the LA methodology equally has its own distinguishing features that makes it more suitable to meet the aims of this research project, in particular the formation of the LA networks between the participants of the study and its role in policy and practice development, to support the co-development of the study intervention, the health literacy framework, using the SCSU-Cs and SCSPs.

3.1.2 The Learning Alliance Methodology:

An alternative research strategy for this project would need to answer both 'what' and 'how' research questions. According to Robson (2011) and Blaikie (2007, 2010), an abductive research strategy is able to answer both these types of research questions. The first part of the study, the 'what' research question: *What are the influencing factors affecting the integration of nutrition in SCD*, is through a phenomenological lens. Therefore, like the inductive strategy (of which phenomenology is an example), the abductive research strategy

is able to understand the meanings and interpretations of the participants of their everyday life (Blaikie, 2007, 2010). The second part of the study considers the 'how' research question: *How can a health literacy framework support the integration of nutrition in SCD*. In this way, the abductive research strategy accommodates both a phenomenological view to understanding the main phenomenon of the research project, whilst also providing reasons (how) rather than causes (why) to explain and understand the barriers to nutrition and how they influence the integration of nutrition into standard care in SCD.

In abductive research strategies, the participants are elevated to a central place (Blaikie, 2007, 2010) as seen in action research projects (Robson, 2011). Elevating participants to a central role are a key aspect of the LA methodology. Pertinent to consider is the fact that the LA methodology has itself evolved from action research and may therefore be relevant to the critical theory paradigm, which has an emancipatory and empowerment function. In particular, the critical theory paradigm focusses on research, which is done 'with people' rather than 'on people', following a non-traditional approach to research where participants are not normally considered central to the research project (Scotland, 2012).

The LA methodology (Moreno-Leguizamon et al., 2015) is thus both a qualitative and a participatory research methodology. Furthermore, the LA methodology as a research approach also embodies dialogic theory and promotes empowerment, partnership working, mutual communication and collaborative working. However, the LA, is an emerging methodology in healthcare research (Moreno- Mguilzamo, 2017) and has several principles in common with PAR. One of the common principles between PAR and the LA methodology is that both approaches focus on the importance of acknowledging the sociocultural turn of the participants or groups (Ozaane and Saatcioglu, 2008) and promote collaboration and partnership working between the researchers and the participants. More specifically, Ozaane and Saatcioglu (2008) refer to an array of factors influencing marginalised groups, such as inequitable distribution of economic resources and opportunities, as well as the unjust power distribution in the social, cultural, and political dimensions of their daily lives, (Thompson

2004), all of which are relevant to the SCD population, a marginalised group, subject to health inequalities.

The LA methodology has been successfully used in research projects with marginalised communities and those from BEM backgrounds. The Learning Alliance Palliative Care and End of Life (LAPCEL) study (Smith and Moreno- Leguizamo, 2017, 2019) demonstrates how stakeholders, including a variety of service providers and service users (from a variety of BEM backgrounds) have joined to form an alliance. The aim of the joint alliance is to discuss issues pertaining to how members of various ethnic groups, through a process of knowledge co-development and sharing, can work towards a common strategy for the best course of action to better manage the needs of the communities receiving palliative care and end of life treatment.

Similarly, the study by Moreno – Meguilzamo et al., (2015) investigating the challenges in health planning of multi –ethnic communities in Kent, demonstrates the value of knowledge co-constructing and sharing in order to achieve a common and joint strategy to support improved health service provision and related outcomes for a range of ethnic minority groups in the region. In order to allow participants of the LA to identify for themselves what is most important to them, Moreno- Leguizamo et al., (2015) recommends conducting independent focus group meetings before the formation of the LA. This approach was adopted by me in this research project, to facilitate the co-development of the study intervention, the health literacy framework.

A key aspect of the LA methodology is the multi-stakeholder involvement supporting an emancipatory and empowerment function to address the power imbalance that can exist between patients and providers or stakeholders in a healthcare context (Dearfield, Barnum and Pugh-Yi, 2017). Since the abductive research strategy places participants in a central role in the research project, the LA methodology is appropriate as it enables the involvement of both the SCSU-Cs and SCSPs as central players of the research project. This allows me as the researcher to gauge the collective knowledge and interpretations of both groups of

research participants to better understand the 'phenomenon of the influencing factors affecting nutrition in SCD'. This in turn supports an integrated view of understanding the problem and how best to address the problem from a bottom-up perspective, to support policy and practice development. Not only does the LA methodology support integrated knowledge (Moreno-Meguizamo, 2017), it also supports a health literacy perspective in relation to the importance of the patient-provider relationship, a stakeholder approach. The patient-provider relationship is identified as a key element of effective health literacy in the context of education and empowerment (Dearfield, Barnum and Pugh-Yi, 2017).

The LA methodology is well suited towards supporting a collaborative and partnership working to inform capacity and policy and practice development in marginalised communities (Moreno-Meguizamo et al., 2015; Smith and Moreno-Meguizamo, 2017, 2019). Establishing the importance of education, emancipation and empowerment, as it relates to stakeholder integration and involvement, is essential to the co-development of the health literacy framework. Hence, co-development depends on the establishment of the Learning Alliance Network (LAN) formed between the SCSU-Cs and SCSPs. The LA methodology therefore not only supports a constructionist paradigm, but it also supports a critical realist paradigm based on the emancipation and empowerment function inherent in collaborative and partnership working. The formation of the LAN between the SCSU-Cs and SCSPs is central for the co-development of the health literacy framework needed to support social actions to transform nutrition service provision. Thus, the recruitment of suitable participants to form the LAN is an important part of the overall research strategy to achieve the aim and objectives of the study (chapter 1, section, 1.2).

3.2 Sampling and Recruitment

3.2.1 Sampling Method:

Purposive sampling is often used in qualitative research projects (Devers and Frankel, 2000; Robson, 2011; Blaikie, 2007, 2010), and as such was chosen for this research enquiry. A purposive sampling method also known as judgemental sampling (Robson, 2011), or non-random sampling, according to Devers and Frankel (2000), allowed me as the researcher to do the following:

- factor in selection sites and subjects, with the aim of identifying suitable sites and subjects which are best suited to answer the research questions,
- enable and judge the suitability of the participants (Robson, 2011, Blaikie, 2010) and the unique characteristics of the research participants (Devers and Frankel, 2000),
- select 'information-rich' individuals or groups or behaviours to elicit the deepest insights into the research questions.

The choice of a purposive sampling method for the research project influenced the decision to approach a local general hospital in East London. The local hospital had an established sickle cell service in which I worked for more than four years, albeit in a non-dietetic capacity. Through this liaison, I was able to access suitable participants, including both SCSU-Cs and SCSPs. It also gave me immediate access to potential gatekeepers.

Sample size:

The sample size of the research participants was not only determined by the sampling method used but also dependent on the research methods used in the research enquiry, as well as the nature and type of data to be collected. The sample sizes were based on the average sample size for conducting focus groups, which are usually set between 8–10

participants (Robson, 2011), although they can also be as small as 2 participants (Leueng and Savithiri, 2009).

The sample size set was aimed at addressing both Objectives 1 and 2 of the research project. The sample sizes set for the four phases of the research project included the following:

- Phase 1 (independent SCSU-C and SCSP focus groups, n=10)
- Phase 2 (formation of the LAN, n=8)
- Phase 3 (co-development of the health literacy framework, n=8)
- Phase 4 (evaluation questionnaire, n=8).

The LAN meetings, sample size (n=8) was designed to reflect an equal number of both SCSU-Cs and SCSPs, to promote equal representation. However, to address the second part of Objective 2, which was the evaluation of the health literacy framework, and to simplify the recruitment process, the sample size was eventually determined by the size of the LAN, at the time.

Of note, the SCSP sample pool, included a range of different Health and Social Care professionals to reflect the range of service provision required in the management of people living with SCD. These professionals included the following: a SCD specialist paediatric nurse, a paediatric Dietitian, a community-based adult Dietitian, a SCD community engagement officer, a SCD social liaison officer, a nutritionist and a GP.

3.2.2 Inclusion and exclusion criteria:

To support the recruitment of the most suitable participants, explicit inclusion and exclusion criteria were selected and included in the participant information sheet (Appendix 1). The inclusion criteria for the SCSU-Cs included:

- any adult over the age of 18 with SCD, with or without responsibilities for children and adolescents with SCD, including adults or carers without SCD who have responsibilities for children or adolescents with SCD.

The inclusion criteria for the SCSPs included:

- any professional who provided health and social care services to people living with SCD and those professionals who come into contact with people living with SCD.

This excluded from the study children and adolescents with SCD and those professionals who did not provide any services to people living with SCD.

Whilst a range of SCSPs were included in the study to reflect the wide range of services provided to support the management of people living with SCD (Okpala, 2004), specific characteristics for SCSU-Cs such as employment status, qualifications and age, were not collected to promote inclusion. However, the data collection allowed for the participants to reflect the impact of these factors in relation to their knowledge, views and experiences of the medical and nutritional management of SCD.

The inclusion of only adults in the study may limit the scope of hearing directly from the children and adolescents living with SCD; however, the voice of the children and adolescents living with SCD would be represented by the adults living with SCD and the carers of affected children and adolescents. It is thought that the adults would be in a prime position to give an account of the nutritional needs and habits of the children and adolescents living with SCD and those in their care.

3.2.3 Recruitment of Participants:

(i) Recruitment process:

The recruitment process for the research project commenced after full ethical approval was obtained in compliance with the Ethics Regulations of (Anglia Ruskin University (ARU), 2016). The original ethics approval for the project was granted in September 2019, by the ARU School Research Ethics Panel (SREP) – (Appendix 2). A first of three amendments to the ethics approval was granted in January 2020 (Appendix 3), following which the awareness and engagement activities for the project could commence to support recruitment. Gatekeepers were informed and active recruitment was set in motion to recruit suitable participants for the study to include both SCSUs and SCSPs. The recruitment of the independent participant groups, however, proved to be very complex. Originally, the service user participant pool only included adults with SCD, however, after slow uptake in the recruitment process, and an ethics approval amendment (March 20 -Appendix 3), carers of children with SCD were also included in the participant pool. The service user participants are therefore called Sick Cell Service User and Carers (SCSU-Cs).

A third amendment to the ethics approval was submitted, requiring a decision via Chair's action (Appendix 5), directly after the first face-to-face focus group occurred, due to the national implementation of lockdown measures in March 2020 that restricted physical gatherings. The third amendment requested a change from face-to-face meetings to video-facilitated group meetings (using Zoom as the chosen video communication support tool with added recording option) to facilitate the verbatim transcription of the discussions during the focus groups. Once all the ethics approvals (with amendments) were in place, the recruitment process for phase one data collection could commence and continue to conduct the independent focus groups.

Awareness and engagement:

Poor knowledge and awareness of nutrition in SCD (SCS, 2018) amplified the need to raise awareness of the research project as widely as possible. Therefore, in addition to the liaison with the Sickle Cell Board and the sickle cell service of the local East London-based hospital, a range of recruitment engagement activities were planned and advertised to raise awareness of the research project (see Appendix 6). A meeting was organised by the SCD community engagement officer with a member of the local borough council, providing a further opportunity to raise awareness and support for the research project. The local borough council member agreed to provide the venues used in the borough, free of charge.

Gatekeeper involvement:

The purpose of the gatekeepers was to identify and approach eligible SCSU-Cs in their networks and provide them with an overview of the research project. Within qualitative research projects, gatekeepers are used to build trust by vouching for the researcher to support the recruitment process (Devers and Frankel, 2000). The lead nurse for SCD, a Clinical Commissioning Group (CCG) commissioner for long term conditions and the chairperson of the local SCD Board were examples of gatekeepers who were consulted.

The SCD Board, commissioned by the local CCG, operated in collaboration with the members of the local hospital and SCD service. Through the liaison with the SCD Board, I was able to have direct access to SCSPs, including the hospital based SCD consultant, who was helpful in allowing me the opportunity to attend a SCD-MDT meeting to share information about the research project to support the recruitment of prospective participants. In addition, liaison with the SCD board enabled access to the Chair of the local patient support group, who connected me with other support groups in the surrounding areas. The ongoing liaison with the SCD Board steering meeting provided valuable buy-in and support for the research project from a variety of stakeholders within the local SCD service provision.

Although gatekeepers were selected from within the NHS and related organisations, data collection sites were held in non-NHS locations to include people living with SCD who were not inpatients in the hospital but those who were living in the community.

(ii) Research Participants – Recruitment:

Interested individuals who were identified by the gatekeepers were asked to email a designated email address or call a designated telephone number to confirm their interest and participation in the research project. In response to each confirmatory email, a copy of the study participant information sheet (Appendix 1) and consent forms (Appendix 7) were emailed to prospective participants. During the initial email contact with the prospective participants, they had the opportunity to ask any further questions about the study and the research project; dates and timelines were also confirmed. Due to the switch from face-to-face data collection to online data collection – and to promote voluntary participation – final consent was obtained via email before the data collection took place.

The recruitment of SCSU-Cs was challenging for a variety of reasons, notably because of the surge in COVID 19 infection rates and people being fearful of becoming unwell (APPG-SCAT, 2020). Additionally, the data collection was scheduled in community venues and not on the hospital's premises, which patients may have expected. As a result, barriers included low levels of trust on the part of the SCSU-Cs, a lack of incentives or the required time commitment for the duration of the research project. Together, these challenges may have affected people's choice to participate in the study.

To circumvent the slow recruitment to the study, I was advised by the SCD consultant from the East London based hospital, to liaise with a SCSU, who worked in conjunction with the Sickle Cell Society. This liaison, which involved speaking with the SCSU to inform them of the study and the problems experienced with recruiting suitable SCSU-C participants, resulted in them personally reaching out to their network of SCSU-Cs encouraging them to

consider participating in the study. The SCSU was happy to participate in the study, and in effect, vouched for me as the researcher and the value of the research project, resulting in an increase in interested participants who emailed me to confirm their willingness to participate in the study, to commence phase one data collection.

Focus group participants – phase one:

The recruitment levels for SCSU-Cs, increased dramatically midway through March 2020, following on from the liaison with the SCSU, recommended by the East London hospital based SCD consultant. Following full ethical approvals, recruitment progressed well.

Table 3.1 below lists the characteristics of both the SCSU-C and SCSP participants. On the day of the SCSU-C focus group, the sample size for the SCSU-Cs (n=10) was exceeded, as indicated in Table 3.2, with a final sample size of n=11.

With a change in data collection from face-to-face to online meetings, the scheduled focus group for the SCSU-Cs was moved from a weekday to a Saturday morning (28/03/2020). The choice of a Saturday helped to ensure a high attendance rate (n=11), all of whom consented before attending the focus group.

By middle of March (2020), just before the national lockdown measures were in place, participants in the SCSP group were recruited, as indicated below, in Table 3.2. Recruitment followed the same format as described for the SCSU-Cs, with interested participants self-selecting onto the research project. The full recruitment of the SCSPs was delayed due to the impact of COVID-19 and staff being redeployed from community services to acute services, to address the increase in hospital admissions of people with COVID 19 infections. As a result, the first scheduled data collection date (19th March 2020) for the face-to-face SCSP focus group only had three participants (a SCD social liaison officer, a SCD community engagement officer and a SCD specialist paediatric nurse). Following a successful amendment to the ethics approval (see Appendix 3), in response to the curtailment of face-to-face focus groups, two further smaller online focus group meetings

were scheduled for the remaining SCSP participants (n=4). The remaining two focus groups took place as follows:

- 31/03/2020: n=2 [a GP and a nutritionist (the latter is also a SCSU)]
- 21/04/2020: n=2 [a paediatric Dietitian and a community based adult Dietitian]

Taken together, a total of three focus groups were held for the SCSP pool.

Table 3.1: Characteristics of the independent focus group participants

Participant Group	n = No of Males	n = No of Females	n = Group Size	Characteristics
SCSU –C	2	9	11	N/A
SCSP	2	5	7	*Adult community Dietitian Paediatric Dietitian Nutritionist Specialist Paediatric Sickle Cell Nurse Sickle Cell Community Engagement officer GP/Chair of CCG Sickle Cell Board

*The range of SCSP – recruited to the study

Learning Alliance Network (LAN) participants – phase two:

The recruitment of the members of the LAN, followed on from the SCSU-Cs and SCSPs who participated in the focus groups in phase one data collection, as mentioned above. To ensure the LAN reflected the voices and perspectives of both participant groups equally, I selected the following participants, in a non-random way, using a purposive sampling technique as mentioned in 3.2.1. Below in Table 3.2, the characteristics of the joint members of the LAN are illustrated to ensure an equal representation of both participant groups in order to support a consensus perspective about the main phenomenon, which is the influencing factors affecting the integration of nutrition in SCD (research question 1, see chapter 1, section 1.2) to support the co-development of the health literacy framework. Eventually, there were two participant withdrawals from this phase of the study, due to personal reasons.

Table 3.2: Characteristics of the participants of the Learning Alliance (LA) Network

Participant Group	n = No of Males	n = No of Female	n = Group Size	Characteristics
SCSU	2	2	4	Sickle cell service users
SCSP	2	2	4	Community based Adult Dietitian Paediatric Dietitian Sickle Cell liaison officer Paediatric Sickle Cell Nurse Specialist
Learning Alliance Network (LAN)	4	4	8	Two withdrawals from the sickle cell provider group: Phase one – Paediatric nurse specialist Phase three – Sickle Cell Liaison Officer

3.3 Research data collection methods and processes

Overview of the four phases of the LA Methodology:

The research project has been designed as a qualitative study, using a four phased sequential participatory LA methodology. Figure 3.1 below illustrates the series of data collection methods (phase one and phase four), with respective thematic data analysis, including a sequence of three LA network meetings (phase two, three and four) processes, to collectively achieve the main aim and objectives of this research project. The sequential nature of the LA methodology is geared towards each phase building on and reliant on the action from the preceding phases. Below is a breakdown of the main aims of each of the four phases that make up the LA Methodology:

Phase one: A data collection phase using independent focus groups including SCSU-Cs and SCSPs participants respectively, with each group aimed at identifying the knowledge, views and experiences of the medical, nutritional and socio-ecological factors and facilitators affecting nutrition integration in SCD.

Following the completion of the data collection from each of the respective independent focus groups, the data will be analysed collectively using a thematic data analysis method and a summary of the analysed data is produced in preparation of phase two (Appendix 9).

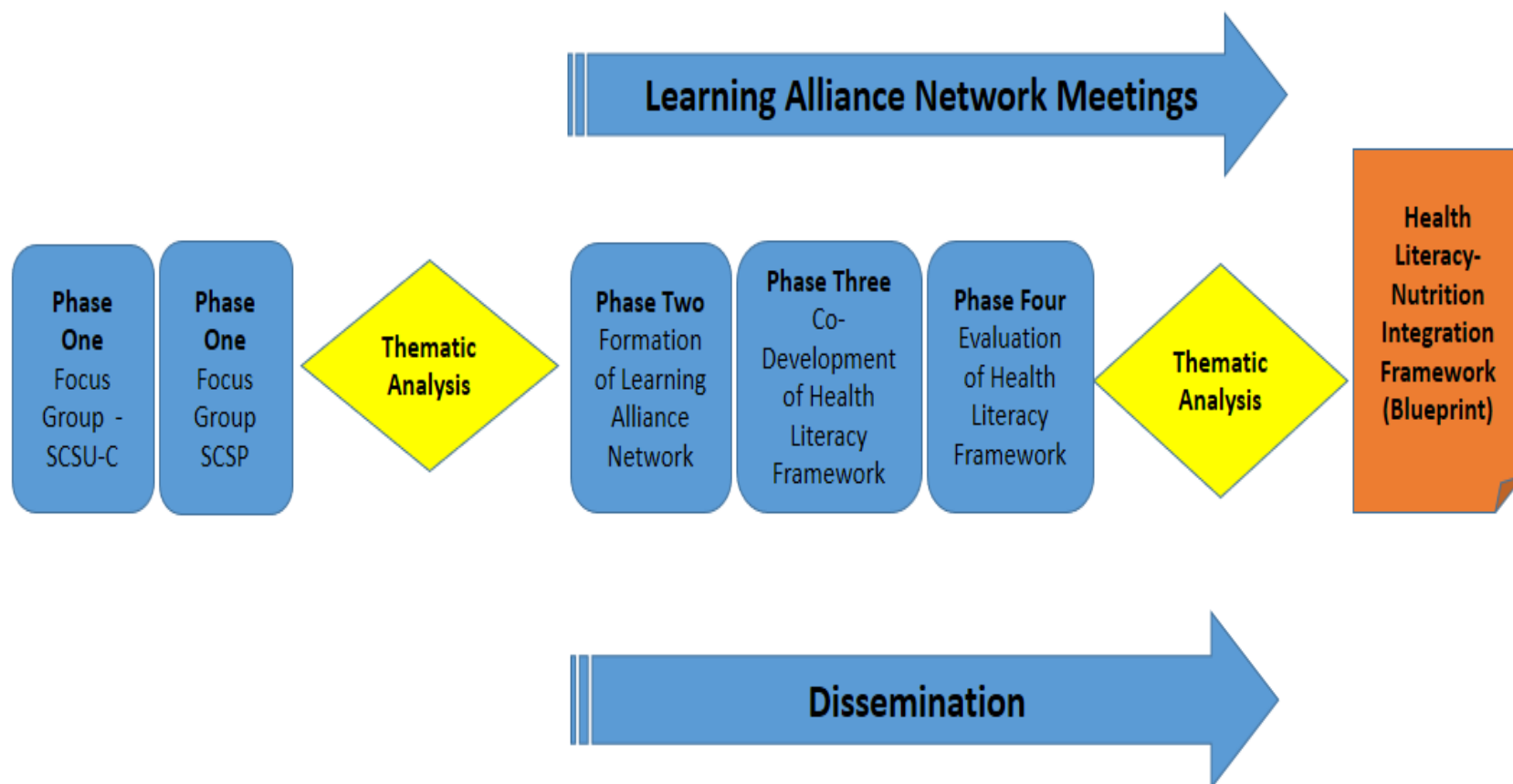
Phase two: The main aim of this first LA network meeting is the formation of the LAN between the SCSU-C and SCSP participants, who together, will discuss the summary of the data analysed in phase one, to reach a consensus viewpoint on the data summary (Appendix 9) to be used in phase three.

Phase three: The main aim of this second LA network meeting is to co-develop the study intervention, the health literacy framework, using the data summary agreed on in phase two – reflecting any additional points raised by the members of the LAN in phase two and three as coordinated by the researcher, that is different from what is already previously collected.

Phase four: The aim of this phase is two-fold, firstly to evaluate the co-developed health literacy framework by the members of the LAN, using an evaluation questionnaire (Appendix 12) as the data collection method, followed by the thematic analysis of the data collected. Secondly, to hold a third LA network meeting to disseminate the initial findings of the research study to study participants, a key part of the LA methodology.

Each of the four phases, thematic data analysis and the three LA network workings will be explored in more detail below.

Figure 3.1 The Four Phase Research Process



3.3.1 Phase one – Conducting Independent focus groups:

Phase one is the main data collection phase using independent focus groups including SCSU-Cs and SCSPs participants respectively, with each group aimed at identifying the knowledge, views and experiences of the medical, nutritional and socio-ecological factors and facilitators affecting nutrition integration in SCD.

(i) Rationale for focus groups:

The value and use of independent focus groups are important observations and recommendations by (Moreno-Meguizamo et al., 2015) as a first step when using the LA methodology in Health and Social Care. The use of independent focus groups enabled me as the researcher to gauge both the independent and collective knowledge, views and experiences of the SCSU-C and SCSP groups. Not only did the independent focus group facilitate knowledge co-production and sharing, but it enabled me to elicit any underlining problems that may have been missed in a mixed group discussion, e.g. because participants may not feel free to speak. In this way, the independent focus groups were a critical first step into facilitating the co-development of the health literacy framework, which is the outcome of Objective 2 (section 1.2) of this research project.

The use of focus groups as part of the research project therefore facilitated the sharing, acquiring and contesting of knowledge (Luheux, Poland and Daudelin, 2006), which are key principles underpinning both Freire's (1970) dialogic approach to education and the knowledge co-production and sharing inherent to the LA methodology: the focus groups facilitating 'social interaction' to elicit rich communication and discussions, helping me to gauge a wider understanding of the research topic.

Although the focus groups were appropriate as a data collection method, the volume of data generated from the four focus group meetings added to the complexity of the research process (Leueng and Savithiri, 2009). On the other hand, it also provided a richness and deeper insight into the question topics explored (Appendix 9) which was valuable to support the co-development of the health literacy framework. Conducting independent focus groups, allowed the participants to speak freely, which allowed me to gain a richer insight into the question topics from the participant's perspectives, but also into aspects such as the impact of COVID 19 and the scepticism it elicited from some of the participants, in both the SCSU-C and SCSP focus groups. Moreover, the focus group discussions allowed the participants to clarify their contributions and take the opportunity to explain, disagree and share their true views with their fellow participants.

Hence conducting independent focus groups were appropriate for the research project; however, allowances had to be made for the online focus groups in regards to managing potential technical challenges, internet connectivity which posed a problem during the SCSU-C focus groups and the 1st online SCSP focus group meeting.

An important consideration, however, was how to overcome the lack of generalisability of qualitative data, which I compensated for through Objective 2: the co-development of the health literacy framework, which serves as tangible knowledge base to inform future policy and practice development.

(ii) Development of the focus group question schedule:

The question schedule would not be complete without first assessing the participant's knowledge and understanding of SCD management, as poor knowledge of SCD, by service users, service providers and stakeholders were previously reported as a problem (SCS, 2018).

Phase one of this research project, i.e. to conduct the independent focus groups involving the SCSU-Cs (n=11) and the SCSPs (n=7), as illustrated above in Figure 3.1, commenced with the design of the question schedules for both the SCSU-C and SCSP focus groups (see Appendix 8). The design of the questionnaire was scheduled ahead of time, as it is essential to collect high quality data from the participant's experiences, knowledge, attitudes and views (Hyden and Bulow, 2003, p 308; Lueng, Poland and Daudelin, 2006). The effective design of the focus group question schedule was based on the main focus of Objective 1, as described in section 3.1.1 and Chapter 1, (to identify and understand the influencing factors affecting nutritional management and service provision in SCD). The design of the question schedule enabled the simplification of the formatting of the question schedule, into four main question categories (and corresponding questions, see Appendix 8) which included:

- Knowledge of SCD management - (Question (Q) 1
- Knowledge of nutritional management (optimum nutrition) – Q 2 to Q 5
- Influencing socio-ecological factors affecting nutrition and service provision – Q 6 to Q 9
- Facilitators to address the socio-ecological factors – Q 10.

The question schedule was designed as a structured approach using predetermined open-ended questions (Robson, 2011) to allow for a more standardised approach to conducting the focus groups. There were minimal differences to the questions dependent on whether it was the SCSU-C group or SCSP focus group. Moreover, the focus group question schedule was developed to collect data that could answer research question 1 and address Objective 1 (see chapter 1, section 1.2) of this research project, as comprehensively as possible.

(iii) Focus group data collection:

With the exception of the first focus group held with the SCSP (n=3), which was held face to face, all the online focus groups, both SCSU-C and SCSP groups followed the same

process to maintain uniformity, as outlined in (Appendix 16). However, due to the size of the SCSU-C focus group (n=11), compared to the two smaller SCSP focus groups, n=2 respectively, a facilitator (who was an SCSU, with prior research experience) was identified from the SCSU-C group, at the start of the focus group. The purpose of the facilitator was to help identify participants who wanted to contribute to the conversations and to prevent interruptions in the level of engagement of the participants. During all the online focus groups, the participants could also use the chat function to make further comments or ask questions, whilst participants were speaking.

Throughout the focus groups, I asked follow-up questions to help clarify the points made by the participants, as well as to ensure that the participants understood the questions explicitly. Each question's answer was then summarised by myself to take into account all participant views, at which point participants had an additional opportunity to add to the answers as appropriate. In this way, the participant engagement facilitated a social constructionist epistemology throughout the focus group data collection process. However, time management is an important consideration when hosting larger focus groups online, such as the SCSU-C group (n=11), to ensure all the questions are asked and answered fully. A lack of time, meant that one question in the SCSU-C, was not answered, leaving a gap in the data collection (compensated for, by the comments of the members of the LAN, in phase two – see Appendix 15).

The breakdown of each of the SCSU-C and SCSP focus group meetings are illustrated below:

- Part one of the SCSU-C group (n=11) was held online (via zoom) and lasted one and half hours, whilst part two lasted one hour.
- The first of three SCSP groups (n=3) was held face to face and lasted two hours.
- Both the second and third SCSP groups (n=2) were held online (via Zoom) and each lasted one and a half hours.

3.3.2 Analysis of Independent focus groups data:

(i) Rationale for thematic analysis:

Thematic analysis according to Boyatzis (1998) bridges the gap between quantitative and qualitative research. In addition, the process of thematic analysis is about (working with and making sense of the data) and as a method, collectively made up of values, assumptions and practices (Braun and Clark, 2006). In this way, as the researcher I play an active role in the analysis process, through an in depth engagement with the data. In this way, thematic analysis supports a constructionist epistemology which is relevant to the research project, as explained above. Thus, I have used thematic analysis as a systematic and subjective process (bias) as I seek to answer the theoretical and conceptual (why) and (how) of the data to be analysed to answer the study questions, as the researcher.

Reflexive thematic analysis (Braun and Clarke, 2019, 2022) is appropriate and relevant as an analysis method for the research project as it fosters a constructionist epistemology and recognises the role of the reflexivity of both the researcher and the research participants in the analysis process to achieve co-development. In this way, reflexive thematic analysis, allowed me as the researcher to draw on my own lived experiences of marginalisation, to adopt a corresponding philosophical paradigm, during the analysis process of the research project. In this way, I was placed in a central role in the analysis process, however, adopting an ethical approach acknowledging the power, knowledge and struggles of the researcher and the participants in the data analysis process. Moreover, adopting a reflexive approach to the thematic analysis allowed me as the researcher to tell a story through the data analysis and the active development of codes and themes. As such, reflexive thematic analysis considers the impact of the themes of the research questions and the research problem, through an active process (Braun and Clarke, 2019, 2022).

This permitted me to fully immerse in the data sets accommodating the need to go back and forth and revisit the data set as new nuances and interpretations developed during and after the analysis process. The ability to go back and forth, is a key part of the reflexive thematic analysis. This flexibility allowed me to change initial interpretations but moreover, enabled me to further analyse the data to espouse health literacy thematic themes from the original themes of phase one (See chapter 4, section 5.4) to develop the health literacy nutrition integration conceptual framework and the Health Literacy – Nutrition Integration Framework (HL-NIF) blueprint. The use of both semantic and latent themes enabled me to answer research question 1 and achieve Objective 1 of the research project (see chapter 1, section 1.2), without detracting from the quality of the analysis process. Thus, thematic analysis therefore aligns well with the purpose of the LA methodology, which fundamentally promotes joint learning (constructionist approach) to understand problems from the perspective of the stakeholders involved in the LA to achieve the goals of the LAN.

(ii) Focus group data analysis process:

Phase one: Data familiarisation

This enabled me to:

- identify initial ideas and insights
- highlight key quotes
- gather initial interpretations/notes
- gain an understanding of the whole dataset.

Phase two: Coding

This allowed me to collate the initial interpretations/notes/ideas relevant to answer research question one/Objective 1, using the following process:

- transferred all the information onto an Excel spreadsheet (see an Extracts 1-3 Appendix 17)

- recorded the information into designated columns to reflect the initial code categories/interpretations/ideas and quotes
- recorded all the quotes separating the SCSU-C quotes from the SCSP quotes
- added an additional column for relevant key literature sources.

Phase three: Generating initial themes

This allowed me to:

- condense the initial code categories before developing the themes
- collate the quotes to support the initial condensed code categories
- actively search and develop the possible themes inherent in the initial code categories from the quotes to answer research question one and Objective 1
- add a separate column to the Excel spreadsheet to list the content to be used for phase two and three from the original data collected in the focus groups contained in the quotes

Phase four: Developing and reviewing themes

Actions taken included:

- check that the themes made sense and fit the initial code categories in relation to the research question and objective to refine the codes
- check that the quotes fit the initial themes developed and added any other relevant quotes as appropriate
- check for the central organising concepts of each theme, highlighting shared perspectives and differences in perspectives between the two participant groups in the quotes used to support the initial themes developed
- update and cross-check quotes included in the spreadsheet listing the content to be used in phase two and three to reflect the changes to the themes and quotes supporting the themes

Phase five: Defining, refining and naming themes

The defining process of the theme development and analysis, remained unchanged at this stage and was able to support the content to be used in phase two and three of the research project. The themes were able to reflect the perspectives of both the SCSU-C and SCSP of the current nutrition landscape in SCD summarising the multi-level influencing factors affecting nutrition integration in SCD.

Phase six: Writing up

The final step of the process was to:

- summarise the main themes and supporting sub-themes (quotes)
- identify differences (see chapter 4, section 4.2.1 and 4.2.2) or similarities (chapter 4, section 4.2.3) or unexpected findings adding to the richness of the analysis and overall findings
- overall, the themes answered research question one and Objective 1 (see chapter 4) to provide the content to support the co-development of the health literacy framework.

A total of four main themes were developed from the data analysis process. The four main themes, as listed below in Table 3.3, provided a summary of the influencing factors – reflecting the main knowledge and care gaps affecting the integration of nutrition into standard care in SCD. The four themes were developed from the active data analysis process to identify the central organising concepts/codes based on the views of both the SCSU-Cs and SCSPs in response to the question schedule used in phase one (Appendix 9).

Table 3.3: Summary of the four themes of the focus groups in phase one:

Question Category	Theme
Knowledge of SCD	1. Invisibility of SCD
Knowledge of nutritional management (Optimum Nutrition)	2. Under-recognised importance of nutrition 3. Lack of priority to nutrition
Influencing socio-ecological factors and facilitators	4. Multi-level factors affecting nutrition and service provision

(iii) Trustworthiness of the data:

The trustworthiness of the data is discussed below in the context of the assumptions I have about the current nutrition landscape in SCD. Of significance, is the fact that despite working as a Dietitian in clinical practice prior to commencing this research project, I did not have any clinical dietetic experience working with patients living with SCD. However, the assumptions I held were influenced by a number of factors including the following;

- the multitude of invitations to speak about nutrition in SCD, from both healthcare professionals in SCD and people living with the condition,
- the existing scientific literature on the role of nutrition in SCD and its associated risks to SCD patients and
- my professional knowledge, skills and experience of working as a Dietitian in a range of other long term conditions, where nutrition played a paramount role in standard care provision.

Based on the above, the assumptions I have about nutrition in SCD is grounded in the following;

- that the nutrition needs of people living with SCD are real,
- nutrition is important to improve the health and wellbeing outcomes of people living with SCD,
- SCD has both medical and nutritional implications,
- a lack of nutrition service provision in SCD exists and
- SCD is a marginalised condition.

Thus, despite having my own assumptions about the current nutritional management landscape in SCD, what was missing was the first-hand dietetic practice experience of working with this patient population. Hence the data collected, would be the first direct data based on clinical practice and experiences of both SCSU-Cs and SCSPs of nutritional management in SCD.

Furthermore, being a qualitative study, my positionality as the researcher, as discussed in Chapter 1, section 1.4, cannot be removed from how the study is designed and how the data is collected or analysed. As such, my own lived experience of marginalisation and oppression coupled with my professional knowledge, skills and practice working as a Dietitian, cannot be ignored as a potential source of bias, or how this has shaped my perspectives and the values I hold about the research project. For this reason, the decision making processes throughout the research project, has been influenced and guided by the lens through which I understand the research problem (the lack of nutrition service provision in SCD), the study population, being from a marginalised background, and the inherent social injustice, lack of advocacy and inequality that defines the current lack of nutrition service provision in SCD.

However, to circumvent the inherent bias of my positionality as the researcher and maintain trustworthiness of the data, the seminal framework by Lincoln and Guba (1985), is

considered in respect to the theoretical, methodological and analytic processes used in the research project. The framework consists of four distinct components namely credibility, transferability, dependability and confirmability, to be discussed below.

Credibility, the first of the four components in the trustworthiness framework (Lincoln and Guba, 1985) relates to how well researchers are able to persuade themselves and their readers of the worth of the research findings (Nowell et al., 2017). Lincoln and Guba (1985), speaks of prolonged engagement and persistent observations and data collection triangulation as evidence of credibility in the data. To this end, I chose to include both SCSU-Cs and SCSPs, knowledge, views and experiences to understand the current nutritional management and the socio-ecological factors and facilitators affecting nutrition and service provision in SCD. By holding independent focus groups as the main data collection methods, there was an opportunity for the respective participants to speak freely about the problems and their experiences of nutrition in SCD, and provided me as the researcher with an independent perspective of the findings thus presenting the opportunity to assess the similarities and the distinctions within the data collected between the two participant groups. This ability to identify similarities and distinctions in the data, promotes transferability of the data, which is a second component of establishing trustworthiness according to Lincoln and Guba (1985). More specifically, transferability speaks to the generalisability of the enquiry which was possible since both SCSU-Cs and SCSPs were included in the data collection process, providing me the opportunity to extract the similarities and distinctions in the data analysis, which is reported in Chapter 4, section 4.3.

Moreover, the LA methodology, more specifically the formation of the LAN in phase two, supported prolonged engagement with the participants as there was now an opportunity for both participant groups to learn more about their respective knowledge, views and experiences of nutrition management in SCD, but equally have the opportunity as members of the LAN, to discuss, clarify and add to the data already collected, promoting prolonged engagement with the data, thus more opportunity to verify the data amongst the participants.

This prolonged engagement by the LAN, also enabled me as the researcher to check the data collected from the respective participant group perspectives and the acceptance of the data collected by the independent participant groups. Inadvertently, the prolonged engagement with the data supports the dependability of the data, a third component of the trustworthiness framework (Lincoln and Guba, 1985). Dependability, demonstrates that the findings are consistent, and this was evident in the discussions of the members of the LAN, in phase two and phase three, the co-development of the health literacy framework respectively. In addition, the LAN meetings also provided opportunity for the members of the LAN to contest the data collected or call out any discrepancies which may have emerged in the independent focus groups.

The value of the ongoing LAN meetings, leads on to confirmability of the data, the fourth component of the trustworthy framework (Lincoln and Guba, 1985). Confirmability of the data collected relates to the degree of neutrality or objectivity of the data, evident in the prolonged engagement of the data by the members of the LAN, throughout the various phases of the research project. This prolonged engagement by the LAN, further reduced the opportunity for researcher bias in the data and thus establishing the researcher's interpretations of the findings, as it relates to the data collected in phase one. Moreover, Novell et al., (2017) highlight the importance of an audit trail of the data collection and analysis to underpin decisions and choices of the researcher, and as such, I have saved the original transcripts of each of the four focus groups, provided evidence of the excel spreadsheets used in Appendix 9, and have exercised reflexivity throughout the data collection processes and analysis, which has been influenced by the assumptions I had about the research problem and nutritional management in SCD, as mentioned above.

Therefore, as a marginalised group, the lack of nutrition service provision available to people living with SCD can be considered a health inequality impacting on the experience, access and outcomes of this patient population, and choosing a methodology that is suited to research conducted in marginalised groups, such as the LA methodology was of paramount

importance, to generate data that accurately reflects the voice of the participants, mirroring the social injustice, lack of advocacy and inequality in nutrition service provision in SCD, a benefit of the LA methodology in conducting research with marginalised groups.

In this way, the study is the first opportunity to generate concrete evidence on the scope of the problem of the lack of nutrition in SCD management. For this reason, acknowledging my positionality as the researcher with regards to my professional and personal lived experience and how this may have influenced the analysis of the data, the validity and accuracy of my interpretations to reflect the research problem. Acknowledging and mitigating for the potential bias of my positionality, furthermore, underpins the quality and trustworthiness of the data and its contribution to knowledge. Moreover, this transparency contributes to the credibility of the data to support policy and practice development in support of the integration of nutrition in SCD, the main aim of the research project.

3.3.3 Phase two: First Learning Alliance (LA) network meeting

Phase two is predominantly a process step of the LA methodology to facilitate the formation of the Learning Alliance Network (LAN) between the SCSU-C and SCSP participants recruited for this part of the study. During this process, the first of three LA network meetings, the main aim is for the newly formed LAN to discuss the summary of the data analysed in phase one, add any additional points raised during the LA network meeting, to eventually reach a consensus viewpoint on the data that will be used in phase three.

(i) Formation of the LAN:

The LAN is the partnership and collaboration between the SCSU-Cs and SCSPs. The formation of the LAN is what distinguishes the LA methodology from action research and is what supports the ongoing work towards practice and policy development (health literacy

framework), (Moreno-Leguizamon et al., 2015), a key output of this research project.

Participation in the LAN was open to all the participants and a purposive sampling approach was used to identify equal representation (n=4) from each of the SCSU –C and SCSP participant groups; to include (n=4 males – two from each participant group) and (n=4 females – two from each participant group), see Table 3.2 in section 3.2.3. In this way, there remained a balance between the two groups to allow for the co-development and sharing of knowledge and, ultimately, the co-development of the study intervention - the health literacy framework. On the day of the first LAN meeting, all the prospective participants were consented before the network meeting commenced to check that they were still willing to participate in the research project. A summary of the agenda set and used during the LAN meeting is provided in Appendix 16.

(ii) Consensus development:

The purpose of phase two was to agree on the main findings of the analysis of phase one data collection (see Figure 3.1 above). The content extracted from the comments, which encompassed the four themes of Phase 1 data collection (Table 3.3 above), were presented to the LAN in this first of three LAN meetings. Appendix 9 – a complete copy of the data summary - was emailed ahead of the first network meeting and shared via screen share function on Zoom, as a visual support during the LAN meeting.

To support the development of a consensus viewpoint, the content of each of the questions asked as part of the phase one question schedule (Appendix 8) was shared with the members of the LAN. Summaries of the data collected, including Themes raised, from both participant groups were shared during the meetings, so each participant group could get a sense of the main points raised from both participant group perspectives (see example extract in Appendix 9). In particular, the LAN was informed of the question that was not fully answered during the SCSU-C focus group, giving those participants a chance to add to the data collected by the SCSPs. The SCSU-Cs, commented positively on the points raised by

the SCSPs, as they recognised the value of the need for SCSPs to have an appropriate level of knowledge about SCD to manage or treat patients/carers effectively. In this way, every member of the LAN was given the opportunity to understand the content shared and ask clarifying questions, adding to what was already presented to achieve a more consensus viewpoint.

For each question, members of the LAN were allowed to add anything else, in addition to what was in the Excel spreadsheet, and further discussion enabled. This process was followed to agree on the content for each of the 10 questions asked (Appendix 8); additional points raised by the members of the LAN were also collated (Appendix 15). The LAN was therefore able to agree on a joint vision for the content to be used to co-develop the health literacy framework. Before the close of the first LA network meeting, I summarised the additional data collected from the members of the LAN, to ensure all the points were captured and double checked that the members of the LAN were happy with the consensus view reached during the network meeting. The date was agreed for the next network meeting before I thanked the participants for their time and their valuable participation and contribution.

3.3.4 Phase three: Second LA network meeting:

Phase three, the second LA network meeting is predominantly a process whereby the members of the LAN work in partnership to co-develop the study intervention, the health literacy framework, using the data summary agreed in phase two – including any additional points raised during the LA networking meeting discussions, co-ordinated by the researcher.

(i) Co-development of the health literacy framework:

The shared consensus viewpoint of the LAN, established in phase two, informed the co-development of the health literacy framework in phase three. As Butterworth et al., (2011) points out, the network promotes knowledge innovation – knowledge generated from the focus groups and it's scaling up in time (sustainability), important factors informing policy and practice development. Policy and practice guidance will help towards addressing the effects of the invisibility of SCD and nutrition, necessitating the development of tailored policy and practice based on the data collected in phases one and two.

(ii) Co-development process:

The second network meeting followed a similar pattern to the first network meeting. The members of the LAN were consented before the network meeting to establish their ongoing interest in the research project. Similarly, to the first network meeting, each member of the LAN was sent a digital copy of the Excel spreadsheet agreed on in the first network meeting.

Unlike in the first network meeting, I allocated each of the remaining members to smaller groups, each comprising of an SCSU and SCSP pair.

Each pair was allocated 30 minutes in a Zoom breakout room to decide on the main points they wanted included in the health literacy framework, before returning to the main meeting group to discuss each of their points with the whole LAN. A short discussion by the whole LAN followed once all the pairs had a chance to share their main points to be included. Extra time was allowed for members of the LAN to add any other further points which may not have previously been mentioned in the preceding phases. Each of the pairs made notes of their main points and this was sent to me to add to the final draft of the data for inclusion in the co-development of the health literacy framework (see Appendix 11). Before the end of the second network meeting, I summarised the main points raised during the network meeting and checked if there was anything else to be added, before thanking the LAN for

their time and valuable contributions. The date of the third network meeting was agreed at the end of the meeting.

3.3.5 Phase four: Third LA network meeting

Phase four is a process with two distinct parts. The aim of this phase is two-fold, firstly to evaluate the co-developed health literacy framework by the members of the LAN, using an evaluation questionnaire (Appendix 12) as the data collection method, followed by the thematic analysis of the data collected. Secondly, to hold a third LA network meeting to disseminate the initial findings of the research study to the study participants, a key part of the LA methodology.

Part one of phase four, is to evaluate how well the health literacy framework is able to contribute to policy and practice development to address the following:

- the knowledge and care gaps in nutrition in SCD
- serve as an educational resource to support the development of future educational and training curricula about nutrition in SCD to help organisations tailor nutrition service provision in SCD in the future.

(i) Evaluation of the Health literacy framework:

Evaluation questionnaire design and process:

An evaluation questionnaire (Appendix 12) was designed to establish whether the health literacy framework was able to support the integration of nutrition into standard care in SCD, as well as address the knowledge and care gaps identified in phase one data collection. Below are four respective question categories of the evaluation questionnaire – with the corresponding questions reflecting its aim:

The levels of nutrition knowledge of the members of the LAN (Questions (Q) 1, 2 and 3):

- Q1 - establish the previous nutrition knowledge of the members of the LAN,
- Q2 - establish the current knowledge of the members of the LAN of the role of nutrition in SCD.
- Q3 - establish if there had been a change in the nutrition knowledge of the members of the LAN and what has contributed to the change, if any.

If there was a change in the nutrition knowledge of the LAN since participating in the research project and what has contributed to that change (Q 4, 5 and 6):

- Q4 – determine how the health literacy framework informed the knowledge of the members of the LAN about the role of nutrition in SCD.
- Q5 and 6 - assess how the health literacy framework informed the knowledge of the LAN about the influencing factors affecting nutrition service provision and the facilitators to address these barriers.

How membership of the LAN has impacted on their knowledge of the influencing socio-ecological factors affecting nutrition Q 7:

- Q7 was to determine how valuable the health literacy framework was to improve the knowledge, awareness and understanding of the role of nutrition in SCD.

The value and impact of the health literacy framework as a tool to support the integration of nutrition into standard care in SCD Q 8:

- Q8 was to determine how the health literacy framework can be used to support the integration of nutrition in healthcare provision in SCD, through structure and systems change.

Although the evaluation questionnaire did not directly enquire about the role of the LAN in the co-development of the health literacy framework, the responses to the evaluation questionnaire aimed to provide insights into how well the LA methodology, more specifically

the formation of the LAN, had contributed to the overall achievement of the aim and objectives of the research project (see chapter 1, section 1.2).

Evaluation process:

The evaluation questionnaire was sent to the members of the LAN two weeks after the second network meeting. All six members of the remaining LAN returned a completed questionnaire.

The findings of the evaluation questionnaire were analysed using thematic analysis. In keeping with a qualitative research approach, thematic analysis was appropriate to establish how well the health literacy framework as a tool can address the influencing factors reflecting the knowledge and care gaps in nutrition in SCD to support the integration of nutrition into standard care in SCD, using a LA methodology.

Thematic analysis process:

The analysis of the findings of the evaluation questionnaire was based on the Braun and Clarke (2006) six-phase thematic analysis process, that was later changed to reflexive thematic analysis (Braun and Clarke, 2019, 2022).

The first phase of the analysis process began with reading through the responses to the questions from each member of the LAN, to familiarise myself with the data and gauge the initial insights from the data. Phase two focussed on identifying the initial codes reflecting the developing patterns of meaning resulting from the interpretation of the responses of the participants to answer research question 2 and Objective 2 (see Chapter 1, section 1.2). Phase three continued with the development of the initial themes inherent in the codes from phase two to reflect the shared views of the participants and to establish the central organising concepts from which the themes were developed, in response to the evaluation questions. Phase four was useful in further developing the initial themes, reflecting the central organising concepts informed by the data extracted from the participant responses.

This analysis process, which was appropriate to achieve Objective 2 to determine how effective the health literacy framework is as a tool to support the integration of nutrition into SCD care provision.

Inadvertently, the responses to the evaluation questionnaire also provided insights in support of the value of the LA methodology to promote the development of the health literacy framework.

During phase five and six of the analysis process, the core content of the themes developed in phase three and four of the six-step analysis process remained unchanged, although the descriptions of the themes were refined somewhat. Overall, the themes reflected the reasons underpinning how well the health literacy framework is able to support the integration of nutrition in SCD. The evaluation analysis used mainly semantic codes and phrases based on the number of times a word or phrase was used by the participants about a particular idea or response. A total of four themes were developed through the data analysis process, illustrated below in Table 3.4.

Table 3.4: Summary of the four themes of the evaluation questionnaire in phase four:

Question category	Theme
1. Level of knowledge	1. Improve knowledge and understanding of nutrition
2. Change in nutrition knowledge	2. Raise awareness of influencing factors affecting nutrition
3. Engagement and impact in knowledge development	3. Valuable tool for communication and training
4. Impact and value of the health literacy framework	4. Improve policy and service provision outcomes

(ii) Dissemination of the initial research project findings:

Following on from the evaluation questionnaire, the third and final network meeting was held on the 12/12/20, a month after the analysis of the evaluation questionnaire was completed. The final network meeting was opened to all the participants from the focus groups and the LAN; however, only four of the six members of the remaining LAN could attend and one from the wider participant pool. Despite the low turnout, there was still representation from both participant groups $n=3$ (SCSU) and $n=2$ (SCSP). Having representation from both SCSUs and SCSPs who were part of the study from the beginning, provided an element of credibility to the data collected and consensus viewpoints that informed the co-development of the health literacy framework.

The third network meeting commenced with the same format as the previous two network meetings. All the participants were consented before the network meeting.

The aim of the final network meeting was to disseminate the preliminary findings of all the data collected with the participants and to gauge any other feedback or comments to add to the data already collected. As such, I shared the Excel spreadsheet summarising the main points used to co-develop the health literacy framework (Appendix 9), the main themes from phase one data analysis (Table 3.3), a digital copy of the first draft of the health literacy framework (Appendix 11) and the themes of the evaluation questionnaire (Table 3.4). There were no further comments or questions from the network meeting to add to the data.

However, all the participants present expressed their gratitude for the work I have done as the researcher, towards the development of nutrition service provision. As the researcher, I used the opportunity to express my thanks and gratitude to all the participants for the invaluable contributions to the successful completion of the data collection process for the research project.

3.4 Ethical Perspectives

The ethics application process illuminated several learning opportunities when considering research projects with marginalised groups, to protect the integrity of the participants and compliance with research ethics boards specifications. The commencing of a 6-month intermission period in 2018 meant that I had to reapply for ethics approval and given a new reference number.

Research with marginalised groups and informed consent:

Miller *et al.*, (2012, p62) state that in qualitative research, gatekeepers act as a route of initial access to participants and are thus in a position to permit access to others, as such, they can be perceived as having potential to 'exercise power' over individuals. Since conventional adverts to recruit suitable participants proved to be unsuccessful, I resorted to using gatekeepers to help with recruitment, as mentioned previously (section 3.2.3). In this instance, despite the perceived 'power' gatekeepers may have over participants being included in the study or not, they provided a way to identify suitable participants for the study. To circumvent the potential power imbalance between the gatekeepers and myself as the researcher, all the prospective participants were asked to self-select onto the study via email or a dedicated telephone. Receipt of the email was interpreted by me as the participant's willingness to participate in the study.

Upon receipt of the email, all the prospective participants were provided with a copy of the participant information sheet (Appendix 1), that explicitly stated the aim of the research project and what the expectations for the participants were. They were also sent a copy of the consent form (Appendix 7) with details about the study. The researcher encouraged those participants who self-selected onto the study to ask any questions about the study or about the information included in the participant information sheet. In order to maintain the prospective participants control over participating in the study, the researcher asked all the

participants to complete a consent form on the day of data collection, to ensure they were still willing to participate in the study of their own free will. In this way, the participants were free to change their mind about participating and consent was informed and voluntary, a key part of the ethics process, Social Research Association (SRA, 2003; Robson, 2011).

Confidentiality, anonymity and privacy:

The project involved the inclusion of different groups of people working collaboratively and in partnership with each other, which could jeopardise confidentiality (Grix, 2010). In this way, group-based research amongst participants can identify several issues with confidentiality (Grix, 2010) and, as such, confidentiality was discussed before every data collection event (SRA, 2003). For this reason, the importance of upholding confidentiality was highlighted in the participant information sheet that was provided to all the prospective participants at the point of recruitment. Every effort was made to remind the participants of the need to respect each other and the contributions made by each other to preserve individual confidentiality.

A specific clause was included in the participant information sheet, a recommendation by the chair of the SREP, to remind participants that, although they may withdraw from the study at any time, their contributions in focus groups, may not be removed, to uphold the confidentiality of the other group member contributions. In the same way, anonymity is an important part of the ethical process, which involved the omission of identifiable data including names, and details of conversations had as part of the data collection processes. All the data was stored in accordance with the requirements of the Data Protection Act (1998, 2018). Electronic data (transcriptions, zoom recordings) was stored on a password-controlled personal computer.

Protection from possible harm:

Human research has the potential for harm or emotional upset (SRA, 2003, Miller, 2012). Therefore, the participant information sheet (Appendix 1), contained details of organisations

in the local area where participants could receive further advice and support they may need in the event they were at risk of distress, anxiety or embarrassment. Being a collaborative and partnership-based research project, where participants may or may not know each other, there is a risk that sharing details and personal knowledge and experiences may trigger emotive memories from current or past experiences. These emotive memories can have a detrimental effect on the mental health and wellbeing of the participants and inadvertently impact on the quality and integrity of the data collection (SRA, 2003; Robson, 2011; Miller, 2012).

Dissemination of findings:

Dissemination of the preliminary findings of the research project is an important part of research with marginalised groups. As such, the process of dissemination also adds to the emancipation and empowerment of marginalised groups, providing them with knowledge to increase their level of autonomy and self-determination as well as building a more trusting relationship with the researcher (Robson, 2011). Hence, the preliminary findings of the research project were fed back to the participants throughout the research project.

Information outlining the key data and themes were circulated to participants of the LAN, a demonstration of how their contributions has been used and how it has influenced the potential impact of the overall research project.

Reflexivity, transparency and flexibility:

Considering the use of incentives in research projects is an important element in the ethics application process. Small scale research projects, with limited budgets, can highlight potential hurdles for researchers with regards to the inclusion or omission of incentives.

Particularly when you have a slow recruitment process, with a target population that has low engagement levels, incentives may be needed to support participant recruitment. However, there is a need to consider how the incentives may influence the integrity and quality of the

data collected, which is a fine line for a researcher to juggle. With a limited budget, I was not able to offer any incentives to the potential participants and relied heavily on the gatekeepers to support the recruitment process and identify suitable participants for the research study who would self-select to be part of the research project. Moreover, my values as a researcher are of paramount importance. Not only in data protection, but in controlling for my own bias throughout the research process, accounting for my own reflexivity, which is reflected in my choice of research problem, research strategy, paradigm, methodology and analysis. Therefore, transparency on my part as the researcher was of utmost importance, evidenced in the ethics application planning and process, and how I endeavoured to conduct the entire research project.

Summary:

This chapter provided an overview of the justification for the choice of philosophical and methodological principles underpinning the research project design and related decision making relevant to conducting research with marginalised groups, such as the SCD population. In addition, the value of collaborative and partnership working to promote emancipation and empowerment were key elements demonstrating the appropriateness of the LA methodology, when working with marginalised groups, and how it supported the critical realist constructionist paradigm of the research project.

The main research processes, methods and analysis used to accurately reflect the participant views, knowledge and experiences of the medical and nutritional management of SCD was discussed that need to be aligned with the research paradigms and strategies to achieve the study aim and objectives. The chapter concluded with the ethical considerations that reflected important principles relevant to reduce inequalities and risk of harm, when conducting research with marginalised groups. The next chapter will outline the main results from each of the four phases of the research project.

Chapter 4

Results

Chapter 4 is divided into two parts. Firstly, part one provides background information of the research participants and reports the findings of the thematic analysis of the focus groups conducted in phase one of the research project, including reflections of the Learning Alliance Network (LAN) meetings in phase two, three and four. Secondly, part two reports the findings of the evaluation questionnaire conducted in phase four. Collectively, the findings of part one and part two aim to answer research question 1 and objective 1 and research question 2 and objective 2 (see Chapter 1, section 1.2), respectively. An overview of the health literacy thematic concepts, derived from the four main themes of the focus group analysis of phase one, is provided, which is used to develop the Health Literacy -Nutrition Integration Framework (NIF) blueprint, the main study output.

Part One: Findings of the thematic analysis of the focus groups

4.1 Background of the research participants

This research project is the first attempt at integrating nutrition into standard care in Sickle Cell Disease (SCD), hence the background information of the prospective research participants provided helpful insights to support the credibility of the data collected. To this end, it is helpful to consider the direct and indirect lived experiences of SCD of both participant groups. It is the collective lived experience of the SCSU-C group participants, the indirect lived experience of those participants who care for people with SCD, and the SCD provider knowledge and experience, that places the total participant (SCSU-C and SCSP) pool in a prime position to provide a representable account of the knowledge, views and experiences of the medical and nutritional management and multiple influencing factors affecting nutrition integration in SCD, albeit a smaller sample size.

From table 4.1 below, it is clear that SP-(3) of the SCSP group and the majority of the SCSU-C group, bar one SC-U (10) that has direct lived experience of living with SCD. However, SU-C (10) from the SCSU-C and SP-(4) of the SCSP group have indirect lived experience of living with SCD, as carers of children living with SCD. Moreover, those SCSP's who work with SCSU-Cs additionally provide a different perspective on the lived experiences of the SCSU-Cs they support in their professional roles. This is helpful in understanding the research problem from a wider perspective, that is from both a service provision perspectives (SCSP) as well as from a perspective of those in receipt of care (SCSU-C) and the importance of this provider-service user relationship and how it impacts patient experience and outcomes.

In addition, considering the gender distribution of the two participant groups, Table 4.1 reflects a higher participation rate amongst the females, which is similar in both participant groups, and this may require some attention to observe any potential differences between the responses of the different gender representations. Noting, also the fact that the two males in the SCSP group do not have direct lived experience of SCD, but indirect lived experience as providers of care to those with lived experience of SCD. Similarly, observing the contrast between the lived experiences in relation to seeking care and engaging with care provision between the males and females, further adds an interesting perspective to the data collected and how it can be tailored to the unique needs, risks and challenges of those living with SCD.

Moreover, it is important to understand the differences in care provision and access to care available to patients depending on the locations of service provision and providers, for example if care is provided in community settings or if care is provided in acute settings as this may influence the overall level and quality of the care provision available to those patients accessing the care. In this way it is helpful that amongst the SCSP group, the participants work in both the acute and community care settings adding further insights to

the level of care provision to be considered in the development and provision of nutrition in SCD. An important consideration amongst the SCSP group is to distinguish whether the participants are part of the SCD MDT or if their service is contracted in, meaning they do not work as part of the SCD MDT, as this may influence their knowledge of SCD and possibly the level and quality of care provision.

Thus, the locations of the SCSU-C participants are relevant, and since the gatekeepers who supported the recruitment of the participants were attached to the local East London hospital mentioned in Chapter 3, section 3.2.1, a large majority of the SCSU-Cs were from the wider London area, with a known higher prevalence of SCD. It is noted that one of the SCSP's lived outside of London, as there may be differences in access and care provision and therefore patient experiences of care depending on the location.

As the researcher, it is important to act with transparency upholding rigorous ethical principles to the choice of sampling, recruitment methods use, choice of participants and the quality and type of data collection to achieve the research aim and objectives and answer the respective research questions, as reported in Chapter 1, section, 1.2. In this way, the sample size for the SCSU-C group participants being mostly those with a direct lived experience of living with SCD, and having insights from a carer with indirect lived experience of living with SCD. As such, these participants are able to provide a credible account of the knowledge, views and experiences of the medical and nutritional management of people living with SCD, shared experiences as well as unique and individual experiences of living with SCD reflecting the complexity and individuality of living with SCD. Equally, the diverse SCSP participants, reflect the varied healthcare, social and nutritional needs of those living with SCD.

All in all, albeit, a mostly homogenous SCSU-C pool, with regards to the lived experience of the participants, there was some differences in reported ages of the SCSU-C pool, some that reported to be employed etc, although this was not formerly assessed in the demographic data as a variable. However, the majority of the participants could provide

some insight into the medical and nutritional management of people living with SCD, albeit drawing on experiences of working with other conditions.

Therefore, as a primary qualitative study, the inclusion of a wide variety of both SCSU-Cs and SCSP participants (recruitment impacted by COVID 19), with varied experiences and levels of service provision, facilitated a good representation and a diverse voice reflecting the knowledge, views and experiences of the medical and nutritional management, and the socio-ecological factors and facilitators affecting nutrition and service provision in SCD, evident in the findings of the analysis of the data collected, to be discussed below.

Table 4.1: Background data and non-identifiable codes of independent focus group participants

Sickle cell service user/carers (SCSU-C)	Gender (M/F)	Lived experience/carers	Sickle cell service provider (SCSP)	Gender (M/F)	Profession (Lived experience or Carer)
Participant 1: SU-C (1)	F	Lived experience	Participant 1: SP (1)	M	Community-based Dietitian
Participant 2: SU-C (2)	F	Lived experience	Participant 2: SP (2)	F	Paediatric Dietitian
Participant 3 SU-C (3)	M	Lived experience	Participant 3: SP (3)	F	Nutritionist (Lived experience)
Participant 4: SU-C (4)	F	Lived experience	Participant 4: SP (4)	F	GP in primary care (Carer)
Participant 5: SU-C (5)	F	Lived experience	Participant 5: SP (5)	M	Community SCD Social liaison officer
Participant 6: SU-C (6)	F	Lived experience	Participant 6: SP (6)	F	SCD- Community engagement officer
Participant 7: SU-C (7)	M	Lived experience	Participant 7: SP (7)	F	Paediatric SCD nurse specialist
Participant 8: SU-C (8)	F	Carer			
Participant 9: SU-C (9)	F	Lived experience			
Participant 10: SU-C (10)	F	Lived experience			
Participant 11: SU-C (11)	F	Lived experience			

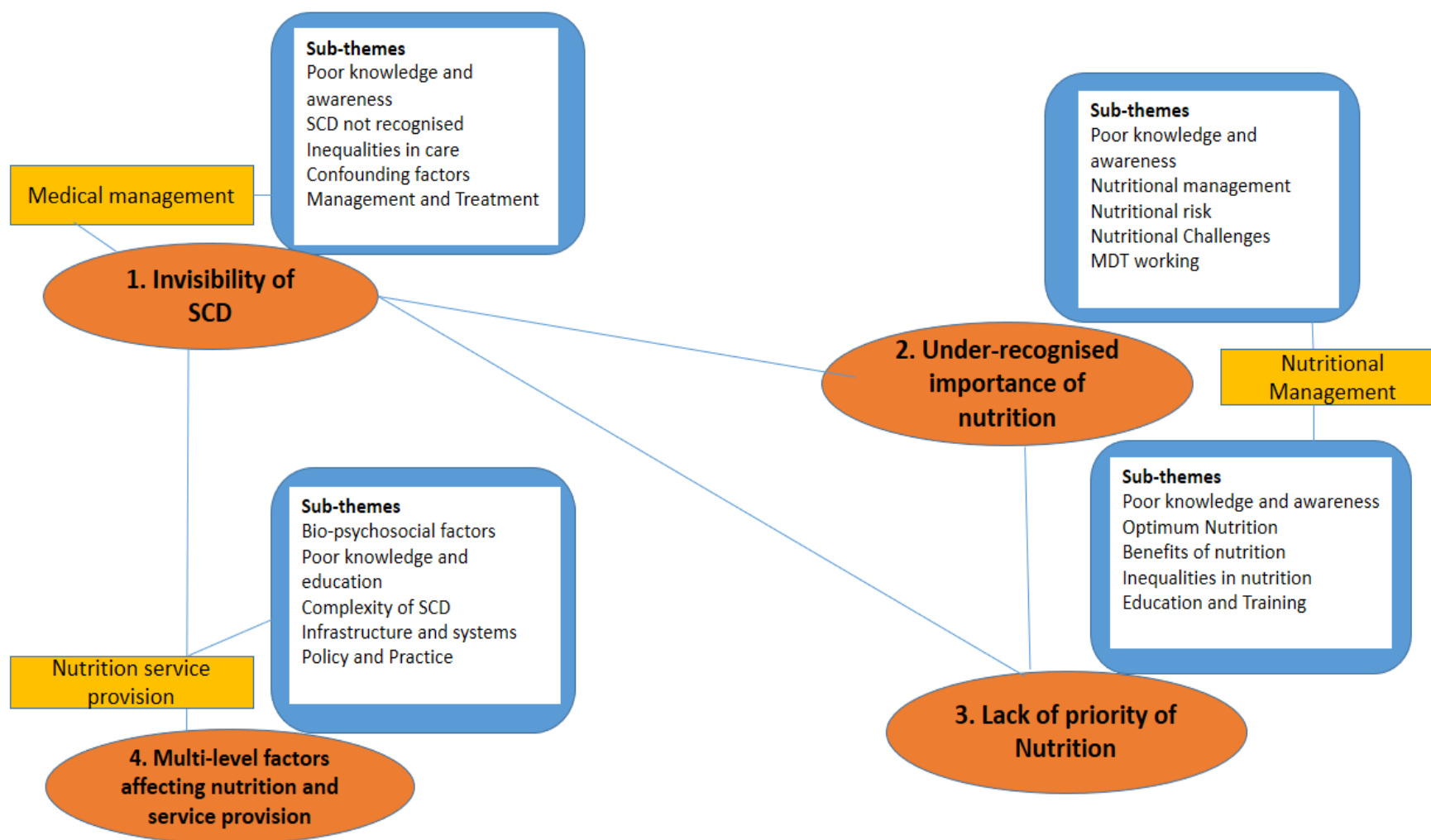
4.2 Focus group findings and themes

The collective findings of each of the separate SCSU-C and SCSP participant focus groups (see extracts in Appendix 17) were analysed to determine Objective 1 and 2 (Chapter 1, section 1.2) of this research project. These findings represent the answers to each of the focus group questionnaire questions (see Appendix 8), representing the four questionnaire categories as mentioned in Chapter 3 (section 3.4.1) with the corresponding questions, as demonstrated below:

- Knowledge of SCD management - (Question (Q) 1
- Knowledge of nutritional management (optimum nutrition) – Q 2 to Q 5
- Influencing socio-ecological factors affecting nutrition and service provision – Q 6 to Q 9
- Facilitators to address the socio-ecological factors – Q 10.

To answer Objective 1, four themes and corresponding sub-themes were developed as illustrated in Figure 4.1 below, representing the final thematic mapping from the data analysis. The four themes include (1) The invisibility of SCD, (2) Under-recognised importance of nutrition, (3) Lack of priority to nutrition and (4) Multi-level factors affecting nutrition and service provision. Below are the findings supporting each of the four main themes and subthemes, in this way identifying the influencing factors affecting the integration of nutrition in SCD, to explicitly answer research question 1.

Figure 4.1 – Thematic Mapping of the analysis of the focus groups



4.2.1 Medical management – Findings:

The influencing factors affecting the medical management of SCD will be reviewed below, as identified by the knowledge, views and experiences of SCSU-Cs and SCSPs in the focus groups in phase one.

Theme 1 – Invisibility of SCD:

The invisibility of SCD is closely related to the poor knowledge and awareness of the condition, that impacts on the level and quality of care provision available to people living with SCD when accessing care in the NHS. This theme evidences the disparities and confounding factors influencing the experiences and medical management of SCSU-Cs of care provision in SCD, reflecting the health inequalities associated with SCD as a marginalised patient population. The sub-themes supporting theme 1 (Figure 4.1), reflects the impact of poor knowledge on the recognition of SCD as a long term condition and the level and quality of care provision available to people living with SCD when accessing care in the NHS. In this way, the theme provides evidence of the disparities in the healthcare experiences and medical management, including some of the confounding factors, reflecting the health inequalities associated with SCD as a marginalised patient population. Poor knowledge is a common thread interspersed into all the themes of phase one, both from a SCSU-C and SCSP perspective.

To this end, nearly all of the participants from both the SCSU-C and SCSP participant pool reported that they had to self –research what they know about SCD and nutrition (irrespective of their location), as mentioned in Figure 4.1 above, this extended to friends living with SCD, as reported by SP (3). Two examples of those who reported to having to self-research what they know about nutrition, is SU-C (6) and SU-C (7). Both SU-C (6) and SU-C (7) resorted to taking matters into their own hands by self –researching what they

needed to know about nutrition, with SU-C (6) reporting that they were never directly asked about nutrition even though nutrition was important to them to stay well.

In this way, the lack of nutrition knowledge and poor access to nutrition information and service provision is connected, indicating a possible link between the lack of knowledge of SCD and nutrition in SCD that needs to be addressed.

SU-C (6) – “nutrition for me I think plays a major part in staying well - ...I’ve never directly been asked about my nutrition and how I eat...I’ve kind of looked up myself - I’ll go and do this research”.

SP (3) – “a lot of my sickle friends who are actually interested in nutrition taking it upon themselves to look up things”

SU-C (7) – “it really is about doing your own research and understanding”

Interestingly, both groups of participants had some level of knowledge about the medical management of people living with SCD, depending on their role in SCD healthcare provision. As such the depth of knowledge of SP (7) (paediatric SCD specialist nurse) who works on a day to day basis with patients living with SCD and works as part of the SCD MDT, was more comprehensive, as illustrated in their response below. In their response, they were able to confidently highlight the complex symptomology of SCD and the complexity of the treatment and management of SCD, essential to adequately meet the medical needs of the patients living with SCD.

SP (7) – it’s a hemoglobin disorder, disease of the red blood cells, and people living with SCD have a lack of oxygen, so anemia, painful crises episodes, affect their daily lives, there’s blood transfusions, top up transfusions, and exchange transfusions, which help to manage the sickle cell, then also medications, like hydroxycarbimide, so this is like the three main treatments for sickle cell, others are in trial at the moment.

Equally most of the SCSU-Cs were able to compile a list of the various complications associated with SCD, that reflected both medical and nutritional problems, further evidence of the interconnectedness between the medical and nutritional management of SCD. One of

the SCSUs [SP (6)], identified poor immunity as a complication of SCD raised as a poignant problem in SCD during COVID 19 (APPG-SCAT, 2020), to add to the list of complications provided by the SCSUs.

SCSUs— “delayed growth, retinopathy, avascular necrosis, fatigue, priapism, side effects of medication, itching, constipation, stroke, epilepsy,

SP (6) – “it affects the immune system”

In contrast, the knowledge of SP (1) (community based adult Dietitian), was markedly limited, as all they knew about SCD they had to self-research, and more so, they were not part of the SCD MDT. As such, SP (2) (paediatric Dietitian), identified the positive effect of their involvement in the SCD MDT, on their growing knowledge of SCD, evident in the response below.

SP (1) – “I work in the community, it’s not very well managed to be honest, because I don’t know much about it in our team, nobody really knows what I know, it was just what I’ve kind of researched myself”.

SP (2)- “because we got a new consultant at the moment, we are getting more and more referrals because she seems to be quite nutrition pro, so I’m starting to learn a little more about it on a day-to-day basis”.

Furthermore, SU-C (8), raised a concern about healthcare professionals working with people living with SCD having miss-information, and how this may add a further layer of complexity to the level of knowledge required to adequately manage the medical and nutritional needs of people living with SCD.

SU-C (8)- “We were not eating meat at the time and we got a lot of flak for that. The health visitor used to say, you’re going to make your child malnourished if you don’t give them meat...it didn’t change my mind because I had to do my own research”.

In addition, SP (5), pointed to the fact that SCD management is not straight forward but is all-encompassing and able to affect the physical, social and mental health needs of patients, impacting both the medical and nutritional management of SCD.

SP (5) - "I guess it is an all-encompassing thing – it affects people's lives physically, socially and mentally".

Reflective of the all-encompassing nature of SCD, SU-C (1) pointed to the increasing life expectancy of people living with SCD as people are now living longer. Hence, there may be other healthcare needs that older people living with SCD may have on top of their SCD related problems and symptoms, impacting on their overall health and wellbeing outcomes.

SU-C (1)- "I have different medical needs as I get older, I find that I get more medical needs because as we were living longer with SCD, we also have age related issues to deal with...I have been a guinea pig all my life because of my age".

Pertinently, SU-C (7) emphasised the importance of healthcare professionals noting that there are differences between the presentation of SCD amongst people living with SCD, depending on their genotype and phenotype, as reflected in the comments below.

SU-C (7) - "A lot of us have differences between us – sickle cell patients have different phenotypes and genotypes – responsible for the variation and this can be present in families as well".

Whilst theme 1 focused on the medical management in SCD, SP (2) recognized the variability in SCD management, in relation to the need for nutrition to be considered in SCD management. Thus the comments by SP (2) points out the absence of the role of the Dietitian as part of SCD healthcare provision. In their comment, SP (2) highlights the invisibility of nutrition in SCD, due to the disregard of the importance of nutrition and nutrition management and the science behind nutrition in the medical management of SCD.

However, SP (2) does draw attention to the value of having a SCD consultant who is pro-nutrition and the positive effect that has on nutrition service provision in SCD.

SP (2) - "I think sometimes we are sort of forgotten about in the sense where people not just doctors but I think everyone kind of thinks nutrition and diet is something that you naturally just know and think about anyway so they don't necessarily think that they need a dietitian because they don't necessarily understand the education or how you could possibly have to study it to understand".

SP (2) - "because we got a new consultant at the moment, we are getting more and more referrals because she seems to be quite nutrition pro, so I'm starting to learn a little more about it on a day-to-day basis".

In summary, based on the collective as well as individual comments reported above from both the SCSU-C and SCSP participants in relation to the lack of knowledge about SCD and nutrition in SCD, theme 1 – the invisibility of SCD is plausible, as most of the participants reported having to self –research what they know, unless they were part of the SCD MDT. In this way, the knowledge, views and experiences of the SCSU-Cs and SCSPs of the medical management of SCD, provided helpful insights into the knowledge and care gaps in SCD, contributing to the invisibility of SCD and subsequent invisibility of nutrition in SCD and how it impacts on the nutritional management of people living with SCD, to be explored below.

4.2.2 Nutritional management - Findings:

The lack of nutrition knowledge, awareness and availability of nutrition service provision in SCD will be explored in theme 2 below. However, despite the array of nutritional needs, challenges and risks which have been identified through the focus group findings to be explored below, nutrition remains an under recognized management option in SCD.

Theme 2 – Under-recognised importance of nutrition:

Theme 2 reflects the under recognition of the array of nutritional needs, challenges and risks as experienced by people living with SCD, when exploring the knowledge, views and experiences of both participant groups, of the nutritional management of SCD. In this way, the theme reflects the impact of poor knowledge and understanding of the link between the medical management and the nutritional management of SCD, and how this impacts on the patient's experience, access and outcomes of nutrition in SCD, and the need for more MDT working, which includes Dietitians.

The under recognition of nutrition in SCD as reflected in the comment by SU-C (9), is shared by many of the SCSU-C participants, and relate to the fact that they never have conversations about nutrition, although they would welcome conversations about nutrition as part of their standard care. Equally the comments from [SU-C (3); SU-C (4)], emphasize the importance of nutrition being included in the standard care provision available to people living with SCD, as part of their standard care provision.

SU-C (9) - "we never have a conversation like you have heard from so many others, we never have a conversation about nutrition".

SU-C (3) – "I feel like there needs to be a bit more discussions on nutrition... and that discussion is never had with my consultants or nutritionist that come around...a lot of discussions about you have to eat more, but never more of what, so yeah".

SU-C (4) – "it does need to be a big conversation".

As such, SU-C (10) calls on the need to have increased opportunity to have 'bigger conversations' about nutrition as part of their medical consultations and that these opportunities should be available on a regular basis allowing patients the opportunity to discuss their nutritional needs and concerns. As such, SU-C (6) recommend these conversations about nutrition happening earlier rather than later to reinforce the message of

good health and nutrition, similar to what is being done with folic acid and fluid intake, recognised as part of the current medical management in SCD.

SU-C (10) – “in general there needs to be a bigger conversation from initial doctors to consultants...the biggest like nutritional conversation I’ve ever had with any of my doctors was you need to drink more water...’there needs to be a bigger conversation because that would have been a lot more beneficial’...when we have done like private research”.

SU-C (6) – “The conversation needs to happen earlier, and continue to happen, so the reinforcement of the message of good health and nutrition, just as they reinforce you must take your folic acid and you must take your penicillin, it should be reinforced that you should be eating”.

The unexpected diagnosis of osteoporosis, as reported in the comment from SU-C (2), reflects the problem of the late diagnosis of nutritional problems in people living with SCD, which is a significant finding of the research project. This was also the case for a paediatric patient whose vitamin D levels were extremely low, and if it wasn’t for a Dietitian asking about their Vitamin D level, it would have gone unchecked, negatively impacting the health outcomes of this patient, a clear risk when nutrition is not part of standard care in SCD.

SU-C (2)- “I for instance was diagnosed with osteoporosis recently and that was a bit of a shock to me and then they’ve now said oh yeah, we are going to start treating it now - oh well why wasn’t it tracked before we got to this level...”.

SU-C (8) “...she asked me what his Vitamin D is like and I said well, I don’t know. So, she said - ‘you’re saying that no one has ever checked it’ and I said yeah, no one has ever checked his vitamin D - so we went to see her she asked me the question we were able to get a blood test for him and absolutely his vitamin D was like nothing”.

This oversight in nutritional care provision to people living with SCD, is ascribed to the fact that the medical management (which includes medication and pharmaceutical management) takes priority, as indicated by SU-C (1) below.

SU-C (1) – “because their role is not to deal with nutrition but it’s to deal with medication, pharmaceutical and stuff”.

Moreover, SU-C (7) connects this oversight of nutrition to how the medical management of SCD systems have been set that does not consider nutrition. Inadvertently, this overlooks nutrition as a viable management option in SCD care provision, that disadvantages patients living with SCD who may have nutritional concerns and needs that they may want to discuss with their SCD healthcare professionals. In this way, as articulated by SU-C (7), ‘they push nutrition to the side and it’s not considered’, directly impacting on the patient’s experience, access and health outcomes in nutrition in SCD, a health inequality impacting on the patient’s ability to manage their nutritional needs.

SU-C (7) – “it’s just not factored, not factored at all into their I guess um medical evaluation of someone with sickle cell” ...so the system is just not considerate what so ever...” ...” they push nutrition to the side and it’s not considered”.

The under recognition of nutrition in SCD management is echoed in the view by SP (4) (a GP) that nutrition has never been mentioned in SCD management and doesn’t feature anywhere in how SCD is managed. From SP (4)’s perspective, SCD management is protocol based, with protocols for pain and blood transfusions and strategies to manage pain. However, nutrition is not acknowledged as having a role to play in the standard management of SCD. This view is supported by SP (6), that points out within their role, health and wellbeing isn’t discussed and more emphasis is placed on what services are available to patients.

SP (4) – “so I think in terms of a medic, I think that it doesn’t necessarily feature anywhere...for the treatment of sickle is usually kind of protocol driven and no one in the past has ever mentioned nutrition... So, we all have the pain protocols, we all have the transfusion protocols, we all have you know what you do in this sort of crisis but no one’s ever mentioned nutrition in anything else you know”.

SP (6) - “So we really don’t talk much about health and wellbeing and the food side of things its more about asking what services are available”.

Yet SU-C (7), clearly identifies inflammation as a nutritional problem for people living with SCD that requires nutritional interventions. Furthermore, the nutritional advice given to SU-C (4) by their consultant, provides evidence of the existence of the nutritional needs of people living with SCD, but the timing of the advice reflects a disregard for the value of the role of nutrition as a viable management option in SCD. The comments of these SCSU-Cs thus provide strong evidence of the existence of the nutritional needs of people living with SCD.

SU-C (7) - “we have a lot of inflammation in our bodies and various other anti-oxidants enhancing blood flow – medicating yourself on a day to day basis with foods that you can eat and try to maintain a descent level of wellbeing so when the inevitable crisis come, it is not so severe as it may be”.

SU-C (4) - “so my consultant told me that I was meant to be on a high protein high carb diet - so I tried it for a while and it worked - but then I questioned why he didn’t tell me earlier...because he only told me 2-3 years ago -it was just like kind of a bypass comments”.

A such, SU-C (9), indicated that there needs to be a more holistic approach to SCD healthcare provision, that includes both the medical and nutritional management in SCD, delivered by expertly trained healthcare professionals in order to tailor service provision to the needs of the individual patients.

SU-C (9) – “I think when you are looking at sickle patients in a more holistic way if the clinical standards are now saying that every patient should be entitled to a psychologist and pain therapist or physiotherapist then why can’t we also have nutrition as part of that and the evidence should also support that...”.

Moreover, a number of nutritional related problems were identified by both the SCSU-Cs and SCSPs. In particular, the SCSU-Cs collectively identified anemia (associated tiredness and fatigue), delayed growth, pica, dehydration and poor appetite, particularly when they were unwell, as key nutritional problems in SCD.

SCSU-Cs- ‘Anaemia, tiredness and fatigue, dehydration, pica, delayed growth, poor immunity’.

In addition, some of the SCSPs [SP (1) and SP (2)] pointed out the problem of fussy eating in children, the gastrointestinal problems (in particular constipation), and the range of vitamins and minerals ('Vitamin D deficiency, Vitamin A, Vitamin C, Calcium (Ca) Omega 3, low iron levels'), required that may be deficient in people living with SCD.

SP (1) and SP (2) - faltering growth, poor appetite, constipation, gastrointestinal problems' 'Vitamin D deficiency, Ca, Vit A, C, Omega 3, low iron levels'

The problem of poor appetite and poor intake during periods of ill-health was identified by a number of SCSU-Cs [SU-C (2); SU-C (6); SU-C (3)] that augments the knowledge of the nutrition needs of patients, and the increased nutritional requirements during these phases of ill-health, particularly when they are an inpatient in hospital.

SU-C (2) – “when I’m ill food isn’t really an interest to me”.

SU-C (6) – “I don’t tend to eat when I am unwell, I don’t eat when I am in hospital, if I do eat it’s because I am getting better”.

SU-C (3) – “I know that for me I don’t eat well at all when I am in hospital – the free meals in hospital doesn’t really do anything for me....”.

It was however, recognised by SU-C (11) that there needs to be an emphasis on addressing the knowledge gaps of healthcare professionals of nutrition in the management of SCD. It was recommended that more education and training be made available for healthcare professionals in SCD tailored to the specific needs of the patients, as currently there is an imbalance of knowledge of nutrition in SCD between the SCSU-Cs and SCSPs, as indicated in the comment from SU-C (2) below.

SU-C (11) – “so in terms of doctors, I don’t know if they have that much training on nutrition for people with SCD because you see you are deficient in a lot of things”.

SU-C (2) – “Well for me, most of the time I find that I know more than the doctors”.

The nutritional management of people living with SCD, is influenced by a number of challenges one of which is the variability in the severity and presentation of the symptomology in people living with SCD, indicated in the comment by SP (4), meaning SCD should be managed on an individual basis.

SP (4) – “I see it not only from the provider side but from the stakeholder side (parent)...I’ve got 2 children with SCD both have varying degrees - one whose extremely brittle and very very poorly all the time and one who has never had a crisis”.

SU-C (6) reflects the care gap in nutrition in SCD, and says they have had to self-research what they know, as they have never been asked about their nutrition or how they eat. In support of this comment, SP (3) concurs that they have had zero nutritional input as part of their standard care in SCD and the advice that is provided is mostly generic and not specifically tailored to patient’s needs. SP (3), also identified that referrals have also not been done, further perpetuating the nutritional challenges for people living with SCD.

SU-C (6) - “I’ve done my own private research; “I’ve never directly been asked about my nutrition and how I eat...I’ve kind of looked up myself...”.

SP (3) – “I am taking the stance from my own view of it like me as well what I’ve experienced for myself um and there’s been, it’s been like zero...there’s really not been any sort of input”.

SP (3) – “You do get a lot of generic advice, there’s nothing specific, there’s not been any sort of input; even the referrals don’t even happen”.

The lack of nutrition service provision in SCD thus further fails to meet the needs of people living with SCD especially those who understand the value of nutrition in helping them to stay well [SU-C (6)]. SP (4) points out the importance of linking the benefits of nutrition to the outcomes of the disease management of patients and that nutritional management needs to be more holistic and not simply focused on pain management in SCD, but it should be recognising both the medical and nutritional needs of people living with SCD.

SU-C (6) – “nutrition for me I think plays a major part in staying well”.

SP (4) – “unless people are able to turn around and say, there is a link between what you eat and how you feel in your medical condition that’s probably the only time we get more motivation from our patients”.

SP (4) - but I don’t think its managed in a kind of holistic way and say ok we need to look at it not just their pain we also need to look at their nutrition”.

Theme 3- Lack of priority to nutrition:

Theme 3 reflects the under prioritization of nutrition in SCD, evident in the paucity of nutrition resources and poor knowledge of both groups of participants, and the inequalities in nutrition service provision as part of standard care in SCD. In this way the theme reflects the limited understanding of optimum nutrition, the benefits of nutrition in SCD healthcare provision and the need for education and training in nutrition in SCD to be tailored to the unique needs of people living with SCD, to provide patient centered and personalised care provision in nutrition in SCD. Thus, the lack of prioritisation of nutrition as a management option in SCD, affects the importance of the role of nutrition in the management of people living with SCD.

It was acknowledged by SP (4), that nutrition may not be a priority when treating people living with SCD, compounding the ignorance around nutrition in SCD, negatively impacting on those patients who do not rely on medications to manage their condition, but have remained reasonably well by eating well. This view of the SCSP is echoed in the comment from SU-C (9), who relies on nutrition to manage their SCD, and to remain well.

SP (4) - “Ignorance around nutrition in many kind of service providers – it’s not at the top of their list when you are looking after these patients”.

SU-C (9) – “in the management of my sickle it’s been central - I’m not on any medication so that’s my medical position I do not take a fixed medicine except penicillin when unwell so outside of that this is how I was raised; my mother was a paediatric nurse”.

To address the lack of nutrition knowledge amongst SCD healthcare providers, SU-C (1) has been providing nutrition education to SCD healthcare providers to highlight the value of good nutrition as a management option in SCD, alongside the medical management and treatment, which often times comes with an array of side effects that need to be managed to improve patient outcomes.

SU-C (1)- “we are campaigning for doctors and nurses to know about basic nutrition apart from the other things. We believe that it’s something that they should know you know when treating us and not just dealing with our medication that has side effects causing other issues, meaning more medication. Nutrition is paramount”.

It was evident from the responses of nearly all of the SCSU-Cs and SCSPs that their understanding of the role of nutrition (optimum nutrition) in SCD was limited to food, and therefore did not reflect the wider factors that may have a role to play in the nutritional needs of people living with SCD. For instance, the common understanding as reported by the SCSU-C and SCSPs was that optimum nutrition is about having a good balance of nutrients needed in the diet SP (3), eating regularly SP (5) to optimize nutritional intake and status of the people living with SCD, meaning they need the support of a trained healthcare professional to be able to do that as part of their standard care provision. However, SU-C (2) considered optimum nutrition to be an ideal and not something that can be achieved on a regular basis, particularly during times of ill health and when they may not be able to access food or prepare food for themselves.

SP (3) – “Optimum nutrition is getting a good balance of everything one has in the diet; getting people to put things back in their diet; optimising the needs for that person”.

SP (5) – “I think a balanced diet eating 3 times a day; like a personal nutritional plan depending on how their condition manifest”.

SU-C (2) – “so for me optimum nutrition is an ideal that is not necessarily something I can keep up with all the time because when you are not feeling well or have no energy to prepare food and anything else you just grab the nearest thing you can...”.

This limited understanding of optimum nutrition was challenged by a comment provided by SP (4), who recognised the importance of considering the wider factors that may influence how well a person living with SCD is able to provide for their nutritional needs. Thus, they said that healthcare professionals in SCD should not make assumptions of people living with SCD and their ability to buy, store and prepare foods, and regard should be given to the socio-economic factors affecting people living with SCD.

SP (4) - "if I am asking them to eat a healthy diet, I am assuming that they can afford to buy the food that we are asking them to eat...I'm making an assumption they have a kitchen that they can prepare the food in some of my patients live in hostels so there are a whole host of socio-economic reasons that those patients face".

Table 4.2 below is a summary of the main perceptions held by the participants about their understanding of optimum nutrition in SCD, reflecting the narrow view of nutrition in SCD held by both the participant groups, being all about food and not necessarily the wider factors influencing a patient's nutritional needs and status.

Table 4.2: Perceptions of optimum nutrition held by SCSU-Cs and SCSPs

Knowledge of optimum nutrition
➤ Getting a good balance of everything one has in the diet ➤ Getting people to put things back in their diet ➤ Know good sources of food ➤ What the food groups are ➤ Vitamins through fruit and vegetables ➤ Balanced eating

Considering food in the context of optimum nutrition in SCD is important, and in this way, it was overwhelmingly clear that many of the SCSU-Cs believed that nutrition was fundamental [SU-C (1)], a key component of them remaining well [SU-C (3)], and should be brought up a lot more in patient consultations [SU-C (10)], adding to the narrative about the

importance of the role of nutrition in SCD to be considered alongside the medical management of people living with SCD. More importantly, that nutrition is valuable in SCD management in all stages of the life course, as highlighted by SU-C (4) who required Percutaneous Endoscopic Gastrointestinal (PEG) feeding when they were a child to augment their nutritional intake at the time, aimed at reducing the frequency of their sickle cell crisis; in line with the findings of the Heyman et al., (1985) study, the first study to demonstrate the benefits of nutrition as a management option in SCD to improve on the clinical outcomes of people living with SCD.

SU-C (1)- “I think it is fundamental”.

SU-C (3) – “nutrition I think plays a major part in staying well”.

SU-C (10) – “I think it’s something that can be brought up a lot more cos for me for a lot of years it wasn’t really addressed” – ‘sickle cell has been my own private research – definitely something that should be more spoken about”.

SU-C (4) – “early in my life I had PEG feeding because my nutrition wasn’t very good so I kept going into crisis”.

In order to identify the benefits of nutrition in SCD, SP (3) pointed to the need to link and connect nutrition and health in SCD.

SP (3) - “there just needs to be a bit more clarity - making the connection between nutrition and health in relation to sickle cell”.

In this way, SP (6) emphasized the need for nutrition to be made mandatory in SCD, providing timely follow up and onward referral, whilst making nutrition a topic that is talked about in consultations, giving patients the opportunity to ask questions and talk about their nutritional needs.

SP (6) - “making it mandatory and then looking at the persons making sure that we are following up and making the right referrals... make it a topic that we talked about its not talked about if they kind of bring that to the light then that can maybe change or will help to change things”.

For this reason, SP (1), suggested that more time is invested to train members of the wider MDT who may also come into contact with people living with SCD, to have knowledge of SCD and be in a position to refer relevant patients to the Dietitian as needed, thus prioritising nutrition alongside the other care needs of the patients.

SP (1) - "I think more kind of training more awareness of it like the wider MDT like physios, OT's psychologists if they are more aware then I could identify more probably referrals to the Dietitian".

In support of the prioritization of nutrition in SCD, Figure 4.2 below, illustrates a range of concerns raised by a number of SCSU-Cs to express their experiences associated with a lack of nutrition service provision, compelling evidence of the lack of prioritisation of nutrition in SCD, impacting on the patient's experiences, access and health outcomes in nutrition in SCD, motivating for the need for more tailored education and training in nutrition in SCD.

Figure 4.2 – SCSU-C concerns about the lack of nutrition service provision available to them as part of their standard care in SCD



In summary, the individual and collective comments reported above, reflect the knowledge, views and experiences of both the SCSU-C and SCSP participants, underscoring both the under recognition of the importance of the nutritional needs, challenges and risks and the lack of priority to nutrition. As such, the overall lived experiences of people living with SCD in relation to their nutritional needs, risks and challenges impact on their experience, access and health outcomes in nutrition in SCD.

4.2.3 Nutrition Service Provision - Findings:

A number of a multi-level factors affecting nutrition and service provision in SCD were identified by the SCSU-C and SCSP groups to be reviewed below.

Theme 4 – Multi-level factors affecting nutrition and service provision:

The multi-level factors identified in the independent focus groups includes: a range of bio- psychosocial factors, poor knowledge and education in nutrition in SCD, a need for improved infrastructure, systems, policy and practice reflective of the nutritional needs of people living with SCD. Theme 4 thus provides an overview of the range of knowledge and care gaps necessitating a population level, whole systems approach to improve nutrition service provision in SCD, to address the effects of the marginalization of SCD and the growing health inequalities associated with the medical and nutritional management of people living with SCD.

Personal factors:

The bio-psychosocial factors identified by most of the SCSU-C and SCSP groups as influencing the nutrition and service provision needs in SCD included the following: the role of family, family culture and traditions impacting on the dietary habits and choices of people living with SCD. In addition, a few of the SCSU-Cs [SU-C (5) and SU-C (9)] identified

emotional and psychological factors such as variances in a patient's mood, motivation, attitudes and beliefs towards nutrition and food choices. Further confounding personal factors identified by some of the SCSU-Cs as playing a role in nutrition in SCD included: the impact of families, family structures, religion and the environment (home and work). In particular, SU-C (1) shared how their clinical symptomology of SCD affected their ability to access and prepare foods, increasing their reliance on family members to support them with their nutritional needs and intakes, especially when suffering from the effects of chronic anemia and fatigue or when they are unwell and do not have the energy or strength to prepare food.

SU-C (5) – “I am also guilty of comfort eating and that also applies when I am not feeling well”.

SU-C (9) – “Sometimes when I am really moody then I would tend to eat stuff that I naturally wouldn't eat”.

SU-C (1)- “I suffer with lethargy and tiredness and I don't feel like cooking everyday...I'm really reliant on my family at home to do a lot of the cooking you know... my daughter keeps things chopped up in the fridge so its easily accessible”.

SU-C (5) reported how their working pattern and environment has posed a problem in relation to what and when they are able to eat and the availability of healthy options.

SU-C (5) - “Because of my job with long hours, I have most of my meals through work, for my breakfast, lunch and dinner, as a result I eat what's available there”.

Moreover, many of both the SCSU-C and SCSP groups reported poor nutrition knowledge to be a personal factor, and in particular SP (4) suggested teaching parents about nutrition, who in turn can support their children, raising awareness of the need for more nutrition education and training in SCD. On the other hand, SP (5), identified the impact of the social factors, including finances, that can impact on what, how well and when people living with SCD are able to eat. SP (1) also identified the difficulty of some young people transitioning

from being looked after to adulthood and having to fend for themselves, when studying, findings jobs and not always fitting in with the rest of their friends.

SP (4) – “Yeah I think the education needs to start in the home, that’s where it needs to start”.

SP (5)- “I think they know...obviously social factors affect how well they can eat; how much time they can commit to cooking”.

SP (1) – “Some of the patients I’ve had, young people going from being looked after through the family and then they’re on their own studying, jobs, so like social life, not fitting in with the rest of your friends or your social circle...”.

The comments below from a few of the SCSP’s including SP (2), indicated that some children present with depression or low mood because they may already be singled out due to having SCD. Puberty was also stated as a personal factor encountered by the younger people living with SCD [SP (2)], that may influence their nutrition needs. Other factors (such as) loneliness, depression and not fitting in with peers were also influencing factors identified by SP (1).

SP (2) – “I think sometimes we get a lot of depression in the kids or low mood because they already feel singled out all the time so food isn’t necessary a priority to them...”.

SP (2) – “or sometimes we get the issues around puberty age where girls especially start becoming more concerned about their bodies...”.

SP (1) – “...maybe the depression and low mood feeling depressed and lonely, your motivation is going to be quite low...”.

Interpersonal Factors:

In line with some of the personal factors already identified, peer support, family traditions and culture, and family structure and support were also identified by some of the SCSU-Cs and SCSPs as influencing interpersonal factors to be considered. Some participants amongst both the SCSU-C and SCSP groups identified the need to address the poor

nutrition knowledge of patients, family and friends, including the need for healthcare professionals to improve their knowledge of both SCD and nutrition in order to raise awareness of SCD and the nutritional needs of people living with SCD. In particular, SP (1) identified the need to not only educate people living with SCD, but also to recognize that some of them may need additional help with activities such as collecting their nutritional supplements for example. In addition, SU-C (4) commented on the value of conversations about nutrition happening prior to admissions to raise awareness and for it to become second nature to eat well.

SP (1) - "Trying to educate them on the importance of nutrition; often patients are too tired to pick up prescriptions from the chemist".

SU-C (6) – "So if the conversation happens prior to being hospitalised, then you kind of, you know like the more you hear something, the more it sinks in and becomes second nature. Then you are already aware of the things that you should be eating".

One SCSP, SP (2), suggested extending nutritional education to schools, to address the stigma of SCD in this environment through activities such as 'show and tell', allowing the children living with SCD to educate others about their condition and how it impacts their life and health and wellbeing to reduce the stigma of living with SCD.

SP (2) - "Sometimes I also speak to schools about maybe do like a show and tell sort of thing at the end of the day trying to get people, like they would do for allergies for example teach the rest of the class not to share food...teaching them that everyone may have their own thing...because if you teach them at a younger age they not, gonna have that stigma as they get older when someone says they have sickle cell they will sort of know what to do".

Institutional Factors:

A few of the SCSPs, in particular SP (3) and SP (4), identified the need to make a closer link between health and nutrition and the nutritional problems in SCD and how nutrition in SCD

and the health outcomes of people living with SCD are linked, as reflected in the comment below.

SP (3) - “there just needs to be a bit more clarity - making the connection between nutrition and health in relation to sickle cell”.

SP (4) – “You know so there is a huge divide about what people really understand by nutrition when you’re looking at it from an institutional level... so in health you need to understand the link between nutrition and health”.

One of the SCSU-Cs, SU-C (9) in particular, called for SCD service provision to take a more holistic and MDT approach to include nutrition, because nutrition is paramount to improve the health outcomes of people living with SCD. This supported the point raised by a SCSP, SP (2), of the need for SCD consultants to be pro-nutrition to help raise awareness of the importance of nutrition in the management of people living with SCD, with the benefit of having Dietitians working as part of the SCD MDT.

SU-C (9)- “well I think joint clinics would be great for both paediatric and adult patients...just sort of have that joint model of care, but we also educating either the nurses or the doctors and enhancing their skills as well”.

A few of the SCSPs identified institutional factors; SP (3) raised awareness of the inequality between the service provision to oncology patients and patients living with SCD, clear evidence of the health inequalities patients living with SCD have to contend with. Furthermore, SP (1) called for more joint up working in community settings in SCD to address the disjointed working styles and practices in this setting, which may include developing a MDT clinic in the community. Furthermore, SP (1) suggested that regular CPD sessions could be organized in community teams to educate and raise awareness about SCD in order to facilitate effective signposting to relevant services. Moreover, SP (1) added, in support of SU-C (2)’s views, that for this to happen, a top down approach in leadership is necessary to raise awareness of nutrition in SCD, but also recognising the importance of

identifying SCD patient numbers in the community setting, to justify funding and service development.

SP (3) – “there’s lots going on in oncology that we can have ample access to but they don’t provide it, it is not open to us; priority is given to oncology patients over sickle cell patients, even though the patients are clumped together”.

SP (1) – “I think it’s like you said, its raising awareness and getting the management to work with the 8A and 8B to know more about it...highlight how many patients we have in the community...we just haven’t identified them working with the CCG and the GP practices as well”.

SP (1)- “it’s all about costing isn’t it, you’d have to look at how many patients they have, the cost that they are to the community hospital, someone would need to do a business plan...”.

SU-C (2) – I think you need to educate people from the top level”.

A couple of the SCSPs, for example SP (7), acknowledged that nutrition can be included in the training they provide to schools, and not just simply to focus on the fluid requirements in SCD, but the overall nutritional needs of the children living with SCD can be monitored. On the other hand, SP (5) emphasized the need for all healthcare professionals in both health and social care settings to lead on spreading the message of the importance of healthy eating, as a way to influence them to take on board the message of nutrition in SCD.

SP (7) – “I do go into schools and do education for all teachers...there isn’t much focus on, well there isn’t any focus on like in the teaching that I do in nutrition, its more on the fluid intake, I guess there could be some information on nutrition so that at least it could be monitored in school”.

SP (5) - “I think if the medical professionals do it, health and social care professionals do it in the schools the families will probably listen and do it, I think it’s when everybody has that information that we will all do it”.

Moreover, a couple of the SCSPs (both Dietitians), SP (1) and specifically SP (2), identified the need for nutritional guidance or recommendations to support their clinical practice and nutritional management of people living with SCD.

SP (2) - "I think any recommendations you can make out of this would be amazing because I think from my end, trying to find like any evidence base is quite hard, I mean asking my seniors for advice and they've straight away been like I don't know, so it would be nice to have something set in stone".

Community/Policy Factors:

A few of the participants of both the SCSU-C and SCSP groups identified an array of community/policy factors affecting nutrition and service provision in SCD, including the lack of support structures and infrastructure to provide healthy food options in the community. The lack of nutrition resources available to service users and providers in the community, was also mentioned by SU-C (6); the need for more information about nutrition in SCD was highlighted by a couple of SCSPs, including SP (7) who highlighted the need for more evidenced based research and more studies to be conducted on the role of nutrition in SCD, to support and inform patients about the types of food to eat specific to their condition and nutrition needs. In this way, a collective approach was recommended by SP (5), in support of advocating for more nutrition service provision in standard care in SCD.

SU-C (6) – "I think it boils down to availability of the information".

SP (7) – "I think if we have the evidenced based research, so there is more research being carried out more studies and say information to say to patients this is the kind of nutrition you need to eat, making sure they stick to it when they are unwell, still keeping up with their nutrition...".

SP (5)- "yeah I think it will have to be a joint collective where both the service providers and service users both together petition to the organization".

Moreover, some of the SCSP group, in particular SP (4,) identified the need for a national campaign to raise awareness of nutrition in SCD on a larger scale (adverts on buses and TV) and using a variety of social media platforms, supported by the Sickle Cell Society (SCS) and the British Dietetic Association (BDA). In addition, SP (4) suggested a digital nutrition algorithm be developed for GPs in primary care to support the nutritional

management of people living with SCD in primary care, as well as possibly a leaflet about nutrition in SCD that GPs can hand to patients when they attend appointments.

SP (4) – “It would be nice to see on a large scale, you know, large scale adverts, on buses, on adverts on the TV, the same way that the diabetes campaign started.”

SP (4) - “suppose the easiest way for me to even remember to do it would be to have some sort of tool that I can use yeah some sort of digital template that I could then ask those questions”.

Some participants from the SCSP groups, SP (2), identified the similarities in the factors influencing food choices in people living with SCD and ordinary people, in respect to their socio-economic status and social class, and whether they live in a more deprived area and have access to healthy shops (that is quite expensive). So for people living on the poverty line, they will be restricted to what they can and can't have, and therefore more prone to buying chicken and chips, as opposed to fruit and veg, for example. In addition, SP (1), suggested people living with SCD to be referred to social prescribers for additional advice and support in their local areas, when they attend for GP appointments. Similarly, some of the SCSU-C participants, SC-U (6), identified a lack of available healthy and affordable food options in the local areas where people living with SCD live. Moreover, SU-C (9) identified the need for people living with SCD to have improved access to cooking workshops, healthy talks, health promotion activities, healthy food shops and markets and using dynamic celebrities to promote nutrition in SCD, in the community.

SP (2) – “...it's just a matter of what areas they live in, is it a deprived area and do they have access to healthy shops which is quite expensive so straight away if the family is on the poverty line they are going to be restricted to what they can and can't have...”

SP (1)- “...right now social prescribing is the big thing, so when these patients go to the GP, they look at different activities like they can link up with these social prescribers them put on classes and support group activities”.

SU-C (6) – “there is not much in the way of healthy food options like in my surrounding areas there's a lot of chicken shops - like whole foods is a very expensive shop”.

SU-C (9) – “...but also those cooking classes are really really important because I know they do it for so many other conditions in other boroughs and it is free”.

A couple of the SCSU-C participants, for example SU-C (8), identified the need to engage with the topic of nutrition in a more collaborative way within the community, but also on a strategic level that involves key stakeholders such as the BDA, Sickle Cell Society (SCS), NHS England and the CCG. SU-C (2) further recommended people living with SCD are to be empowered to write to their local MP's and hospital boards to raise awareness of nutrition and the quality of hospital food, to improve patient experience, access and health outcomes in nutrition in SCD. SU-C (9) recommended that nutrition be part of clinical policy in the same way that people living with SCD, access other non-medicalised therapies.

SU-C (8) – “I think that um the SCS has got lots of connections with the CCG, with all the NHS England groups, so I think they are quite well placed to raise this issue support people also”.

SU-C (2)- “I think possibly people writing to their local MP and writing to the hospital board...to find better ways of giving people better healthy choices”.

SU-C (9) - “I think there is something about how to sort of push from a policy perspective and clinical policy, nutrition to be part of these non -medicalised therapies that we can have access to whether we are outpatients or inpatients”.

4.2.4 Socio-ecological Facilitators:

Below is a summary of key socio-ecological facilitators from the SCSU-C and SCSP groups reflecting the knowledge and care gaps in the nutritional management of SCD, that have influenced the core recommendations developed by the researcher, to be discussed below.

Knowledge gaps:

- There is need of expertly trained healthcare professionals in SCD, tailored to the needs of individual people living with SCD
- SCD healthcare provider's knowledge of SCD is a must
- There is need of evidenced based resources to reflect the nutrition needs of people living with SCD
- More education and training for healthcare professionals in SCD to provide individualised treatment to people living with SCD

- Providers need the right level of knowledge about nutrition to support the ongoing nutritional needs of people living with SCD who rely on nutrition to remain well
- Knowledge of nutrition to reflect the complexity of SCD and nutrition
- Need more tailored education and training

Care gaps:

- Consider commissioning arrangements in SCD management
- There is need of committed SCD consultants who are pro-nutrition
- Nutrition to be talked about in consultations giving patients opportunity to ask questions and about their nutritional needs
- Provide a holistic approach to healthcare in SCD including both the medical and nutritional management
- Dietitians to work as part of the SCD MDT
- Patients to have access to nutrition service provision and have a chance to speak about nutrition and not have to rely on their own private research
- Refer people living with SCD onto specialist dietetic support

A collective synthesis of the SCSU-C and SCSP views and shared perspectives will be explored below, to provide more helpful insights and context to understand the varying perspectives of the two participant groups, when reviewing the four main themes which have been developed from the overall analysis of the focus group findings.

4.3 Synthesis of the SCSU-C and SCSP views

A synthesis of the overall SCSU-C and SCSP views, as illustrated in Table 4.3 below, provide helpful insights into the different perspectives of the two participant groups about their knowledge and experiences of the medical and nutritional management of SCD.

4.3.1 SCSU-C views:

Interestingly, the collective responses of the SCSU-Cs were from the point of expressing their need for improved access to nutrition service provision in SCD. The SCSU-C group identified a number of key factors and gaps that needed to be considered to address the current care gaps and how best to overcome these gaps in service provision. For example, SU-C (1) identified an important oversight as it relates to how SCD is not being recognized as it should be as a long term condition and can therefore be considered to be invisible.

Although nutrition was identified as being beneficial in improving patient's mental health [SU-C (3)], concerns were raised by SU-C (4) about the impact of good nutritional care in the context of nutrition being a potential 'cure' for SCD. In this, SU-C (4) expressed concerns about how a SCD patient's eligibility for Personal Independent Payment (PIP) may be impacted by an improvement in a SCD patient's symptomology, because of nutrition.

Reports of the late diagnosis of nutritional problems in people living with SCD, for example SU-C (2) reported to be diagnosed with osteoporosis, a diagnosis they were not expecting, was a potential significant finding of the research project. In this way, calls from SU-C (5) to have timely access to nutrition service provision, and for nutrition to be provided proactively and not reactively during annual review meetings for example, is needed to prevent delays in the diagnosis of nutritional problems in people living with SCD.

Moreover, SU-C (3) raised concerns about the quality of hospital food available to people living with SCD, thus collectively, the SCSU-C group, were keen for nutrition service provision to be made available to them as part of their standard care provision.

SU-C (1)- "I believe sickle cell is not recognised as it should be" + "sickle cell is overlooked it's an invisible illness so you can walk around and people cannot be recognising or realising there is something wrong".

SU-C (3)- "I myself find that nutrition is very important for my health, even in terms of mental health nutrition has been a massive part of my mental health".

SU-C (4) – “Mine is socio-economic factors and nutrition...if you eat and cure yourself as in you don’t get as many crises and you’re feeling better...but then when it comes to things like PIP, they need to see that you are unwell to give you the money”.

SU-C (2)- “I for instance was diagnosed with osteoporosis recently and that was a bit of a shock to me and then they’ve now said oh yeah, we are going to start treating it now - oh well why wasn’t it tracked before we got to this level...”.

SU-C (5) W- “having a more proactive approach...maybe if we had regular meetings with a nutritionist or a dietitian it wouldn’t take me having the condition in the first place - I would have already you know prevented it before it happened”.

SU-C (3) – Um the food that is provided in hospitals, I can’t really eat it in terms of number 1, flavour, and two, nutritional value because a lot of it is carbs and a lot of it is pre-heated and packaged process food, and I go into hospital to try to get better and I feel like I come out losing weight”.

4.3.2 SCSP views:

Distinctly, the responses of the SCSPs were from the perspective of the reasons why nutrition service provision was not part of standard care in SCD. For example, SP (3) identified the ‘inequality of care provision’ between care provided to oncology patients versus care provided to those living with SCD and SP (4) highlighted that care is protocol based. SP (6) reflected how the disparity in SCD and potential of stigma, extends to the experiences of school children living with SCD, especially as the symptomology of SCD may be invisible to people, meaning there is grounds for people to overlook or misunderstand the needs of school children living with SCD, for instance when reporting pain or when they need to access the bathroom on a more frequent basis.

SP (1) identified the need for people living with SCD to be empowered to take responsibility for their health, and social prescribers are now available to all patients through GP surgeries. Thus, empowering people living with SCD needs to happen in conjunction with the support of the SCSPs, including SCD consultants who are pro-nutrition [SP (2)] and have ultimate responsibility for the patients in their care.

Additional points raised by the SCSP’s, for example SP (3) and SP (4), recognise the value and the role of social media to broadcast uniform messaging to a wider audience, in

particular the younger people living with SCD. In so doing, the invisibility of SCD can be partially addressed and the role of nutrition in SCD to improve patient experience, access and outcomes in nutrition in SCD can be promoted via social media.

SP (3) – “there’s lots going on in oncology that we can have ample access to but they don’t provide it, it is not open to us; priority is given to oncology patients over sickle cell patients, even though the patients are clumped together”.

SP (6)- “I think sometimes it’s a matter of promoting in schools’ certain conditions if a child had allergies, they have a meeting with the school straight away...but they won’t necessarily do that for sickle cell...physically they look the same or most of the time you won’t necessarily know - whereas if someone had a broken leg you would know straight away”.

SP- (1) - “being young adults - they don’t really understand why nutrition is important for them as well often they’ve been told to do that but they don’t know why or they are not as empowered”.

SP (1)- “...right now social prescribing is the big thing...”.

SP (2)- “because we got a new consultant at the moment, we are getting more and more referrals because she seems to be quite nutrition pro, so I’m starting to learn a little more about it on a day-to-day basis”.

SP (4) – “I mean like social media, definitely, definitely your Instagram, your snapchat, Facebook, twitter, all of those things definitely, they are always in there...particularly the younger ones”

Table 4.3- Summary table to show views of SCSU-Cs versus SCSPs about the nutritional management of SCD

SCSU-C group views	SCSP group views
1.Recognition of sickle cell – invisibility	1.Inequality in care provision
2.Nutrition Reactive versus Proactive	2.Protocol based care
3.Annual Reviews	3.Consultant who is pro-nutrition
4.Clinical Standards in Sickle Cell	4.Buyin from consultants
5.Late diagnosis of nutrition problems	5.Stigma
6.Nutrition important for mental health	6. Patients need to be empowered
7.Nutrition as a cure for sickle cell	7.Social prescribing
8.Nutrition miss information	8.Uniform message
9.Taking self –responsibility	9. Social media
10.Quality of hospital food	10.Disparities in schools

4.3.3 Shared participant perspectives:

The synthesis of the SCSU- C and SCSP views provided a helpful insight into the shared perspectives between both participant groups, as illustrated in Figure 4.3 below. In particular, the following six shared perspectives that shed light on key influencing factors pertaining to the medical and nutritional management of SCD, is demonstrated below.

COVID 19, was identified by both participant groups as a factor impacting on the medical management of SCD, particularly in relation to the SCD being brought under the health radar. Interestingly, for SU-C (1) bringing SCD under the health radar elicited feelings of skepticism, and in contrast SP (3) was relieved that their employers could finally understand the impact of COVID 19 on a person living with SCD.

Closely related to COVID 19, both groups identified the invisibility of SCD, more so in relation to the inequalities between SCD care provision and other long term conditions in haematology, such as oncology patients, and called for more and bigger conversations about nutrition as part of their routine consultations.

A significant finding of the research project, was that both participant groups reported to have self-researched what they know about nutrition and the medical management of SCD, in particular those SCSPs who were not part of SCD MDTs. For this reason, both participant groups identified the need for education and training in nutrition in SCD, to address the knowledge gaps of both groups.

The paucity in the knowledge of nutrition in SCD, underpins the limited views of optimum nutrition in both participant groups, as both understood nutrition to be all about food and both groups identified that more emphasis is placed on fluid as opposed to nutrition as a wider topic.

Figure 4.3: A summary of the shared perspectives between SCSU-Cs and SCSPs about nutritional management in SCD



As seen above, the collective main views (Table 4.3) and shared perspectives (Figure 4.3) of the SCSU-C and SCSP groups enhanced my understanding of the focus group findings. The next stage of the research project, the formation of the Learning Alliance Network (LAN) between the SCSU-C and SCSP, was aimed at facilitating joint conversations amongst the members of the LAN towards reaching a consensus viewpoint on the myriad of influencing factors reflected in the multiple knowledge and care gaps and facilitators identified in the independent focus groups, to be explored below.

4.4 Reflections on the process of the LAN meetings

Below is an overview of the reflections on the process (discussions and actions) that took place during phase two, three and four of the LA methodology, providing insights into how consensus was reached about the data collected in phase one of the research project and how this data was used to co-develop and evaluate the health literacy framework in phases three and four respectively, by the members of the LAN.

4.4.1 First LAN meeting reflections- (12/10/20 - online):

The first LA network meeting involved the formation of the Learning Alliance Network (LAN) between the SCSU-C and SCSP, which followed after the independent focus groups, and was the first opportunity for both groups of participants to meet online. Not only did the LAN provide the members with an opportunity to work collaboratively and in partnership, it enabled the members of the LAN to talk about and jointly discuss the data collected in phase one. The LAN comprised of the following participants: SCSU-Cs (SU-C – [2, 3, 7 and 9]) and SCSPs (SP – [1, 2, 5 and 7]). The joint working between the participant groups also allowed them to develop a common viewpoint and shared critical consciousness and praxis about the problems encountered by both the SCSU-C and SCSP, in relation to the medical and nutritional management of people living with SCD, and the myriad of influencing factors affecting nutrition and service provision. Moreover, the mutual dialogic discussions were an opportunity to add new perspectives to the data already collected, to simultaneously work towards reaching a consensus viewpoint about the data that will be used to co-develop the health literacy framework in phase three of the research project. Additional points raised by the members of the LAN in the first network meeting (Appendix 15), added to the themes developed from the analysis of the focus group data collection. These additional points raised by the members of the LAN, added to the knowledge and care gaps underpinning the

four main themes of the research project to achieve objective 1, and answer research question 1: *What are the influencing factors affecting nutrition integration in SCD?*

4.4.2 Second LAN meeting reflections- (22/10/20 - online):

The purpose of the second LA meeting was to co-develop the health literacy framework by the members of the LAN. The co-development of the health literacy framework was a collaboration between the members of the LAN and myself, as the researcher. In this way, the members of the LAN co-developed the initial draft of the health literacy framework (Appendix 11) based on the data collected in phase one of the research project, and the discussion and consensus viewpoint reached about the data in phase two of the research project. For the initial co-development of the framework, the question categories used in the focus group questionnaire (Chapter 3, section 3.4.1) were used to provide structure for the health literacy framework. The content of the framework contained the knowledge and care gaps which defined the medical and nutritional management of people living with SCD, including the socio-ecological factors and facilitators affecting nutrition and service provision in SCD collected in phase one.

The benefit of the collaborative and partnership working between the members of the LAN, further enhanced their critical consciousness and praxis about the knowledge and care gaps identified during the focus groups, in order to help tailor future policy and practice change in nutrition in SCD. With nutrition not yet integrated into the standard care in SCD, it was important to evaluate how well the health literacy framework was able to support the integration of nutrition in SCD, to answer research question 2: *How can a health literacy framework support the integration of nutrition in SCD?* Hence, phase four of the research project had two parts, the first part, to evaluate the health literacy framework and part two, to hold a third LA meeting, as reviewed below.

4.4.3 Third LAN meeting reflections- (12/12/20 - online):

The purpose of the third LAN meeting was to disseminate a summary of the preliminary findings of the research project, in line with the collaborative principle of the LA methodology to promote knowledge sharing, co-production and empowerment. No additional points were identified during this meeting. The participants expressed their gratitude to me for my efforts in undertaking the research project towards improving the nutrition service provision for people living with SCD. The third LAN meeting followed the evaluation of the Health literacy framework by the members of the LAN, presented below.

Part 2: Findings of the thematic analysis of the evaluation questionnaire

4.5 Evaluation questionnaire findings and themes

The findings of the evaluation questionnaire by the LAN, as illustrated in Table 4.4 below, were analysed to determine objective 2 and research question 2 (Chapter 2, section 1.2), as mentioned above. These findings represent the answers to each of the questions of the evaluation questionnaire (see Appendix 12), in response to the four questionnaire categories as mentioned in Chapter 3 (section 3.4.5). The four question categories and corresponding questions are found below;

- Current nutrition knowledge – Question (Q) 1, 2 and 3
- Change in nutrition knowledge – Theme Q 4, 5 and 6
- Influencing factors affecting nutrition and service provision – Q 7
- Value of the health literacy framework – Q 8

To answer objective 2, four themes and corresponding sub-themes were developed, see (Appendix 18), representing the final thematic mapping from the data analysis. The four

themes include (1) Improve knowledge and understanding of nutrition, (2) Raise awareness of influencing factors affecting nutrition, (3) Valuable tool for communication and training (4) Improve policy and service provision outcomes, effectively answering research question 2.

The purpose of the framework was to address the knowledge and care gaps in the nutritional management of people living with SCD, to improve nutrition service provision in SCD. The knowledge and care gaps in the nutritional management of people living with SCD has already been established in Part 1 above, to answer objective 1, to identify and understand the nutritional management of SCD and the socio – ecological factors and facilitators affecting nutrition and service provision in SCD. Below is an exploration of each of the four main themes, as listed in Table 4.4 below.

Table 4.4 – Summary of the findings and themes of the evaluation questionnaire:

Themes	Findings
Theme 1: (Q1+Q2): Improve knowledge and understanding of nutrition	➤ Not fully aware of the role of nutrition in SCD ➤ Limited knowledge ➤ Unaware of role of nutrition in SCD ➤ Now knowledge is better and bit broader ➤ Knowledge now improved ➤ Greater awareness
(Q3):	➤ Different viewpoints ➤ Learnt new things ➤ Knowledge further improved ➤ Learning from other patients ➤ Interesting to hear about different side effects of medication
Theme 2: (Q4): Raise awareness of the influencing factors affecting nutrition	➤ Learn how nutrition can be included in treatment ➤ Role of the focus group powerful ➤ Learning from others ➤ Need a more tailored approach ➤ Wider perspectives
(Q5+Q6):	➤ Problems are shared with other sickle cell patients ➤ Identification of gaps in care and policy ➤ Further work needed to improve training and awareness of nutrition ➤ Importance of nutrition education ➤ Nutrition not a priority and often overlooked ➤ Many barriers both internal and external ➤ More awareness of barriers in the community service provision ➤ The effect of money ➤ Importance of nutritional education for the patient, community and MDT ➤ Further work needed to improve training and build awareness
Theme 3: (Q7): Valuable tool for communication and training	➤ Valuable tool ➤ Clear guided conversations ➤ Improving awareness in primary and secondary care ➤ Vital and can lead to better training ➤ Provide value to many clinicians
Theme 4: (Q8): Improve policy and service provision outcomes	➤ Educational tool ➤ Standardising starting point ➤ Government lobbying ➤ Global coverage ➤ Needs to be deeply imbedded in clinical standards, academia, clinical practice, education, schools, colleges ➤ Provide to sickle cell patients, GP's, hospital doctors, parents, healthcare workers, social services

4.5.1 Theme 1- Improve knowledge and understanding of nutrition:

Theme 1 was developed from the amalgamation of the responses of the members of the LAN to Question (Q) 1, 2 and 3 (Chapter 3, section 3,4,5) of the evaluation questionnaire, illustrated in Table 4.4 above. It was apparent from the participant responses to Q1 and Q2, that the previous knowledge of the members of the LAN, about the role of nutrition in SCD, may have been substandard. Based on the responses of the members of the LAN to Q1 and Q2, it can be inferred that the knowledge before participation in the research project may have been limited with some members being unaware of the role of nutrition in SCD.

Judging by the responses to Q2 about the current knowledge of SCD, it can be implied that the current knowledge of the members of the LAN about the role of nutrition has improved and they now have a greater awareness of the role of nutrition in SCD.

It can be inferred from the responses in row one and two in Table 4.4 above, that the nutrition knowledge of the members of the LAN has changed. Furthermore, the responses to Q3 demonstrate that the involvement of the members of the LAN in the study, has contributed to the change in their knowledge and awareness about nutrition in SCD. In particular comments such as 'learnt new thing's, 'different viewpoints', communicated the value of being part of the LAN and hearing and learning from the different viewpoints of the other participants. It was evident that the involvement in the LAN was helping to raise awareness of the role of nutrition in SCD amongst the members of the LAN to promote shared learning.

Furthermore, it was clear that engagement in the study and the wider interactions of the members of the LAN as part of the study, influenced and promoted the collective learning of all the members of the LAN. The value of learning with and from others, was an important finding in support of the usefulness of the LA methodology reflected in the comment, 'interesting to hear about different side effects of medication' indicating that the participant may not have previously been aware of the side effects of the medication but their

involvement in the research project has added to their wider knowledge. Thus, the involvement of the members of the LAN, led to increased levels of nutrition knowledge and awareness, providing an opportunity for the members of the LAN to learn from each other.

4.5.2 Theme 2 – Raise awareness of influencing factors affecting nutrition:

Theme 2 was developed from the amalgamation of the responses of the members of the LAN to Question (Q) 4, 5 and 6 of the evaluation questionnaire, as illustrated in row three and four in Table 4.4 above. It was evident from the responses of the LAN, that their involvement in the co-development of the health literacy framework by the LAN, contributed to their learning about how nutrition could be included in the management and treatment of people living with SCD. In this way, the framework was therefore able to provide a more comprehensive understanding of the role of nutrition in the management and treatment of SCD, as well as the factors that affect the nutrition needs of people living with SCD. In addition, these responses clearly demonstrate how the framework is able to promote the education and learning of the members of the LAN. Their responses make a strong case to support the value and use of the health literacy framework in education and training in nutrition in SCD, potentially becoming an educational resource and reference for healthcare providers in SCD.

The responses from the members of the LAN in Table 4.4 furthermore provide clear insights into how the participants awareness of the influencing socio-ecological factors have enhanced their learning and thinking about the role of nutrition in SCD, offering them a real depth to their understanding of the problem. This is evident in these responses; ‘problems are shared with other sickle cell patients’, ‘the identification of gaps in care and policy’, ‘the need for further training and awareness of nutrition’, ‘the differences between community care provision and the acute’ and ‘the realization that nutrition is not seen as a priority’. Furthermore, the realization that many of the barriers or influencing factors to nutrition

integration were both 'internal and external', added to the depth in understanding of the factors influencing the nutritional management of people living with SCD. Therefore, the LAN's involvement in the study led to increased and enhanced levels of nutrition knowledge and awareness, and the need for tailored education and training about the specific factors affecting nutrition.

4.5.3 Theme 3 – Valuable tool for communication and training:

Theme 3 was developed from the responses of the members of the LAN to Q7 of the evaluation questionnaire, in row five of Table 4.4 above. Compelling evidence is provided in Table 4.4 to support the valuable role the framework is able to play as an educational and communication tool, thereby enhancing the engagement between the members of the LAN. This is evident in the collective responses of the members of the LAN which include the following: the framework is able to guide conversations clearly, 'it is able to improve awareness of healthcare professionals in both primary and secondary care', 'it is vital as an educational tool and can lead to better training' and significantly, 'it is a valuable tool that can be used by many clinicians'. In this way, the health literacy framework increased and enhanced the levels of knowledge and awareness amongst the members of the LAN, and in so doing, was deemed a valuable tool to support education and training, communication and policy development.

4.5.4 Theme 4 – Improve policy and service provision outcomes:

Theme 4 was developed from the responses of the members of the LAN to Q8 of the evaluation questionnaire in row 6 of Table 4.4 above. The responses from the LAN in Table 4.4 above, provide helpful insights to demonstrate how the framework is able to accommodate multi-stakeholder involvement in supporting the integration of nutrition into healthcare provision in SCD. Moreover, the responses also demonstrate how the framework

can work towards the improvement of policy and practice change to improve nutrition service provision in SCD.

The responses suggest that the framework can enhance outcomes through global coverage of the need to integrate nutrition into standard care in SCD in the following responses: 'it is standardized starting point for discussions', 'it can support government lobbying', and 'it can be embedded in a range of relevant policy and practice developments, in academia, clinical practice, schools and clinical standards'. Based on these responses, it is evident that the framework can be used by a range of people from sickle cell service users, carers, stakeholders and extending all the way through to health and social care professionals and clinicians to support the integration of nutrition in standard care in SCD, to achieve objective 2 and answer research question 2 (Chapter 1, section 1.2), mentioned above in (section 4.3).

Following the evaluation of the initial version of the health literacy framework by the members of the LAN (Appendix 11), I was able to conceptualise and develop a more comprehensive version of the initial health literacy framework, to demonstrate how nutrition can be integrated into standard care in SCD management.

4.6 Conceptualisation of health literacy as an expansive and integrative concept

4.6.1 Health literacy thematic concepts:

The consideration of health literacy as an expansive and integrative concept was a significant pivoting point of the research project (Chapter 1 and 2, see Appendix 13), and demonstrates my conceptualisation of health literacy as an expansive and integrative concept. As illustrated in Table 4.5 below, I used the four main themes developed from the focus group analysis (column one in Table 4.5 below), to espouse four corresponding health literacy thematic concepts (column 2 in Table 4.5).

Below is the theoretical reasoning underpinning my choice for each of the health literacy thematic concepts, to develop the health literacy Prevention Education Empowerment Contextual-factors Engagement (PEECE) model.

- Theme 1 and Theme 2 (focus group), represent the concept of Prevention (raising awareness of nutrition to reduce the nutritional challenges and risks of people living with SCD),
- Theme 3 (focus group), represent the concepts of Education and Empowerment (addressing the knowledge and care gaps in the nutritional management of SCD),
- Theme 4 (focus group), represents the concept of Contextual-factors (identifying and addressing the multilevel factors affecting nutrition and service provision)
- The fourth component Engagement, was based on the merging of the LA principles of Collaboration and Partnership (bringing together all the relevant stakeholders and service users, carers needed to co-produce policy and practice guidelines).

In this way, I used the four main themes of the focus group analysis as the theoretical basis for the conceptualisation of the novel health literacy PEECE model. The PEECE model, therefore, illustrates the expansive and integrative conceptualisation of health literacy, depicting a new way to understand health literacy and its role in policy and practice development in nutrition in SCD. As such, the corresponding health literacy thematic concepts formed the basis of the health literacy nutrition integration conceptual framework, illustrated below in Figure 4.4. In turn, the health literacy nutrition integration conceptual framework formed the conceptual basis of my input in the development of the Health Literacy – Nutrition Integration Framework (HL-NIF), a blue print for the development of nutrition service provision in SCD, to be discussed below.

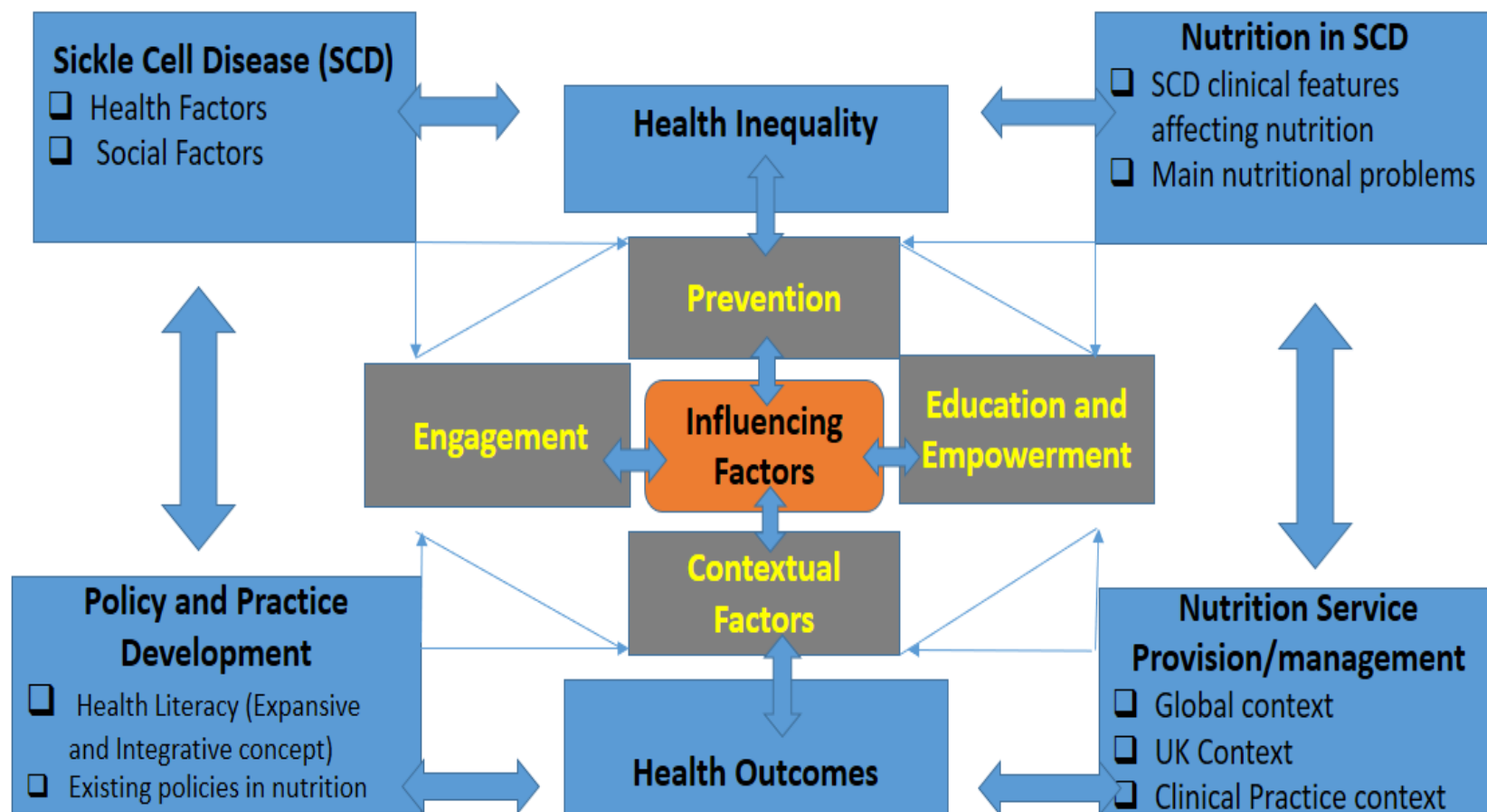
Table 4.5 - Espousing health literacy thematic concepts from the main themes of phase one

Main Themes	Health literacy thematic concepts (PEECE Model)
1. Invisibility of SCD 2. Under-recognised importance of nutrition	1.Prevention
3. Lack of priority to nutrition	2.Education and Empowerment
4. Multi-level factors affecting nutrition and service provision	3. Contextual Factors
5. Collaborative and partnership working	4. Engagement

4.6.2 Health Literacy Nutrition Integration Conceptual Framework:

The health Literacy - nutrition integration conceptual framework Figure 4.4 below, outlines the main theoretical perspectives included in Chapter 2, reflecting the current nutrition landscape in SCD, highlighting the main gaps from the critical evaluation of the literature. In this, the nutrition integration conceptual framework illustrates the main components required to build a foundation on which the nutrition in SCD landscape is to be underpinned, necessary to support the integration of nutrition into standard care in SCD. What the conceptual framework therefore does, is display the interconnectedness of the various components underpinning nutritional management in SCD. In this way, the conceptual framework: identifies the role of SCD (and the relevant health and social factors), Nutrition in SCD (emphasising the link between the clinical features and nutrition and the main nutritional problems), the need for nutrition service provision/management (and the related global and UK contexts) and the need for policy and practice to support nutrition service provision in SCD, a recognised gap in the nutrition in SCD landscape. Without policy and practice, the development of nutrition services in SCD is not possible, negatively impacting on the health outcomes of people living with SCD. In this way, the health literacy PEECE model, reflects the multiple influencing factors to be considered to contribute to policy and practice development, in support of nutrition service provision in SCD that is tailored to the nutritional needs of people living with SCD.

Figure 4.4 - Health Literacy - Nutrition Integration Conceptual Framework



4.6.3 Health literacy – Nutrition Integration Framework (HL-NIF) Blueprint:

Addressing the lack of nutrition service provision in SCD, by supporting the integration of nutrition in standard care in SCD is fundamental to the aim of the research project.

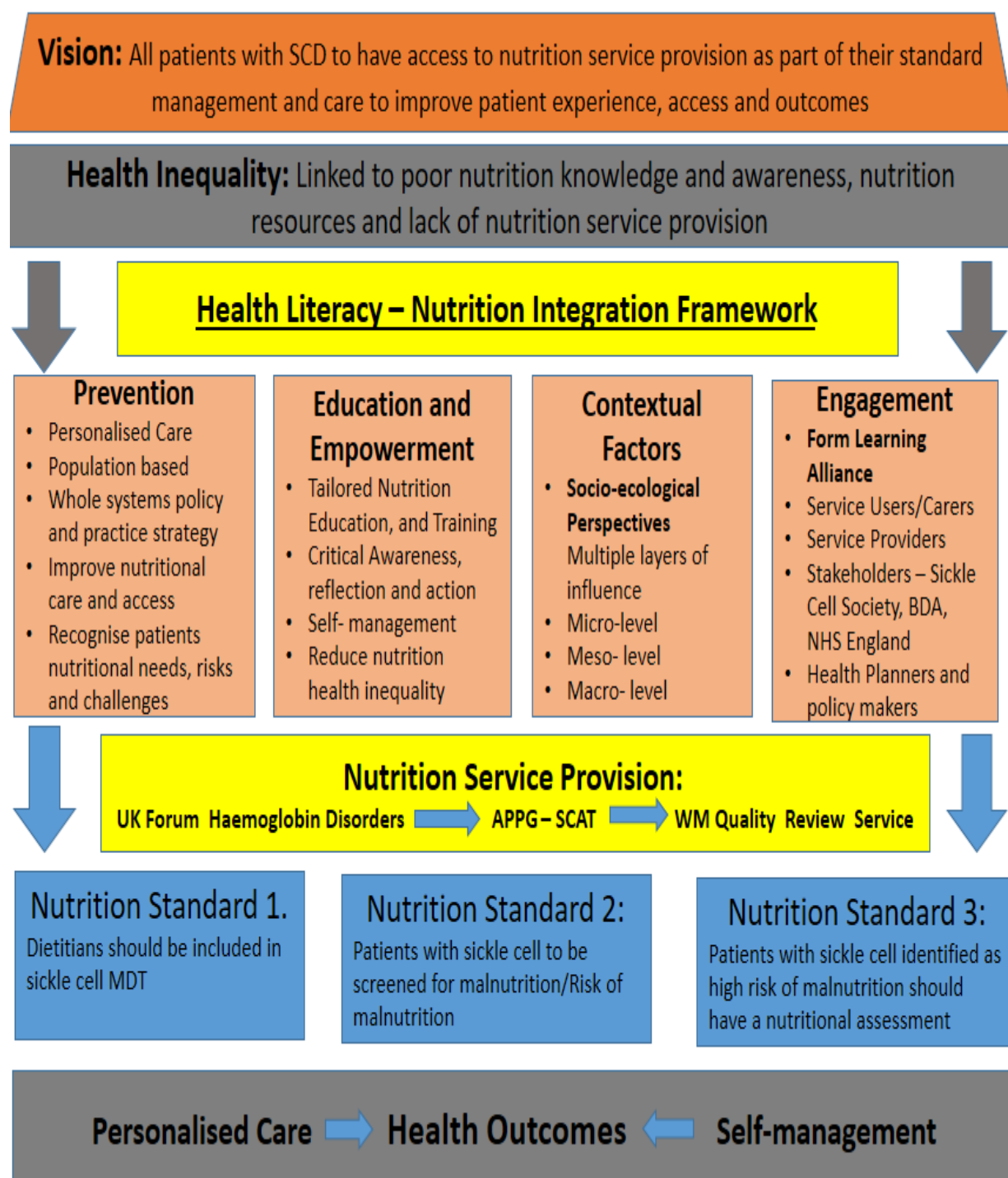
Therefore, the results reported thus far, illustrate the multiple influencing factors affecting nutrition and service provision in SCD reflecting the overall knowledge and care gaps that encompass the nutritional management of people living with SCD, collected from the knowledge, views and experiences of both the SCSU-Cs and SCSPs. My development of both the focus group themes, reflected in part 1 of this chapter and the evaluation questionnaire themes in part 2 of this chapter, collectively demonstrate how objective 1 and objective 2 has been achieved.

Thus the espousing of the health literacy thematic concepts from the main themes of the focus group analysis (Table 4.5 above), coupled with themes of the evaluation of the health literacy framework, underscore the theoretical basis of the development of the HL-NIF (blueprint). Inadvertently, this demonstrates the value of the initial health literacy framework (Appendix 11), in supporting the integration of nutrition into standard care in SCD. Building on the construction of the initial health literacy framework by the LAN, I used the health literacy PEECE model, reflecting the overall knowledge and care gaps of the research project informed by the main gaps in the literature (summarised in Chapter 2, pg66), to develop the HL-NIF (blueprint) illustrated in Figure 4.5 below. The HL-NIF is a blueprint to support the development of nutrition service provision in SCD, tailored to the needs of the SCD population. Simultaneously, the HL-NIF (blueprint) is able to highlight the marginalisation of SCD (invisibility and health inequalities) that forms a key part of the fundamental reasons for the lack of nutrition service provision in nutrition in SCD. In this way, the health literacy PEECE Model (see Table 4.5) is a central feature of the HL-NIF (blueprint), demonstrating the expansive and integrative conceptualisation of health literacy, integral to the development of nutrition service provision in SCD.

More importantly, the need to acknowledge nutrition in all three health domains is of paramount importance, if nutrition in SCD is to become more visible and integrated into standard care. Therefore, not only is a collaborative and partnership approach essential, nutrition service provision needs to be developed on a population level, adopting a whole systems policy and practice strategy tailored to the unique nutrition needs, risks and challenges of the people living with SCD. Moreover, the development of the HL-NIF (Blueprint) is aimed at addressing the myriad of knowledge gaps identified through the critical evaluation of the literature, as reported in the summary of Chapter 2 (pg66).

Hence, to bring all this information together, I will be developing a Health Literacy Nutrition Integration Report, titled '*Invisible and Overlooked*' to be presented to the APPG-SCAT (following the completion of research project) to raise the priority of nutrition in SCD management, but in the meantime, I have compiled a supporting letter (Appendix 10) and sent an email request to the APPG- SCAT to consider placing nutrition in SCD on their agenda for open discussion (Appendix 19, Example 2). The aim of the HL-NIF (blueprint) is reflected in my vision for nutrition service provision to address the health inequalities associated with a lack of nutrition service provision, that is patient centred, based on the national nutrition standards to promote personalised care and self-management to improve the health outcomes of people living with SCD.

Figure 4.5 Health Literacy – Nutrition Integration Framework (Blueprint) – Matthews, 2023



Summary:

The chapter provides a clear overview of the key influencing factors affecting nutrition and service provision reflecting the main knowledge and care gaps and recommendations drawn from the views, knowledge and experiences of the SCSU-Cs and SCSPs about the medical, nutritional and socio-ecological factors affecting nutrition and service provision in SCD.

The four themes demonstrated the impact of the marginalisation of SCD and the impact it had on the medical and nutritional management of people living with SCD. In addition to the four themes becoming the theoretical basis for the co-development of the health literacy framework and the HL-NIF (blueprint), the themes were instrumental in providing the theoretical basis for the espousing of the health literacy thematic concepts, to demonstrate the expansive and integrative conceptualisation of health literacy to support policy and practice development in nutrition integration into standard care in SCD.

The next chapter provides an in depth discussion of the main findings of the research project in the context of the marginalisation of people living with SCD, the influencing factors affecting nutrition, the socio-ecological factors affecting nutrition and the need for policy and practice guidance to support the integration of nutrition into standard care in SCD.

Chapter 5

Discussion

This chapter critically evaluates and discusses the findings of the research project, as reported in Chapter 4, the main influencing factors affecting nutrition integration – reflected in the core knowledge and care gaps in relation to existing literature. The four main points to be discussed include:

- the marginalisation of people living with Sickle Cell Disease (SCD)
- the influencing factors affecting nutrition in SCD
- socio-ecological factors impacting the integration of nutrition management in SCD
- supporting evidence for clinical and nutritional policy and practice change in the integration of nutrition management in SCD.

A discussion of the main limitations and strengths of the research project and reflections on my learning is provided, including the recommendations aimed at achieving the overarching aim and objectives of the study.

5.1 The marginalisation of people living with SCD

The marginalisation of SCD has serious health and management implications for people living with SCD in the UK and needs further recognition and better consideration. As such, patients living with SCD need to be recognised and considered appropriately in healthcare service provision. The avoidable death of a patient living with SCD as reported in the APPG-SCAT (2021) report, bears witness to the impact of the marginalisation and underlying racism and invisibility (poor recognition and lack of consideration) of people living with SCD in relation to their medical management. Similarly, the findings of this research project brings the lack of nutrition service provision available to people living with SCD under scrutiny, thus potentially adding to the pre-existing marginalisation, as experienced in the low level and

quality of medical provision reported in the APPG-SCAT (2021) report. When the medical needs, as well as the nutritional needs, of people living with SCD are ignored and not considered, the patient voice is lost; this is in direct conflict with the call from the NHS Long Term Plan (2019) for personalised care.

Personalised care is aimed at giving people control over their healthcare needs and provision, giving them a voice, thus recognising and considering them as having a central position in healthcare provision. Central to personalised care are the values upheld by the NHS constitution. According to these values, healthcare professionals are to work closely with patients, treat patients with dignity, respect and compassion, where all patients count and care provision is geared towards improving lives, and not resulting in avoidable patient deaths. Both the values and standards of the NHS Constitution therefore present clear guidance relating to the quality of care provision to ensure all patients, including patients living with SCD, receive a high level and quality of care. Unfortunately, the untimely death of the SCD patient (APPG-SCAT, 2021) provides the opportunity to reflect on how poorly the NHS values are being applied to this patient group in healthcare services, as a result of the marginalisation and lack of recognition and consideration given to patients living with SCD.

In conjunction with personalised care, the House of Care model (The Kings fund, 2013) demonstrates the need for patient centred care, where the providers and patients work collaboratively and in partnership with each other to tailor service provision to the needs of the patients. This highlights the fact that all the influencing factors affecting the patient needs (both medical and nutritional, as highlighted in this research project) need to be considered in this mutual healthcare relationship. From this research project, it is evident that the nutritional needs of people living with SCD are not considered as part of standard care provision as witnessed in the lack of nutrition knowledge, resources and nutritional care provision available to the SCSU-Cs in the research project. However, the APPG-SCAT (2021) report, as did the findings of this research project, reveal failings in opposition with the NHS Long Term Plan calling on personalised care, evident of the inequalities reflected in

poor patient experiences, access and poor health outcomes in the medical management (APPG-SCAT, 2021)– and the inequalities identified in this research project in relation to nutritional management in SCD. Evidently, the APPG-SCAT (2021) report does not include the nutritional management of people living with SCD, and in this way, the research project findings both confirms the nutritional needs of people living with SCD and adds to the findings of this important report. Therefore, taken together, people living with SCD are not able to take control of their healthcare needs (both medical and nutritional) and as such, not empowered to call out inequalities in their care provision.

In this research project, the disparity in access to care and support between cancer patients and people living with SCD, with cancer patients having more access to services, was identified by a SCSP (who is also a SCSU-C), despite the two patient groups commonly being banded together and being under the same service providers (section 4.2.2).

However, these discrepancies in healthcare may reflect the lower level of advocacy for specialised care for people living with SCD. In this way, these healthcare disparities are impacting on the quality of healthcare provision available to people living with SCD. These disparities in healthcare access are also reported in a paper by Lee et al., (2019) in which a range of health care disparities in SCD were identified, where SCD was compared to chronic conditions like cystic fibrosis and haemophilia, in relation to funding and having more services available for these patients to manage their condition. In comparison to cystic fibrosis, a genetic condition that has a smaller prevalence to SCD both in the UK and US, SCD lags behind on so many levels (Power-Hayes and McGann, 2020; Lee et al., 2019; APPG-SCAT, 2021 p37). In the US for example, cystic fibrosis receives more funding and pharmaceutical investments (3.5 times more - National Institute for Health) and 440 times from national foundations when compared to SCD (Lee et al., 2019), although there have been recent changes to legislation in the US and the UK. The UK, now has 12 Haemoglobinopathy Coordinating Centres (HCC's), to lead, co-ordinate, support and promote how hemoglobinopathy services are delivered UK wide.

The marginalisation of people living with SCD is not however limited to the medical healthcare needs, as identified in this research project. From the study findings, a range of social and wider determinants of health are identified that influence the nutritional needs of people living with SCD, which has not been explored in the existing scientific corpus on the role of nutrition in SCD, hence adding to the complexity of the nutritional management of people living with SCD. In acknowledgement of the social and wider determinants of health at play in SCD, sociologists implore the value of considering the effect of race and ethnicity (Taylor and Field, 2007; Walsch and Tait, 2014; Craig et al., 2012; Chattoo et al., 2019) as well as social class when considering healthcare provision to marginalised patient groups, such as people living with SCD. For this reason, the holistic lived experience of people living with SCD is of paramount importance and the compartmentalisation of the various needs, for example the medical versus nutritional versus ethnicity, should not be done in isolation but be considered holistically. Therefore, it is imperative that all the needs (medical, nutritional, social and economic) of people living with SCD are considered, to accurately reflect their lived experiences in order for the breadth of these influencing factors to be recognised and considered and deemed significant enough, and reflected in their overall care provision.

Important to note, is the fact that over 60 years' worth of existing scientific research on the role of nutrition in SCD and the nutritional needs of people living with SCD already exists, showing explicit causal links between the clinical features of SCD and the nutritional implications in SCD (Hyacinth, Gee and Hibbert, 2010). However, it has been recognised by Hyacinth, Gee and Hibbert (2010) that, whilst the medical management of SCD has developed over the years, the nutritional management in SCD has largely remained overlooked and ignored with little consideration given to the significance of the nutritional needs of people living with SCD, evident in the findings of this research project, reflecting the under recognition of the importance of nutrition in SCD management. In this way, the people living with SCD need to be empowered to play a central role in how their nutritional needs are recognised, considered and managed, when personalised care is emphasised in SCD

management. Thus, in order for this to happen, the nutritional needs of the people living with SCD need to be recognised as part of their overall holistic lived experience.

Importantly, the findings of this study provided clear evidence in support of the fact that nutrition is not recognised and considered as part of the standard care in SCD (Prasad, 1997; Hyacinth, Gee and Hibbert, 2010; Khan et al., 2016; Ohemeng and Boadu, 2018; Umeakunne and Hibbert, 2019; Kamal et al., 2021), evident in the number of calls from SCSU-Cs for more and bigger conversations and opportunity to talk about their nutritional needs. Moreover, reports of the delay in highlighting the diagnosis of nutritional problems (Chapter 4, section 4.2.2) by both an adult SCSU and SCSU-C, (a parent of a child with SCD) albeit only 2 participants, points to the inherent potential risk open to several other patients living with SCD, who may also have undiagnosed nutritional risks, overlooked in the absence of nutritional service provision as part of standard care in SCD. Hence, adopting a reactive approach to nutritional care potentially places vulnerable people living with SCD at an even greater risk of a late diagnosis of nutritional problems – thus adding to the increased risk of morbidity, mortality, disability and poor QOL outcomes (Fitzburgh et al., 2005; Lucas and Mason, 2008; Aljuburi et al., 2012; Piel et al, 2021). Hence, a SCSU-C suggested nutrition service provision in SCD take a more proactive and preventative approach, rather than a reactive approach, as mentioned in Chapter 4 (section 4.2.1).

For example, if the paediatric SCD patient's vitamin D levels (reported in this research project) were checked as part of ongoing nutrition service provision, the risk of becoming dangerously low would have been avoided. Similarly, osteoporosis is a known co-morbidity among the adult SCD population (Eskiocak, Yilamz and Ilhan, 2022), however in the case of this particular SCSU-C, there was an oversight in highlighting their risk for developing osteoporosis, that came as a shock to them, which could have been averted if the value and importance of nutrition was part of standard care provision, including nutritional surveillance of people living with SCD. Furthermore, a late diagnosis of osteoporosis can add to the already high cost, healthcare and co-medication burdens, but this could possibly be averted

if diagnosed and addressed in a timely manner and if patients' vitamin D levels and other nutritional needs were checked as part of nutrition service provision among others. Low vitamin D, diet, haemolysis, inflammation, being underweight, and hypogonadism are key risk factors for the development of osteoporosis in patients living with SCD (Eskiocak, Yilamz and Ilhan, 2022). In this way, this finding serves to highlight the need for more surveillance of the nutritional risks in SCD, and that nutrition should not be considered in isolation of the clinical features inherent in SCD.

This amplifies the calls inherent in the responses of the SCSU-Cs that they wanted more nutrition service provision. Similar to the SHAPE study (2022), a multinational qualitative study conducted with nearly 1000 people living with SCD, a few of the SCSU-Cs identified the need for SCD to be recognised and considered as a long-term condition. Additionally, one of the SCSU-Cs acknowledged the importance of nutrition for their mental wellbeing (Chapter 2, section 4.2.1), since in particular, COVID-19 was found to be a significant influencing factor in regards to the mental health and wellbeing of the wider population (Ham, 2020) as well as of the SCD patient population (APPG-SCAT, 2020). Moreover, the interest in SCD and its infection risk being brought to the forefront of healthcare management raised surprise with one of the older SCSU-C participants (section 4.2.1). On the other hand, a SCSP (service user) shared that they were grateful because, during that time, their employer was able to understand the significance of their long-term condition and could finally understand the impact of a person living with SCD (section 4.2.1). Furthermore, the APPG-SCAT (2021) report highlighted the levels of mistrust, which exists between service users and providers due to previous negative experiences in their care provision.

Paradoxically, a SCSU-C reflected on their concerns that if nutrition could 'cure' SCD symptoms, it would have a negative impact on a patient's eligibility to apply for personal independent payments, as reported in Chapter 4 (section 4.2.1) and in so doing, significantly reduce access to disposable income, especially since the cost of living crisis and food insecurity are already a problem in marginalised groups (Coleman- Jenson, 2020). A further

concern raised was around the quality of hospital food and the lack of access to nutritionally-adapted meals when being hospitalised, particularly as the appetite of people living with SCD is prone to be reduced as a consequence of upregulated pro-inflammatory interleukin 6 due to disease modifying inflammation (Hyacinth, Gee and Hibbert, 2010). Poor appetite among SCSU-Cs, when feeling unwell, was also a key finding reported in this research project in Chapter 4 (section 4.2.2), in line with existing scientific literature (Hyacinth, Gee and Hibbert, 2010).

In contrast to the SCSU-C responses calling for more nutrition service provision, the responses of the SCSPs were instead geared towards explaining why there was a lack of nutrition service provision (chapter 4, section 4.2.2). A key finding, they highlighted was the importance of having a SCD consultant who was 'pro-nutrition'. When SCD consultants are 'pro-nutrition', they are open to inviting Dietitians to be part of the SCD MDT, thereby increasing the visibility of nutrition as part of ongoing SCD management. More so, the availability of Dietitians to identify and manage the nutritional risks of people living with SCD offer patients a central role in their care provision. Therefore, the call for Dietitians to be part of the SCD MDT, as reflected in Standard 1 of the National Nutrition Standards in SCD (SCS, 2018, pp. 75-77) is possible when SCD consultants are 'pro-nutrition'. This means that SCD consultants are important gatekeepers to integrating nutrition into standard care provision in SCD and placing the patients at the heart of the care provision. In this way, other members of the SCD MDT, may also have a role in advocating for the nutrition needs of the people living with SCD in their care.

Taken together, poor access to appropriate nutrition or nutrition management amounts to compelling evidence in support of the need for nutrition service provision. Thus, the findings of this research study suggest that both SCD and SCD nutrition are impacted by marginalisation, as evidenced in the lack of knowledge about SCD and nutritional needs of people living with SCD, which themselves may stem from the invisibility of SCD (under recognition) and lack of consideration in the wider healthcare landscape. Poignantly, these

findings mirror those of the APPG-SCAT (2021) and may suggest that our results are also a consequence of the marginalisation of people living with SCD, with the similar risk of poor health outcomes and health inequalities affecting the experiences, access and outcomes of people living with SCD. However, distinctly, the APPG-SCAT report does not recognise or consider the significance of the role of nutrition in SCD management or how the nutritional needs of patients affect their health and wellbeing outcomes. The findings of this study complement those of the APPG-SCAT report by adding the nutrition component: the nutritional needs of people living with SCD are currently being overlooked, not considered and therefore neglected within the standard healthcare provision in SCD. Hence, the research project is able to add to the breadth of influencing factors affecting the integration of nutrition in SCD as well as those factors impacting the health and wellbeing of people living with SCD, over and above their medical needs. Similarly, the myriad of social and wider determinants of health factors already identified provide compelling evidence to adopt a patient centred, personalised care approach in SCD management., which would equally serve to reduce the incidence of the marginalisation of people living with SCD.

5.2 Influencing factors affecting nutrition in SCD

Despite the existing literature highlighting the existence of the nutritional needs of people living with SCD, evidence of the neglect of nutrition in SCD is reflected in a number of papers calling for nutrition to be part of the standard care provision in SCD (Prasad, 1997; Hyacinth, Gee and Hibbert, 2010; Khan et al., 2016; Ohemeng and Boadu, 2018; Umeakunne and Hibbert, 2019; Kamal et al., 2021), and is reflected in the findings of this research project. This research project is therefore the first attempt to actively address this problem. In this way, the findings of this study as reported in Chapter 4, provide primary data to highlight the influencing factors and gaps in nutrition in SCD, with the aim to address this significant oversight in the nutritional management in SCD. A range of influencing factors

reflected in knowledge and care gaps were identified in the medical and nutritional management in SCD, from the analysis of the focus group data as reported in Chapter 4, clearly marking the need for policy and practice tailored to the unique nutritional needs of people living with SCD, to be discussed below.

5.2.1 Medical management:

(i) Links between SCD and SCD nutrition:

The lack of knowledge about SCD and SCD nutrition, to support the nutritional management of people living with SCD in healthcare management, was reported in both the SCSU-C and SCSP groups. Nearly all of the participants in both groups reported having had to self-research what they now know about both topics, that has enabled them to self-report a good level of knowledge about the complications of SCD, demonstrating the value of having readily available medical and nutritional resources in SCD. A key distinction was made by a SCSU-C and SCSP who emphasised the variability of SCD and that management in SCD is not a one-size-fits-all approach. As such, a personalised care approach in SCD management is indicated to help tailor the care provision to the patient's individual needs. In keeping with Aljuburi et al., (2012), a SCSU-C raised the point that people living with SCD are now living longer, thanks to improvement in healthcare provision in SCD. However, this means that people living with SCD may experience 'different' medical needs related to ageing, over and above the SCD-related complications that in turn, may exacerbate their increased risk of morbidity, mortality, poor QOL and disability (Fitzburgh et al., 2005; Lucas and Mason, 2008; Aljuburi et al., 2012; Piel et al, 2021).

In relation to knowledge gaps, it was evident that when SCSPs were not part of the SCD MDT, their knowledge of the condition was limited. In contrast, when the Dietitian for example worked as part of the SCD MDT, their knowledge about SCD was increased, and moreover, they were able to educate the SCD MDT about the role of nutrition in SCD

(section 4.2.2). Whilst the medical knowledge of a SCSP who worked as part of the SCD MDT was good, their knowledge of nutrition in SCD was significantly reduced, this was also true for the SCSP, a GP who worked in primary care, reporting nutrition was an overlooked management option in the existing management protocols. The lack of knowledge of nutrition in SCD amongst the wider healthcare professionals was also identified as a problem by a SCSU-C, and reported that healthcare professionals were at risk of providing misinformation to the people living with SCD and carers (section 4.2.1).

Moreover, a paediatric SCD nurse specialist (as part of this research project) shared that more effort is needed to include nutrition education in schools to help raise awareness of the value of nutrition in SCD to enhance patient outcomes (section 4.2.3) and not only focus on fluid, which is current practice, but also include the nutritional needs of the school children living with SCD. This in turn may help to reduce the stigma experienced by children living with SCD. Stigma amongst SCD pupils was also indicated by a SCSP (section 4.2.2), who suggested to have more awareness of SCD in schools through, for example, show and tell activities (section 4.2.3).

Thus the knowledge gaps identified in this research project, confirm the need for education and training about nutrition in SCD, that should not to be limited to healthcare professionals in Health and Social Care but extended to educational institutions and places where people living with SCD are likely to be encountered, as reflected in the national clinical standards for education in SCD (SCS, 2018, p78). Education and training should also be available to people living with SCD, as part of their standard care provision. This underscores the need for more knowledge and awareness of the impact of SCD and SCD nutrition to support the health and wellbeing outcomes of people living with SCD, especially considering the effect this can have on the school attainment of school children living with SCD. Poor education attainment in SCD was recognised as a significant problem in SCD (APPG, SCAT, 2009), whilst education is also identified as a wider determinant of health (Dahlgren and Whitehead,

1991), impacting on the potential long term employment prospects of people living with SCD (Berghs, et al. 2021; Berghs and Dyson, 2022; Pirez et al., 2022).

Thus, the lack of knowledge of SCD and SCD nutrition is a key factor affecting the recognition and consideration given to the role of nutrition in SCD management. More importantly, the link between SCD and SCD nutrition needs to be more explicitly emphasised, as noted in improvements in patient access to nutritional management when Dietitians are part of SCD MDTs. In this way, the research findings, as reported in the knowledge, views and experiences reflecting the SCSU-Cs voice, alongside the voice of the SCSPs, need to be acknowledged in the calls for more nutritional management in SCD. This is emphasised in the national nutrition standards, (SCS, 2018, pg 75-77; Matthews, 2019k), the only policy document about nutrition in SCD presently, calling for Dietitians to be part of the SCD MDT.

5.2.2 Nutritional management:

(i) Care gaps:

The individual and collective responses of the SCSU-Cs and SCSPs highlight a range of care gaps in the nutritional management of people living with SCD to be discussed below. The care gaps identified through the analysis of the findings, in particular Theme 2: ‘under-recognised importance of nutrition’ (section 4.2.2), reflect the implications of when nutrition is not part of the standard care provision for people living with SCD. As a consequence, the nutritional needs, risks and challenges linked to nutrition were not identified by SCD clinicians and healthcare professionals, as meaningful clinical outcomes to be included in the overall management of people living with SCD.

The importance of demonstrating the connection between nutrition, health and SCD (section 4.2.2), in facilitating the recognition of the nutritional needs of people living with SCD, was

identified by a few of the SCSPs. Nutrition amongst others have been identified by Tanabe et al., (2010) as an important self-management practice of people living with SCD to improve health and wellbeing outcomes. Similarly, the relevance of this connection was highlighted in the first direct study on the role of nutrition in SCD, conducted by Heyman et al., (1985), albeit a small study with only five participants who had growth delays. The findings of this study provided compelling evidence of the benefit of nutrition in addressing the nutritional deficits of the study subjects when given an enteral nutritional supplement. Inadvertently, the study showed a clear link between the clinical features of SCD and the improvement in clinical outcomes when nutrition support was offered as a management option. Interestingly, among our research participants, two of the SCSU-C participants received Percutaneous Endoscopic Gastrostomy (PEG) feeding, due to delayed growth. Yet, although the clinical and nutritional outcomes were improved in both these study participants, they commented on the timing of the feeds, for example interfering with their day time eating, indicating again the need for a personalised approach to care, which Dietitians can offer. Moreover, one of the SCSU-Cs also pointed out that the beneficial nutritional advice they were given, ie. to eat more energy rich and more protein rich foods, was not done in a timely manner and could have been addressed earlier in their care (section 4.2.2), more evidence in support of adopting a proactive nutritional care provision than a reactive care provision.

In the review by Hyacinth, Gee and Hibbert (2010), the authors identify clear links between the nutritional implications in SCD and the pathophysiology of SCD. Indeed, the authors describe the under nutrition in SCD as being 'a critical feature' and a 'significant complication' (pg 58) of SCD. This description of the under nutrition in SCD means that in the same way that the clinical features of SCD (chronic haemolysis, or vaso-occlusion) for example are significant problems in SCD management, under nutrition also warrants management as part of the standard care in SCD. Equally, if under nutrition is a significant complication in SCD, as is avascular necrosis or leg ulcers which are part of standard management, then it needs to be considered as such.

Unfortunately, according to a few SCSU-Cs, the focus of the medical management of SCD is more on the medical and pharmaceutical treatment of the condition. In particular, one of the SCSPs (a GP) indicated that the medical management in SCD is protocol-driven, with protocols for pain and blood transfusions for example, but without any mention of the nutritional needs of the patients at all (section 4.2.2). As one SCSU-C put it, *'nutrition is pushed aside and not factored into the medical management of SCD'*, and the outplay of this under-recognition and lack of consideration given to nutrition has resulted in one of the SCSP (a service user) reporting that they had received 'zero' nutritional input as part of their standard SCD care (section 4.2.2). In addition, they reported that referral to Dietitians also does not happen for people living with SCD, further increasing the risk of their nutritional problems worsening, which may contribute to their risk of malnutrition (both under and over nutrition), necessitating the need for more nutritional surveillance in standard SCD management. For this reason, nutrition standard 2 of the national nutrition standards call for people living with SCD to be screened for any possible nutritional risk, by adequately trained staff such as Dietitians (SCS, 2018, pg 75). The research findings therefore providing evidence that people living with SCD may not be routinely screened for their nutritional risk, further evidence that people may not be aware of the existence of the national nutrition standards in SCD management.

What is overwhelmingly clear is that the SCSU-Cs were keen for nutrition service provision to be part of their standard care. A number of the SCSU-Cs reported that nutrition is paramount and fundamental to their health and an important part of remaining well (section 4.2.2). This was particularly true for one specific SCSU-C, who was not on any medical treatment for their SCD and relied on eating well to remain well. However, many of the SCSU-Cs reported that nutrition was not asked about at their routine appointments - they were not asked about their nutritional needs or concerns, and subsequently, there was no scope to talk about their nutritional needs or concerns. Instead, the only mention markedly related to nutrition was the emphasis on drinking enough fluids to prevent dehydration,

recognised as a trigger for vaso-occlusive crises, in line with existing health promotion advice (NICE, 2021b; NHS choices, 2022), illuminating the limited view to the importance of nutrition as a management option in SCD care provision.

This observation was in alignment with the findings of other research (Mitchell, et al., 2004), where caregivers of children with SCD were given water at the expense of nutrition, because in that moment, the risk of vaso-occlusive crisis was more of a priority for them. This is an important point to consider when advice and support are being offered to caregivers of children experiencing or recovering from sickle cell crisis, and having Dietitians trained in SCD and working as part of the SCD MDT can play a valuable role in reducing a patient's risk of malnutrition, thereby enhancing patient outcomes, by adopting a personalised approach to care. For this reason, the nutritional needs of all people living with SCD need to be recognised and considered in SCD management. In the article by Ievers and Landis (2001), 93% of the parents of SCD children in the study reported nutritional problems, however no advice or support was offered to these parents in response to their children's nutritional needs, highlighting a more serious concern. Nutrition standard 3 of the national nutrition standards calls for the nutritional management of those people living with SCD deemed to have or be at risk of malnutrition, by adequately trained staff such as Dietitians (SCS, 2018, p 75-77), and in this way, the findings of the research project affirm the nutritional needs of people living with SCD and the need for nutrition to be considered as part of standard management in SCD.

Thus, calls for bigger conversations about nutrition in consultations and follow-up appointments (Section 4.2.2) from a number of SCSU-Cs draws attention to the need for nutrition to be part of standard care provision in SCD. More importantly, for these conversations to happen earlier, and regularly, for example to be included in the annual review appointments, as a way of reinforcing the benefit of nutrition in good health. These calls came from both the male and female SCSU-Cs, although it is believed that men and women differ in their beliefs and ultimate engagement in preventative health behaviours

(Korin et al., 2013), demonstrating the importance assigned to nutrition in SCD care provision. Moreover, Umeakunne and Hibbert (2019) argue that nutrition be considered an ‘adjunct’ to the medical management of SCD and not an ‘either/or’ scenario; in this way nutrition is able to complement the medical management in SCD. Kamal et al., (2021), the most recent paper to call on nutrition to be part of the standard management of SCD, identifies nutrition as an independent indicator of the severity of SCD that warrants inclusion in the standard care of people living with SCD. In the article, the identification of the macro and micro nutrient deficiency was encouraged, further evidence of the importance of the nutritional needs of people living with SCD, as confirmed by the nutritional care gaps identified in this research project findings.

As previously mentioned, several articles support the view that despite the ‘obvious’ presence of the nutritional needs of people living with SCD (Prasad, 1997; Hyacinth, Gee and Hibbert, 2010; Khan et al., 2016; Ohemeng and Boadu, 2018; Umeakunne and Hibbert, 2019; Kamal et al., 2021), nutritional management is overlooked and it is time for SCD clinicians to consider the role of nutrition alongside the medical management of SCD. The review article by Hyacinth, Gee and Hibbert (2010) further suggests a nutritional supplement that contains both macro and micro nutrients to improve the nutritional management of people living with SCD, whereas Khan et al (2016) specifically mentions the need to advise patients on how to eat well on a low income, recognising the impact of the socio-economic status of people living with SCD. Collectively, these authors illustrate the variety of nutritional needs and interventions required by people living with SCD. In this way, the research project findings reflect the need for SCD management to be more holistic and not only focus on pain management for example, but for nutrition to be part of SCD clinical and nutritional policy and practice. A SCSP further suggested that nutrition be mandatory, whereby nutrition is a topic addressed at medical appointments, patients can be followed-up and referred to Dietitians for identification, assessment and treatment of the individual’s nutritional needs (section 4.2.3).

(ii) Knowledge gaps:

The development of Theme 3: lack of priority to nutrition (section 4.2.2), is based on the fact that nutrition is not considered a viable problem evident in the lack of nutritional management, education and resources reported by many of the participants in both the SCSU-C and SCSP groups. Furthermore, the influencing factors reflected in the care and knowledge gaps identified from the data analysis give the impression that the nutritional management in SCD is also not considered important enough to be treated as part of standard care, evident in the knowledge gaps to be discussed below.

As previously mentioned, nearly all of the participants of both the SCSU-C and SCSP focus groups reported having to self-research what they know about SCD and nutrition in SCD. For SCSU-Cs having to self-research what they know, also means there are no structures and services available to provide this essential information to help them self-manage their nutritional needs, risks and challenges, as part of their standard care provision. The lack of structures, resources and services available to people living with SCD will have a negative impact on their experience, access and outcomes in relation to the role of nutrition in SCD. Furthermore, the quality and accuracy of the resources that are being considered add to the level of risk to these patients, in the nutritional choices that they make to self-manage their nutritional needs. It was clear that more nutrition resources are needed about nutrition in SCD, however, collectively the SCSU-C and SCSP participants were able to mention a few of the more common nutritional problems in SCD, such as anaemia, fatigue, dehydration, and vitamin deficiencies (vitamins A, C, D). The SCSU-Cs identified pica as a problem that occurs in paediatrics and pregnancy in SCD. Pica is a problem identified as having an unusually high prevalence in people living with SCD with a recognised correlation to lower Haemoglobin levels (Ivascu et al., 2001), further evidence of the need for nutritional management to extend across the life course of people living with SCD. The Dietitians, in the SCSP group, also added the gastrointestinal problems associated with the side effects of the

analgesia used to treat the sickle cell crisis, such as constipation, usually treated with regular laxatives (NICE, 2021b), as a nutritional problem to be considered.

The findings above, therefore add to what is reported in an article previously published, (Matthews, 2015b), about nutrition in SCD. The article, written by a Dietitian with an interest in SCD nutrition, highlights the nutritional assessment of people living with SCD to be multi-factorial, and mentions frailty as a potential risk in people living with SCD, particularly young adults requiring hip replacement due to complications of SCD such as avascular necrosis. Frailty is not commonly linked to younger people, and therefore young people living with SCD can fall through the loop and the impact of their disability not adequately considered, in a nutritional context. As such, the research findings provide evidence in support of the multi-factorial nature of nutrition in SCD and the complexity of nutritional assessment and management of people living with SCD.

In the present research, the lack of nutrition knowledge among clinicians was identified as a problem, in particular, one SCSU-C shared that they arrange training for healthcare professionals about the role of nutrition in SCD, because they mostly focus on the medications which often times have side effects for patients (section 4.2.2). Moreover, a second SCSU-C reported that they often times know more than the healthcare professionals they see, attesting to the knowledge gaps that may affect the level and quality of care experience for those attending and accessing care provision in SCD. Taken together, links between nutrition and health outcomes of people living with SCD need to be made as part of standard medical care provision.

The fact that nearly all SCSU-C and SCSP participants reported having to self-research what they know about SCD and nutrition in SCD, is also evidence to the lack of priority of patients' nutritional needs in their care provision. In addition, the lack of awareness by health policy planners and clinicians, despite a rich literature on the role and importance of nutrition in SCD, is reflected in the nutrition knowledge gaps identified in this research project. Furthermore, despite the national nutrition standards for SCD being included in the national

clinical standards for the management of adults with SCD (SCS, 2018), the nutritional management in SCD remains overlooked as a management option, and not part of standard of care. This continuous under recognition of nutrition as a management option in SCD, is extended to the UK Forum on Haemoglobin Disorders Quality Standards (2021) report, used to assess the quality of service and care provision to people living with SCD in the UK. In these standards, reference is made to Dietetics (identified as support services in the report), to be requested as required. However, there are no specific management guidelines included in the quality report, and there is no mention about the need for the nutritional needs of people living with SCD to be identified, assessed through screening or if any risk is identified for these patients to be treated and managed by adequately trained staff such as Dietitians (identified to be included in the SCD MDT (SCS, 2018, p75-77).

Therefore, from a professional practice perspective, the value of the role of the Dietitian is illustrated in the document by the (BDA and BSNA, 2013) as a way of helping the NHS to meet Domain 2: improving the QOL outcomes of patients living with long-term conditions (DHSC, 2022). Since SCD is a long-term condition, with existing poor QOL outcomes secondary to the pathophysiology of their chronic condition, it warrants the service provision by Dietitians, to help improve patient outcomes through appropriate nutrition screening, assessment and intervention. Similarly, a previously published scoping cross-sectional survey (Matthews, 2016g) found that 96% of the responding Dietitians reported that SCD warrants Dietetic input. However, the survey also identified the poor knowledge of these Dietitians (78%) of SCD, exposing the need for more evidence-based resources to be available to support the nutritional management of people living with SCD. However, for this to become a reality, closer links between research findings and clinical practice are needed (Nartey et al., 2021), which this research project aims to achieve through the main study output: the health literacy framework co-developed by the Learning Alliance (LA) network formed between some of the members of the SCSU-C and SCSP groups respectively.

This significant oversight of the recognition of the need for nutritional management in SCD, adds to nutrition in SCD becoming a health inequality that needs to be addressed, urgently. Similarly, to how the APPG-SCAT (2021) report relies on the SCD patients' voices to make a sound argument of the failures in their treatment, this research project uses the SCSU-C and SCSP voices to provide sound evidence of the current *status quo* of the nutritional management landscape in SCD in the UK. As such, the findings of this research project report the knowledge gaps of nutrition in SCD and the requirement of urgent development of healthcare professional education and training on the nutritional management in SCD, tailored to the unique nutritional needs of people living with SCD. Currently, advice given to people living with SCD is reported to be more generic as mentioned by both the SCSU-C and SCSP groups. In response to improving the generic advice given to people living with SCD, the participants suggested that training be extended to the wider MDT to raise awareness of SCD and SCD nutrition and in so doing, support onward referrals of patients who may be at risk of nutritional problems. Hence, the time is now to listen to and acknowledge the SCD patients' voices and their calls for their nutritional needs, risks and challenges to be recognised and considered. When the lack of nutrition service provision in SCD is acknowledged, then the importance of the nutritional needs of people living with SCD can be validated and acted upon, and their nutritional needs met as part of their standard care provision in SCD that is personalised and tailored to their individual needs. An additional benefit of the research project is adopting a comprehensive view to reflect the complexity of the nutritional needs and management of people living with SCD to be discussed below.

5.3 Socio-ecological factors

This study used a socio-ecological lens to explore nutrition and service provision in nutrition in SCD, to provide a comprehensive lens to identify and understand the myriad of influencing

factors affecting nutrition and service provision in SCD. The development of the socio-ecological model, by McLeroy et al., (1988), depicts a number of layers of influence including the personal, interpersonal, institutional, community and policy levels, that influence the healthcare needs and experiences of patient populations. Moreover, a socio-ecological lens is advocated when considering health in populations from a BEM background (Greenhalgh, 2009; Early, 2016).

Since SCD is considered as a marginalised group (APPG –SCAT, 2021), it is equally important to consider the key social concepts that are pertinent to the health and healthcare experiences of the SCD population. In addition, Berghs et al., (2020) calls for renewed efforts to not only consider the biological factors associated with SCD, but to extend this understanding of SCD to consider the effects of the social determinants of health and how it impacts on the medical management of people living with SCD and its associated health outcomes (Sonnenklar et al., 2022; Khan et al., 2022). Similarly, SCD is also affected by the wider determinants of health ((Dalghren and Whitehead, 1991) and in particular, education and access to health services have already been established as influencing factors in the nutritional management of SCD. SCD is predominantly considered as a bio-medical problem. The researcher (Berghs et al., 2020) thus recognises that there is more to SCD than its pathophysiology and that there is a need to consider the health and wellbeing outcomes of people living with SCD across the life course of the patients. In adopting a socio-ecological lens to the research, the value of nutrition in SCD is clearly illustrated to be more than simply an environmental problem (Rees, Williams and Gladwin, 2010), but a problem that is more complex and requires a more comprehensive exploration.

The health and healthcare experiences of the SCD population is situated in society, and although nutrition management in SCD has not been explicitly explored in this way, a socio-ecological lens lends itself to the sociological situating of nutrition in SCD. This approach is not explicit in existing research on the role of nutrition in SCD, thus these socio-ecological factors identified through the research project adds to what can be known about nutrition in

SCD and the multiple influencing factors affecting nutrition and service provision. More importantly, also illustrates the complexity of nutritional management in SCD from a multi-level perspective. Thus, social and structural concepts such as race, ethnicity, social class, economic status and health inequalities (Taylor and Field, 2007; Craig et al., 2012; Walsch and Tait, 2014; Chattoo et al., 2019) are also pertinent concepts to consider in the lack of nutrition management in SCD. A socio-ecological lens facilitates a more comprehensive approach to identifying and understanding the nutritional management landscape in SCD. Therefore, the nutritional management of SCD needs to be reframed in order to tailor policy and practice in support of the development of nutrition service provision in SCD. In this way, the adoption of a whole systems lens is facilitated in how policy and practice in nutrition service provision can be tailored to the unique nutritional needs of people living with SCD. The personal and interpersonal (micro level), institutional (group level) and community/policy (community/government level) factors and facilitators in nutrition in SCD will be discussed below.

5.3.1 Micro level factors:

The data collection and analysis of the research findings have identified a number of personal and interpersonal (micro level) contextual factors in relation to the nutritional management of SCD. Based on the individual and collective SCSU-C and SCSP comments, a number of micro level factors emerged including personal internal factors (psychological, emotional, attitudes, beliefs, culture, stigma, religion and health symptomology) and external factors (environmental, time, knowledge, family culture, food traditions, peer support) that determine the nutritional needs and choices of people living with SCD and how well these needs can be met. Both these internal and external factors demonstrate the interconnectedness between the psychological and sociological factors that are at play in how well people living with SCD are able to navigate their nutritional needs and how these

nutritional needs are to be managed. Whilst the national clinical standards of SCD highlight key contextual factors in relation to the social welfare of people living with SCD, such as housing and employment (SCS, 2018, p81), and the need for people living with SCD to have psychological support (SCS, 2018 p73; NICE, 2021b), these contextual factors need to be extended to consider its impact on the nutritional needs of people living with SCD, which is what this research project aims to achieve. The interconnection between the social and psychological factors affecting the nutritional needs of people living with SCD, is highlighted in the article by (Matthews, 2015b), giving weight to the findings of the research project as factors impacting nutritional management in SCD. Inadvertently, healthcare professionals such as those working in social welfare and psychology become important advocates to call out the need for the nutritional management of people living with SCD as part of their standard care, who may be talking to these healthcare professionals about how their social and psychological factors are impacting on their nutritional needs and health and wellbeing.

As such, Parnell (2016) highlights the importance of the relationship between the person receiving care and those providing care. In the absence of nutrition service provision, existing clinicians and healthcare professionals in SCD have an important role to advocate for the nutritional needs of people living with SCD, and this may require them to challenge the *status quo*, in order for these people to receive the adequate nutrition advice and support they need. When this does not happen, people living with SCD are left to self-research what they need to know about nutrition in SCD, evidenced in this key finding of the research project, amongst both the SCSU-C and SCSP groups. More importantly, Parnell (2016) highlights care provision as a two-way transaction, where service users and service providers interact with each other to ensure good health outcomes (Berkman, Davis and McCormack, 2010) for the person receiving the treatment. However, the absence of nutrition service provision (supported by the findings of this research project), confounded by the range of internal and external (social and psychological) factors affecting nutritional management, perpetuates the marginalisation of SCD, as their nutritional needs are not

deemed significant enough in the standard care provision. Sadly, to date, whilst the social welfare and psychology service provision has already been integrated into standard care for people living with SCD, the link between the social and psychological factors in nutrition remains overlooked and a source of ignorance in SCD care provision, which this research project aims to illuminate.

The social (family support structures) and cultural (family, food and traditions) factors identified in the health literacy tapestry model (Parnell, 2016), considered as contextual factors, are noted to influence an individual's sphere of life, work and play. Within these social and cultural factor groups highlighted by Parnell (2016), there are individual internal and external factors identified as micro level factors in this research project, influencing the nutritional needs of people living with SCD, but in a broader sense, also impact on the life, work and play of the people living with SCD. In the same way, these micro level social, psychological, cultural and environmental factors add a layer of complexity to the nutritional needs of people living with SCD, some of which are recognised to cause Disease-Related Malnutrition (DRM) (Waitzberg et al., 2001). Disease-related malnutrition (particularly under nutrition in SCD) is a term not explicitly recognised in SCD literature (Matthews, 2019); however, these collective internal and external micro level factors may work together over time to increase the risk of disease-related malnutrition in SCD, necessitating the recognition and prioritisation of nutrition in SCD. In particular, a confounding factor increasing a patient's risk of disease-related malnutrition is the health and related clinical symptomology of SCD. These factors can impact on the ability of people living with SCD to undertake their activities of daily life, such as cooking, cleaning, shopping and food preparation, when the patients are in pain or unwell, potentially placing pressure on family and peers to offer the help that is required, demonstrating the interconnection between the micro and meso level factors. Poor nutrition knowledge of SCSU-Cs also limits their ability to make sound nutritional choices. Similarly, the structural social factors of social class and ethnicity can invariably impact on the SCD patient's ability to self-manage their condition, in other words, their ability to take

greater control of their health choices (Nutbeam, 2008). Thus, the range of micro-level factors identified in the study, provides proof of the impact these factors have on the nutritional needs and management of people living with SCD.

Amongst both the SCSU-Cs and SCSPs, the need to address poor nutrition knowledge was recognised in order for people living with SCD to make better nutritional choices to self-manage their condition but moreover, that this knowledge is extended to family, friends and peer support who often times have to assist the person with SCD with meal preparation. Improved nutrition knowledge was also identified as a key facilitator to address the stigma surrounding SCD, in particular amongst school children living with SCD. In so doing, improving the person's ability to access help as and when they may need it. An important point raised by the members of the LA Network (in phase two) was the role of the partners in caring for people living with SCD, and the additional support and education for those who support patients living with SCD, especially those patients who live alone including the elderly patients living with SCD (see Appendix 15). Unsurprisingly, the importance of addressing the nutrition knowledge gaps was a dominant personal and interpersonal facilitator to better support the nutrition care needs and support of people living with SCD. Acknowledging these micro level factors will go a long way to improve the health and wellbeing outcomes of people living with SCD. Hence, it is also important to consider the institutional factors (meso level) factors, affecting nutrition management in SCD.

5.3.2 Meso- level factors:

The institutional level factors are paramount in nutrition service provision. It is at this level that the decision making to implement service provision takes place, and where the unmet nutrition needs of people living with SCD are either acknowledged or overlooked. Failing to acknowledge the unmet nutritional needs of people living with SCD only serves to add to the already high levels of morbidity and mortality associated with the pathophysiology of SCD

(Fitzburgh et al., 2005; Lucas and Mason, 2008; Aljuburi et al., 2012; Piel et al, 2021) and associated health inequalities in the condition. A number of institutional factors have thus been identified in this study to be discussed below.

As previously mentioned, poor levels of nutrition knowledge and lack of resources are a serious oversight in nutrition service provision in SCD, as poor knowledge place limits on the ability of those affected to establish and clarify the link between nutrition and health outcomes in people living with the condition, maintaining the knowledge/power ratio in marginalised groups (Freire, 1970). This oversight potentially increases the healthcare burden of people living with SCD, which in turn may carry increased healthcare utilisation and care costs (Aljuburi et al., 2012; Aljuburi, 2015). The clinical management of SCD already carries a high care cost burden, in terms of SCD crisis and care provision (Aljuburi, et al., 2012; 2015; Pizzo, et al., 2014; Jobanputra, 2021) and the unmet nutritional needs may inadvertently, be adding to this healthcare cost burden in SCD, which may be due to the late diagnosis of nutritional problems in SCD, a significant finding of the research project as previously mentioned. Since the nutritional needs of people living with SCD are treated more reactively and not pro-actively or preventatively, it fails to comply with the calls of Domain 1 of the NHS Outcomes framework (2022), in regards to acknowledging the importance of prevention in improving patient outcomes, which includes greater surveillance of the nutrition needs of people living with SCD, only possible when nutrition service provision is part of standard care in SCD.

In current healthcare provision in SCD, patients with SCD are predominantly reliant on healthcare professionals to advocate for their healthcare needs. As such, the knowledge and care gaps in nutrition in SCD, calls for greater critical consciousness (Freire, 1970) of both the people living with SCD and providers about the social discomforts that are purporting the *status quo* in nutrition in SCD. Knowledge/power gaps partly explain the marginalisation of the people living with SCD, maintaining them in a state of dependence and powerlessness to develop the necessary praxis (Freire, 1970) to take action to change their health experience

and outcomes. Being in majority from a BEM background, people living with SCD are subject to unfair and unequal treatment commonly attributed to racial discrimination and identified as a critical factor influencing health in people from ethnic minority backgrounds (Taylor and Field, 2007; Walsh and Tait, 2014). On a bigger scale, institutional racism is identified as a problem in SCD healthcare provision (Anionwu and Atkin, 2001), reflected in the lack of policies and procedures, and lack of access to appropriate and effective care (Macpherson, 1999; Walsch and Tait, 2014). In this way, the lack of nutritional management in SCD, as evidenced in the collective findings of this research project discussed above, portrays the potential unfair and unequal treatment in regards to nutritional management in SCD, recognised as racism, which leads to the denial of equal opportunities based on the belief that one ethnic group is more superior than another (Taylor and Blakemore, 2007; Walsch and Tait, 2014).

It is sufficing to say that the marginalisation of the people living with SCD provides a deeper context to explain the lack of knowledge and the resulting power imbalance between the 'patient and provider' on the one hand, and the wider structural factors such as race and ethnicity in how people living with SCD experience their healthcare provision on the other hand. In the context of the lack of nutrition service provision observed in the current nutritional management of SCD, institutional racism is evident when the SCD consultants and other healthcare professionals do not advocate for the nutritional needs of the patients in their care. When people living with SCD lack the knowledge, critical consciousness and praxis to take action to change their healthcare choices, they are at risk of poor experiences, access and health outcomes, driving their experience of health inequalities, especially since they are already prone to social inequalities on the basis of their race and ethnicity. A SCSP indicated the need for people living with SCD to be empowered to take more control over their nutrition, but reflected on the fact that they may not have the knowledge and understanding of the link between SCD and nutrition (section, 4.2.2), compounded by the

other influencing factors such as the social, cultural and psychological factors at play in nutrition in SCD.

Further examples of institutional factors (both direct and indirect) pertain to instances whereby SCD patients are hospitalised when receiving treatment for a painful crisis, as hospitalisation can increase a patient's risk for malnutrition (Elia, Zellipor and Stratton, 2005), exacerbated by length of admission, poor appetite during admission and increased nutritional requirements etc. However, people living with SCD may have an increased risk of malnutrition secondary to the pathophysiology of their condition, therefore, awareness of nutrition should not be limited to people living with SCD who are hospitalised, but should be extended to all healthcare settings where people living with SCD are treated, live, work and play, and family members need to have awareness and knowledge of nutrition too to best support the person in their care (Harris and Gore, 1996). Other institutions to consider are schools; as emphasised by a SCSP who said, education on nutrition should not be confined to focussing solely on fluid intake as is the current status quo, but that education and training to schools should also include nutrition and the nutritional needs of children living with SCD. This means that staff in schools should also be made aware of the role of nutrition as a management option in SCD. Moreover, the health differences between SCD and other conditions, for example, children living with long term conditions such as diabetes or allergies, who standardly may have care plans that include nutrition, was highlighted by a SCSP. These differences, yet again reflect the potential health inequalities children living with SCD have to contend with, and their reliance on healthcare professionals to adequately advocate for their healthcare needs. This research projects thus serves to draw attention to these instances of inequalities which is flagged in the APPG-SCAT (2021) report. For this reason, both the SCSU-Cs and SCSPs comments pointed to the need for nutrition education to be available to all the key stakeholders in SCD healthcare provision, integrated into all areas of SCD management and patient pathways. This includes all key stages of life such as

patients transitioning from paediatrics to adult care as well as being incorporated into existing SCD educational conferences to include a wider audience.

5.3.3 Macro level factors:

Understanding and exploring the macro level factors in nutritional management in SCD plays a significant role in contributing to policy and practice development. Community infrastructures, resources and systems play an important part in how well communities are able to support the nutritional health and social care needs of various populations, including the SCD population. Nevertheless, it is understanding how these macro level factors affects the SCD population within the wider society, that needs to be considered.

Race and Ethnicity:

Being predominantly from a BEM background, ethnicity and race are structural variables at play in the healthcare provision and experience of people living with SCD. Race in particular is identified as a critical factor influencing the health of ethnic minority groups (Craig et al, 2012; Walsch and Tait, 2014; Chattoo et al., 2019), hence racism or unequal treatment - already established in SCD (APPG-SCAT, 2021) - adds to the complexity of the healthcare provision and experience of people living with SCD. Importantly, ethnicity is multifactorial in how it affects the health experiences of people from ethnic minority groups. Relevant to people living with SCD, Walsch and Tait (2014) highlight a number of these factors. Among these factors relevant to SCD, the following are included: genetic or biologic factors (as SCD is a genetically inherited condition), cultural and individual factors and structural or material factors (such as occupation, income, housing, diet and social position). A number of structural factors were identified in this research project, for example patients not having access to affordable healthy food options - this translates into factors such as those affecting

the diet and lifestyle choices, social position and income potential of people living with SCD. In particular, social position and income equate strongly to poverty and deprivation, best understood in the context of social class (Walsh and Tait, 2014).

Social class:

The British Society is identified as being divided and structured into social classes (Walsch and Tait, 2014). Social class is how people are grouped in terms of their occupation, income and parental education and how this impacts on people's life chances and their experiences of health (Walsh and Tait, 2014). In keeping with social class and its effect on health experience, patients who experience low income and poverty, for example, are prone to social inequalities (Walsh and Tait, 2014) and material inequalities (Taylor and Field, 2007). Since social class is based on a person's occupation, it stands to reason that if the education attainment of people living with SCD are compromised due to ill health (APPG-SCAT, 2009), this in turn may have a negative effect on their employment prospects (Berghs, Cronin-Chavez, and Ebenso, 2020); Berghs et al., 2021; Berghs and Dyson, 2022) and their eventual occupation and social class stratification.

In support of the impact of social class in the SCD population, poverty and deprivation have been identified in this research project as two macro level factors influencing the nutritional needs of people living with SCD. Poverty and deprivation limits peoples access to affordable healthy options, for example, people may buy cheaper food options, such as high fat and sugary foods, as this is more affordable to them, inadvertently impacting on their overall health risks and nutritional wellbeing, particularly those on limited budgets (see Chapter 4, section 4.2.3). Therefore, considerations for families and households are equally important, as the choices made by the family, based on the social class stratification and socio-economic status, have a direct bearing on the health and health choices and access of the individuals who make up the families and households (Walsch and Tait, 2014).

Paradoxically, a SCSU-C, raised a concern about the possibility of nutrition being a cure in SCD and this negatively affecting their personal independent payment applications, as this may be their only source of income – impacting on their life chances, material circumstances and inequalities (Taylor and Field, 2007). Thus, access to finances and structural factors such as housing, is an important consideration in people living with SCD and their nutrition as identified by a SCSP, highlighting the fact that some people living with SCD, may be living in hostels, where there may not have access to fridges, stoves etc (section 4.2.2) and forced to buy food daily and cheaper options, such as chicken and chips, that are perceived as being more affordable in light of the limited finances and material resources. The geographical location and access to healthy affordable food was also identified by a few SCSU-Cs, further influencing the buying patterns and food choices in the people living with SCD.

Furthermore, the health literacy of individuals living with SCD from a lower social class is also an important factor especially as it relates to how they use information to help them make decisions to maintain good levels of health (Nutbeam, 2000). This is especially relevant in regards to their choices of food and eating habits. As such, the level of health literacy (high or low) of a person impacts on their health outcomes (Paasche- Orlow and Wolf, 2007). Thus a SCSP in this research project, indicated that people living below the poverty line, with limited access to finances, may have problems with affordability, poor food security and may also be more prone to these inequalities. In this way, the nutritional management of SCD is complex, and benefits from being considered in a socio-ecological context, as there are a number of social and structural factors that have a direct bearing on the individual SCD patient's choices, access and ultimately health and healthcare experiences, as reflected in the findings of this research project.

Education:

Poor nutrition knowledge and education, were key findings throughout this research project. In addition to needing more education about nutrition, people living with SCD could benefit

from having increased access to support structures within the community. For example, community structures such as SCD centres, SCD support groups where people living with SCD can access more help, advice and support, were identified by both the SCSU-Cs and SCSPs as being important (section 4.2.3). In addition, the availability of other community structures such as libraries, leisure centres and GPs, where people living with SCD can access information about SCD and where these people can access help and advice on a range of topics including nutrition and healthy eating advice and support, need to be made available when nutrition service provision in SCD is planned in health and social care.

However, evidence of the lack of these valuable support structures in the community was evident in the fact that nearly all the SCSU-Cs and SCSPs reported having to self-research what they know about nutrition, healthy eating, emphasising the stark reality that for many people living with SCD, nutrition may not be considered a problem. On the other hand, SCSPs too may not consider the need to improve their knowledge about nutrition. Suffice to say, more work is needed to raise awareness of the importance of nutrition as a viable management option in SCD, that takes both a health and sociological perspective, as a range of social structures are at play in the SCD population. More importantly, in ethnic minority groups, lack of access to appropriate and effective care – is reflected in higher morbidity and mortality rates (Taylor and Field, 2007). Hence, the type of infrastructure and the distance to the infrastructure, such as shops, supermarkets and vendors, all have a role to play in accessibility and affordability for people with limited funds and who may be living on the poverty line. More emphasis may have to be given to develop the social capital in nutrition in SCD, evident in the calls for more stakeholder involvement, more policy level changes and more lobbying for nutrition to be considered in the standard care provision of people living with SCD.

5.3.4 Socio-ecological – facilitators:

Collectively, the research project findings identify a range of influencing factors including knowledge and care gaps that define the current nutrition landscape in SCD. Of note,

although the national nutrition standards in SCD (SCS, 2018 pp 75-77) provide a helpful starting point to inform and support nutrition service provision in SCD, the research project presented an ideal opportunity to gauge first hand, how the identified knowledge and care gaps in nutrition in SCD could be addressed and improved, from the SCSU-C and SCSP perspectives (section 4.2.3). Thus a number of socio-ecological facilitators were identified to address the socio-ecological factors influencing nutrition and service provision in SCD, affecting the integration of nutrition in SCD.

Amongst the facilitators identified, a SCSU-C suggested that nutrition service provision in SCD needs to be holistic, with joint MDT, whilst a couple of SCSPs recommended a national awareness campaign using a variety of social media platforms to be considered to call on the need for improved nutrition service provision in SCD. Both SCSU-Cs and SCSPs recommend involvement of stakeholders, in strategic positions and organisations, as being essential to drive forward change in policy and practice in nutrition in SCD. In addition to having multi-stakeholder involvement, these participants suggested that the commissioners of dietetic services need to be aware and have the knowledge and backing of policies to support the provision of nutrition services for people living with SCD, and should be extended to community and primary care enabling GPs to make these services available to people living with SCD and offer dietetic input as needed. Furthermore, two SCSPs (Dietitians) suggested that the British Dietetic Association (BDA) make available evidence-based practice to support the nutritional treatment and management of people living with SCD, tailored to their unique nutritional needs. Therefore, if the inequality of the lack of nutrition service provision in SCD is to be addressed, a collaborative approach to service provision is required, to reinforce uniform messaging about the role of nutrition in SCD.

Furthermore, SCSU-Cs and SCSPs identified the need for more commitment from all the relevant strategic organisations and stakeholders, such as the Sickle Cell Society (SCS), The UK Forum on Haemoglobin Disorders, APPG-SCAT, the reformed NHS England and the BDA, if changes are to be made at policy and practice level. Furthermore, these

stakeholder bodies need to pay attention to and engage in conversation to include nutrition in the management of SCD, thereby acknowledging the value of nutrition in SCD management. A reasonable recommendation towards raising awareness of nutrition in SCD, is therefore to recommend that nutrition be placed on the agenda of these strategic stakeholders, where there is opportunity for these stakeholders to engage with people living with SCD and use their voices to ensure their unmet nutrition needs are met within standard care provision. All in all, the findings of this research project provide evidence of the need for a patient-centred, personalised care approach (NHS, 2019) to clinical and nutritional policy and practice development and change.

5.4 Clinical and nutritional policy and practice development in SCD

The formation of the Learning Alliance Network (LAN) between the SCSU-Cs and SCSPs as described in objective 2, is aimed at using the findings from Objective 1 (knowledge and care gaps representing the influencing factors affecting nutrition integration) to co-develop and evaluate the health literacy framework to support the integration of nutrition into standard care in SCD. Below is a discussion of the evaluation of the health literacy framework in support of clinical and nutritional policy and practice development in SCD, an overview of the value of the LA methodology in developing the study and future healthcare interventions and how the health literacy PEECE model enhances the outcomes potential of healthcare interventions.

5.4.1 Evaluation of the Health literacy framework:

The purpose of the evaluation of the health literacy framework is focussed on developing clinical and nutritional policy and practice to support the integration of nutrition into standard care in SCD, towards achieving the main aim of the research project. Despite calls for nutrition to be included in standard care provision for people living with SCD over the years,

to address the growth and nutritional deficiencies caused by the clinical features of SCD, this research project is the first attempt at embedding nutrition into standard care provision in SCD on a national and professional basis. The current lack of clinical and nutritional policy and practice in nutrition in SCD, serves to maintain the status quo, impacting patient outcomes in nutrition in SCD. The co-development of the health literacy framework by the LAN, discussed in Chapter 4 (section 4.3) is therefore the first attempt to address the lack of clinical and nutritional policy and practice in nutrition in SCD at population level using a whole systems approach. The evaluation of the health literacy framework was essential to assess the efficacy of the health literacy framework to address the knowledge and care gaps, reflecting the key influencing factors affecting nutrition integration in SCD.

With the co-development of the health literacy framework being the first attempt at clinical and nutritional policy and practice level change in SCD, there are very little examples to compare. The most relevant document of comparison is the APPG-SCAT (2021) report, as there are many correlations between the findings of this report and the research project, as demonstrated above, in section 5.1. Firstly, the participant profile of the APPG-SCAT (2021) report included people living with SCD, carers, service providers and politicians. With exception to the politicians, the participant profile of the research project included both SCSU-Cs and SCSPs. More importantly, the research project used a Learning Alliance (LA) methodology to support the formation of a LAN (between the SCSU-Cs and SCSPs) to reflect the individual and collective voices of both the SCSU-C and SCSP participant groups. In the same way that the statements of the participants formed the basis of the conclusions about the failings of healthcare provision in SCD (APPG-SCAT, 2021), the themes of this research project represents the breadth of influencing factors – reflected in the knowledge and care gaps in nutrition in SCD affecting the integration of nutrition in SCD.

It was evident from the responses of the LAN following the evaluation of the health literacy framework, that the health literacy framework was able to improve the knowledge and understanding of nutrition of the members of the LAN, as reported in Chapter 4 (section 4.4).

In addition, the responses of the LAN revealed knowledge gaps were variable, with some members of the LAN having more knowledge and others having less knowledge of the nutritional needs of people living with SCD. The APPG-SCAT (2021) report identified the need for more healthcare professionals in SCD to have greater knowledge of SCD, whilst the health literacy framework enquired about the medical and nutrition knowledge of the members of the LAN. It was clear that nutrition was not a topic considered in the APPG-SCAT (2021) report, further perpetuating the invisibility of nutrition in SCD, but equally emphasising the neglect of nutrition as part of standard care in SCD management. However, the members of the LAN were able to benefit from shared learning, as those with limited nutrition knowledge could learn from those with more nutrition knowledge – a beneficial outcome of adopting a LA methodology.

The shared learning between the members of the LAN contributed to raising awareness of the influencing factors affecting nutrition integration in SCD (section 4.4.2). In this way, the members of the LAN developed a critical consciousness about various aspects of nutrition, demonstrating the value of engaging in the research and having an opportunity to speak to others about nutrition in SCD. Hence, the collaborative and partnership working supported by the LAN contributed to not only increasing the nutrition knowledge and management of SCD, but it also supported the learning and development of the members of the LAN. This learning included the myriad of influencing factors affecting the nutritional management of SCD, from different perspectives, to what they may have known previously, thus broadening their knowledge base in nutrition in SCD. In this way, the members of the LAN developed an understanding of the complexity of nutrition in SCD, and that nutrition in SCD was more than 'just food and fluid'. In so doing, the members of the LAN were able to engage in a time of praxis to reframe nutrition and develop confidence in care providers to support the nutritional needs of people living with SCD.

As a consequence, the members of the LAN recognised the benefit of the health literacy framework as a valuable tool for communication and training (section 4.4.3), evident in the

improvement in their own learning and development. Thus, they were able to see how the increased engagement and education developed their ability and confidence to recognise how the health literacy framework could be used to steer conversations with others about nutrition in SCD, and also how the knowledge and care gaps reflected in the framework can inform future education and training in nutrition in SCD. The participants found that they were able to learn from each other's viewpoints and perspectives and the wider discussions during the overall data collection activities. Suffice to say that engagement in the study has contributed to improving the nutrition knowledge of the members of the LAN (section 4.4.1).

In addition to being a valuable tool for communication and training, the benefit of the health literacy framework can be extended to its role in improving clinical and nutritional policy and service provision outcomes in nutrition in SCD (section 4.4.4). This perspective was clearly articulated by the members of the LAN as they recognised the value of the framework to contribute to structure and policy change. Moreover, the LAN recognised the importance of having multi-stakeholder involvement to promote the integration of nutrition into standard care in SCD, including organisations such as the UK Forum for Haemoglobin Disorders, SCS, APPG-SCAT, NHS England, Health Education England and the BDA and other relevant leaders in SCD, as well as the people living with SCD. In this way, there needs to be national and professional level policy and practice change to address the myriad of influencing factors affecting the integration of nutrition in SCD, identified as knowledge and care gaps in this research project. In order to do so, buy in and commitment from all the key players are required to see improvements in the experience, access to and health outcomes in nutrition in SCD, for people living with SCD. A fresh perspective to address the neglect of nutrition in standard care in SCD was the suggestion by a SCSP to make nutrition in SCD mandatory and a topic to be talked about, within Health and Social Care in SCD, ensuring that people living with SCD are followed up and referred to adequately-trained healthcare professionals in nutrition such as Dietitians, known diet and nutrition experts (BDA, 2017).

In this way, the evaluation of the health literacy framework was able to add to the knowledge of the what can be understood about the benefits of the study intervention at improving the awareness, knowledge and learning about the influencing factors affecting nutrition integration in SCD management. In addition, the evaluation demonstrates how collaborative and partnership working between the SCSU-Cs and SCSPs can enhance shared learning and positively contribute to addressing the knowledge and care gaps to promote the development of clinical and nutritional policy and practice development in nutrition in SCD care provision.

5.4.2 The Learning Alliance Methodology in healthcare intervention development:

It is evident from the discussion in section 5.4.1 above, that the LA methodology using the LAN contributed to how well the health literacy framework was able to address the influencing factors –reflected in the knowledge and care gaps in nutrition in SCD, and moreover, how the health literacy framework was able to contribute to clinical and nutritional policy and practice development in nutrition in SCD. However, in addition to the above, some consideration is needed to evaluate how well the LA methodology is able to develop the patient voice in clinical and nutritional policy and practice to benefit the development of future healthcare interventions using this methodology. Below follows a discussion providing a critical exploration of how the LA methodology is able contribute to the development of the patient voice in healthcare interventions.

Value of the LA methodology process and experience:

Two previous studies using the LA methodology in marginalised populations, the first considering the challenges and contributions of multicultural health planning (Meguilzamo et al., 2015) and the Learning Alliance Palliative Care and End of Life study (Smith and Moreno-Meguilzamo, 2017), provide insightful perspectives on the value of developing the patient voice in healthcare intervention planning. Although these studies demonstrate the

value of the LA methodology, in particular how collaborative and partnership working between the members of the LAN contributed to the planning and development of the respective healthcare interventions, the service user/patient groups were not offered the opportunities to meet independently to consider what was most important to them and what were their main challenges and problems they wanted to address. However, a recommendation that came out of the study conducted by Moreno-Meguizamo et al (2015) recognised the value of holding independent focus groups with the service user/patient groups first, before the formation of the LAN, as a way to ensure that they are able to speak freely about the problems that are most important to them. In so doing, the service user/patient groups are empowered and allowed to have greater control over the topics to be considered for discussion and thus be in a position to influence the discussions on policy and practice change towards the development of the healthcare intervention.

By empowering the service user/patient groups in this way, a greater critical consciousness of the problem is promoted, through knowledge co-production and sharing inherent of the LA methodology, thereby enabling the participants to develop a wider understanding of the problems. Moreover, knowledge co-production and sharing importantly facilitates a greater opportunity for praxis, through critical reflection to develop a range of ideas that can contribute to the wider discussions that will take place in the LAN meetings with representation of all the stakeholders. Greater empowerment amongst the service user/patient groups also promotes increased autonomy and motivation (Nutbeam, 2008) to want to engage in more discussions, empowered with a deeper understanding of what the problems are and initial ideas of how the problem can be tackled. However, the holistic lived experiences and circumstances of the respective service user/patients, as in the case with this research project, also needs to be factored in. To harness the benefits of empowering the service user/patient groups in the way as described above, this study chose to conduct independent focus groups between the SCSU-C and SCSP participants, to learn first-hand what their knowledge, views and experiences of the medical and nutritional management

and socioecological factors and facilitators of nutrition and service provision in SCD, before the formation of the LAN. First identifying and understanding the problem, from the respective participant groups, inadvertently helped to even out the potential knowledge/power gap which is commonly experienced in marginalised groups (Freire, 1970).

Thus, holding independent focus groups before the formation of the LAN, was helpful in empowering the patient voice, in so doing allowing them to take greater control and make a positive contribution to how their respective problems can be understood and should be addressed. Importantly, these discussions should reflect their holistic lived experience and circumstances, but still allow them to be part of a group contributing to community assets and cultural perspectives in healthcare intervention planning and development. Adding this initial step to the research project does however add time and cost implications to the research project and can affect the level of engagement of the participants. However, working with empowered and engaged individuals who feel part of the solution, having the opportunity to make a positive contribution to the solution, feeling listened to and valued as an equal partner to changing the status quo, is so much more rewarding to the marginalised populations who may already be feeling that they do not have a voice, they do not matter and less than human. The value of the LA methodology in developing the patient voice in healthcare intervention development is therefore enhanced by conducting independent focus groups, giving the service user/patient groups the opportunity to use their voices freely to share their lived experiences and moreover make a positive contribution to social change through collaborative and partnership working, to promote emancipation, empowerment, enabling the service user/patient groups to have greater involvement and take greater control when engaging in their own healthcare management as well as healthcare planning and provision.

5.4.3 Health literacy in policy and practice development in nutrition in SCD:

(i) The Health literacy PEECE model:

It was clear that the health literacy framework, as evaluated by the members of the LAN, was valuable to address the influencing factors reflected in the knowledge and care gaps identified through the research project, to support clinical and nutritional policy and practice change in nutrition in SCD. However, to demonstrate how health literacy was used as an expansive and integrative concept to contribute to clinical and nutritional policy and practice change, the Prevention Education Empowerment Contextual - factors Engagement (PEECE) model was developed. The health literacy PEECE model was developed from the health literacy conceptual thematic themes, which are derived from the four main themes of the focus group analysis (see Chapter 4, section 4.6.1, Table 4.5), and built on the evaluation of the health literacy framework co-developed by the LAN, in phase three, forming the theoretical basis for health literacy as an expansive and integrative concept. The health literacy PEECE model is thus a novel approach to developing clinical and nutritional policy and practice to improve nutrition service provision outcomes in SCD. The main theoretical components of the health literacy PEECE model underpin the three health domains considered in the integrated conceptualisation of health literacy by Sorenson et al., (2012).

The three health domains in health literacy relevant to SCD – healthcare management, public health and health promotion (Sorenson, et al., 2012) – provide further theoretical underpinning to the health literacy PEECE model, as Prevention is paramount in public health, and Education and Empowerment and the Contextual factors, are important concepts in health promotion. In this way, the health literacy PEECE model reflects the expansive and integrative conceptualisation of health literacy, providing compelling theoretical support to contribute to national and professional policy and practice development at a population level, using a whole systems policy and practice strategy to address the knowledge and care gaps in nutrition in SCD. Thus, the health literacy PEECE model has been central to the

development of the health literacy nutrition integration conceptual framework (Chapter 4, Figure 4.4) – reflecting the gaps in the literature (as summarised in Chapter 2, pg66) and the Health Literacy-Nutrition Integration Framework (HL-NIF) blueprint, as discussed in Chapter 4, (section 4.6.3, Figure 4.5), that collectively aims to address the influencing factors affecting nutrition integration in SCD, to support the development of clinical and nutritional policy and practice in nutrition in SCD.

An important consideration for adopting health literacy in SCD, is the identified link between health literacy and health outcomes. This link between health literacy and health outcomes is established in health, public health and health promotion (Dearfield, Barnum and Pugh-Yi, 2017; Parnell, 2016, Nutbeam, 2008; Paasche-Orlow and Wolf, 2007) and can play a significant role in the health outcomes of people living with SCD. Since the nutritional care provision in SCD is yet to be established as part of the standard care provision available to people living with SCD, there is an opportunity to explore the integration of nutrition into SCD healthcare provision through a health literacy lens. There is sufficient evidence available in the APPG-SCAT (2021) report of the effects of poor health literacy on the part of healthcare professionals working in SCD, e.g. resulting in the avoidable death of a 21-year-old person living with SCD in April 2019. Hence, linking health literacy to health outcomes in SCD is both plausible and important when considered as an expansive and integrative concept in nutrition in SCD, to support clinical and nutritional policy and practice development.

In addition, health literacy is a determinant of health (WHO, 1998; Nutbeam, 1998; 2000; 2008; Rudd, 2015; Passche and Lowe, 2007; Sorensen et al., 2012; Parnell, 2016; Berkman, Davis and MacCormack, 2010), and as such is plausible to consider in the context of clinical and nutritional policy and practice development in nutrition in SCD. In this way, health literacy is key in preventing ill-health, as prevention is recognised as a fundamental principle of public health. The invisibility of SCD and associated poor healthy literacy has already been established, meaning many people living with SCD are at increased risk of poor health outcomes (APPG SCAT, 2021). For example, the late diagnosis of nutritional

problems holds serious repercussions for patient outcomes and can lead to people living with SCD not trusting healthcare professionals, which was a finding reported in the APPG-SCAT (2021). In this report, patients were afraid to go to hospital for medical treatment, as they did not trust the healthcare professionals to provide the appropriate level of care (APPG-SCAT, 2021 p36).

The quality of the care provision in turn has repercussions for people living with SCD who may have unnecessary risks related to their care due to no fault of their own. The late diagnosis of the nutritional problems in both a paediatric and adult SCD patient in this research project, are clear examples of the risks open to people living with SCD due to a lack of nutrition service provision in standard care in SCD. The care provision should as far as possible offer a personalised care approach to care provision (NHS England, 2013; Kings Fund, 2013; NHS England, 2019) supported by the white paper, 'Our Health, our Care, our Say' (DOH, 2006). Hence, in addition to clinical and nutritional policy and practice reflecting the complexity of SCD, it should promote patient-centred care that is tailored to the unique needs of people living with SCD to support patient safety and high quality care provision.

Health literacy is also linked to patient safety, and ultimately patient outcomes (Weis, 2007; Parnell, 2016). Weis (2007, p13) identifies this about poor health literacy: "it is a stronger prediction of a person's health status than age, income, employment skills, education level and race". Hence, the late diagnosis of nutritional problems in people living with SCD needs to be addressed to prevent it from happening in the first place, and nutritional care provision should be proactive and not reactive, as previously mentioned. Therefore, to enable people living with SCD to take greater control of their health (Nutbeam, 2008), an essential feature of health literacy, attention should be given to developing nutrition service provision in SCD that is underpinned by education and empowerment, important concepts in health promotion. Health promotion is connected to health literacy, as reflected in the definition of health literacy by the Institute of Medicine (IOM, 2004): a "concern for everyone in health promotion, protection, disease prevention and early screening and maintenance and policy

making". In other words, health literacy should be incorporated into all aspects of health and healthcare provision.

However, in nutrition in SCD, this is prohibited due to the lack of nutrition knowledge, poor availability of nutritional resources, information and poor nutrition service provision (Matthews, 2016g). Hence, health literacy is defined as highlighting the importance of people being able to make appropriate health decisions, reliant on their ability to obtain, process and understand basic health information and services (Ratzan and Parker, 2000). The knowledge and care gaps identified through this research project, provide compelling evidence in support of the need for the development of nutrition service provision, in order to address the lack of basic health information to support positive health related decision making. Moreover, the formation of the LAN is representative of the value of collaborative and partnership working in healthcare provision in providing a platform for knowledge sharing and co-developing. The LA methodology promoted engagement and joint working between the members of the LAN, and helped to improve the knowledge and awareness of the LAN of the nutritional needs of people living with SCD.

Nutrition is considered as a non-invasive treatment option (Hyacinth, Adekeye, Yilgwan, 2013) in SCD, however, the current lack of nutrition service provision in SCD management is indeed contributing to the poor health outcomes of people living with SCD, an example being the late diagnosis of nutritional problems in people living with SCD. Thus collectively, the health literacy PEECE model, built on the collaborative and partnership working of the LA methodology, provides all the elements to address the knowledge and care gaps in the medical and nutritional management of people living with SCD, supporting a population level, whole systems management strategy to support the integration of nutrition in SCD. For this reason, there is a priority to address the myriad of influencing contextual factors that ultimately impact on people living with SCD, and their ability to take more control of their health and health outcomes.

5.5 Nutrition integration in SCD

The value of the health literacy framework would not be complete without first evaluating it against the existing relevant national practice documents such as the national nutrition standards (SCS, 2018, p 75-77) and the UK Forum on Haemoglobin Disorders Quality Standards (2021) for SCD, how it informs what is included in the Annual Review Pro Forma as well as the GP Outpatient letter and national health policies and outcomes frameworks in Health and Social care.

5.5.1 Evaluation of clinical and nutritional policy and practice documents in SCD:

Relevant practice documents:

The neglect of nutrition as part of standard care in SCD, impacting patient outcomes, is reflected in some of the omissions in practice documents in SCD management. In this way, the nutrition service provision needed to identify, assess and treat the nutritional needs, risks and challenges of people living with SCD, caused by the clinical features of the condition, is overlooked and neglected. For this reason, the appraisal of the existing clinical practice documents in SCD management such as the Annual Review Pro forma and the GP Outpatient letter, will go a long way in correcting the neglect of nutrition in standard care in SCD to reduce health inequalities and promote improved patient outcomes in nutrition in SCD. In this way, this research project adds to the evidence base in support of the nutritional needs of people living with SCD.

From earlier discussions, it was clear that MDT working was beneficial to support efficient nutrition service provision in SCD, reflective of nutrition standard 1 (calling on Dietitians to be part of the SCD MDT). The benefit of this collaborative working was successfully demonstrated in the comments from a paediatric Dietitian who was working as part of the SCD MDT in their trust with a SCD consultant who was 'pro-nutrition', although this would

only be effective if the SCD consultant is 'pro-nutrition'. This collaborative working meant patients benefited from having their nutritional needs recognised, assessed and managed by a Dietitian, resulting in positive health outcomes for the patients, which was extended to Dietitians being able to offer wider support that included schools to support school children living with SCD.

In this way, when Dietitians are part of the SCD MDT, a more holistic level of care and support can be offered to the people living with SCD improving their overall health and wellbeing outcomes. For this reason, more effort is required to ensure that Dietitians are included in the steering committee supporting the West Midlands Quality Review Service (commissioned by the UK Forum on Haemoglobin Disorders). In so doing, the quality standards will be able to provide more in-depth guidance on how best to screen for, assess and manage the nutritional needs, risks and challenges of all patients living with SCD, in order for nutritional care provision to be tailored to their needs and of a high quality. Thus, the UK Forum on Haemoglobin Disorders Quality Standards (2021) report, needs to be amended to comprehensively reflect the (reframed optimum nutrition) needs of people living with SCD, where optimum nutrition refers to the medical, nutritional and socio-ecological factors affecting the nutritional needs of people living with SCD. This also means that the West Midlands Quality Review Service (commissioned by the UK Forum on Haemoglobin Disorders), can more comprehensively assess how well the nutritional needs of the people living with SCD are being met in healthcare systems and services in the NHS.

The Annual Review Pro forma provides a comprehensive management plan and health evaluation tool, however it does not consider the close link between the clinical features and related complications and its link to the nutritional complications as experienced by those living with SCD. A helpful example of the successful integration of a service in SCD care provision is new born screening as highlighted by Davies and Oni (2001), that demonstrates the potential value of integrating nutrition into standard care in SCD. In this way, the Annual Review Pro forma, is ideally placed to link nutrition to the medical care of patients. For

example, the Pro forma only checks the height and weight of patients, but fails to record the Body Mass Index (BMI) (ratio of the weight in relation to the height of an individual). This additional information is necessary to assess the malnutrition risk (both over and under nutrition) of patients, providing important information to guide nutritional treatment and management plans as recommended by the Malnutrition Universal Screening Tool (MUST) – a key action required in Standard 2 of the national nutrition standards for SCD and NICE guidance (2006, 2012). Furthermore, the breadth of complications listed in the Annual Review Pro forma highlights not only the complexity of the medical management of the condition, but many of these complications for example: acute chest syndrome, osteoporosis, leg ulcers and pulmonary hypertension - to name but a few – may add to the nutritional risks and nutritional requirements of people living with SCD. Subsequently, patients with these complications may benefit from early referral and assessment from a SCD trained Dietitian or nutritionist to prevent worsening of the nutritional risk and related risk of morbidity. It is a known fact that infection risk can increase the nutritional requirements and risks of people living with SCD, and by making MUST screening a priority as part of the Annual Review Pro forma (ideally at each review appointment), this risk can be identified earlier and potential complications prevented and associated cost and disease burden reduced through nutritional surveillance. Therefore, adding nutrition and diet to the discussion checklists section of the Annual Review Pro forma, would take a more proactive approach to reducing the nutritional risks to patients and address the health inequality associated with the invisibility of nutrition, as it aligns with wider health policies in other long term conditions, such as cystic fibrosis, oncology and diabetes for example. Even adding diet and lifestyle in the holistic assessment (in the Pro forma) will raise awareness of the importance of nutrition in the standard management of SCD.

The solution to support the building blocks for nutrition service provision, at national and professional policy and practice level, includes the national nutrition standards for SCD (SCS, 2018, p75-77) with Dietitians playing a central part in care. The findings of a cross

sectional survey looking at the knowledge, involvement, and attitudes of Dietitians towards SCD, found that 96% of the responding Dietitians believed they had a role to play in managing the nutritional needs of people living with SCD, and 87% believed that SCD warrants nutritional interventions (Matthews, 2016g). Furthermore, Dietitians have been identified to help the NHS Outcomes Framework (2021/22) achieve domain 2 (improving the quality of life of people living with long term conditions) according to the report by the BDA and BSNA (2013.). Dietitians have also been shown to play an important role in primary care, as part of the Additional Roles Reimbursement Scheme (ARRS) roles (NHS Plan, 2019; BDA, 2015), meaning they are best placed to provide nutritional care to patients who attend their GP practices, as identified in a small project, conducted by myself in my role working as a primary care Dietitian (Appendix 19, Example 1).

NHS Health Outcomes Frameworks:

The NHS outcomes framework (2021/22) provides national level accountability for the outcomes the NHS delivers. Five domains are provided, geared towards reducing health inequalities in care provision for all populations. The findings of the evaluation of the health literacy framework thus aligns well with the five domains of the NHS outcomes framework as it has shown to address the knowledge and care gaps identified in the research project, to reduce the health inequalities associated with SCD as a marginalised group but more importantly, the effects of poor nutrition service provision, in supporting policy and practice in order to integrate nutrition in standard care in SCD.

Similarly, the NHS Public Health Outcomes Framework (2020/21) indicators help health and care professionals and the public to understand trends in public health, and that provides guidance to improve health outcomes in all the patient populations. Although the health literacy framework does not directly provide information about health trends, the information does reflect the current *status quo* in the nutrition landscape in SCD, and the specific needs, risks and challenges that impact on the health outcomes of the people living with SCD. In particular, the HL-NIF (blueprint) (section 4.6.3, Figure 4.5) is geared towards providing a

pathway to support the development of nutrition service provision, using the health literacy PEECE Model that reflects the findings of the research project, highlighting both the problem, but equally offering a tailored solution to support national and professional policy and practice development in nutrition in SCD.

5.5.2 Policy and practice recommendations to integrate nutrition in SCD:

As discussed in Chapter 2 (section 2.1.1 Figure2.1), the paucity of research about the lack of integration and nutritional management in SCD, coupled with the ongoing lack of nutrition service provision in SCD management, is affecting the nutritional health and wellbeing outcomes of people living with the condition. These poor health and wellbeing outcomes need to be challenged at national and professional clinical and nutritional policy and practice levels, with people living with SCD being placed at the centre of SCD care provision. To facilitate this change, the lack of translation of the existing corpus on the role of nutrition in SCD into meaningful evidence base, also needs to be challenged if future nutrition policy and practice guidance in SCD is to be developed and for the current status quo to be changed. However, if the current status quo in nutrition service provision in SCD is to be disrupted, existing, new and emerging research conducted in this area, (including this research project), should be working towards linking their research findings to clinical practice, with the view to contribute to the development of policies to improve nutritional practice in SCD, as called for by Nartey et al., (2021).

This research project is the first attempt at integrating nutrition into standard care provision in SCD and as such, aims to break the cycle of poor nutrition service provision in SCD management to reduce the risk of the late diagnosis of nutritional problems in people living with SCD, a key finding of this study (Chapter 4, section 4.2.2). It can therefore be inferred, that the identified risk of the late diagnosis of nutritional problems in this patient population can be attributed to the lack of access and risk of poor outcomes associated with the lack of

nutrition service provision in SCD. In this way, urgent action is needed to not only increase the critical consciousness and praxis about the importance of the role of nutrition in SCD management, but importantly to consider nutrition alongside the medical management of SCD with Dietitians working as part of the SCD MDT, as called for in the UK –based national nutrition standard one (SCS, 2018, pp. 75).

Equally, there is much to learn from conditions such as cystic fibrosis (a genetic condition with a smaller prevalence than SCD), where globally, nutrition has already been successfully integrated into standard care provision for this patient population (Spielberg and Clancy, 2016), albeit that there are more resources and research available to this patient population, when compared to SCD. Nevertheless, the successful integration of nutrition in standard care in cystic fibrosis, provides compelling evidence of the value of this research project aiming to support the integration of nutrition into standard care in SCD management. In doing so, this research project has sought to better understand the nutrition landscape in SCD, that has come to be defined by the lack of nutrition service provision, but has also provided clear evidence of the nutritional needs, risks and challenges facing people living with SCD.

In addition, distinct to existing research on the role of nutrition in SCD, this study is the first to identify a wide range of influencing factors that may have contributed to the lack of integration of nutrition in SCD, with further evidence demonstrating the complexity, invisibility and inequalities linked to nutrition in SCD. These findings summarised in Chapter 4 (Figure 4.1) and the themes developed from the thematic analysis, identified the invisibility of SCD and nutrition in SCD, knowledge and care gaps reflecting the lack of nutrition service provision in SCD, the health inequalities associated with the poor nutritional experiences, access and outcomes as reported by the participants and the many influencing socio-ecological factors affecting nutrition and service provision. In this way, the research project provided primary data that could be used to inform the development of tailored policy and

practice guidance in the UK and potentially globally, to improve nutrition service provision in SCD management.

Hence, the co-development of the health literacy framework and the subsequent development of the health literacy PEECE model (see section 5.4.3) and HL-NIF (Blueprint – see Figure 4.5), was an attempt to facilitate policy and practice development in the UK, drawing on the findings and themes of the data collected in phase one of this research project. This is the first known attempt to co-develop a health literacy framework in the UK in SCD, with these frameworks explicitly summarising the main influencing factors that maintain the status quo in nutrition in SCD, although it may be transferable for use with other marginalised groups and conditions – presenting an opportunity for further application and study. These influencing factors, will need to be addressed in order to improve nutrition service provision for people living with SCD. In a UK context, the frameworks also provide undeniable evidence, reflecting the voice of both the SCSU-Cs and SCSP's acknowledging the marginalisation of SCD, similar to the APPG-SCAT report (2021), '*No one is listening*', a watershed document that is shaping the current clinical pathways in SCD care provision.

Moreover, the evaluation of the health literacy framework by the LAN (see section 5.4.1 of this chapter), the theoretical basis of the HL-NIF (Blueprint), was found to be a valuable education, communication and policy tool, to support the integration of nutrition in SCD, in the UK. In so doing, these frameworks, in particular the HL-NIF serves as a tailored policy and practice blueprint to organisations and highlights the various components and key stakeholders in SCD, that require consideration when integrating nutrition into SCD management. Hence, the recommendations reported below are drawn from and are reflected in the HL-NIF (Blueprint – Figure 4.5), in this way linking the study data (findings and themes) to the study recommendations, to address the invisibility, inequality and promote the integration of nutrition in SCD.

Therefore, if nutrition is to be successfully integrated into standard care, as we see in the case of people living with cystic fibrosis, the recommendations of the study need to be

reflecting the marginalisation and associated inequalities experienced by the SCD population, as discussed in section 5.1. In practice this means, making recommendations to address the invisibility of nutrition in SCD, the health inequalities associated with the lack of nutrition service provision in SCD, addressing the knowledge gaps that has resulted in participants having to self-research what they know about SCD and nutrition in SCD, and creating the opportunity for people living with SCD to have more opportunity to talk about nutrition as part of standard care. It means having Dietitians who work as part of the SCD MDT, in primary and community level settings and it means that existing policies in SCD reflect both the medical and the nutritional needs of people living with SCD.

In this way, sickle cell patients will be in a better position to have a positive experience, access to and able to have improved health and wellbeing outcomes because both their medical and nutritional needs are acknowledged and managed well. Below is a list of the recommendations for the research project to facilitate these changes to the nutritional management of patients living with SCD, at policy and practice level.

Recommendations to integrate nutrition in SCD:

Nutrition service provision is not established as part of the standard practice in SCD care provision and the recommendations below aim to address the influencing factors identified through the research project. These influencing factors are reflected in the four main themes below and the corresponding recommendations, are small steps towards developing nutrition services that can be available to all patients living with SCD, thereby supporting the integration of nutrition into standard care in SCD.

Theme 1 - Invisibility of SCD:

- 1. To acknowledge the lack of nutrition service provision in SCD - affecting patient experience, access and outcomes.*

Practice - based action - to liaise with the secretariat and chair of the APPG- SCAT for nutrition to be placed on the APPG - SCAT agenda as a standing item to be discussed.

Recommendation 1, will serve to increase the critical consciousness of healthcare professionals in SCD care provision about the nutritional needs, risks and challenges facing people living with SCD, as identified through the research project. Adding nutrition to the APPG-SCAT agenda as a standing item, will not only bring the problem of the invisibility of nutrition in SCD to the forefront of the members of the APPG -SCAT and the SCS in the short term but enable ongoing advocacy and conversations to ensure the uptake, inclusion and integration of nutrition into future national and professional policy and practice guidance in SCD management.

Theme 2 - Under-recognised importance of nutrition:

2. *To improve nutrition service provision available to patients living with SCD - to reduce the risk of the late diagnosis of nutritional problems in people living with SCD.*

Practice- based action: to liaise with the UK Forum on Haemoglobin Disorders, for the inclusion of the national nutrition standards in the UK Forum Quality Standards, serving as a point of reference for the West Midlands Quality Review Service.

3. *To improve the access of patients living with SCD to nutrition service provision - allowing them more opportunity to speak about their nutritional needs and concerns.*

Practice -based action: to liaise with the UK Forum on Haemoglobin Disorders and SCS to consider how holistic nutrition management information (BMI and MUST), nutrition and diet related factors, including a trigger for referral to the Dietitian can be included into the existing clinical documents such as the Annual Review Pro forma and GP OPD letter.

4. *To recognise the nutritional needs, risks and challenges of patients living with SCD, by SCD consultants and the wider SCD health and social care team, in order to improve patient outcomes.*

Practice-based action: to work with the SCS, BDA and HCCs to encourage and support SCD clinicians and health and social care professionals to ask patients about their diet and

nutrition (such as their food intake, appetite, concerns or questions), as part of their standard care.

Recommendations 2 and 3, allows for the streamlining of future clinical and nutritional policy and practice documentation in SCD management to facilitate the nutritional management of people living with SCD, in this way recognising the nutritional needs, risks and challenges of the patients (Recommendation 4). In so doing, SCD clinicians will be able to ask about the nutritional concerns of patients and SCD patients will have the opportunity to speak about their nutritional needs, risks and challenges, as part of their standard management and care. This will also help to raise awareness of the importance of nutrition in the management of SCD, inadvertently bringing nutrition to the forefront of the SCD clinician's minds including GP's, with the added benefit of increasing the surveillance of nutrition in the management of SCD to improve the patients experience, access and outcomes in nutrition.

Theme 3 - Lack of priority to nutrition:

5. *To develop tailored nutrition policy and practice guidance to support the development and implementation of nutrition service provision in SCD, to provide pro-active rather than reactive nutrition service provision.*

Practice-based action: to liaise with the SCS, UK Forum on Haemoglobin Disorders, BDA on how best to use the HL- NIF (Blueprint) to guide health planners, policy makers, NHS and related organisations about the components required for nutrition service provision in SCD.

6. *To increase the involvement of Dietitians in the nutritional management of patients living with SCD as part of the SCD MDT in community and primary care settings, to facilitate the nutritional assessment, diagnosis and treatment of SCD patients.*

Practice-based action: to liaise with members of the reformed NHS England and the BDA, with support from the SCS and APPG – SCAT, to call for the implementation of the nutrition standards for SCD.

Recommendation 5, provides a helpful framework to contribute to the development of tailored nutrition policy and practice guidance to support nutrition service provision in SCD. Recommendation 6, is well supported by the nutrition standards in SCD as the standards provide evidence base in support of the need for increased involvement of Dietitians in the nutritional management of patients living with SCD, as part of the SCD MDT. Moreover, in this role the Dietitian is able to provide nutritional screening, assessment and management of the nutritional needs, risks and challenges of patients living with SCD, as identified through the research project, to improve the patient experience, access and health outcomes in nutrition. The standards also serve as a call to action to the BDA, to support the role of the Dietitian in the nutritional management of people living with SCD.

Theme 4 - Multi-level factors affecting nutrition and service provision

- 7. To develop tailored rather than generic nutrition education, resources, policies and training curricular based on the specific needs, risks and challenges of the SCD patient population, to reduce the need for SCD patients to self-research nutritional information thereby reducing their risk of miss-information.*

Practice-based action: use the data generated in phase one of the research project (summarised in the co-developed health literacy framework), highlighting the complexity of nutrition in SCD to develop tailored nutrition related education and training resources including policy and practice guidance.

Recommendation 7, highlights the need for tailored nutrition education and training programmes in SCD that are essential to add to the nutrition knowledge and skills of a range of healthcare professionals, including Dietitians. Without evidence based education and training, health and social care professionals will continue to have suboptimal knowledge and information of how best to assess, diagnose and treat the nutritional needs of patients living with SCD. There is a need for the reformed NHS England to commission high quality evidence-based nutritional training in SCD, to increase the literacy and empowerment of

both the patients and healthcare providers. In this way, support can be provided to people living with SCD to better self-manage their nutritional needs, risks and challenges to improve their health outcomes.

Challenges in implementing the recommendations:

Being the first attempt at working towards the integration of nutrition in standard care in SCD, the implementation of the recommendations are challenging, not only in relation to the lack of policy and practice guidance but more so historically (as nutrition is not seen as a priority), culturally (requiring a shift from the traditional medical focus) and financially (the cost implications - workforce, Information Technology (IT) and changes to existing institutional structures and systems).

Main Challenges:

Historically, nutrition is not seen as a priority in SCD care provision and there remains a lack of awareness and critical consciousness of the role of nutrition in SCD - despite the existence and inclusion of the national nutrition standards in the national clinical standards for adults living with SCD (SCS, 2018). In addition, there is a need to convince key policy and strategic stakeholders of the importance of nutrition in SCD management. This will require a cultural shift as traditionally, medical management is the focus of care provision in SCD - and the '*No one is listening*' report (APPG – SCAT, 2021), bears witness to this reality. As such, there are competing priorities between the medical and nutritional management of SCD, especially in the absence of nutrition policy and practice guidance in SCD.

The invisibility of nutrition in SCD is thus driving the status quo of the lack of nutrition service provision, despite the existing literature and the findings of this research project confirming the existence of the nutritional needs, risks and challenges of people living with SCD. Whilst the SCD population remains disempowered to ask for what they need (from policy makers) in relation to their nutritional needs, there is overwhelming evidence of their nutritional

needs, as identified through the findings of this research, from the perspective of both the SCSU-Cs and SCSPs. In addition, the lack of awareness and praxis on the part of the BDA, with regards to the level of consideration they have given to the nutritional needs of the SCD population, a shift in culture is required in relation to their interest and commitment to consider nutrition in SCD as an emerging speciality in the profession, moving forward.

Herein lies the importance of firstly, improving the knowledge and awareness of the causal link between the clinical features of SCD and nutrition in SCD, and secondly at a structural level, acknowledging the impact of the marginalisation of SCD. However, in the current financial climate, the cost implications of increasing nutrition service provision are a major factor in regards to workforce development, IT and institutional structures and systems. These are truly challenging factors but if we neglect to change, then we are contributing to the health inequality linked to the lack of nutrition service provision in SCD, that are affecting the patients experience, access and outcomes, thereby potentially increasing the cost and health burden of SCD.

Mitigating the challenges:

Even though the challenges (historical, cultural and financial) in implementing the recommendations seem insurmountable - the recommendations reported above, provide opportunities to bridge some of these structural/macro level challenges, becoming small stepping stones towards policy and practice level change, in the absence of nutrition service provision. These structural/macro level challenges will however, continue to impact the change required in policy and practice in nutrition in SCD. For this reason, the findings of this research project are important as it provides a tangible evidence base to question some of these structural hurdles, giving value and worth to the recommendations being made and the need for change. Moreover, without understanding the context of the current clinical pathways and priorities in the medical management in SCD- triggered by the '*No one is listening* report' (APPG-SCAT, 2021), overcoming these challenges would seem to be impossible. Thus, in light of the difficulty inherent in these macro level factors, I have

endeavoured to engage in a number of albeit micro level steps/actions, to help me mitigate the potential challenges in the implementation of the study recommendations. Below is a list of these micro level actions:

- Being visible in the SCD space to increase knowledge and raise awareness of nutrition in SCD– for example engaging in education sessions (17/06/22), attending SCD conferences (26/10/23), presenting online international seminars on nutrition in SCD (4/12/22), and leading on a Mayoral school's education campaign to raise awareness of SCD and nutrition in secondary schools, in Waltham Forest (July 21 – May, 2022).
- Networking by keeping in contact with key colleagues working in SCD who have understanding of the challenges in SCD management, via zoom calls, regular phone calls working etc.
- Being solution focussed, which was the motivation to develop an evidenced based framework such as the HL-NIF (Blueprint) to support the implementation of the recommendations for the study.
- Asking for help from mentors and colleagues in navigating these challenges.
- Undertaking this doctoral research project, with its focus on changing policy and practice in nutrition in SCD management - partly draws attention to the historical and cultural challenges in nutrition in SCD, which the recommendations are aiming to address.
- Adopting a participatory and collaborative research/learning methodology to the research project involving the patient and provider voice, was an attempt to provide insider knowledge, views and perspectives on nutrition in SCD, thereby drawing attention to the need for changing the historical and cultural perspectives held about the value of nutrition in SCD care provision, impacting patients experience, access and outcomes.

Impact of the research project:

In summary, the impact of this research project, of which the recommendations play a big part, is constantly unfolding. As previously mentioned, the challenges are huge however, the recommendations are stepping stones to facilitate change and bridge the gaps in working towards improving nutrition service provision available to all people living with SCD, as highlighted in the HL- NIF (Blueprint – Figure 4.5).

For this reason, the research project has intended to highlight the marginalisation of SCD both in light of the SCD population and nutrition in SCD, these insights in particular playing a pivotal role in challenging the status quo - the lack of nutrition service provision in SCD.

Below is a list of the practice-based actions I have already undertaken to enhance the impact that this research project will have in the future in regards to achieving the overall aim of the research project - to support the integration of nutrition in SCD:

- I was able to speak directly with the chair of the APPG- SCAT (17/10/23) who agreed for me to have a 10-minute speaking slot at the next available APPG-SCAT meeting.
- I sent a supporting letter to the Chief Executive Officer (CEO) of the BDA (19/6/23) – see Appendix 19 (Example 4) and am currently having meetings with the incoming BDA chair (9/10/23) about how my research can be implemented to support the involvement of Dietitians in the nutritional management of people living with SCD.
- I sent a supporting letter to the Deputy Director and Director of health inequalities (NHS England) (25/9/23) – see Appendix 19 (Example 5) and had an initial introductory meeting (11/10/23) with the national lead for health inequalities (NHS England) about my research and its aim.
- I had two short follow up meetings with the chair of the SCD Patient Advisory Group (PAG) (17/10/23) with opportunities to speak with SCD patients to increase their awareness of the role of nutrition in their standard care provision, to improve their health outcomes.

- I was invited to speak about nutrition (5/10/23), as part of an NHS England sponsored education series alongside an NHS medical professional working in SCD.
- I have registered my non-profit organisation (24/10/23) with the aim of after successfully completing my doctoral degree, using it as a platform towards the implementation of the study recommendations - a conference is being planned for September 2024 to disseminate the findings of the research and promote the involvement of Dietitians as part of the SCD MDT.
- I have had a meeting (18/10/23) regarding a community improvement project in SCD– to support the recruitment of the first Dietitian to work as part of the SCD MDT- in line with nutrition standard one (SCS, 2018 p 75).

5.5.3 Reflexive perspectives on my personal and professional learning and development:

My debut book (Matthews, 2019i) starts with the chapter titled '*Looking back to go forward*', and in looking back, I can see how living in Apartheid South Africa has influenced my beliefs, judgements and values and how this in turn has affected my experiences, perceptions and limitations (Finlay, 1998). In the same way these experiences, beliefs, judgements, values as well as my faith, have shaped the decision making and choices throughout my life, including my doctoral journey. In stepping out into the world of academia, qualifying to work as a Dietitian, first in South Africa and then relocating to the UK in 2000, opened up my perspective of the world, the patients I looked after and my capability to achieve beyond the limitations of my beliefs and values. I was drawn to working with people from marginalised backgrounds, and worked for four years in a SCD service in East London. In this way, people living with SCD became a natural choice to work with, when the opportunity arose in 2011.

Moreover, the ongoing personal and professional learning and development process has allowed me to develop my research competencies in nutrition in SCD, backed up by my professional knowledge, experience and expertise as a Dietitian. Evidence of my evolving learning and development as a researcher, was being able to synthesise new ways of understanding existing concepts, for example redefining how I used health literacy in nutrition in SCD, to be an expansive and integrative concept to support the development of policy and practice in SCD, different to its traditional use in literacy and information sharing. Earlier on in my doctoral journey I was also able to pivot, from my original aim (developing a nutritional prompt sheet in nutrition in SCD) to instead consider – the integration of nutrition in standard care provision, that had never before been done in SCD. To account for any bias in the study, based on my reflexive positionality in this research project, I have used the Lincoln and Guba (1985) seminal model to ensure the trustworthiness of the data generated in the research project, as discussed in Chapter 3 (see section 3.3.2 (iii)), and have chosen a participatory Learning Alliance (LA) methodology, deemed to be appropriate in research with marginalised patient groups, such as the SCD patient population.

The clinical features of SCD were known to have a devastating impact on the health and wellbeing outcomes of this patient population. I was inspired to see how I could use my professional knowledge and nutritional and clinical experience working as a Dietitian, to address some of the health challenges of this patient population, since nutrition played a critical role in improving the health and wellbeing outcomes in the patients I was already managing. As a direct correlation existed between the clinical features of SCD and nutrition, and how nutrition is able to improve the clinical and health outcomes in other long term conditions such as diabetes and heart disease for example, I was compelled to investigate the role of nutrition in SCD. I felt obligated to take the 'bull by the horns' and undertake this doctoral level research despite my lack of research experience, despite the limited interest in nutrition in SCD, despite the lack of support and resources from the BDA and SCS and despite nutrition in SCD being a neglected part of standard care provision.

Preceding, the doctoral journey, I wrote a number of articles on the topic to increase critical consciousness of nutrition in SCD and create an opportunity for praxis (critical reflection towards action), both within and outside of SCD. In this way, I established my interest in SCD and the need for change. The doctorate became the vehicle for policy and practice level change in nutrition in SCD, hence the decision to undertake a professional doctorate degree. What became clear to me was the benefit of working in primary care in a Primary Care Network (PCN) role, within a group of GP practices. This role enhanced my knowledge about the current institutional services and systems (such as the Integrated Care System (ICS), IC Boards (ICB) governing health planning, policy development and integrated care delivery in the NHS.

It is in this role, that I advocated for the opportunity to conduct a project in nutrition in SCD, to assess and support the health and wellbeing outcomes of people living with SCD in the PCN (see Appendix 19- Example 1). This project was instrumental in highlighting how nutrition can be integrated in primary care, what are some of the challenges that need to be addressed but more importantly, ways in which Dietitians are able to improve the health and wellbeing outcomes in the SCD patient population. These insights have added to the perspectives of how I am able to understand nutrition in SCD, and the possibilities of how, with the support of the findings of this research, Dietitians are well placed to manage the nutritional needs of people living with SCD in primary care and in the community.

Dietitians can therefore play an important part in how well the nutritional needs of the SCD patient population can be managed, as part of standard care. Thus the theoretical contribution of the research project (the knowledge and care gaps, the socio-ecological factors and facilitators) provide insights into how service provision ought to be developed and implemented, tailored to the specific needs of the SCD patient population. A key finding from a cross sectional study (Matthews, 2016g), highlighted knowledge gaps, and a lack of nutritional guidelines and standards as reasons for low involvement of the Dietitian in SCD care. It was thus imperative that this research project, aimed to address these gaps in

service provision. Moreover, key messages identified through the research and literature review, added a helpful perspective in support of the existence of both the medical and nutritional needs of people living with SCD.

What was also emerging through the research was the role of the social and wider determinants of health, adding to the complexity of the nutritional needs, risks and challenges of the SCD population. In response to this emerging complexity of nutrition in SCD, I was able to redefine optimum nutrition, as the cumulative causatum of the nutritional, medical and socio-ecological factors affecting the nutritional needs of the SCD population, impacting their health and wellbeing outcomes helpful for Dietitians to tailor the nutritional management of patients living with SCD. Developing tailored nutrition education and training resources are therefore essential to communicate these theoretical conclusions, that should be ingrained in the development of future nutrition policy and practice guidance in SCD. In this way, the HL-NIF (Blueprint), reflects the complexity of nutrition in SCD, becoming an instrumental tool to communicate these essential findings of the research project in support of the development of nutrition service provision in SCD.

Working in my current PCN role and undertaking this professional doctorate, was favourable in me being nominated to be part of the NHS England SCD clinical pathway review, in August 2022. This involvement not only helped me to understand the complexity of national level policy and practice development, but provided me with key contacts to facilitate future meetings, such as the introductory meeting with the lead of health inequalities (NHS England). As such, I have had the opportunity to directly share some of my research findings in support of the integration of nutrition into standard care in SCD, with the view to raise awareness of and reduce the risk of the late diagnosis of nutritional problems in the SCD patients, as identified through the research project. Amidst pressing medical management priorities, more conversations are planned in the future to highlight the health inequality of poor nutrition service provision in SCD. This would of course be strengthened with increased

support and backing from the BDA and the SCS, key strategic bodies who can facilitate change in nutrition in SCD at national policy level.

The learning process was not always easy as, I would often revert to my default ways of thinking, especially when I felt confused or unsure of my next steps, or when I received constructive feedback from supervisors, reviewers and peers. Thus developing personal leadership empowerment, was key to help me navigate the complexity and challenges of doctoral level research. As such, my biographical narrative, my faith, my dietetic clinical and professional experience and world view of social justice, advocacy and empowerment, has laid the foundation to undertake this novel research - in a marginalised patient population where nutrition is invisible, under recognised and lacks priority. It is my vision that the patient and provider voice will be heard through this research project and be the catalyst for change in policy and practice in nutrition in SCD.

5.6 Limitations and Strengths

5.6.1 Limitations:

A significant limitation of the study relates to the fact that demographic information for both groups of participants were not included. This missing data lead to a gap in the understanding of the direct impact of the ethnicity, racism, and socio-economic standing of particularly the SCSU-Cs. However, some of these background insights have emerged through the participant responses, although collecting this information would have provided grounding for the discussion on the sociological implications of conducting research in a predominantly marginalised patient population. Insights into the participant's ethnicity, racial discrimination and prejudice, and how this has played out in their experiences of healthcare and healthcare provision would have added a richness to the data.

However, the overall responses relating to the knowledge, views and experiences of the SCSU-Cs in particular, of the medical and nutritional management of SCD, and those of the SCSPs, reflect the impact of the under recognition, and lack of importance ascribed to the nutritional needs and management of people living with SCD. With engagement and recruitment in SCD research being a problem, this was done to increase recruitment level. However, the impact of the marginalisation in SCD emerged organically, evident in the responses of both participant groups.

The LA methodology is regarded as a multi-stakeholder methodology; as such, a potential limitation of the study was the under representation of particularly strategic bodies such as the SCS and the BDA. Having representatives present from these two organisations would have provided a wider perspective to the discussions and the overall data collection.

However, this creates an opportunity to research further, possibly within a multi-location study. The inclusion of a SCSP (a GP), who worked in primary care with connections to the local Clinical Commissioning Group (CCG) at the time and secondary care services, provided a helpful perspective from a policy and practice and health planning perspective, although they were not able to be part of the LAN.

Using a two-step process to the co-development of the health literacy framework meant that the initial version of the framework (Appendix 11), co-developed by the LAN, rather than the final version of the framework, the HL-NIF, blueprint, (section 4.6.3, Figure 4.5) was evaluated as part of the research project. In addition, the evaluation of the framework was done by the LAN, and not by all the participants who attended the focus groups. This could have influenced the findings of the evaluation questionnaire, as those participants who were not part of the LAN may not have been able to answer the questions as comprehensively, since they were not part of the co-development of the health literacy framework. In the same way, it may have provided an opportunity to compare the findings between the two groups. Furthermore, the scaling and generalisation of the findings may be limited and may therefore benefit from future studies being multi-location, for example using the HCCs as a focal point

to assess how well the findings of the study and evaluation of the framework reflects others knowledge, views and perspectives on the medical and nutritional management of people living with SCD.

Although the study considers health literacy as a key concept in policy and practice development, the study did not explicitly assess the health literacy skills of the participants, as this was not part of the aims of the study. Instead, through the study identifying the knowledge views and experiences of the participants, the study implicitly gauged the health literacy of the participants about the medical and nutritional management of SCD and the influencing factors affecting nutrition and service provision. Thus, the study did not explicitly determine the participants' ability to navigate healthcare services or their knowledge of public health and health promotion. Overall, the participants' responses provided some valuable insights into this through their responses, and the collective viewpoint reached by the LAN helped to overcome the potential individual poor health literacy of the group in improving their knowledge of nutrition and the influencing factors affecting nutrition in SCD and related policy and practice needed to improve nutrition service provision.

5.6.2 Strengths:

The Medical Research Council (MRC) promotes the use of the Knowledge to Action (KTA) process as a measurement of the methodological rigor of new innovations (Graham et al., 2006). The development of the HL-NIF (blueprint) (Figure 4.5) can be considered an innovation as currently this is a gap in the nutrition in SCD corpus. In particular, the use of health literacy as an expansive and integrative concept in policy and practice development in nutrition in SCD. As such, the theoretical framework of the research project provides sound theoretical basis for the study and draws on principles of the critical social theory and critical pedagogy underpinning the seminal work of Paulo Freire (1970). Key gaps informing the research project are the lack of education about the role of nutrition in the management of

SCD and that education, according to Paulo Freire (1970), is a tool of emancipation and empowerment contingent on critical consciousness (an awareness of the social discomforts of the oppressed) and praxis (critical reflection leading to action). Both education and empowerment are concepts of the HL-NIF (blueprint). In this way, the HL-NIF (blueprint) provides evidence base to raise critical consciousness and praxis of the need to integrate nutrition in standard care in SCD.

Additional strengths of this research project includes the following:

- Challenges theory underpinning the nutritional management in SCD from the perspective of oppression and marginalisation.
- Brings the people living with SCD into the forefront of nutrition service provision planning giving them a 'voice' in nutrition integration in SCD.
- Calls out the injustice of the lack of nutrition service provision from both a service user/carers and provider perspective.
- Provides a practical framework to support the integration of nutrition into standard care in SCD adopting a whole systems policy and practice strategy.
- Provides a comprehensive lens to identify and understand nutritional management in SCD and facilitates the reconceptualization of optimum nutrition in SCD as being more than food but multi-factorial.
- The research project methodology provides a modified version of the LA methodology – being the first LA study to use independent focus groups before the formation of LANs.
- As a Dietitian, I was able to use my group facilitation skills during the focus group data collection meetings, a clear advantage in qualitative fieldwork.

Overall, the study facilitated a multi-disciplinary perspective as it drew on theoretical concepts from health, sociology, nutrition, education and psychology to achieve the aim and objectives – and formulate the recommendations for the study.

Summary:

A discussion of the main influencing factors reflected in the knowledge and care gaps identified through the research project, provided helpful insights to understand how these gaps impact on the marginalisation and the lived experience of nutrition in SCD, of people living with SCD. In particular, the late diagnosis of the nutritional problems in people living with SCD, is evidence of how the lack of nutritional management (a health inequality) in SCD is impacting on the experience, access and health outcomes, in nutrition in SCD. The emergence of the social structural factors and how it impacts on the nutritional needs and management of SCD cannot be ignored, and clearly illustrates the health inequalities inherent in SCD healthcare provision.

Taken together, all these factors reflect the complexity of nutrition in SCD, requiring a comprehensive lens at population level, to fully identify and understand the myriad of influencing factors affecting the nutritional management in SCD. In this way, health literacy is conceptualised as an expansive and integrative concept to tailor policy and practice and the co-development of the health literacy framework and the consequent development of the HL-NIF (blueprint), important to addressing the multiple knowledge and care gaps in nutrition in SCD. The next Chapter will discuss the main conclusions of the study, including the contribution to knowledge and future directions of the research project.

Chapter 6

Conclusions and Recommendations

This chapter provides an overview of the key conclusions based on the main influencing factors reflecting the core knowledge and care gaps underpinning the medical and nutritional management of people living with Sickle Cell Disease (SCD), as reported and discussed in Chapter 4 and 5, respectively. As well as providing a summary of the contribution to knowledge, this chapter highlights the central policy and practice recommendations towards achieving the integration of nutrition in SCD, including the next steps to extend the impact of the research project.

6.1 Conclusions

The findings of the research project identified a range of knowledge and care gaps that reflect the current nutritional management and the existing nutrition landscape in SCD. The knowledge and care gaps reflect the myriad of influencing factors that need to be addressed to improve nutrition service provision in SCD. Although not explicitly explored in the research project, the marginalisation of SCD was found to play a significant role in the lack of nutrition service provision in SCD, a neglected concept identified in the existing scientific literature exploring the role of nutrition in SCD, as reviewed in Chapter 2. However, the impact of the marginalisation of SCD, influences our understanding of what matters most to the SCD patient population when placing people living with SCD at the heart of care provision. In developing future nutrition service provision in SCD aimed at integrating nutrition into standard care in SCD, it is paramount that the impact of the marginalisation of SCD on the level and quality of care provision in both the medical and nutritional management in SCD, is considered.

Hence the co-development of the health literacy framework, the main output of the research project, should equally acknowledge the central role of the people living with SCD and how the framework depicts and addresses the knowledge and care gaps reflective of the challenges and risks of people living with SCD, with unaddressed nutritional needs. In this way, the evaluation of the health literacy framework played an important role in establishing how well the framework was able to contribute to policy and practice change, to support the integration of nutrition into standard care in SCD. Thus, the implicit dual intension of the health literacy framework as a tool to address the knowledge and care gaps in nutrition in SCD, serves also to improve the nutrition service provision in SCD, whilst inadvertently also serves to improve the experience, access and health outcomes in nutrition, for people living with SCD. The following conclusions shed light on the how the research project findings and analysis helped to support the achievement of the overarching research project aim and objectives and address the knowledge gaps of the research project.

The marginalisation in nutrition in SCD:

The foundation for advocating for the nutritional management of people living with SCD, was largely dependent on understanding the factors which influence the medical management of people living with SCD. Thus the marginalisation of SCD, as evidenced in the medical management of SCD, can be linked to the lack of nutritional management in SCD. This same patient population experiencing the effects of marginalisation as part of their medical management, will at some point, require access to nutrition service provision. It is not a stretch to extrapolate that whatever affects the medical management in SCD – as seen in the reports of the APPG-SCAT (2021) that racism led to failings and lack in the level of care to people living with SCD – will most likely affect the failings and lack of nutritional management in SCD. So too, will the health inequalities in the medical management in SCD influence the health inequalities in nutrition in SCD.

However, the priority given to the medical management of SCD, in existing SCD care provision, still overshadows the role of nutrition in SCD, augmenting the inequalities to people living with SCD who may require nutritional management. Thus the lack of nutrition service provision in SCD, negatively impacts on the experiences, access and outcomes of people living with SCD, in the same way that the health inequalities in SCD medical management affects the experience, access and outcomes of this patient population. The clinical symptomology inherent in SCD requires medical management, however, the same clinical symptomology in SCD is also responsible for the nutritional implications in SCD. Notwithstanding, these clinical symptoms and features in SCD often result in a range of serious complications which are known to contribute to the high levels of morbidity, mortality, disability and poor QOL outcomes associated with SCD, being a multi-system and multi-organ, long term condition. The effects of the lack of nutrition service provision in SCD, may likewise be adding to the healthcare burden in SCD. Thus, the invisibility of nutrition service provision may be directly affected by the marginalisation of SCD and the medical management in SCD.

Invisibility of nutrition in SCD:

Acknowledging, the invisibility of SCD as a significant problem is therefore important if the invisibility of nutrition in SCD is to be addressed. Although COVID 19 has had a positive effect on bringing SCD under the health radar in raising awareness of the infection risk of people living with SCD, it has also raised a level of scepticism amongst the SCSU-C group about the motives behind this action taken, as the APPG-SCAT (2021) report refers to the ongoing invisibility and lack of trust between the SCSU-Cs and SCSPs. Pertinent to nutrition in SCD, the invisibility relating to the causal links between the clinical features of SCD and the nutritional needs of people living with SCD needs to be highlighted. In neglecting the causal link between the clinical features of SCD and the nutritional implications of SCD,

confounded by a lack of nutrition service provision in SCD management, the people living with SCD remains at an increased risk of the late diagnosis of their nutritional problems. This was a significant finding of the research project, with direct impact on patient experience, access and outcomes.

This finding was true in both the paediatric and adult patient cohorts, meaning that potentially, all patients living with SCD, who do not have access to nutrition as part of their standard care, may be at risk at any given time. Hence, the consequence of the late diagnosis of nutritional problems in people living with SCD may ultimately be adding to the high level of morbidity, mortality, disability and poor QOL outcomes associated with SCD.

Thus the implications of the invisibility of SCD is far reaching and could also be linked to the poor knowledge and awareness of the condition, a clear finding of the research project. In particular, the research findings reflected that in those SCSPs whose knowledge of SCD was limited, their limited knowledge had a direct impact on their level of care provision to patients living with SCD. Moreover, when nutrition service provision was provided in isolation and not part of the SCD MDT, the knowledge and potential to learn more about the condition, was limited.

However, for example when the Dietitian in the research project worked as part of the SCD MDT, they acknowledged that the overall patient experience, access and outcomes of the people living with SCD in their care could be greatly improved. The Dietitian also reported that working as part of the SCD MDT increased their learning potential several fold. In the same way, they were able to share their knowledge of nutrition with the SCD MDT, and in so doing was able to add to their nutrition knowledge. The improvement in nutrition care provision, when Dietitians are part of the SCD MDT, means that SCD consultants become important gatekeepers for the integration of nutrition into standard care provision for people living with SCD, so too are other members of the SCD MDT, who are in a position to advocate for the nutrition needs of the people living with SCD in their care. This action by SCD consultants, and wider SCD MDT members, could have positive ramifications on the

impact of the invisibility of SCD and the invisibility of nutrition, to improve the inequalities in nutrition service provision in SCD management.

Nutrition as a health inequality in SCD:

As already established, poor knowledge in SCD and nutrition can be linked to the level and quality of care provision in SCD. Thus a significant finding of the research project, amongst both the SCSU-C and SCSP groups, was the fact that the majority of the participants had to self-research what they know about SCD and nutrition. Depending on their level of involvement in the SCD MDT, some of the SCSPs had more knowledge than others, demonstrating the value of healthcare professionals working as part of the SCD MDT, and not in isolation. The fact that the majority of the participants had to self-research, was in itself an indicator of the lack of nutrition knowledge and resources to support the nutritional management of people living with SCD. The disparity in the levels of knowledge about nutrition in SCD, therefore negatively impacts on the quality of care provision to the SCD patient population. As a result, the lack of nutrition knowledge and resources compounds the lack of knowledge of other supporting healthcare professionals in their ability to adequately support the nutritional needs of people living with SCD.

The lack of knowledge about nutrition in SCD, highlights the need for more tailored evidenced based resources to be made available to the SCD patient population and healthcare professionals. The availability of tailored nutrition resources, education and training courses and information would also enable Dietitians in particular, to learn how to enhance and improve their management of the nutritional needs of people living with SCD, which are varied and can be complex. The SCSU-Cs identified the value of having Dietitians as part of the SCD MDT, however, referrals of patients made to the Dietitian were often not done. Thus, although Dietitians are recognised as diet and nutrition experts, they are not routinely providing nutritional care to people living with SCD, and when referrals are made, the advice given to patients may be more generic and not necessarily tailored to SCD, due

to the lack of available tailored education and training and nutritional resources to support dietetic practice in nutrition in SCD.

For instance, SCSU-C participants highlighted the fact that people living with SCD, do not have a good appetite when unwell. This in turn has a substantial impact on their ability to eat well to meet their increased nutritional requirements, during periods of ill-health. A SCSU-C mentioned that, '*all they wanted from the Dietitian was to know what type of foods to eat*', but this information was not always provided. The findings of the research project confirm the existence of the nutritional needs of people living with SCD in line with existing research. As such, the patients wanted more opportunity to talk about their nutritional needs during their review appointments with the SCD consultant. However, time constraints confounded by nutrition not being part of standard care in SCD, has limited the opportunity for patients to discuss any nutritional concerns or questions. This means that, nutrition in SCD is not adequately recognised and prioritised in standard SCD management, leaving the nutrition needs of people living with SCD unmet and becoming a health inequality impacting patients experience, access and health outcomes.

The complexity of nutrition in SCD:

Adopting a socio-ecological lens to the research project, exploring the micro, meso and macro level factors affecting nutrition and service provision, reflects the complexity of nutrition in SCD management. What can be gleaned about the micro level factors raised reported in Chapter 4, is that nutrition in SCD is more than the reported under nutrition and nutrient deficiencies, and although fluid intake to prevent dehydration is important, it was important to reframe optimum nutrition in SCD, to reflect that myriad of influencing factors affecting the nutritional needs in people living with SCD. A number of personal and interpersonal factors have a direct bearing on how well people living with SCD are able to eat and provide for themselves, especially in times of poor health and disability. More than just gaps in knowledge about nutrition in SCD, the patients required a lot of support from

family, peers and friends to overcome some of the key challenges they experience in relation to food choices, storing food, food preparation and having regular access to money and food. Thus the range of the social, mental, psychological, economical, emotional, religious and spiritual care needs of the SCD patient population, not previously explored in the context of nutrition in SCD, provided primary insights into the micro-level factors affecting nutrition in SCD.

The meso-level factors affecting nutrition in SCD, highlighted key concerns for SCSU-Cs in relation to the level and quality of care provision in SCD. It was obvious that nutrition was not part of the standard care available to people living with SCD, however, the quality of hospital food was a key issue raised as an important influencing factor, in particular how this impacted on their nutritional intake when in hospital in relation to their recovery times. So too, SCSU-Cs identified the impact on the level of support provided by the healthcare professionals in SCD who had a limited knowledge of nutrition in SCD. The SCSPs on the other hand, focussed on problems related to nutrition service provision.

One SCSP identified the need to highlight the role of nutrition in SCD management in a school context, including reinforcing the value of school meals available to pupils living with SCD, who may have increased nutritional needs. Both the SCSU-C and SCSP groups suggested the improvement of nutrition knowledge in the context of transitioning from paediatric to adult services, improving staff knowledge about nutrition in SCD and improving nutrition knowledge and training at schools. In addition, a SCSU-C, suggested using existing SCD conferences to raise awareness of nutrition and its role in standard SCD care.

A number of macro level factors and facilitators were identified by both the SCSU-Cs and SCSPs. Both participant groups focussed mostly on the lack of availability and access to healthy affordable food shops and markets in their local areas. The effects of deprivation and socio-economic status of the population were identified as influencing factors, and the need for additional support within the local community and environments, raising awareness of the impact of the social structural factors in the nutritional behaviours, choices and access to

healthy food options. Both SCSU-Cs and SCSPs highlighted the need to emphasise support available to patients living with chronic conditions, providing support for those who are affected by food insecurity, having help and support available to patients who are unwell. The involvement of statutory and voluntary national organisations such as the reformed NHS England, SCS, APPG -SCAT, and the BDA, were identified as being crucial to support policy change and that service provision be holistic, promoting joint MDT working. Calls for raising awareness of nutrition in SCD, using national social media awareness campaigns, and having policies in place to support nutrition service provision, was emphasised within the SCSP group.

Cumulatively, these multi-level factors contribute to a powerful narrative to reflect the complexity of nutrition and service provision in SCD that necessitates policy and practice change on a population and whole systems level to support nutrition service provision, tailored to the unique needs of people living with SCD.

A whole systems approach in nutrition in SCD:

The complexity of nutrition in SCD is what warrants a whole systems strategy in SCD. In developing this, the LAN formed between the SCSU-C and SCSP groups provided the opportunity for both participant groups to collectively increase their critical consciousness and praxis about the current state of nutritional management of SCD. In this way, the LAN provided a valuable opportunity for both groups of participants to understand the nutritional management needs and challenges of people living with SCD, from a common viewpoint, that acknowledges their different perspectives. A key purpose of the evaluation of the health literacy framework, was to address the knowledge and care gaps in the nutritional management of SCD, as well as determine how effective it can be to support the integration of nutrition into standard care in SCD.

The thematic analysis of the framework provided clear evidence of the benefits of the collaborative and partnership working of the LAN, and how well the framework was able to

address the knowledge and care gaps in the nutritional management of people living with SCD, as reported in Chapter 4. Overall, the development of the health literacy framework and the eventual HL-NIF (blueprint) was based on a multi-level, whole systems approach, tailored to the unique and specific nutritional needs of people living with SCD. The HL-NIF (blueprint), showcased the expansive and integrative conceptualisation of health literacy, espoused from the core knowledge and care gaps of the research project, to encompass a population based, whole systems strategy to support the integration of nutrition into standard care in SCD management.

6.2 Contribution to knowledge

To date nutrition is not part of standard care in SCD and currently no framework exists to support the integration of nutrition into standard care in SCD. Thus, the gaps as identified in Chapter 2 (summary, pg66) inform the main contributions to knowledge of the research project, pertaining to the influencing factors affecting nutrition integration in relation to theory and policy and practice development in nutrition in SCD:

- The co-development of the health literacy framework was found to be a valuable educational, training and policy tool, which has formed part of the theoretical basis for the development of the HL-NIF (blueprint), as an example of a whole systems management strategy informing policy and practice change, tailored to the needs of people living with SCD. In so doing, the HL-NIF (blueprint) is aimed at addressing the influencing factors reflected in the many knowledge and care gaps evident in the lack of nutrition service provision available to people living with SCD.
- A number of socio-ecological factors and facilitators relating to nutrition and service provision were identified through the research project, contributing towards the evidence base supporting the complexity of nutrition in SCD, relevant when

considering the planning and provision of future nutrition services tailored to the unique needs of people living with SCD.

- The novel use of health literacy as an expansive and integrative concept to contribute to policy and practice development, that considers health literacy as more than health education, but incorporates the three health domains relevant in SCD, that parallels between health literacy and education and empowerment, key principles in the LA methodology and Freire's (1970), '*Pedagogy of the oppressed*'.
- The socio-ecological factors facilitated the reconceptualisation of optimum nutrition as I define it to be – a cumulative causatum of the medical, nutritional and socio-ecological factors affecting the nutritional needs of people living with SCD that impact on their health and wellbeing outcomes.

In this way, the research project is the first attempt at integrating nutrition into standard care in SCD and adds to existing literature by providing primary data on the influencing factors – reflecting knowledge and care gaps affecting the nutritional management and service provision in nutrition in SCD. Moreover, the research explores nutrition in the context of marginalisation, which is neglected in existing literature on the role of nutrition in SCD.

6.3 Recommendations for national and professional policy and practice change in nutrition in SCD

The vision driving the focus for policy and practice change in the research project is simple;

- *'for nutritional management to be available to every patient living with SCD as part of their standard care in SCD to improve their experience, access and outcomes'.*

The findings of the research project, reflected in the core knowledge and care gaps identified, illustrate the paucity of policy and practice guidance in nutrition in SCD, upholding the lack of nutrition integration in SCD, in the context of the marginalisation of SCD. The

knowledge and care gaps identified in the research project define the current nutritional management and nutrition landscape in SCD. To address these knowledge and care gaps, a number of recommendations have been suggested, as discussed in Chapter 5 (section 5.5.2), to work towards achieving the overarching aim of the research project,

- *‘to support the integration of nutrition into standard care in SCD, using a health literacy framework’.*

For change to be possible in transforming nutrition service provision in SCD, policies in nutrition in SCD need to acknowledge the marginalisation of SCD and how this has impacted on both the medical and nutritional management of SCD. For this reason, the development of the HL-NIF (blueprint) is specifically aimed at not only informing policy development, but is aimed at underpinning practice change towards developing nutrition service provision that would be tailored to the unique nutritional needs, challenges and risks of the SCD patient population, as identified in the findings of the research project. Thus the invisibility of nutrition in SCD and resulting health inequalities needs to be reflected in both policy and practice change, adopting a population based, whole systems strategy. Hence, the recommendations below (as discussed in chapter 5.5.2) , are linked to the four themes of the research project (see Figure 4.1) reflecting the multi-level influencing factors affecting nutrition integration in SCD, that need to be addressed at policy and practice level in order for nutrition to be successfully integrated into standard care in SCD management.

Theme 1 - Invisibility of SCD:

Policy Change:

1. *To acknowledge the lack of nutrition service provision in SCD - affecting patient experience, access and outcomes.*

Theme 2 - Under-recognised importance of nutrition:

Policy change:

2. *To improve nutrition service provision available to patients living with SCD –to reduce the risk of the late diagnosis of nutritional problems in people living with SCD.*

Practice change:

3. *To improve the access of patients living with SCD to nutrition service provision - allowing them more opportunity to speak about their nutritional needs and concerns.*
4. *To recognise the nutritional needs, risks and challenges of patients living with SCD, by SCD consultants and the wider SCD health and social care team, in order to improve patient outcomes.*

Theme 3 - Lack of priority to nutrition:

Policy change:

5. *To develop tailored nutrition policy and practice guidance to support the development and implementation of nutrition service provision in SCD, to provide pro-active rather than reactive nutrition service provision.*

Practice change:

6. *To increase the involvement of Dietitians in the nutritional management of patients living with SCD as part of the SCD MDT, in community and primary care settings, to facilitate the nutritional assessment, diagnosis and treatment of SCD patients.*

Theme 4 - Multi-level factors affecting nutrition and service provision

Practice change:

7. *To develop tailored rather than generic nutrition education, resources, policies and training curricular based on the specific needs, risks and challenges of the SCD patient population, to reduce the need for SCD patients to self-research nutritional information thereby reducing their risk of miss-information.*

6.4 Future Directions

Currently, limited knowledge and guidelines exists on how to integrate nutrition into SCD. Thus, the findings and the research project output, the HL-NIF (Blueprint), offers a helpful starting point for future research in the area of nutrition integration in SCD. To this end, the following research project ideas have been elicited:

- To conduct a multi-location study (possibly using the HCCs)

The multi-location study will be aimed at assessing the wider SCSU-C, SCSP and stakeholder knowledge, views and experiences about the nutritional management of SCD. Thus potentially, the opportunity is provided to identify any differences between the various locations and SCD patient populations in regards to nutrition knowledge, awareness, resources and service provision, infrastructure and systems. Effectively, this may also be able to identify any differences between urban and rural locations in nutritional management and service provision and what might be contributing to these differences.

- To evaluate the HL-NIF (Blueprint)

This can be part of an independent implementation study or either as part of the multi-location HCC study, to learn more about the efficacy and/or weaknesses of the framework. It would be interesting to assess how well the framework can be adapted to developing nutrition service provision in a range of countries, other than the UK and establish the potential transferability of the health literacy PEECE model, in improving health outcomes in other at risk or marginalised patient groups.

Summary:

The neglect of nutrition service provision available to people living with SCD can no longer be ignored. No longer can people living with SCD be deprived of access to nutrition service provision as part of their standard care. It is time to tailor nutrition service provision to the unique needs of the SCD patient population. Nutrition in SCD is a priority and as such, its current invisibility and neglect as a health inequality, need to be addressed. Thus, immediate attention should be given to include nutrition in SCD at national and professional policy and practice level to address the myriad of influencing factors reflected in the knowledge and care gaps defining the current nutrition landscape in SCD.

If nutrition service provision is to be integrated and embedded into standard care in SCD, there will need to be commitment from all the relevant stakeholders involved in the medical and nutritional health and service planning of people living with SCD. The connection between the marginalisation of SCD and the medical and nutritional management in SCD must be acknowledged in relation to patient experience, access and outcomes. To this end, the co-development of the HL-NIF (Blueprint) as a multi-stakeholder, population level, whole systems national and professional policy and practice management strategy, is required to integrate nutrition into standard care in SCD, thus providing a more patient centered and personalised care provision in nutrition in SCD.

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(h) Appendices:



Appendix 1: Participant Information Sheet (PIS)

PARTICIPANT INFORMATION SHEET

Improving Health Outcomes in Sickle Cell Disease: A small -scale qualitative study to develop a Learning Alliance Nutrition Health Improvement Plan (LANHIP)

Purpose and value of the study:

The purpose of the study is to establish and understand the nutrition knowledge and awareness, nutrition service provision and access and nutrition policy needs in Sickle cell and how this information can be used to improve nutrition service provision in Sickle Cell Disease. There is a lack of research on this topic in Sickle Cell Disease. However, the research will provide valuable information as well as allow for sickle cell patients and a wide range of healthcare providers and those with a special interest in sickle cell, to work together to find solutions to address the nutritional needs of sickle cell patients. The data collected in the study can be used to improve the nutrition and health outcomes in Sickle Cell Disease.

Invitation to participate:

You are invited to participate in this study if you are a parent of a child or adolescent with Sickle Cell Disease, an adult with Sickle Cell Disease, you provide health and social care services to people living with Sickle Cell Disease and if you are interested to know more about Sickle Cell Disease. You will **not** be eligible to participate if you don't have Sickle Cell Disease (and you're not interested in learning more about Sickle Cell Disease), if you're not a parent of a child or adolescent with Sickle Cell Disease or if you do not provide services or care for people with Sickle Cell Disease.

Before you decide whether or not you wish to participate please read the following information and if you have any questions, please contact the researcher (contact details are provided at the end of the document). Your participation is entirely voluntary; you are free not to take part if you so wish. If you do decide to take part, you are free to withdraw from the study at any time during the research process, though it will not be possible to remove your input from focus groups, as the focus group will form part of a group discussion. Thank you for reading this information sheet.

Who is organising the research?

The research is being conducted by Claudine Matthews who is a Professional Doctoral student from Anglia Ruskin University. She has previously worked in a sickle cell service and is a Dietitian by background.

What will happen to the results of the study?

A summary of the findings of the study will be presented to participants as part of the research project, and if you consent to providing the researcher with an email address or home/work address, it can be sent to you at the end of the study. The results will also be written up as a paper with the aim of publication and will be used by the researcher as part of her Doctoral Thesis. The findings will inform future research projects and bids to develop nutritional care service provision for Sickle Cell Disease in other London Boroughs and other areas with a high Sickle Cell Disease prevalence in the UK.

In relation to this specific research project, we need to make you aware of the following

☐	We do not need your personal data at any stage of this research project		
We are responsible for the personal data you give to us as a:			
☒	Data Controller (We are in sole control over the research)	Who are we?	Claudine Matthews
☐	Joint Controller (Where ARU and another organisation are working together on research)	with:	N/A
☐	Data Processor (Where the data will belong to another organisation and ARU is being engaged under contract/ agreement to conduct the research and provide an outcome but has no rights over the personal data)	on behalf of:	N/A

I will be asking you for the following information:

Personal Data				Sensitive Personal data	
☒	Name (For consent purposes only)	☐	Image (Photo or video)	☐	Racial/ Ethnicity data
☐	Age	☒	Experiences	☐	Political/ Religious beliefs
☐	Address/ location data	☒	Opinions	☐	Trade Union membership
☐	Employment & Earnings	☒	Knowledge	☐	Genetic/ Biometric data
☐	ID Numbers (e.g. NHS)	☐	[Other]	☐	Health
☐	Online identifier	☐	[Other]	☐	Sex life/ orientation data

We do not intend to collect data held about you from existing records. All data collected will comply with the GDPR and Data Protection Act (2018).

What will happen to your data?

All data collected will be anonymised which means we will remove any personal identifiers that may lead to you being identified from the information collected or that may be combined with any other information. Instead of using the actual names of the participants, codes will be assigned to participants and the data collected from each participant.. The data will, however, still be able to be linked together by the researcher should this be required. The personal data collected such as your experiences, opinions and knowledge, will only be to meet the research objectives and for no other purpose.

The research data will remain in the European Economic Area (EEA). All the data collected, will be securely held in password protected computers. The data collected will only be shared with my supervisor and will remain in an anonymised format. Every attempt will be made to ensure the data remains anonymous and this includes the data used in my research thesis, subsequent publications and quotes used to support the data collected. There may however, be a slight chance that participants may be identified by their peers or colleagues.

As part of the research project, data will be collected using focus groups, telephone interviews and network meetings, which will be digitally recorded. Each focus group, the telephone interviews and network meetings will be allocated a number and documents and files relating to each data collection activity will be saved under that number. All data collected from participants will remain anonymised, and codes will be used instead of names. The digital voice-recordings of each data collection activity, will be uploaded to a password-protected computer. Once the audio files have been uploaded to the computer, it will be deleted from the digital voice recorders.

The data collected as part of the focus groups, telephone interviews and network meetings will be transcribed by a professional transcription service provider, registered with the university, with a non-disclosure agreement in place to comply with the GDPR and Data protection Act (2018) legislation. During transcription, all potentially identifying information will be anonymised. Transcripts will be saved with the same number as the audio file. Only the researcher will have access to your personal data collected, that can be associated with your identity. Any hardcopy documents (e.g. consent forms) will be scanned to a password-protected computer.

Will I be reimbursed travel expenses?

Due to limited funds, your travel expenses will not be reimbursed.

Source of funding for the research:

N/A

What will happen if you agree to take part? (Identify, approach and obtain consent)

You have been informed by gatekeepers about the study and have been identified by the gatekeepers to be suitable to take part in the study. You have subsequently decided to contact me because you want to take part in the study. After contacting me, I have emailed you a copy of this Participant Information Sheet to read through, before signing the consent form. You can only take part in the study, if you have signed the consent form. Once I have received confirmation of your consent, you will be included in the study. You are however, free to withdraw from the study at any point, without giving any reasons.

If you agree to take part in the study, you will be allocated to a relevant study group as follows: Group 1 - (service users – adults with, and parents of children and adolescents with sickle cell), Group 2 – (service providers), Group 3 – (non-service providers). An expectation of the study is that all participants in the study groups will take part in a focus group. In addition, at the end of each focus group, participants can choose to take part in a telephone interview which will happen on a separate date, and join other focus group participants to form a nutrition network, which will represent the participants of the three different study groups. You can contact me at any time if you have further questions

As part of your participation in a focus group, you will be asked to share your experiences, opinions and knowledge about the nutritional needs of sickle cell patients and how its management can be improved. The focus groups will last approximately 90 minutes (but this will vary depending on how much you would like to talk about). The telephone interviews, will happen on a separate date and time agreed by you, with a smaller number of participants; the aim is to address some of the issues raised at the focus groups. It will last approximately 30-40 minutes.

If you agree to form part of the nutrition network, there are two meetings to attend. The first nutrition network meeting will be scheduled, to share and discuss the initial findings of the study; the second meeting -to share the final findings of the study which will be used to improve the nutrition and health outcomes of sickle cell patients. As part of data collection, the researcher will take notes and make observations throughout the study. The focus groups and nutrition network meetings will be held at a centrally located, local community-based hall, at a convenient time for the participants and refreshments will be available. The focus groups, telephone interviews and network meetings will be digitally voice recorded and transcribed for data analysis.

Are there any risks involved and if so, what will be done to ensure your wellbeing/safety?

The focus groups and telephone interviews will not discuss your personal health but will focus on general questions around your nutrition knowledge, awareness, needs and related concerns of Sickle Cell Disease. However, there is a slight chance that the interview could raise difficult and upsetting issues around your experiences of nutritional problems due to your sickle cell. You do not have to answer any questions that you do not wish to and you may take a break from or leave the focus group/network meeting or telephone interview at any point, without giving a reason for doing so. Any data collected during focus groups cannot be withdrawn after it has taken place as data is amalgamated and it will not be possible to remove data from individuals. If any significant distress is experienced or you would like more information or support you have the choice of contacting your healthcare provider, your GP, or local support services such as one of the following:

Organisation	Contact details	Website
Hackney Citizens Advice Bureau	300 Mare Street, Hackney, London, E8 1HE	http://www.hackneylocaloffer.co.uk/kb5/hackney/localoffer/service.page?id=E
Hackney Sickle Cell and Thalassemia Centre	457, Queensbridge Road, Hackney, London, E8 3AS	http://www.hackneylocaloffer.co.uk/kb5/hackney/localoffer/service.page?id=Hcq2wQ
Social Action For Health	Ment House, Ground Floor, Unit, 1G Mentmore Terrace,	https://www.safh.org.uk/

	London E8 3DQ 0208 5101970	
Mind in the City – Hackney and Waltham Forest	8-10 Tudor Rd, London E9 7SN 0208 985 4239	https://www.mindinhackney.org.uk/
Hackney Social Services	Hackney Service Centre 1 Hillman Street E8 1DY Tel 020 8356 6262	https://hackney.gov.uk/adults-older-people
Hackney Community Foodbank	Florence Bennett Centre, Cherbury St, Hoxton, London N1 6TL 0207 254 2464	https://www.google.com/search?biw=1301&bih=620&ei=lrw9W-m5F4b4gQac6rqqDA&q=hackney+food+bank&oq=hackney+food&gs_l=psvab.1.0.0l10.3859.9011.0.11504.26.20.0.0.0.0.449.1963.8j5j1j0j1.15.0...0...ab..14.12.1145...0i67k1j0i131k1.0.JiD0A9Zgl1w

Are there any benefits from taking part?

There are no direct benefits by taking part but this research will give you the opportunity to talk about your experiences and views on your nutritional knowledge, nutrition service needs and related concerns in Sickle Cell Disease. The research project will give you the opportunity to interact with local service providers and health professionals and discuss the changes you might want to see introduced. The findings will inform future research and help develop evidence that can be used to develop future nutritional services for sickle cell, based on what participants identify as the main issues.

Are there any instances where confidentiality would be broken?

The only time that confidentiality would be broken, is if anything is disclosed which indicates a risk of harm to yourself or others. Should this be necessary it will be discussed with you at the time of interview. It is also important to note that as focus groups are group discussions, there is a chance that confidentiality and anonymity could be broken by others taking part in the same focus group. For

this reason, there is an expectation that each participant maintains and keeps discussions in the focus groups confidential, and this will be emphasised at the start of each focus group.

Contacts for further information:

Claudine Matthews (Researcher)

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Project mobile number: 07871264182

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Project mobile number

This study will be reviewed and approved by the School Research Ethics Panel (Education and Social Care) at Anglia Ruskin University.

If you have any concerns or complaints about any aspect of the study please contact the researchers in the first instance using the contact details above. However, if after speaking with the researchers you wish to complain formally you can do this through contacting the following website complaints@anglia.ac.uk : Postal address: Office of the Secretary and Clerk, Anglia Ruskin University, Bishop Hall Lane, Chelmsford, Essex, CM1 1SQ. Normally your complaint will be acknowledged within five working days and answered as soon as possible thereafter.

Please keep a copy of this information so you can refer to it again if you wish.

Appendix 2: Original ethics approval letter



Chelmsford & Cambridge

16th September 2019

Claudine Matthews

Dear Claudine

Principal Investigator	Claudine Matthews
SREP Number	ESC-SREP-18-334
Project Title	Improving health outcomes in Sickle Cell Disease: a small-scale qualitative study to develop a Learning Alliance Nutrition Health Improvement Plan (LAMHIP)

I am pleased to inform you that your ethics application has been approved by the School Research Ethics Panel (SREP) under the terms of Anglia Ruskin University's Research Ethics Policy (dated 24 July 2019, Version 1.11). This application has also been approved by FREP.

Ethical approval is given for 3 years from 16th September 2019. If your research will extend beyond this period, it is your responsibility to apply for an extension before your approval expires.

It is your responsibility to ensure that you comply with Anglia Ruskin University's Research Ethics Policy and the Code of Practice for Applying for Ethical Approval at Anglia Ruskin University available at www.aru.ac.uk/researchethics including the following:

- The procedure for submitting substantial amendments to the committee, should there be any changes to your research. You cannot implement these amendments until you have received approval from the SREP for them.
- The procedure for reporting accidents, adverse events and incidents.

- The General Data Protection Requirement (GDPR) if your research will take place in the European Economic Area (EEA)¹ or involve sending or bringing any personal data² into it. If your research will take place in the UK or involve sending or bringing any personal data into it, you must also comply with the Data Protection Act (2018). Other countries in the EEA may have further data protection legislation you must comply with. If your research will take place outside the EEA, you must comply with any data protection legislation relating to that country or countries.
- Any other legislation relevant to your research. You must also ensure that you are aware of any emerging legislation relating to your research and make any changes to your study (which you will need to obtain ethical approval for) to comply with this.
- Obtaining any further ethical approval required from the organisation or country (if not carrying out research in the UK) where you will be carrying the research out. This includes other Higher Education Institutions if you intend to carry out any research involving their students, staff or premises. Please ensure that you send the SREP copies of this documentation if required, prior to starting your research.
- Any laws of the country where you are carrying the research and obtaining any other approvals or permissions that are required.
- Any professional codes of conduct relating to research or requirements from your funding body (please note that for externally funded research, where the funding has been obtained via Anglia Ruskin University, a Project Risk Assessment must have been carried out prior to starting the research).
- Completing a Risk Assessment (Health and Safety) if required and updating this annually or if any aspects of your study change which affect this.
- Notifying the SREP Secretary when your study has ended.

Please also note that your research may be subject to monitoring.

Should you have any queries, please do not hesitate to contact me. May I wish you the best of luck with your research.

Yours sincerely,

Professor Jeffrey Grierson (Chair)
School of Education & Social Care
For FHMS Research Ethics Panel

Copy to: David Smith

¹ The EEA includes EU member states and also Iceland, Liechtenstein and Norway.

² Personal data means any information relating to an identified or identifiable natural person ('data subject'); an identifiable natural person is one who can be identified, directly or indirectly, in particular by reference to an identifier such as a name, an identification number, location data, an online identifier or to one or more factors specific to the physical, physiological, genetic, mental, economic, cultural or social identity of that natural person.

Appendix 3: First ethics amendment approval



Chelmsford & Cambridge

7th January 2020

Claudine Matthews

Dear Claudine

AMENDMENT APPROVED (7.1.20)

Principal Investigator	Claudine Matthews
SREP Number	ESC-SREP-18-334
Project Title	Improving health outcomes in Sickle Cell Disease: a small scale qualitative study to develop a Learning Alliance Nutrition Health Improvement Plan (LAMHIP)

I am pleased to inform you that your **AMENDMENT** request has been approved by the School Research Ethics Panel (SREP) under the terms of Anglia Ruskin University's Research Ethics Policy (dated 24 July 2019, Version 1.11). This application has also been approved by FREP.

Ethical approval is given for 3 years from 16th September 2019. If your research will extend beyond this period, it is your responsibility to apply for an extension before your approval expires.

It is your responsibility to ensure that you comply with Anglia Ruskin University's Research Ethics Policy and the Code of Practice for Applying for Ethical Approval at Anglia Ruskin University available at www.aru.ac.uk/researchethics including the following:

- The General Data Protection Requirement (GDPR) if your research will take place in the European Economic Area (EEA)¹ or involve sending or bringing any personal data² into it. If your research will take place in the UK or involve sending or bringing any personal data into it, you must also comply with the Data Protection Act (2018). Other countries in the EEA may have further data protection legislation you must comply with. If your research will take place outside the EEA, you must comply with any data protection legislation relating to that country or countries.
- Any other legislation relevant to your research. You must also ensure that you are aware of any emerging legislation relating to your research and make any changes to your study (which you will need to obtain ethical approval for) to comply with this.
- Obtaining any further ethical approval required from the organisation or country (if not carrying out research in the UK) where you will be carrying the research out. This includes other Higher Education Institutions if you intend to carry out any research involving their students, staff or premises. Please ensure that you send the SREP copies of this documentation if required, prior to starting your research.
- Any laws of the country where you are carrying the research and obtaining any other approvals or permissions that are required.
- Any professional codes of conduct relating to research or requirements from your funding body (please note that for externally funded research, where the funding has been obtained via Anglia Ruskin University, a Project Risk Assessment must have been carried out prior to starting the research).
- Completing a Risk Assessment (Health and Safety) if required and updating this annually or if any aspects of your study change which affect this.
- Notifying the SREP Secretary when your study has ended.

Please also note that your research may be subject to monitoring.

Should you have any queries, please do not hesitate to contact me. May I wish you the best of luck with your research.

Yours sincerely,

Professor Jeffrey Grierson (Chair)
School of Education & Social Care
For FHEMS Research Ethics Panel

Copy to: David Smith

Appendix 4: Second ethics amendment approval



Chelmsford & Cambridge

6th March 2020

Claudine Matthews

Dear Claudine

AMENDMENT APPROVED 6.3.20)

Principal Investigator	Claudine Matthews
SREP Number	ESC-SREP-18-334
Project Title	Improving health outcomes in Sickle Cell Disease: a small scale qualitative study to develop a Learning Alliance Nutrition Health Improvement Plan (LAMHIP)

I am pleased to inform you that your **AMENDMENT** request has been approved by the School Research Ethics Panel (SREP) under the terms of Anglia Ruskin University's Research Ethics Policy (dated 24 July 2019, Version 1.11). This application has also been approved by FREP.

Ethical approval is given for 3 years from 16th September 2019. If your research will extend beyond this period, it is your responsibility to apply for an extension before your approval expires.

It is your responsibility to ensure that you comply with Anglia Ruskin University's Research Ethics Policy and the Code of Practice for Applying for Ethical Approval at Anglia Ruskin University available at www.aru.ac.uk/researchethics including the following:

- The General Data Protection Requirement (GDPR) if your research will take place in the European Economic Area (EEA)¹ or involve sending or bringing any personal data² into it. If your research will take place in the UK or involve sending or bringing any personal data into it, you must also comply with the Data Protection Act (2018). Other countries in the EEA may have further data protection legislation you must comply with. If your research will take place outside the EEA, you must comply with any data protection legislation relating to that country or countries.
- Any other legislation relevant to your research. You must also ensure that you are aware of any emerging legislation relating to your research and make any changes to your study (which you will need to obtain ethical approval for) to comply with this.
- Obtaining any further ethical approval required from the organisation or country (if not carrying out research in the UK) where you will be carrying the research out. This includes other Higher Education Institutions if you intend to carry out any research involving their students, staff or premises. Please ensure that you send the SREP copies of this documentation if required, prior to starting your research.
- Any laws of the country where you are carrying the research and obtaining any other approvals or permissions that are required.
- Any professional codes of conduct relating to research or requirements from your funding body (please note that for externally funded research, where the funding has been obtained via Anglia Ruskin University, a Project Risk Assessment must have been carried out prior to starting the research).
- Completing a Risk Assessment (Health and Safety) if required and updating this annually or if any aspects of your study change which affect this.
- Notifying the SREP Secretary when your study has ended.

Please also note that your research may be subject to monitoring.

Should you have any queries, please do not hesitate to contact me. May I wish you the best of luck with your research.

Yours sincerely,

Professor Jeffrey Grierson (Chair)
School of Education & Social Care
For FHMS Research Ethics Panel

Copy to: David Smith

Appendix 5: Third ethics amendment approval via Chairs action

~~Claudine.Matthews@myaru.onmicrosoft.com~~
Sent: 17 March 2020 13:11
To: King, Peggy <Peggy.King@anglia.ac.uk>
Subject: Ethics Amendments via Chairs action in light of COVID 19

Dear Peggy,

Following on from recent events and after discussion with my supervisors, please find a request for amendments to my ethics form via chairs action.

In light of COVID 19 regulations, I have to make changes to my data collection activities which is mainly face to face focus groups and network meetings. My time frames are very tight and any time delays will have a huge impact on me being able to complete my research project in the stipulated time.

I have attached the PIS and PC forms reflecting the amendments - to use video calling when face to face contact is not permissible.

Please let me know if you require anymore information.

Best wishes,
Claudine

<CM Final Ethics PIS Form (with COVID 19 amendments) 17 3 20.docx>
<CM Ethics - PC Form Final (with COVID19 amendments) 17 3 20.docx>

-- Please click here to view our e-mail disclaimer <http://www.aru.ac.uk/email-disclaimer>

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From: King, Peggy <Peggy.King@anglia.ac.uk> on behalf of SREP-FHEMS-ESC <SREP-FHEMS-ESC@anglia.ac.uk>
Sent: Thursday, March 19, 2020 1:20 PM
To: Matthews, Claudine (Postgraduate Researcher) <Claudine.Matthews@myaru.onmicrosoft.com>
Subject: FW: Ethics Amendments via Chairs action in light of COVID 19

Hi Claudine,

Please see message below from the Chair.

Yes this is fine. Perhaps just mention that she needs to make sure she only makes audio recordings and not video ones.

Wishing you good luck with this research. Keep safe

Best wishes,



Peggy King
School Administrator
Faculty of Health, Education, Medicine & Social Care

Appendix 6: Research project advert

Pioneering Public Health study: Nutrition in Sickle Cell - improving health outcomes - transforming lives!

Do you want to be a history maker?

Do you want to make a difference in Sickle Cell?

Then I want to offer you the opportunity to do so and personally invite you to be a participant in my pioneering Doctoral research project- Developing a Nutrition Health Improvement Plan to improve health outcomes in Sickle Cell Disease.

Are you a service user (parent of a child and adolescent with SCD, or an adult with SCD), or a service provider in sickle cell, then I need your help!

Eligible participants will be recruited to 3 possible study groups: Group1: service users, Group2: service providers, Group 3: non service providers.

What is required as a participant is the following: to partake in a,

- focus group,
- telephone interview
- opt in to form part of a multi- stakeholder network in nutrition in SCD

The focus groups will take place in a community centre at a convenient time and refreshments will be available.

Your participation is completely voluntary but will help to inform the changes we can make to improving nutrition service provision in sickle cell! The final research thesis I hope to present to the House of Commons via the All-Party Parliamentary Group and for it to be part of the Quality Assurance Documentation in SCD- eg UK Forum on Haemoglobin Disorders Quality Standards (2021) report and quality peer review process.

Thank you for your help!

I look forward to hearing from you:

My email: cmnutri@icloud.com

Mobile: 07871 264182

Many thanks,

Claudine Matthews (Researcher)

Appendix 7: Consent Form



Participant Consent Form

Name of participant:

Project title: Improving Health Outcomes in Sickle Cell Disease: a small-scale qualitative study to develop a Learning Alliance Health Improvement Plan (LANHIP)

Researcher: Claudine Matthews, Professional Doctoral Student, Anglia Ruskin University, Chelmsford, Essex, CM11 8Q. Email: claudine.matthews@pdr.anglia.ac.uk Mobile: 07871264182

Please tick the appropriate research option and initial in the box beside each statement below if you agree to participate in a Focus Group ☐ Telephone Interview ☐ Nutrition network meeting ☐ or all three ☐

- | | |
|--|--------------------------|
| 1. I agree to take part in the above research. I have read the Participant Information Sheet and I understand what my role will be in this research. | ☐ |
| 2. I understand that I am free to withdraw from the research at any time before and during the focus group/telephone interview/network meeting, without prejudice, and without needing to give a reason. | ☐ |
| 3. I have been informed that any information I provide will be confidential (except in instances outlined to me in the Participant Information Sheet) | ☐ |
| 4. I understand that I am free to ask any questions at any time before, during and after the study. | ☐ |
| 5. I agree to the focus group/telephone interview/network meeting being digitally voice-recorded for the purposes of transcription and data analysis. | ☐ |
| 6. I agree to the researcher using personal quotes, which have been anonymised as part of the analysis and dissemination of the research, the thesis and future publications. | ☐ |

Data Protection: I agree to Anglia Ruskin University processing personal data which I have supplied. I agree to the processing of such data for any purposes connected with the Research Project as outlined to me.

Participant name Signed.......... Date.....
(print)

Researcher

Name Signed.......... Date.....
(print)



Please keep this form in case you wish to withdraw at a later date. If you do wish to withdraw, you can complete the form and return it to the main investigator (Claudine Matthews).

Title of Project: Improving Health Outcomes in Sickle Cell Disease: a small-scale qualitative study to develop a Learning Alliance Health Improvement Plan (LANHIP)

I WISH TO WITHDRAW FROM THIS STUDY

Name:

Date:

Appendix 8: Phase one question schedule

Research service user question schedule:

1. What do you know about the main clinical problems of sickle cell and how it is managed?
2. What role do you think does nutrition play in the management of your sickle cell?
3. What is your understanding of optimum nutrition and describe what you think are the main nutrition needs of sickle cell patients across the **life span (infants, children, adolescents, adults, pregnancy)**?
4. What are some of the main nutrition challenges you or those you care for face and how do you manage it?
5. How do you think service providers can help to identify, assess and manage your nutrition needs more effectively?
6. In your opinion what are some of the personal (**your social, emotional, psychological, physical, nutrition knowledge**) factors that may affect your food choices/beliefs/habits/behaviours? – why do you think that? – what are some of the implications?
7. What are some of the interpersonal (**interactions with peers, family, service providers**) factors that may have an effect on your food choices/beliefs/habits/behaviours? – Why do you think that? – what are some of the implications?
8. What may be some of the organisational (**the places where you receive health, social care, education**) factors that may be affecting your nutrition needs – (**thinking about how nutrition is assessed, monitored and managed**)? Why do you think that? – what are some the implications?
9. What may be some of the community level factors (**areas where you live, work, have leisure activities, shop**) that may be affecting your nutrition needs – (**thinking about your access to healthy food options, nutrition educational resources, healthy eating advice, tailored nutrition activities, cooking lessons etc**)? Why do you think that? – what are some of the implications?

10. Taking one factor at a time – (**personal, interpersonal, organisational, and community level factors**) – what suggestions do you have to improve the impact these factors may have on your nutritional needs?
11. Before we close this focus group meeting, do you have anything else to add or clarify from the points we have already discussed?

Research stakeholder question schedule:

1. What do you know about sickle cell and how it is managed?
2. Where does nutrition fit into your patient management plan (Do you ask your patients about their nutrition needs/status?)
3. What is your understanding of optimum nutrition and how does this relate to what you think are the main nutrition problems for sickle cell patients?
4. What do you think are some of the main challenges facing sickle cell patients with regards to their nutrition needs and how is it currently managed?
5. How can you as service provider/stakeholders better support the nutrition needs of sickle cell patients with regards to how it is identified, assessed and managed?
6. In your opinion as a stakeholder, what do you think are some of the personal factors that affect, what/when (food choices/beliefs/habits/behaviours) of service users – why do you think that?
7. What do you think are some of the interpersonal factors (peers, family, service providers) that affect, what/when (food choices/beliefs/habits/behaviours) of service users – why do you think that?
8. What do you think are some of the institutional factors (where they receive the health and social care, education) that may affect service users nutrition needs, assessment, monitoring and management? – why do you think that?
9. What do you think are some of the community level factors (areas of work, play, recreation, shopping) that affect service users access to healthy food options, nutrition educational resources, healthy eating advice, cooking lessons etc) – why do you think that?

10. Taking one factor at a time – (personal, interpersonal, institutional and community level factors)-can you suggest any recommendations that would improve the impact of these factors on the nutritional needs of sickle cell service users?
11. Before we close this focus group meeting, do you have anything else to add or clarify from the points we have already discussed?

Appendix 9: Extract from the digital copy and summary of phase one data and developing themes

Extract of digital copy of phase one data summary

A	B
1 Question Two	Question two - Summary
2 Reasons for lack of nutritional management - 16/8/20	Nutrition not a priority - health, social care, education
3 Referrals not made to the Dietitian	Poor nutrition knowledge- patients/providers
4 Clinicians lack of knowledge about nutrition	Poor referral to the dietitian
5 Generic advice rather than tailored or specific	Poor quality hospital food
6 SCD management protocol driven - (pain, transfusions)	nutrition knowledge self researched- patients/providers
7 clinicians don't ask about nutrition	Nutrition- reactive rather than preventative
8 there's no input and nutritional educational resources	More emphasis on fluids (dehydration) rather than overall nutrition
9 Poor quality of hospital food	Confusion about what foods to eat and how food affects the body
10 social factors have a role to play	
11 Don't talk much about health and wellbeing - don't ask about nutrition	
12 Nutrition doesn't really get brought up	
13 Treated like any other nutrition support patient - (generic and not tailored)	
14 Need to educate patients on the importance of nutrition in SCD - with limited knowledge	
15 Lack of family and service provision support structures (schools, parents, ill health)	
16 children's eating habits - fussy eating, food likes and dislikes	
17 Nutrition only slowly being introduced into SCD management	
18 Patients and service providers have to do their own private research	
19 More emphasis on drinking fluids rather than overall nutrition	
20 patients never been asked directly about their nutrition	
21 Disease managed with nutrition alone	

Summary of phase one and two data collection and themes:

Questions (Q)/Themes	Summary of Main Points	Discussion points
Q1. Theme 1. knowledge and management of SCD	Good knowledge from both service users and providers - but had to self-research SCD invisible – symptoms invisible SCD not transmittable Variance of severity of SCD between patients	Providers – <i>(knowledge self-researched) + (need consultants who are pro-nutrition)</i> SCD invisible
Q2,3,4. Theme 2. Nutrition management Sub-themes: Access to nutrition services Digital support	<i>'clinicians lack knowledge about nutrition'; 'Clinicians don't ask about nutrition'; 'Referrals not made to the Dietitian'; 'Poor quality of hospital food'; 'More emphasis on drinking fluids than nutrition'; 'sicklers who are ill don't eat very much'; 'Nutrition not given the same priority as pain, social care'</i>	Nutrition management <i>reactive</i> rather than <i>preventative</i> <i>Fluid (Hydration)</i> recognised as a priority over nutrition as a whole Power imbalance between the medical management of

Risks - late diagnosis of nutrition problems	<i>needs'; 'I've recently been diagnosed with osteoporosis why hasn't anyone told me about the risk before'; 'I've never been asked about my nutrition'; 'Need for a digital healthcare system'; 'disparity in priority given to oncology patients over sickle cell patients, even though the patients are clumped together'; 'Needs an MDT approach';</i> Nutrition plays a fundamental role but not the focus of management – pain Tx a priority	sickle cell and nutrition management Pain Affects appetite and oral intake
Q3. Theme 3. Nutrition Knowledge and awareness Sub- themes: Optimum nutrition The value of nutrition Risk of self - research an	<i>'GP's knowledge of nutrition only on a generic level'; 'Clinicians to reinforce importance of nutrition – having consultant buy in (pro-nutrition); lack of understanding of food groups'; don't want to put more pressure on parents'; 'I don't fully get what optimum nutrition really means'; 'optimum nutrition for me is an ideal, when I'm ill food isn't really an interest to me';</i> Both groups see optimum nutrition as food and balancing nutrients – fluid seen as a priority – advice more generic than tailored	-Narrow view of nutrition Optimum nutrition – <i>mostly understood in relation to food and balancing food intake</i>
Q1-9. Theme 4. Nutrition Education and Training	<i>Everything I know about nutrition I've had to self – research';</i> Poor nutrition knowledge of patients and providers – <i>'need to understand link between nutrition and health – offer holistic care;</i> lack of scientific evidence base, understand the role of nutrition Nutrition overlooked. Delayed identification of nutrition problems – don't understand link between SCD and nutrition	Disempowerment of both groups -due to poor knowledge of nutrition and poor nutrition resources/service provision Poor quality of hospital food
Q6-9. Theme 5. Barriers to nutrition Sub- themes: Social support structures Nutrition support services Community based structures	<i>Nutrition is fundamental in sickle cell'; 'we have more referrals because our consultant is pro-nutrition'; 'Nutrition not a priority – pain a priority';</i> Misconception nutrition is a cure – <i>'If I've cured my sickle cell with nutrition'</i> Nutrition is paramount Poor referrals for nutrition – access to dietitians MDT Approach -include discussion re nutrition <i>Improve the knowledge and awareness of the wider MDT</i> Education and Training – patients do their own research, lack of nutrition educational resources)	<i>Nutrition is a cure</i> – concerns about patient eligibility of benefits Need consultants who are pro-nutrition <i>Health inequalities</i> – oncology a priority <i>COVID 19</i> – infection risk Policy change Use of social media – dynamic celebrities

	Lack of support structures (personal, social, community factors) Integrating nutrition into SCD management Prioritise nutrition as in Fluid/pain Include nutrition in patient Protocols Distance of food shops, availability of healthy food, no of chicken and chip shops Nutrition resources available -locally, of British Dietetic Association (BDA), Sickle Cell Society (SCS) Policy development - Role, Health Wellbeing Board, BDA, SCS, CCG, Consultants – national nutrition campaign in SCD, interested celebrities/chefs COVID 19 – (raise importance of nutrition in SCD)	
Q10. Theme 6. Recommendations to promote nutrition Sub – theme: Developing Nutrition service provision	Holistic Model (MDT) Health policy to mandate nutrition in SCD. MP involvement, Hospital Boards Multilevel approach Digital management algorithm – GP's BDA Sickle Cell Society Health and Wellbeing Board NHS England CCG National Campaign APPG	Health Education Health empowerment Health integration Clinical Governance NHS Principles,

Appendix 10: APPG – SCAT Nutrition in SCD supporting letter

All Party Parliamentary Group (APPG) - Sickle Cell and Thalassaemia (SC+T)

To the Parliamentary Officer: APPG-SC+T

Date: 28/11/22

Ms XXXXX

54 Station Road, London, NW10 4UA

Re: Nutrition in Sickle Cell Disease (SCD)– a growing health inequality

The publication of the report, 'No one is listening' (APPG SC+T, 2021) commissioned by the Sickle Cell Society, bears witness to the impact of racism and the invisibility of SCD in healthcare provision in the NHS. Since its discovery in 1910, much work has been done to develop the medical and clinical management of the multiple symptoms of this genetically inherited long term condition. However, it is only since the 1980's that under nutrition associated with the condition, has been recognised as a serious complication of the condition, but very little has been done to integrate or embed nutrition management into standard care in SCD. As a consequence, nutrition remains an unmet need for many vulnerable patients living with SCD, despite the direct causal link between the clinical features of SCD and the nutritional problems identified in the existing scientific literature. Thus, the lack of nutrition service provision in SCD, has become a health inequality in SCD.

It is a well-established fact that SCD has both medical and nutritional implications, but to date, very little policy and practice guidelines exist to reflect the role of nutrition in SCD. Equally, very little has been done to develop and support nutrition service provision in SCD, perpetuating the poor nutrition knowledge, awareness, lack of evidence based resources and poor nutrition service provision in the condition. In this way, countless individuals living with SCD, are at risk of the late diagnosis of nutritional problems, for which they often have to self –diagnose and self-manage their nutritional needs, which would normally be managed by expertly trained healthcare professionals such as Dietitians. A recent case study conducted in primary care provided compelling evidence of the impact of neglecting nutrition service provision in SCD on the health and wellbeing outcomes of patients living with SCD, and therefore, the time for change is now.

The current nutrition landscape in SCD, is therefore a source of significant health inequality in the nutritional management of patients living with SCD, affecting patient experience, access and health outcomes. The ongoing invisibility of nutrition as an 'adjunct' management option in SCD, can no longer be overlooked and ignored. Many people living with SCD are asking for more support and awareness about the role of nutrition as part of their standard disease management and care provision. However, many people living with SCD and healthcare providers, are not aware of the national nutrition standards, despite the inclusion of the national nutrition standards for SCD as part of the 2018, national clinical standard for the management of Adult sickle cell patients. If the status quo in the nutrition service in SCD is to change, more effort is needed to raise awareness of the role of nutrition as a management option in this chronic condition. Equally, more effort should be made to identify funds to support the development of future nutrition service provision.

As such, there has to be a change in policy, for there to be a change in practice to improve the experience, access and health outcomes of vulnerable people living with SCD, who are reliant on healthcare professionals for their healthcare needs. In recognition of the growing interest and urgency in considering the role of nutrition as an adjunct management option in SCD, I have embarked on a self-funded Professional Doctorate, to identify and understand the myriad of factors influencing the nutritional management in SCD, to eventually use this information to address the poor knowledge and care gaps facing both patients living with SCD and service providers to ensure a high quality nutritional care provision in SCD. The overarching aim of the research project is to support the integration of nutrition into standard care in SCD, through policy and practice development that is tailored to the unique nutritional needs of patients living with SCD.

Preliminary findings of my doctoral research project, affirms the need to change the current nutrition landscape in SCD, using a population level and whole systems approach to integrate or embed nutrition into standard care in SCD. As such, there is need for change on a micro, meso and macro level to address the multiple knowledge and care gaps and influencing factors that affect the integration of nutrition into standard care in SCD. For this reason, the main output of the research project is the co-development of a Nutrition Integration Framework, to inform future policy and practice development to support the integration of nutrition into standard care in SCD, thus aiming to improve the experience, access and health outcomes for patients living with SCD.

In this way, the APPG – SC+T, has a responsibility to place nutrition on the agenda for open discussion in order to address the health inequality of the lack of nutrition service provision in SCD. Following consultation with a number of patients living with SCD and service providers, please find below a range of signatures, in support of the integration of nutrition into standard care in SCD. No more can we overlook or ignore the calls from patients living with SCD, for nutrition to be part of their standard care provision. No more can we stand back and observe the effects of poor nutrition service provision on the experience, access and health outcomes of patients living with SCD. We all have an obligation to take responsibility to do all we can to integrate nutrition into standard care in SCD. Hence, placing nutrition on the APPG – SCD+T agenda, is the first step in changing the nutrition landscape in SCD to address the growing health inequality in the nutritional management of patients living with SCD.

Please accept this letter as a formal request for nutrition in SCD to be placed on the APPG-SC+T agenda. I look forward to receiving a response to this letter.

Yours sincerely,

Claudine Matthews, RD, MSc, PGCE, FHEA

Primary Care Network Dietitian

Professional Doctoral Candidate (Anglia Ruskin University)

Email: cmnutri@icloud.com

Appendix 11: Initial version of the health literacy framework

Health literacy framework – co-developed by the LAN

Guidance for completion:

Please consider the following questions:

1. What needs to be included in the framework?
2. What would you expect to see in this type of document?
3. What else needs to add?

Part 1: What is SCD?

What is SCD? SU-C (7): • Disease of the red blood cells • As a result, it has a number of knock on effects as a result of sickle cells being unable to carry oxygen SU-C (9) • Make definition as simple and as definitive as possible for those who have never heard or know much about sickle cell • Sickle cell is not transmittable • Use simple language that lay people could understand- plain English and non-medical jargon • SCD is a hidden disease • Symptoms not always obvious SP (2): - Inherited from our parents - You can be a carrier but not have sickle cell disease - sickle cell red blood cell shape - red blood cells disease - Not transmittable - Hidden disease - Symptoms are not always obvious - Not also visible who has sickle cell disease	Management: SU-C (7): • Regular check-ups, hemaoc, cardio, renal, ophthalmology 'annual MOT' • Hydration • Staying warm when it's cold • Exercise – not often spoken about • Diet SU-C (9): • Management and treatment options- differentiating between medicalised treatment options and management options because management options could be alternative and non-medicalised SP (2): - Medication - Transfusions - Regular check-ups – medical team knowing what questions to ask Treatment Options: SU-C (7): Depends on severity of condition starting from (minimal intervention): 1. Painkillers when you have a crisis (reactive) 2. Regular supplements i.e. folic acid (proactive) 3. Precautionary antibiotics (penicillin) (proactive) Regular blood transfusions or hydroxyurea (invasive proactive) SP (2): • Hydroxyurea
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	• Nutritional supplements if weight gain is an issues • medication • regular appointment • psych input if needed • MDT input • X-Jade • Physio access • Acupuncture • Self-care
Main clinical Features: SU-C (7): • Sickle Cell Crisis • Anaemia - tiredness and fatigue • Affects the immune system SU-C (9): • Excruciating pain in the body as a result of a crisis • Lack of oxygen in blood vessels • Breathing difficulties • Fatigue /Anemia • Mobility restrictions SP (2): - anaemia - crisis - Pain - Nausea/Vomiting from medication - Breathing issues - Fatigue - Effects it has on the immune system Complications: SU-C (9): • Stroke / neuro conditions • Organ damage ie Cardiovascular, renal, liver • Acute chest syndrome • Retinopathy • MH	• ??should management and treatment be the same thing
Reflections: SU-C (7): There should be more focus on long term health, wellbeing and maintenance, instead of preparing/condemning people to a range of complications o co-morbidities which may not actually happen if long term health is prioritised over reactive approaches SP (2): There should be more focus on long-term health, wellbeing and preparing people for co-morbidities that may not happen if long-term health is focussed on. Wondered whether management and treatment should be separated or jointed? SC-U (2): Keep this section concise to make room for expansion on other sections	

Part 2: What is Optimum Nutrition?

What is optimum nutrition? SU-C (7): • I guess patients getting a wide range of nutrients - of protein, fluid, a range of vits and minerals • getting enough of the micronutrients - drinking enough water to reduce dehydration SP (2): - no deficiencies - Meets calorie and protein requirements - Hydrated - Varied diet - Getting enough micro/macro nutrients -	Main challenges in nutrition management SU-C (7): • Nutrition is overlooked, neglected and undertreated - not integrated into management. • Nutrition not given the same priority as pain, dehydration, social care needs • Impact of medication - side effects of medication impacts on appetite suppression • Socio-economic, cultural issues around diet - salt. SP (2): - finance - health / ability to prepare /eat food - knowledge of what is nutritious foods - Time - Fatigue - Nausea/Vomiting - Ability to feed themselves if they are in a lot of pain - Socio-economic factors Cultural cooking – vegetarian, vegan
Main Nutrition needs SU-C (7): • Vitamin D deficiency and then anaemia • micronutrients so bone health - Calcium, Vit C and D • General supplements i.e. Vit A and omega 3 I've taken all my life • Alcohol within government guidelines SP (2): - Age groups – should this be divided into your age group as your needs vary based on your age. - Where in the treatment cycle are you? - Iron deficiencies – or iron overload - Vitamin D - Calcium for both health - Alcohol	
Reflections: SU-C (7): Any nutrition is better than an overall focus on the right things. There are a lot of myths, unconfirmed theories and fad diets which can affect people's nutritional quality. SP (2): Without nutritional knowledge the concept of what is optimum nutrition is different for everyone.	

Part 3: Socio –economic factors influencing the nutrition needs of sickle cell patients

Personal Factors:	Institutional Factors
--------------------------	------------------------------

SU-C (9): - Consider child poverty and access to nutritional foods in the context of today's campaign with Marcus Rashford SP (2): - Mood e.g depression - Physical challenges - Religion - Finance - Lack of support / awareness - Motivation during crisis or when they are feeling unwell - Image - Physical health / Fitness / - Money and poverty - Financial /affordability	SP (2): - Hospital - Work and the food availability - School meals - Lack of support / Awareness - Food around work
Interpersonal Factors: SP (2): - Family support - Stigma - How to patients see Healthcare professionals – knowing what to ask - How the supporting team can relate and support the patient/person / awareness	Community/Policy: SU-C (9): - Diversity of cities and towns and access to cultural foods SP (2): - Support / Family support - Access to food - Lack of support within their community or environment

Part 4: Facilitators – to address the influencing socio-economic factors

Personal Factors: SP (2): Providing information and knowledge on where to find food/affordability	Institutional Factors: SU-C (9): - consider nutrition in the context of Transition for peads patients into adult service SP (2): - Making sure the staff/knowledge - More options available to help or support patients - Staff knowledge – knowing what to ask - School teaching – knowing what to teach/address - Conferences - Free school meals - Presentation at schools
Interpersonal Factors: SU-C (9): Role of partners in care – consider education in nutrition	Community Factors: SU-C (9): Picking up on SC-U (7) point- the NHS is good at dealing with fixing people/ reactive care and not so

• People who live alone/ without a partner – dealing with a different set of challenges • Elderly sickle cell patients SP (2): - Free school meals - Home visits	much looking after people with chronic long term conditions SP (2): - Coupons e.g. previously with Coeliac disease you would get vouchers for products. Or you have the food banks that can get you certain staple foods. There should be something available to support those that either can afford or don't have the abilities to go to the shops - Deliveries or a supportive community that can help those that are unwell and provide them with the food. Also if you can have medication set to be delivered then this should be available for when you are unwell. SU-C (9): (Lobbying) • Ongoing lobbying with government – link to APPG agenda and parliament support to help put not just sickle cell on the agenda but nutrition in sickle cell as high priority. Sickle cell still an area that experiences health inequalities in society- especially in the context of COVID and the BAME population. • Conferences such as ASCAT, ASH, BSH etc are helpful to use to as a means to raise awareness of nutrition in sickle
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Part 5: Helpful Resources:

SP (2):
BDA
Sickle cell website
NHS website
Presentations at schools

Appendix 12: Phase four evaluation questionnaire

Phase Four: Nutrition in Sickle Cell Research Project

Evaluation Questionnaire:

Thank you for participating in Phase Four of the research project. This part of the project is to evaluate the Nutrition Health Education Framework by answering the following questions. **Completed questionnaires should be returned via email by Friday 14th November 2020.**

Q1: What is your previous knowledge of the role of nutrition and the nutrition needs of sickle cell patients?

Q2: What is your current knowledge of the role of nutrition and the nutrition needs of sickle cell patients?

Q3: What has contributed to the change (if any) to your knowledge of the role of nutrition and the nutrition needs of sickle cell patients?

Q4: Has the Nutrition Health Education Framework informed your knowledge of the role of nutrition in sickle cell health provision, if so how?

Q5: In what way has the Nutrition Health Education Framework informed your knowledge of the barriers (personal, interpersonal, institutional and community factors) affecting the nutrition needs and management of sickle cell patients?

Q6: In what way has the Nutrition Health Education Framework informed your knowledge of the recommendations to address the barriers affecting the nutrition needs and management of sickle cell patients?

Q7: How valuable is the Nutrition Health Education Framework as a tool to improve your knowledge, awareness and understanding of the role of nutrition and the nutrition needs of sickle cell patients?

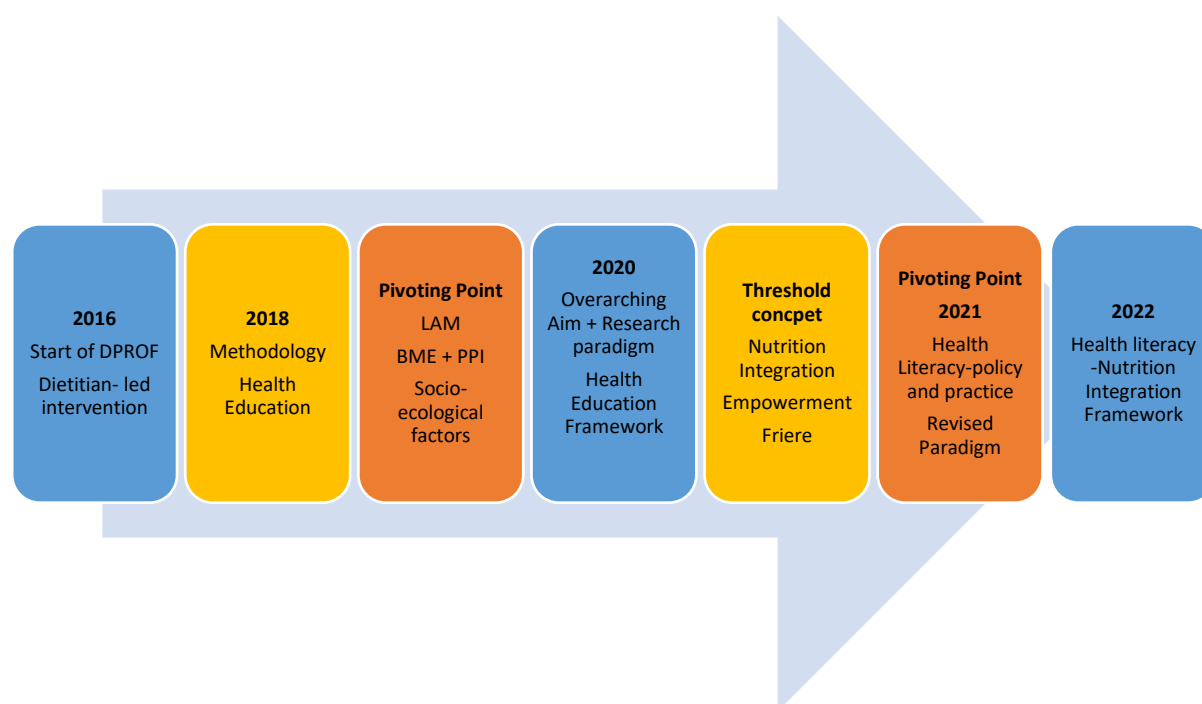
Q8: How can the Nutrition Health Education Framework be used to support the integration of nutrition into sickle cell healthcare provision?

Any comments?

THANK YOU.

Appendix 13: Chronological timeline of key threshold concepts and pivoting moments in the research project

Figure 1: - Chronological timeline of key threshold concepts and pivoting moments in the study



Appendix 14: Overlapping conceptual perspectives of the research project

Table 1: Overlapping conceptual, theoretical and methodological frameworks of the research project – Matthews (2022)

Concepts	HL – Conceptual	Freire - Theoretical	LA–Methodological
Education and Empowerment	√	√	√
Critical consciousness	√	√	√
Mutual learning	√	√	√
Collaboration and Participation	√	√	√
Social Determinants of health	√	√	√
Social Constructionism	√	√	√
Critical Realism	√	√	√

Appendix 15 Additional points raised by the LA Network

Table 1 Additional Points raised by the nutrition network alliance

Additional points raised by nutrition network alliance per focus group question category
1. Knowledge and management of Sickle cell (The invisibility of sickle cell) Additional points: ➤ Fatigue after blood exchanges ➤ Withdrawal after pain killers ➤ Lack of concentration
2. Knowledge and management of Optimum nutrition: Additional points: ➤ Lack of awareness of the types of food to buy ➤ Consider whether people have money to buy the foods they need ➤ People generally confused about how food affects their body ➤ People are also confused about what is good food or not ➤ How will this project bring about change in policy and service provision? ➤ COVID 19 restrictions on hospital menus and selection open to patients ➤ The impact of medication – side effects of medication and how it impacts on appetite suppression ➤ The lack of dietitians as part of the sickle cell management MDT – the effect of the neglect of nutrition ➤ Sickle cell provider's knowledge about sickle cell is a must
3. Socio-ecological factors affecting nutrition: Additional points: Personal: ➤ Fear about combining nutrition and medication should be addressed – on an individual basis ➤ Assess individual patient needs – eg, ability to open foods when unwell or in pain ➤ Considering impact of complications of sickle cell -eg, renal and pregnancy ➤ Consider peoples working patterns ➤ Consider people's environments – impact on their state of health (APPG report) ➤ Consider people's faith – some religions require people to fast and this may impact on their health ➤ Impact of COVID on food availability and food insecurity – eg, access to organic food ➤ Consider increased community trends – veganism and vegetarianism ➤ Impact of peer pressure on food and eating habits Institutional: ➤ Patient DNA appointments – need to rethink how patients can access nutrition services Community/policy ➤ Commissioning arrangements ➤ Consultant input

Appendix 16: Agenda for focus group and network meetings

Table 1: Agenda for focus groups and network meetings

Timing	Session	Objective
Prior to the meeting	Pre-read: Question set	The question schedule was emailed to participants ahead of the focus group to allow participants to get familiar with all 11 questions and be 'mind-ready' on the day of the focus group. A digital copy of an excel spreadsheet was emailed to the members of the NAN ahead of the first LA network meeting - due to the volume of the content to be shared.
On the day	Meet and greet	Introductions to break the ice and create a welcoming atmosphere
	Introductions	To remind participants • to maintain confidentiality • to respect other people's views and participation • the purpose, the process and duration of the focus group • the sessions will be recorded for transcription purposes • they can withdraw at any time although their collected data will be used, as stipulated in PIS (appendix 1) Participants were also asked if they had any questions prior to commencing the focus group

Appendix 17: Extracts of developing themes (phase one) from Excel Spreadsheet

Extract 1: Central codes for developing themes: Eg. (Question 2 of the Focus group questionnaire)

CM DPROF Phase 1 stage 2 -data Analysis- Service Users 28/8/20 - Excel (Product Activation Failed)			
File Home Insert Page Layout Formulas Data Review View Tell me what you want to do...			
Clipboard Font Alignment Number Styles Cells Editing			
B1 X ✓ fx Broad Codes			
A	B	C	D
1 Focus Group Questions	Broad Codes	Stage 2 - condensed themes - 9/8/20	Stage 3 - condensed themes - 16/8/20
2 Question 2:			
3 Where does nutrition fit into the patient	Nutrition knowledge of GPs and consultants	The role of nutrition in SCD	The role of nutrition as a management option in SCD
4 management plan?	Nutrition overlooked, underused and underdeveloped		
5	Psychology - why not Dietitians	Nutrition as a management option	Access to nutrition service provision and support to mana
6	Patient access to nutritional advice and support		
7	Patient access to nutritional information and resources	Nutritional educational resources	Nutrition education and evidence based resources (guide
8	Quality of Hospital Food		
9	Nutrition advice tailored to SCD needs	Nutrition Education in SCD - scientific evidence base	
10	Application of patient knowledge and support to do that		
11	Impact of social factors on nutrition	Nutrition guidelines and standards	
12	Mind the sickle nutritin GAP		
13	Opportunity to ask about nutrition at every appointment	Nutritional challenges	
14	Nutrition education - importance of nutrition		
15	Clinical Standards of Care		
16	Nutritional Standards of Care		
17	Nutrition Support		
18	Support Structures - medical/health - family/community		
19	Support settings - schools		
20	Compliance with treatment - monitoring		
21	Food fortification		
22	Nutritional challenges - (peads and adults)		
23			

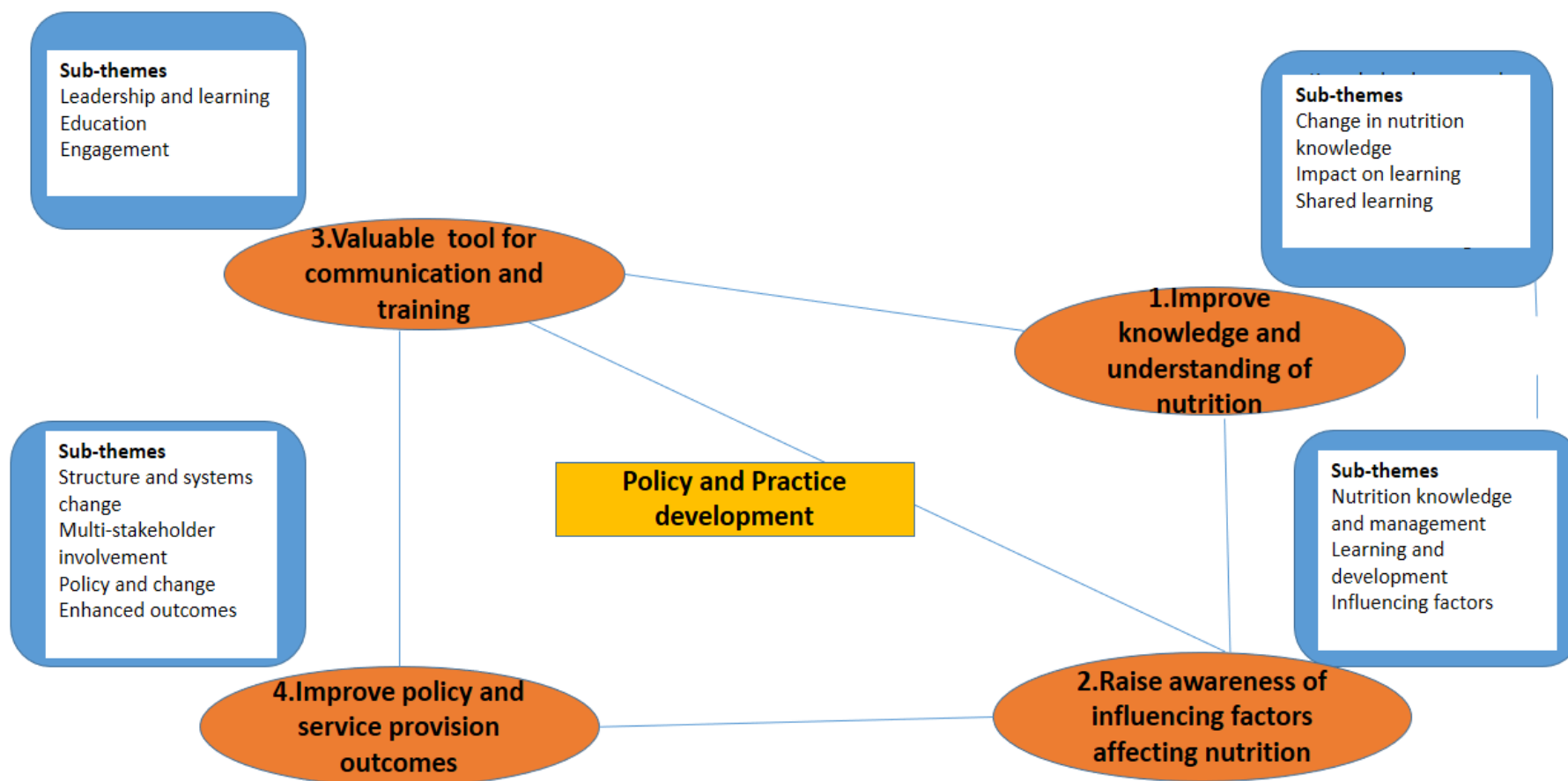
Extract 2: Comments from SCSPs – in response to Question 2 of focus group questionnaire

CM DPROF Phase 1 stage 2 -data Analysis- Service Users 28/8/20 - Excel (Product Activation Failed)			
File Home Insert Page Layout Formulas Data Review View Tell me what you want to do...			
Clipboard Font Alignment Number Styles Cells Editing			
E22 X ✓ fx SP (3) - even the referrals done even happen - so its not like we get referred onto a			
D	E		
1 Stage 3 - condensed themes - 16/8/20	Quotes		Emerging interpre
2			
3 The role of nutrition as a management option in SCD	SP (4) - so I think it terms of a medic, I think that it doesn't necessarily feature anywhere		should nutrition b
4	SP (4) - from the treatment of sickle is usually kind of protocol driven abd no one in		MUST score, nutri
5 Access to nutrition service provision and support to manage nutritional problems	in the past has ever mentioned nutrition. So we all have the pain protocols, we all have		Dietitians knowle
6	the transfusion protocols, we all have you know what you do in this sort of crisis		Article to be writt
7 Nutrition education and evidence based resources (guidelines and standards)	but no one's ever mentioned nutrition in anything else you know		various systems (c
8	SP (4) - only recently that weve started to seek sort of psychological help you know		patient satisfactio
9	SP (4) - do I ask about nutrition - no. Ill be honest with you no, firstly I wont know		Dietitians arent su
10	what to ask and secondly I wouldny know what to do with the answer, you know apart		from me maybe refer on to the dietitian and then I don't know sort of what level of
11	service they will get, I would imagine very generic		Importance of ask
12	SP (3) - Im taking the stance from my own view of it like me as well what ive experienced		All healthcare pro
13	for myself um and theres been, its been like zero.		Education on the
14	SP (3) - theres really not been any sort of input		such a huge priori
15	SP (3) - I try to ask those questions and I have many times said you know is there any		QOL of vulnerable
16	information, is there anything you know after being unwell being hospitlaised for a		SCD is chronic - th
17	number of weeks		major difference is
18	SP (3) - I know that the hospitla food for one has not been up to particualt standard,		pathophysiology f
19	we know there are deficiencies		Nutrition support
20	SP (3) - you do get a lot of generic advise to you which is fine but yes theres nothing specific		
21	SP (3) - even the referrals done even happen - so its not like we get referred onto a		Referrals not alwa
22	specialist nutritionist or anything its just whoever has the generic advice		
23			

Extract 3: Comments from SCSU-Cs – in response to Question 2 of focus group questionnaire

CM DPROF Phase 1 stage 2 - data Analysis - Service Users: 28.8.20 - Excel (Product Activation Failed)		
File Home Insert Page Layout Formulas Data Review View Tell me what you want to do... Sign in Share		
Clipboard Font Alignment Number Styles Cells Editing		
E76	SU-C (3) - I myself find that nutrition is very important for my health - even in terms of mental health has	
	D	E
55		XX
56		SU-C (3)- I think its fundamental its fundamental even in our support group we have nutritionist
57		and we promote like a holistic way of eating um a holistic lifestyle - a lot of things are
58		detrimental to the blood flow
59		SU-C (10) - its something that should be brought up more cos for me a lot of years it wasn't
60		really addressed - only in the last couple f years has my doctors or anything even brought up the
61		nutritional side of sickle cell and how that can impact any type of day to day changes have really
62		helped me improve just the pain ive been in or just how my body reacts - my own private research
63		its defiantly something tha t should be mor spoken about
64		SU-C (2) - as yu get older the doctors always say drink a lot of fluids make sure you are hydrated so that
65		affect the health a lot beause dehydration can cause promotes the onset of sickle cell crisis straingth away
66		SU-C (6) - nunitrition for me I think plays a majoor part in staying well - catering your diet to suit your health
67		SU-C (6) - ive never directly been asked about my nutrition anf how I eat and what I eat it just always been
68		something ive kind of looked up myself - ill go and do this research
69		SU-C (7) - in the management of my sickle its been central - im not on any medication so that's my
70		medical position I do not take a fixed medicone except peniilin when unwell so outside of that this is
71		how I was raised my mother was a peadiatric nurse - its really about doing your own research and
72		understanding and for me understanding the scienc behind the condition - we have a lot of inflammation in our
73		bodies and varous other things anti-oxidants enhaving blood flow - medicating yourslef on a day to day
74		basis with the foods that you eat and try to maintain a descent level of wellbeing so when the inevitable
75		crisis come its not as severe as it may be
76		SU-C (3) - I myself find that nutrition is very important for my health - even in terms of mental health has
77		been a massive part of my mental health

Appendix 18: Thematic Mapping of the evaluation themes



Appendix 19: Research Output – Examples one to six

Example 1: Abstract of the primary care Nutrition in sickle cell project

PCN Dietitian: Abstract of the Primary Care Nutrition in Sickle Cell Project

Background:

Sickle Cell Disorders (SCD) are seen to be a significant public health issue and is not only the most common genetic disorder but the fastest growing in the UK. The National Confidential Enquiry into Patient Outcomes and Death (NCEPOD) Report (2008), highlight the fact that SCD is also a common reason for hospital admissions and is associated with high rates of morbidity, mortality, disability and poor Quality of Life (QOL) in those affected by the condition. Nutrition in sickle cell however, is an overlooked management option in SCD and contributing to the poor health and wellbeing outcomes of vulnerable sickle cell patients attending both secondary care and primary care services. The ongoing poor nutrition service provision serves to widen the nutrition healthy inequality gap for this patient population. A pioneering nutrition in sickle cell project was undertaken, by the NW2 PCN Dietitian (Claudine Matthews).

Aim: To address the health inequality gap secondary to poor nutrition service provision, establish the baseline health and wellbeing needs of the patients and assess the impact of the dietetic input to improve the health and wellbeing outcomes of the patient cohort.

Participant demographics:

Total patients /Age range	Genotypes	Sickle cell crisis	Treatment Modality	Weight classification	Previous Dietetic input
Adults: n= 6 (18yrs – 57yrs)	HbSS – n=3 HbSC – n=3	Yes – n=5 No – n=1	Yes – n=4 (HU, blood transfusion) No – n=2 (folic acid and penicillin)	N= 4 normal weight *N= 1(pregnant) N= 1 Overweight *LD N= 1 Obese	NO - n= 6
Paeds: n = 6 (5yrs – 16yrs)	HbSS – n=2 HbSC –n=2 HbSBeta Thal n=2	Yes –n=3 No – n=3	N=2 (TCD, blood transfusion) N= 4 (folic acid and penicillin)	N= 2 underweight N= 2 Overweight N= 2 Obese	No – n= 6 *delayed puberty

Project Findings/Discussion:

It was evidence that none of the patients in the study had prior dietetic input. The weight classification in the paediatric group highlighted the problem of stunting (short height for age – evident of chronic malnutrition). One paediatric patient had delayed puberty and was significantly underweight, but later improved towards the end of the project. The parents of the obese paediatric patients were happy to receive healthy eating advice and written

information. Follow up calls revealed improved knowledge and awareness of healthy eating and healthy food options. The pregnant patient, had not received dietetic input but was happy to know that she could ask for help if needed and was in full support of the project. The adult patient (with Learning disability) who was overweight, managed to lose 4 kg of weight following dietetic input and the variety of her diet improved markedly. Moreover, all the participants and the parents of the paediatric patients, fully supported the project.

Conclusion:

Overall, the implementation of this project enabled the NW2 PCN to take affirmative action to address the health inequality associated with poor nutrition service provision, thereby improving the health and wellbeing outcomes of the sickle cell patient cohort in the project.

Example 2: Email request for nutrition to be placed on the APPG-SCAT agenda

Request for Nutrition in SCD to be placed on the SCTAPPG Agenda

1 attachment

Good morning

I trust you are well!

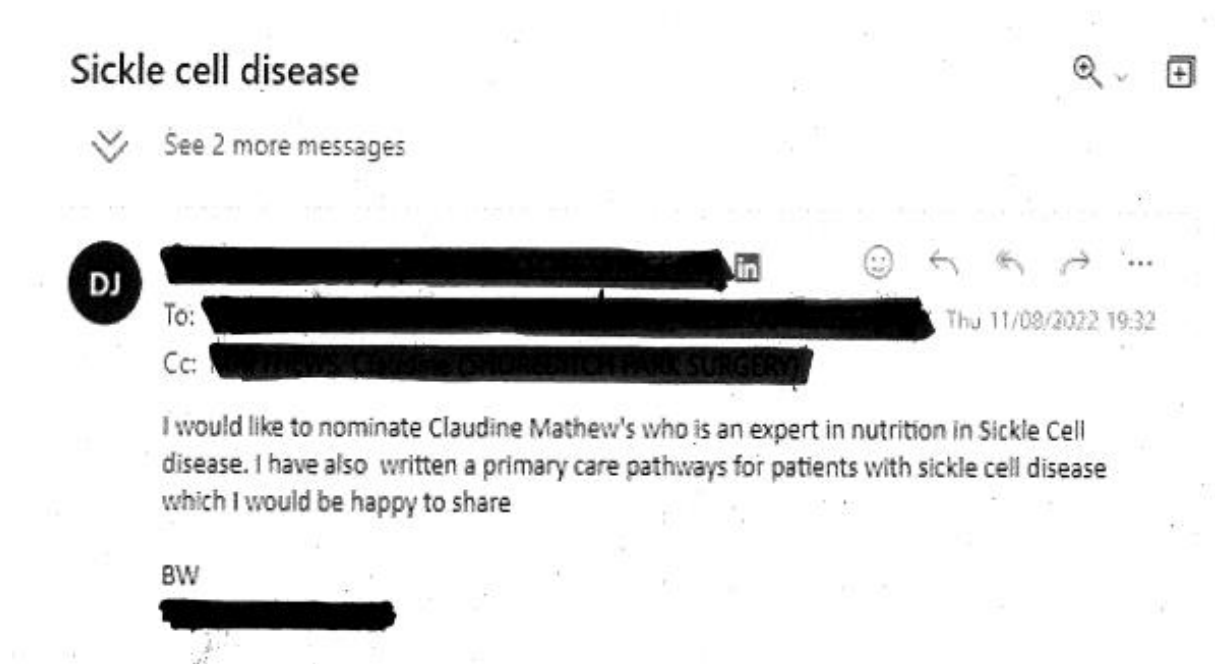
It was wonderful to meet you online at the BPSA Sickle Cell Conference on Saturday 26/11/22. Thank you for sharing the work that you do as part of the SCTAPPG and policy officer for the Sickle Cell Society.

Thank you also for committing to add nutrition to the SCTAPPG Agenda for open discussion, (and question time in parliament) as you will read in the attached letter- nutrition in SCD is a neglected management option in this genetic condition, making it an **unmet need** for many vulnerable patients living with sickle cell and a **growing health inequality** that needs **urgent attention**!

As I mentioned on Saturday, I have dedicated my Doctoral research to the topic - with a focus on **integrating nutrition into standard sickle cell care provision** - as our patients are at risk of the late diagnosis of nutritional problem, a key finding from my Doctoral research project. I have 6 months (from January) until I complete my Doctorate.

Windows Ink Workspace

Example 3: Email Nomination to be part NHS England SCD clinical pathway review



Example 4: Email to the CEO of the BDA

Letter in support of Nutrition in SCD - A developing Health Inequality

MC

Matthews, Claudine (Shoreditch Park Surgery)

To: [redacted]@bda.uk.com

Mon 26/06/2023 10:41

Cc: Claudine-Matthews <cmnutri@icloud.com>



Nutrition in SCD - APPG - SC...
28 KB

Good morning, Ms [redacted]

I trust you are well!

Further to my email, dated 19/6/23 - I am just sending a follow up email to check whether you have received the original, as found below.

I look forward to hearing from you, as convenient.

Best wishes,

Claudine Matthews

PCN Dietitian

Shoreditch Park and City Primary Care Network

Example 5: Email to the Deputy Director and Director of Health Inequalities (NHS England)

Nutrition in Sickle Cell Disease

Nutrition in Sickle Cell Disease



MATTHEWS, Claudine (SHOREDITCH PARK SURGERY)



To: [REDACTED] (NHS ENGLAND – X24); +1 other

Mon 25/09/2023 10:44

Cc: MATTHEWS, Claudine (SHOREDITCH PARK SURGERY)

Nutrition in Sickle Cell Diseas...
22 KB

Good morning, Dr [REDACTED] and Prof [REDACTED]

I trust you are both keeping well!

Thank you for all the amazing work you are doing to highlight and address health inequalities for the minority ethnic groups in the NHS, on a strategic letter.

As such, I am writing to you regarding **the neglect of nutrition as part of standard care provision in SCD, affecting patient outcomes**. I am soon completing my Doctorate degree - aimed at integrating nutrition into standard care in Sickle Cell Disease - an important issue I raised as part of the SCD clinical pathway review conducted in August 2022 (as part of Workstream 2).

Example 6: Research Output – Publications, Conferences, Presentations

Professional Publications:

Articles:

Matthews, C.E., 2021. *Pioneering National Nutrition Standards: How one Dietitian is blazing a trail for the provision of nutritional care for sickle cell patients*. Complete Nutrition (CN) Magazine, 21 (1), pp.54-56. [pdf] Available at:< <https://nutrition2me.com/wp-content/uploads/2021/03/Sickle-Cell-Disease.pdf>> [20 July 2017].

Abstracts:

Matthews, C. et al., 2021. A qualitative study to understand the optimum nutrition needs of sickle cell patients and the influencing socio-ecological factors. Journal of Human Nutrition and dietetics, [e-journal] 34 (1) pp3-71. Abstract from the 2020 BDA Research Symposium, 2 December 2020, Birmingham, UK.

Matthews, C. et al., 2021. A qualitative study to understand the barriers to integrating nutrition into sickle cell healthcare provision. HemaSpere. Abstract book of the 15th Annual Sickle Cell and Thalassaemia, 1st EHA European Sickle Cell Conference and 60th Anniversary of BSH, 26-31 October 2020, London, UK.

Peer Review Publication:

Matthews, C.E., 2019. The role of nutritional care in Sickle Cell Disease: A real phenomenon. ACTA Scientific Nutritional Health, 3 (2), pp.74-80. Available at:< <https://actascientific.com/ASNH/pdf/ASNH-03-0180.pdf>

National Nutrition Standards (SCS, 2018, pp 75-77):

Sickle Cell Society (2018). Standards for the Clinical care of adults with sickle cell disease in the UK. Accessed online: www.sicklecellsociety.org/wp-content/uploads/2018/05/Standards-for-the-Clinical-Care-of-Adults-with-Sickle-Cell-in-the-UK-2018.pdf (Jan 2021).

Conferences:

Development of the inaugural Annual Nutrition in SCD Conferences:

1st Annual Nutrition in Sickle Cell Conference, Online London, UK – 18th June 2021 ***“The Evolution of Nutrition in Sickle Cell Disease: A call to action”***

2nd Annual Nutrition in Sickle Cell Conference, Online London, UK – 17th June 2022
“Nutritional approaches to improve health outcomes for sickle cell patients: a focus on policy and practice”

Presentations:

Centre for Disease Control – Online Webinar: Nutrition in Sickle Cell Disease – 28th July 2021^{8th} ***“Integrating nutrition as a management option in Sickle Cell Disease: a call to action”***

8th Annual Warriors Convention – Nutrition in Sickle Cell Disease- 8th August 2021

“Stronger Together: Building a nutrition in Sickle Cell Ecosystem”

Pan -Midlands HCC Educational Event – Nutrition in Sickle Cell Disease – 24th June 2022
“Why nutrition in Sickle Cell Matters: a call to action”

North West HCC Educational Event – Nutrition in Sickle Cell Disease – 22nd August 2022
“Nutrition in Sickle Cell Disease: an opportunity for policy and practice change”

Appendix 20: List of articles screened in the systematic literature review:

Database	Article
CINAHL	1.Forte, L.R.S. 2000. The meaning of Hope to family care givers of children with Sickle Cell Disease. Doctoral thesis. 2.Harris, L.H. and Gore, S., 1996. Sickle cell disease: care throughout the lifespan. <i>Journal of National Black Nurses Association</i>, 8(1), pp.33-44. PMID:9128544.
MEDLINE	3. Swe, K.M., Abas, A.B., Bhardwaj, A., Barua, A. and Nair, N.S., 2013. Zinc supplements for treating Thalassaemia and Sickle Cell Disease. <i>Cochrane Database Systematic Reviews</i>, 26 (6), Available at:CD009415. doi: 10.1002/14651858.CD009415.pub2. PMID: 23807756; PMCID: PMC9964104.
Psch INFO	4.Gesteira, E.C.R., Bousso, R.S., Misko, M.D., Ichikawa, C.R.F., Oliveira, P.P., 2016. Families of children with Sickle Cell Disease: an integrative review. <i>Online Brazilian Journal of Nursing</i>, 15(2), pp.276-290. Available at: https://www.redalyc.org/pdf/3614/361453979017.pdf. 5.Tanabe, P., Porter, J., Creary, M., Kirkwood, E., Miller, S., Ahmed-Williams, E. and Hassell, K., 2010. A qualitative analysis of best self-management practices: Sickle cell Disease. <i>Journal of National Medical Association</i>, 102(11), pp.1033-1041. Available at: http/ 6.Campbell, A.D., Ross, P.T., Kumagai, A.K., Christner, J.G. and Lypsom, M.L., et al., 2010. Coming of age with Sickle Cell disease and the role of patient as teacher. <i>Journal of National Medical Association</i>, 102(11), pp. 1073-1078. Available at: doi: 10.1016/s0027-9684(15)30735-5. PMID: 21141297. 7. Hilker, K., Jordan, S.S., Jensen, S., Elkin, T.D. and Lyer, R., 2006. Development of a Screening Instrument of Adherence in Pediatric Sickel Cell Disease. <i>Children's Health Care</i>. 35. 235-246. Available at: 10.1207/s15326888chc3503_3. 8.Davies, S.C. and Oni, L., 2001. Sickel cell disease screening programs integrated into management care. <i>Dis-Manage-Health-Outcomes</i>, 9 (6), pp. 295–304 (2001). https://doi.org/10.2165/00115677-200109060-00001.