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Mother–infant interactions with infants with congenital visual impairment and associations with longitudinal outcomes in cognition and language

Elena Sakkalou,[1,2,3] Michelle A. O'Reilly,[1,2] Hanna Sakki,[1] Clare Springall,[2] Michelle de Haan,[1] Alison T. Salt,[1,2] and Naomi J. Dale[1,2]

[1]UCL Great Ormond Street Institute of Child Health, London, UK; [2]Great Ormond Street Hospital, NHS Foundation Trust, London, UK; [3]School of Psychology and Sports Science, Anglia Ruskin University, Cambridge, UK

Background: This study investigated mother–infant interactions, including maternal maintaining of infant attentional focus and sensitivity, with infants with congenital severe and profound visual impairment (VI) and the association with developmental trajectories from one to three years. **Method:** Fifty-five infants and mothers were video-recorded playing together with a standard set of toys at Time 1 (T1) mean age 12.95 months (8.13–17.05 months). *Maintain* was categorized as the mother following and maintaining the child's focus, and *Sensitivity*, the mother's responsiveness and contingency to infant behaviour. Vision level was measured using the Near Detection Scale. Cognition and language were measured at T1, 12 months later (T2) and 24 months later (T3) using the Reynell-Zinkin Scales. **Results:** Cross-sectional analyses showed that mothers of infants with severe VI (basic form vision) produced higher rates of Maintain compared to those with children with profound VI (light perception at best). Linear mixed-effects models examining developmental progression from T1 to T3 (controlling for vision level) showed an average increase of 5 DQ points (CI 95%: 1.03–9.08) in verbal comprehension for higher Sensitivity. No significant findings were found for Maintain. **Conclusions:** The findings suggest that mother–infant interactions (maternal Maintain) are associated with level of vision at infancy, but only maternal Sensitivity has a long-term positive association with advances in verbal comprehension from infancy to about three years. They highlight the need for incorporating strategies related to parent–infant interactions, including increased sensitivity, into early intervention for children with visual impairment. **Keywords:** Visual impairment; blindness; child; infant; mother–child relations; language; cognition.

Introduction

Congenital visual impairment (VI) has a profound effect on early development and the early relationship between the caregiver and child. Although congenital eye disorders are rare (estimated at <3–4 infants with chronic severe VI per 10,000 births in the United Kingdom) (Rahi et al., 2003), the severity of risks for development is of high clinical urgency. Lack of visual input (profound VI – light perception at best) and very low levels of vision (severe VI – some 'form' vision) have been associated with significant developmental delays and challenges in acquiring sensorimotor/cognitive, social-communicative and language abilities with delays of up to 12–24 months (Dale & Sonksen, 2002; Hatton, Bailey, Burchinal, & Ferrell, 1997; Reynell, 1978). Infants with VI may appear very passive and quiet throughout their first year, lacking eye contact, facial expression and other nonverbal means of seeking or responding to parental attention. Bodily movements may be socially undirected and vocal cues infrequent and not necessarily socially responsive. By the second year, the toddler may be more socially responsive, but as independent mobility and play emerge, they are often single-channelled in attention and self-directed. The child may become more difficult to regulate, and these behaviour challenges may impact on the parent's well-being (Alon, Ophir, Cohen, & Tirosh, 2010; Sakkalou, Sakki, O'Reilly, Salt, & Dale, 2018). Parents and infants experience obstacles in achieving coordinated interactions, including parental difficulty in reading infant nonvisual cues, and the infant's inability to pick up parental social intentions or to engage with the parent (Andersen, Dunlea, & Kekelis, 1984; Dale, Tadić, & Sonksen, 2014; Rowland, 1984; Tröster & Brambring, 1992). This is most marked in infants with profound VI or very low vision, and their lack of engagement is associated with parents tending to more directive or one-sided interactions (Andersen et al., 1984; Behl, Akers, Boyce, & Taylor, 1996; Moore & McConachie, 1994; Preisler, 1991). Having some functional vision, even if only low levels of 'form' or residual vision, may lead to more synchronized and coordinated interactions.

In typically sighted children, responsive parenting, which refers to parental behaviours contingently linked to the child's experience and underlying mental states, has positive effects on developmental outcomes. These behaviours include following the child's lead and maintaining the child's interest and focus of attention (Landry & Chapieski, 1989; Spiker et al., 2002), and responding promptly and contingently and with warm sensitivity towards the child

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(Ainsworth, Blehar, Waters, & Wall, 1978; Murray, Fiori-Cowley, Hooper, & Cooper, 1996). Attention-maintaining and sensitivity strategies are associated with enhancements in cognitive development in typically developing and atypical populations (Akhtar, Dunham, & Dunham, 1991; Bakermans-Kranenburg et al., 2003; Deans, 2018; Landry & Chapieski, 1989; Landry, Smith, Swank, & Guttentag, 2008; Tomasello & Todd, 1983).

Attention-maintaining and sensitivity have not been systematically investigated in interactions between mothers and infants with VI, yet these infants rely heavily on support from their mothers. Given the developmental and mother–infant interactional risks found in this population, the present study investigated maintaining and sensitivity strategies during play between mothers and infants with VI aged 8–17 months.

Following-in and maintaining the child's attentional focus allows for the establishment of shared attention and has been shown to promote vocabulary acquisition (Akhtar et al., 1991; Tomasello & Todd, 1983), focused attention, problem solving, exploration and learning (Bono & Stifter, 2003; Landry, Garner, Swank, & Baldwin, 1996). Whereas typically sighted children have the advantage of gaining information from objects not directly within reach, children with VI do not have the same access to objects in their environment. They require objects to be brought near their field of vision or for children with Profound VI, often within immediate contact. By following-in and providing tactile and auditory feedback, mothers provide opportunities which infants would not be able to obtain through vision and eye gaze (Akhtar & Gernsbacher, 2008; Bigelow, 2003) and sustaining child attention to their existent focus of attention may be advantageous for optimal learning.

Whereas maintaining centres on following-in and sustaining the infant's focus of attention, sensitivity focuses on how sensitively a mother is attuned and responds to the infant's signals and mental states (Murray et al., 2008). It refers to the mother's ability to perceive the infant's point of view and to respond contingently and appropriately, reinforcing these in the child. Sensitivity in infancy has been shown to positively influence later language, cognitive and social skills including children with intellectual disability and autism spectrum disorder (Mahoney, Finger, & Powell, 1985; Wan et al., 2013).

Although both maintaining and sensitivity are considered positive parenting strategies, one may argue that maintaining focuses on explicit and directly observable aspects of behaviour, for example the child's focus of attention to an object which they are holding. Sensitivity focuses on the mental states and subtle behaviour conveyed by the child, for example emotions, intentions. Thus, whereas a mother who maintains may follow the child's signals and show the child how to play with an object, a mother with high sensitivity may predict and respond contingently and appropriately to the infant's inferred intentions. We therefore hypothesized that first, maintaining would be associated with advances in sensorimotor understanding (nonverbal cognition), as a consequence of helping the infant focus on and learn about aspects of objects and their properties and concepts. Second, sensitivity would be linked to advances in language development, as perceiving, contingently responding and putting the infant's inferred intentions into actions and words is one of the fundamental requirements for learning language (Rollins, 2003; Tomasello & Todd, 1983; Vallotton, Mastergeorge, Foster, Decker, & Ayoub, 2017).

Because these hypotheses cover immediate and longer-term differential influences, we examined the influence of these parenting strategies on the developmental progression of nonverbal cognition and language longitudinally from 8–17 months to 24 months later (approximately 3 years of age). The developmental risks and vulnerability for young children with VI, especially in the areas of nonverbal cognition (sensorimotor understanding) and language (e.g. verbal comprehension and expressive language), continue across the first three to four years of life (Dale & Sonksen, 2002). The data from this study are part of a national longitudinal project investigating early development and intervention in young children with visual impairment (OPTIMUM project, Dale et al., 2017, 2019).

In summary, this study set out to investigate mother–infant interactions and potential associations between maternal behaviours and their longitudinal impact on nonverbal cognition and expressive and receptive language outcome in young children with congenital VI, with a focus on mothers. These associations have not been investigated before. We propose the following cross-sectional and longitudinal hypotheses:

1. Maintaining and sensitivity are correlated because although they encompass separate parenting characteristics, they both reflect responsive parenting;
2. The severity of the child's VI affects maternal responses; that is, mothers with infants with severe VI (SVI, some 'form' vision) show higher levels of Maintain and Sensitivity than mothers whose infants have Profound VI (PVI, no vision or light perception at best);
3. A higher frequency of maintaining in infancy is associated with increased rate of sensorimotor understanding/nonverbal cognition, compared with a lower frequency;
4. Higher maternal sensitivity in infancy is associated with increased rate of expressive (expressive language structure) and receptive language (verbal comprehension) compared with lower sensitivity.

As in our previous analyses of the OPTIMUM cohort, the demonstrated relationship of vision with longer-term developmental outcome was considered and controlled for (Dale et al., 2017, 2019). This part of the study was designed and conceptualized from outset, with statistical predictions prior to data collection, but was analysed separately from the primary objective of evaluating the effects of the Developmental Journal for babies and young children with visual impairment – DJVI) on developmental outcomes (Dale et al., 2019).

Method

Design and setting

This study is part of a prospective longitudinal observational project (OPTIMUM cohort) recruited between 2011 and 2014 (Dale et al., 2017, 2019). Data collection occurred between 2011 and 2016. Child assessments took place at a research site laboratory room or home at three time points, T1 (baseline), T2 (12 months later) and T3 (24 months later).

Participants

Inclusion criteria were infants aged 8–17 months, with a diagnosis of a congenital disorder of the peripheral visual system (CDPVS), that is ophthalmological disorders of the globe, retina or anterior optic nerve with (potentially complex) or without (potentially simple) known central nervous system involvement in the paediatric diagnosis (Dale & Sonksen, 2002). All children had chronic severe-profound VI (estimated near resolution acuity of logMAR 1.0 or worse at time of entry). Exclusion criteria included diagnosed neurological and/or motor/hearing impairment, retinopathy of prematurity, severe prematurity and parents who spoke insufficient English to complete questionnaires. All participating mothers had sufficient English to fill out questionnaires. There were mothers who spoke in their native language during mother–infant play; however, these mothers had sufficient English to fill out the questionnaires. For videos where we had the expertise in the team to translate the native languages (researcher being a native speaker of the language), we produced a verbatim transcript, which was used alongside the video during coding. The videos that could not be translated were not coded. See Dale et al. (2017) for details of recruitment and sampling information.

Mothers only were invited to participate in this part of the study to ensure consistency of the parents' gender and to avoid a possible gender confound in style of parenting responses if male and female parents were included. This sampling decision is in line with the previous literature on sensitivity and maintaining which also focused on mother–infant dyads and thus would permit comparison of our data results with those from the previous literature. Children with 'potentially simple' CDPVS only were included in this analysis, to eliminate the potential confound of additional brain complexity.

Measures and procedures

Vision level. Best corrected vision level was measured by the developmental psychologist/neuroscientist (ES, MO'R), trained by the consultant neurodisability paediatrician (AS), using the Near Detection Scale – NDS in standard conditions (Sonksen, 1983). The NDS is a 10-point scale ranging from no light perception (0) to 0.1 cm 'lure' (9) according to visual fixation on incrementally sized lures at 30-cm distance; vision level was divided further into Profound visual impairment – PVI (points 0–1, light perception at best) and Severe visual impairment – SVI (points 2–9). The estimated resolution acuity was measured using Keeler Acuity Cards (Keeler, 2014), but is not reported in this study.

Mother–infant play. As part of a longer session involving other assessments, infants and mothers were video-recorded for 10-min playing together with a standard set of age-appropriate toys that were suitable for infants with VI including a jack-in-the box, pull-along toy, toy piano and book. Mothers were asked to play with their infant as they would at home and were allowed to sit as they chose for the duration of the interaction. The majority of interactions took place on the floor and on some occasions where a table was present the child sat on an age-appropriate chair at the table with their mother. The size of the room varied, but the toys were kept in close proximity to the mother and child and they were placed within easy reach of both participants. The rooms were well-lit except in the case of two children who were photosensitive. For these two children, the light was dimmed but the interactions were still visible on camera. The mothers and children were video-recorded by one camera and a microphone, which was placed in near distance from the participants, in order to capture audio sound. A member of the research team video-recorded the play session, and they were sat at a distance of about 1.5–2 m away from the participants. Mothers were told 'We would like you to play with [child name] for 10 min. Here are some toys that you can play with if you would like. Please play with [child name] as you would usually at home.' For Maintaining, videos were coded after the session using Interact (Mangold, 2013; version 9.6.4). The trained rater (HS) had not directly assessed the child and was unaware of the child's developmental level. A different rater (CS) scored Sensitivity by watching the same videos. The investigator (ES) acted as a secondary coder for reliability. Both the first and second raters (HS, CS, ES) received direct training and gained experience of working with infants and young children with visual impairment under the supervision of the principal investigator (ND), who is a consultant clinical psychologist with extensive clinical experience and expertise in infants and children with visual impairment. The first raters were an assistant psychologist (CS) and a research assistant (HS), who had experience participating in the paediatric developmental vision clinic (under supervision of ND) and had undertaken past coding on various scales with this population. The second rater (ES) is a developmental psychologist with expertise in mother–infant interactions and trained the first raters on the Sensitivity and Maintaining coding schemes. Piloting was undertaken to ensure that both raters had a similar understanding of the categories of the mother and infant behaviours, and reliability was calculated following the completion of all videos.

Maternal attention-directing strategies. The coding manual (with minor adaptations for the VI population) from Landry and colleagues (e.g. Landry & Chapieski, 1989) was used to code the first five minutes of the recording of a mother–infant interaction. This scheme consisted of three attention-directing strategy codes: *Maintain, Introduce* and *Redirect.* (a) *Maintain* was categorized as 'the infant has a focus of interest and the mother follows and maintains the child's focus and is an explanatory variable in the longitudinal analysis.' (b) *Introduce* was categorized as 'mother introduces a toy or focus of attention when the child focus is undetermined.' (c) *Redirect* was categorized as 'the infant has a focus of interest and the mother tries to redirect the child's focus onto something else.' The total number of all attention-directing instances observed during the 5-min coding period was summed. The percentage for each attention-directing strategy was calculated by dividing the number of instances of each

strategy by the total number of all maternal attention-directing instances and multiplying by a hundred.

Sensitivity. Mother–infant interactions were coded using the Sensitivity coding scheme (Murray, et al., 1996, 2008). Minor adaptations for VI were included; for example, holding and guiding the child's hand to objects was not considered insensitive. Coding consisted of a 5-point global impression rating scheme (1 low – 5 high), with '5' scored as having *High Sensitivity*, where mothers were viewed as being empathic, accepting and responsive in ways appropriate to the infant's behaviour; '1' – Mothers scored as having *High Insensitivity*, viewed as being geared largely by signals within themselves, with delayed and inappropriate responses towards their infant. Midpoint was '3' – *Inconsistently Sensitive* (average level sensitivity) if they were sensitive at some times or in respect to some aspects of the infant's experience, but not in others (Murray et al., 2008). The global impression and rating were reached after observing the complete 10-minute recording.

Cognition and language. Cognition and language were assessed by the developmental psychologist/neuroscientist (ES/MO'R), trained by the consultant neuropsychologist (ND), using the play-based assessment of Sensorimotor Understanding (SMU), Response to Sound and Verbal Comprehension (RSVC) and Expressive Language Structure (ELS) subscales of the Reynell-Zinkin Scales (RZS) for young children with VI. The sighted age equivalent norms of the RZS were used to form developmental ratio quotients (DQs), and vision level was controlled in the analysis (Reynell, 1978; more information in Dale et al., 2017 and Appendix S1).

Reliability

Intra-class correlation coefficients were calculated for 20% of maternal behaviours for the Maintain coding, which are event-based, and all of the Sensitivity ratings, which depend on global impression and are deemed more subjective. High degrees of inter-rater reliability were found for Maintain (ICC = 0.92) and Sensitivity (ICC = 0.87). Further information about manuals and training of coders can be found in Appendix S1.

Statistical methods

To address hypotheses 1–2 regarding associations between Maintain and Sensitivity, and further associations between these two maternal variables and child vision level at T1, Spearman correlations were used. To address hypotheses 3–4, longitudinal analyses were undertaken based on repeated outcome measurements of SMU, RSVC and ELS DQs at T1, T2 and T3. Linear mixed-effects models were fitted to the data using restricted maximum likelihood allowing for the model specification of assumptions of random effects (random intercept at baseline and random slope from T1 to T3; Baraldi and Enders, 2010 Laird & Ware, 1982). One model per outcome (SMU, RSVC and ELS) was analysed; therefore, three models are reported (Table 2). The explanatory or independent variables were (a) Maintain (percentage maintaining out of 100) or Sensitivity (1–5), (b) Vision level (PVI, SVI) at T1 and (c) Time (three-level categorical variable: T1, T2 and T3). Previous research by our group and others has demonstrated that level of vision is related to developmental outcomes and infants with profound VI (no vision or light perception at best) are the most at risk (Dale & Sonksen, 2002; Vervloed et al, 2000). It was important to control for the effect of vision level at T1 (PVI/SVI) on the different rates of the development of SMU, RSVC and ELS Sighted DQ norms. We have demonstrated elsewhere that vision category level (PVI or SVI) at T1 remained stable at T2 (Salt et al., 2020). Statistical analyses were undertaken using R version 3·3·2 and the package nlme. For further details, see Appendix S1.

Results

Participant characteristics

Data from 55 mother–infant dyads (80% of those meeting the criteria for 'potentially simple' CDPVS in the total sample, $n = 69$) are reported. The OPTIMUM cohort was shown previously to be representative of population study of children with congenital peripheral visual disorders (Table 1), and census socio-economic, parental education and black minority and other ethnic group membership indicators (see Dale et al., 2017). Fourteen children (20%) of 69 with 'potentially simple' CDPVS could not be included (see Appendix S1).

Characteristics of the participants are provided in Table S1. Infants' ages ranged between 8.13 and 17.05 months ($M = 12.95$; $SD = 2.54$ months) at T1. Fourteen infants (25.5%) were PVI, and 41 (74.5%) had SVI.

Maintaining

Percentage of instances of Maintain ranged from 18.75% to 95.65%. Paired-samples Wilcoxon signed-rank tests revealed that mothers produced significantly higher proportion of Maintain strategies ($M = 62.38$, $SD = 19.77$) compared to Introduce ($M = 18.66$, $SD = 16.29$), $z = -5.90$, $p < .001$, $\eta_p^2 = 0.63$ and Redirect strategies ($M = 18.96$, $SD = 15.07$), $z = -5.96$, $p < .001$, $\eta_p^2 = 0.65$. There was no significant difference between Introduce and Redirect strategies $z = -.05$, $p = .96$. The average percentage of Maintain with children with PVI was $M = 53.23$, $SD = 18.17$ and for children with SVI was $M = 65.51$, $SD = 19.52$; with significantly lower percentage of Maintain strategies in the PVI than in the SVI group ($U = 177.00$, $p < .05$, $\eta_p^2 = 0.08$).

Sensitivity

Average level of Sensitivity was $M = 2.93$, $SD = 0.97$ for the total sample. Average Sensitivity for mothers

Table 1 Frequency and percentage of visual disorders according to primary anatomical site[a] affected and vision-level category

Visual disorder (grouped according to primary anatomical site affected)	$n = 55$ (%)	PVI ($n = 14$)	SVI ($n = 41$)
Globe	24 (43.6)	5	19
Optic nerve	4 (7.3)	1	3
Retina	24 (43.6)	8	16
Other	3 (5.5)	0	3

[a]More than one site was affected in some individuals. Vision disorder diagnoses were classified according to a UK national epidemiological framework (Rahi et al., 2003).

of children with PVI was $M = 2.71$, $SD = 0.91$ and for SVI was $M = 3.00$, $SD = 0.99$. There were no significant differences between the PVI and SVI groups, $U = 236.50$, $p = .32$. Eighteen per cent of mothers had scores of 4 and 5 (higher Sensitivity).

Associations between Maintain and Sensitivity

A Spearman correlation between Maintain and Sensitivity showed a weak-to-moderate significant correlation $\rho = .36$, $p < .01$.

Longitudinal outcomes of RZS scores for Maintain and Sensitivity

In the linear mixed-effects models, Maintain was not associated with any estimated increase in DQ on SMU (0.03), RSVC (–0.14) or ELS (–0.09) from T1 to T3 (Table 2). Sensitivity was not associated with an estimated increase in DQ on SMU (–0.20). A small nonsignificant increase in DQ of 1.60 was shown on ELS. However, Sensitivity (in the direction of higher level) was associated with an estimated increase in DQ of 5.06 (CI: 1.03–9.08, $p = .01$) on RSVC from T1 to T3. In all models, Vision level (in the direction of SVI) was associated with an estimated increase in DQ, ranging from 11.13 to 21.12 DQ points compared to PVI, with all models reaching statistical significance ($p < .05$).

Discussion

This study is the first to examine variations in levels of attention-directing strategies, including Maintain and Sensitivity during play in mothers of infants with rare 'potentially simple' congenital disorders of the peripheral visual system and severe to profound VI.

At T1, Maintain and Sensitivity showed a low-to-moderate correlation, suggesting that the two maternal behaviours may share common aspects but are not an identical parenting construct. The longitudinal association of these constructs with cognition and language trajectories from T1 to T3 was examined. The cross-sectional results show Maintain tended to vary between participants and differed according to the child's level of vision at T1. As predicted, mothers who had children with light perception at best (PVI) tended to use less Maintain strategies than mothers who had children with 'form' vision (SVI). However, contrary to prediction, no significant longitudinal association of Maintain strategy was found with the trajectory of sensorimotor understanding/nonverbal cognition (SMU) DQ from T1 to T3; it was also not found for verbal comprehension (RSVC) and expressive language (ELS) DQs.

In terms of Sensitivity, the cross-sectional results at T1 suggest that mothers produced on average the midpoint (inconsistently sensitive) level of Sensitivity and unlike Maintain and contrary to prediction, Sensitivity did not differ significantly according to the child's vision level. However as predicted, the longitudinal results showed a significant positive association of Sensitivity with the estimated increase in verbal comprehension (RSVC) DQ from 12 months to 3 years of age when controlling for vision level.

As predicted, level of vision in the direction of SVI was significantly positively associated with estimated average increases ranging between 15.19 and 20.74 DQ in SMU, RSVC and ELS in Maintain and between 11.13 and 21.12 DQ in Sensitivity from T1 to T3. This highlights the greater developmental delay or slower progress of infants and young children with profound VI, as reported previously. Mothers in this sample had on average a similar level of Sensitivity to that reported in other studies with typically sighted and developing children using the same coding scale (e.g. Murray et al., 1996, 2008). Even with multiple factors likely to be influencing each child's individual rate of development including raised risk of developmental delay and controlling for vision level and time, the longitudinal mixed-effects modelling analysis demonstrated that for one-unit increase in Sensitivity at T1 infants showed on average an estimated increase of 5 DQ points (CI 95%: 1.03–9.08) in verbal comprehension (RSVC) from T1 to T3. This increase in DQ suggests an

Table 2 Results of the linear mixed-effect models with SMU, VC and ELS as the outcome (dependent) variables measured at all time points (T1, T2 and T3) and Sensitivity, Maintain and Vision level (SVI, PVI) as the explanatory (predictor) variables

RZS scales (explanatory variables)	Estimated effect (DQ) ($n = 55$)	95% CI		p value	RZS scales (explanatory variables)	Estimated effect (DQ) ($n = 55$)	95% CI		p value
SMU									
Maintain[a]	0.03	–0.19	0.24	.80	Sensitivity[a]	–0.20	–4.44	4.04	.93
Vision level[b]	20.74	10.96	30.53	**.0001**	Vision level	21.12	11.61	30.64	**.0001**
RSVC									
Maintain	–0.14	–0.36	0.07	.18	Sensitivity	5.06	1.03	9.08	**.01**
Vision level	15.19	5.59	24.79	**.003**	Vision level	11.13	2.17	20.10	**.02**
ELS									
Maintain	–0.09	–0.34	0.17	.50	Sensitivity	–1.60	–6.64	3.44	.5
Vision level	15.87	4.14	27.60	**.009**	Vision level	15.92	4.49	27.34	**.007**

[a]Positive values for higher sensitivity and higher maintain.
[b] Positive values for the SVI group.
Bold valuse are highlighted to indicate significant results.

estimated standard deviation increase of approximately 0.33 *SD*. These results bear some similarity to previous findings from studies with typically developing sighted children and other clinical populations which found cross-sectional differences in longer-term outcome (Baumwell, Tamis-LeMonda, & Bornstein, 1997; Belsky & Fearon, 2002; Paavola, Kemppinen, Kumpulainen, Moilanen, & Ebeling, 2006). For children with VI, RSVC draws on semantic linguistic understanding of communication, objects and routines which they cannot see or in a highly blurred manner, and therefore, sensitivity and attunement of social support and interaction and language may be particularly important for mastery of verbal comprehension.

One possible explanation for the lack of association between vision level and Sensitivity may be that some mothers have similar difficulties in attuning to and identifying subtle communicative cues in their infants whether they have no vision or very low levels of vision. Moreover, many of the infants with SVI would have been previously in the PVI range in the first months of life which could also have impacted on early parenting style. Alternatively, Sensitivity may be a characteristic of the individual mother's personality and own parenting experiences and her ability to pick up mental states and respond to these, rather than a reactive response to her infant's interactive capacities (Belsky & Barends, 2002; Belsky, Crnic, & Woodworth, 1995). Other factors like postnatal or postdiagnostic depression and anxiety may be impacting on parenting strategy and bonding (Murray et al., 1996; Sakkalou et al., 2018). However, when we examined correlations between measures of anxiety, depression (Hospital Anxiety and Depression Scale – Zigmond & Snaith, 1983) and parenting stress (Parenting Stress Index – Abidin, 1995) levels, which we had collected as part of the overall study, with Sensitivity and Maintaining no significant relations were found (Sakkalou et al., 2018). These results suggest that maternal psychological profiles of anxiety, depression or parenting stress are not impacting the relationship found between Sensitivity and levels of verbal comprehension.

This study is part of a larger project that investigated the effects of the Developmental Journal for babies and young children with visual impairment – DJVI (Dale et al., 2019). Parent-mediated early intervention was delivered by qualified teachers of VI across many services in England. Receiving the DJVI, in comparison with home-based intervention with 'other support', was associated with a clinically relevant longitudinal acceleration in expressive language (ELS – advancement of 11.72 DQ on average) from T1 to T3 and of a similar average increase level of nonverbal cognition (SMU) to this analysis (5.06 DQ), but not reaching 95% significance. However, no increase was found of verbal comprehension (RSVC) in those receiving the DJVI, in comparison with this analysis where the estimated increase in RSVC was significantly associated with maternal Sensitivity. This raises important questions for early intervention of why expressive language and verbal comprehension appear to be differentially facilitated, though the study design did not permit further consideration of this possible dissociation. As part of early intervention in the future, direct support to maternal sensitivity may potentially further enhance the effects of the DJVI, but more research is needed to investigate this. Methods of training or augmenting sensitivity in mothers may be relevant here; video-feedback attachment-based parenting (VIPP), adapted for young children with VI, is of interest, but a recent study with 1- to 5-year-olds showed inconclusive results (Platje et al., 2018).

Another issue of interest is that children who are progressing well in RSVC may be less at risk for emerging autism spectrum disorder (ASD), which the population with VI is at high risk for (Do et al, 2017), but this is not yet established. The research cohort is now being followed up at 4- to 7-year-olds in relation to investigation of ASD outcomes and future analysis will examine whether maternal sensitivity differences have any association with likelihood of ASD at late preschool/ early school age. This would support an infancy sensitivity-based intervention to be trialled to improve ASD outcomes in this population (see Green et al., 2017).

Strengths and limitations

This study has various strengths, including the representativeness of the national cohort of children with CDPVS disorders (Dale et al., 2017), precise measurement of vision level and narrow age range at baseline, strong inter-rater reliability and the linear mixed-effects modelling which takes into account explanatory (predictor) outcomes and individual developmental trajectories over time. The study has limitations which may affect its generalizability to the wider population of children with VI. It may be underpowered to show effects for SMU and ELS as there is high heterogeneity within the sample. The short duration of the video may not have provided a true representation of how the mother and child interact at home, and demand characteristics such as being videotaped in front of the assessors may have influenced their mother–infant interactions. Although mothers had diverse educational backgrounds, over half of the mothers had at least a university degree which may not have been totally representative of the general population. It is also not known how natural interactions with other caregivers, for example fathers, may influence the child's developmental progress. Previous literature on interactions with children with VI has focused on mother–infant pairs; therefore, there is no knowledge of differences between mother–child and father–child interactions. This is something that would need to be investigated in the future. The use of the RZS norms, which although derived from the only semi-

standardized developmental scales for children with VI, may have limitations; for example, they consist of broad age equivalents and the norms need to be updated. However, the study used a more standardized administration and used the 'sighted' norms and controlling for vision level using the PVI and SVI categorization (Dale et al., 2017). The proportion of children with PVI compared with SVI was much lower and in line with population expectations, but may have skewed some comparisons. The study is an observational study, and therefore, other potentially confounding factors which have not been accounted for may be influencing the associations. Future investigation controlling for maternal sensitivity through a randomized controlled trial with a parent-mediated sensitivity intervention may lend further light to the impact of the reported association with RSVC. Future research could consider the impact of father parenting style and also examine children with cerebral visual impairment to extend generalizability with the wider VI population.

Conclusions

This study is the first to demonstrate longitudinal associations between maternal sensitive responding to infant cues during play at 8–17 months, and advancement in DQ points in verbal comprehension in their infants with severe vision impairment 12 and 24 months later. The study has important implications for future habilitation as it suggests that maternal sensitivity is a potential facilitatory factor that could be incorporated in early intervention programmes such as the Developmental Journal for Visual Impairment – DJVI, which has already suggested improvements in other areas of development in children with congenital visual impairment (Dale et al., 2019).

Ethical approval

Parents gave informed consent, and the study was undertaken according to IRAS Ethics No. 10/H0713/46. Observational methods are reported according to STROBE checklist guidance.

Supporting information

Additional supporting information may be found online in the Supporting Information section at the end of the article:

Appendix S1. Supplemental information.
Table S1. Participant sample characteristics.

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Correspondence

Naomi J. Dale, Great Ormond Street Hospital for Children NHS Foundation Trust, London WC1N 3JH, UK; Email: n.dale@ucl.ac.uk

Key points

- Congenital visual impairment (VI) has been associated with delays in sensorimotor development and language from infancy; the impact of parent–infant interactions on early developmental progress has not been systematically investigated.
- Maternal maintaining of attention and sensitivity was examined during mother–infant play interactions at 8–17 months, and associations with longitudinal developmental trajectories were investigated.
- Maternal sensitivity was significantly associated with increases in verbal comprehension DQ from infancy to 3 years; Maintain did not show associations with developmental outcomes.
- Maternal sensitivity is a potential contributory factor to early learning, and further consideration should be given to how it could be enhanced to support infants with VI.
- This paper is relevant to clinicians working with infants with VI, parents and practitioners delivering early intervention.

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