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Aggressive paediatric camptodactyly: The evolution of a proposed treatment algorithm

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Summary *Introduction:* It is a long-established teaching to avoid operating on camptodactyly unless there is a failure of non-operative treatment, such as serial splinting and hand therapy, and there is an established proximal interphalangeal joint (PIPJ) contracture of 60°; a recent systematic review reflects this continuing approach, with some papers advocating intervention with a lesser degree of contracture.
Aim: To evaluate whether early flexor digitorum superficialis (FDS) release, followed by gentle passive manipulation (GPM), will correct severe 'congenital' camptodactyly, if undertaken at an earlier age than usual, thus avoiding the more aggressive surgical approach required in the established adolescent cases.
Method: The surgical technique and treatment algorithm are described. A multi-centre case series is presented; data analysis included patient demographics, syndromic association, side/digit affected, ages at onset, progression, referral and at surgery, operation details, pre- and post-operative contracture and range of motion.
Results: There were 12 patients (3 males, 9 females) who underwent 15 operations for 24 involved digits. Patients had surgery by 3 months (median) post-referral, and there was a significant improvement in median (range) PIPJ contracture (90°(30°-90°) vs. 0°(0°-45°); $p<0.001$) and range of motion (0°(0°-60°) vs. 90°(50°-95°); $p<0.001$), at a median post-operative follow-up of 2.5 years. According to the Siegert grade, 87.5% of digits had excellent/good post-operative outcomes and 12.5% had fair outcomes.

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Conclusion: This paper specifically addresses the problem of aggressive and progressive camptodactyly in the young child. By this, we mean patients who have failed non-operative treatment and have PIPJ contractures $\geq$60°, and those whose contractures have increased by 30° within 1 year. All cases responded to early FDS release and GPM, hence correcting the PIPJ contracture. However, cases with multiple digital involvement, whether syndromic or not, and failed previous surgery or the older child, required additional procedures to restore a dynamic dorsal apparatus and active extension.

Introduction

In many centres, the mainstay of camptodactyly treatment remains non-operative based on serial splintage and hand therapy; a strong working relationship between hand therapist and surgeon is essential for successful treatment.[1-3] It appears to be an accepted dogma that surgery should be reserved for failed cases of non-operative management, where a fixed flexion contracture of 60° has been reached to the proximal interphalangeal joint (PIPJ); although recent papers have advocated intervention with a lesser degree of contracture.[1,4] It is not surprising that surgery will perform poorly in a group of pre-selected failures of primary treatment. It has previously been demonstrated that once the PIPJ has achieved a 60° contracture, 80% of such cases will have central slip attenuation, thus complicating the problem by adding extensor incompetence.[5]

It has previously been demonstrated that virtually all structures that cross the PIPJ are implicated in camptodactyly.[6] However, it is important to differentiate the primary underlying causes from the secondary effects of delayed treatment. Primary causative factors include a short flexor digitorum superficialis (FDS), abnormal lumbrical origin and insertion, and shortness of the generic retinaculum cutis. Delayed treatment leads to the problems found in any long established PIPJ contracture, such as adhesions of the dorsal apparatus and lateral bands, central slip attenuation, volar plate contractures, and tightness of the accessory collateral ligaments as they pass over the proximal phalangeal condyles. In longstanding cases, Smith and Grobbelaar have demonstrated that with appropriate surgical technique, good to excellent results are achievable in 83% of patients according to the Siegert grading system; we propose that this unifying theory of camptodactyly and described surgical approach is still appropriate for established adolescent cases.[1,7]

One has to consider, however, whether it is appropriate to wait for fragile periarticular adhesions to become firmly fixed contractures prior to surgical intervention. We propose that early palmar surgical release of the FDS in young children, with gentle passive manipulation to mobilise periarticular adhesions, may avoid the establishment of firmly fixed contractures, and prevent secondary changes, which are more difficult to treat; this strategy was proposed to the authorship group, having first been undertaken by the senior author (PJS) in 2012.

Aim

The aim of this study is to evaluate the outcomes of early release of rapidly aggressive and progressive camptodactyly in children; this is defined by a 30° deterioration in flexion contracture within a 1-year period or less. We hypothesise that the surgery required to achieve release when intervening early will require a less aggressive approach in that it primarily will involve FDS release and gentle passive manipulation; good outcomes should therefore be achieved without a requirement for releasing the joint and associated structures as dense fibrous adhesions will not have developed by this time. The surgical technique and treatment algorithm are described.

Method

All patients were referred due to a failure of pre-existing non-operative management with serial splintage and hand therapy. A retrospective, multi-centre case series was undertaken; relevant institutional review board approval was granted. Data analysis included patient demographics, syndromic association, side and digit affected, ages at onset, progression, presentation and at surgery, operation details, pre- and post-operative contracture and active range of motion. An important distinction was made on clinical examination between flexion deformity and contracture (the former may be present in the absence of the latter). The wrist and metacarpophalangeal joints (MCPJs) are fully flexed and the PIPJ is observed; if the PIPJ is not able to be passively extended to neutral, then a contracture is present. Surgical release of the contracture was undertaken according to the described technique below, with the primary common step being FDS release and gentle passive manipulation. Some comparative analyses of previously published data by Smith and Grobbelaar were also undertaken for discussion purposes and are included within the discussion section of this paper accordingly.[1] Categorical data were analysed using the Chi-squared test. As continuous data were statistically skewed, median and range values were calculated; after confirming non-parametric distribution using the Shapiro-Wilk test, data were analysed using the Mann–Whitney U test.[8]

Surgical technique

A treatment algorithm is presented (Figure 1). All patients within this series met the criteria for surgical management, having already failed non-operative management, including

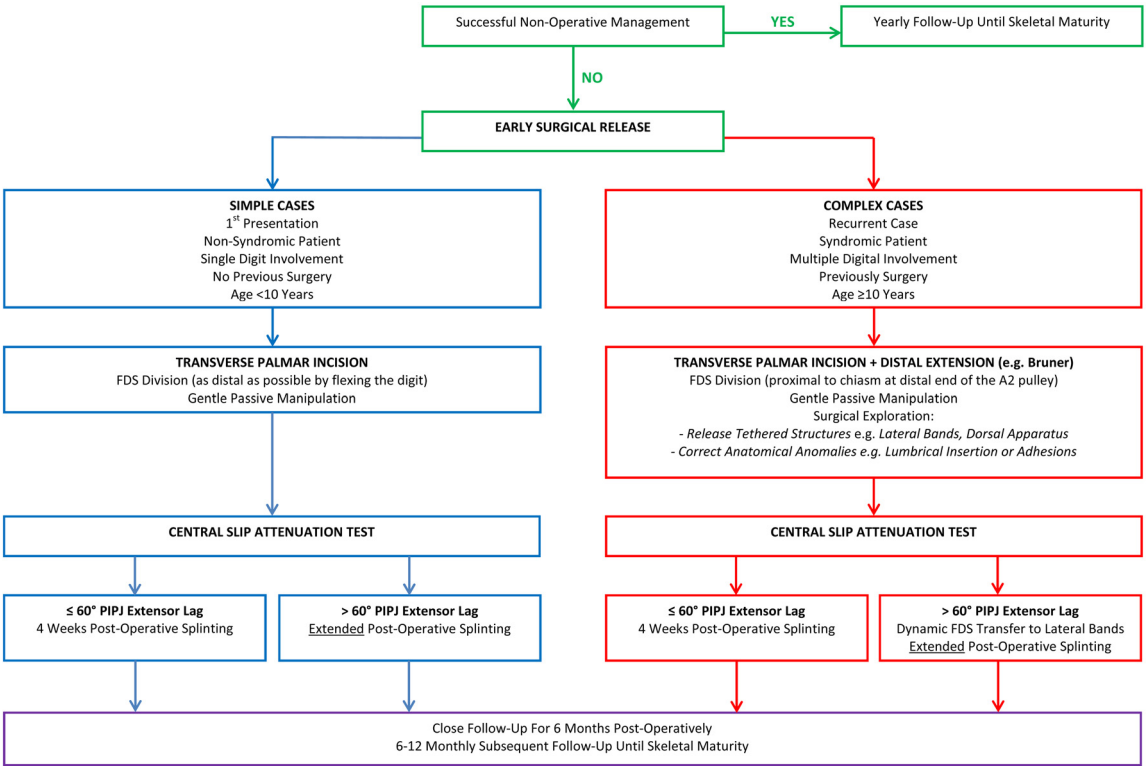

Figure 1 Treatment algorithm for aggressive paediatric camptodactyly surgery.

splintage; failed non-operative management was declared when there was a flexion contracture of 60° at the PIPJ or an increase in flexion contracture of 30° in 1 year or less. Most patients had such aggressive disease that by the time they came to surgery, they had 90° flexion contractures, even though the time between consultation and surgery was short.

Cases for early surgical release are initially considered as to whether they are simple or complex (Figure 1). After intra-operative re-evaluation of the flexion contracture (Figure 2), a transverse incision is made in the distal palmar crease for simple camptodactyly cases; the FDS is located and if intact proximally is released as distal as possible via the palmar incision by flexing the digit (Figure 3). In complex cases, the skin incision is extended distally in a Bruner fashion to release the FDS at the level of the chiasm; this is in order to allow for the release of any tethered structures such as the lateral bands or dorsal apparatus, and correction of anatomical anomalies such as those affecting the lumbrical insertions, if present. It also allows for the FDS to be divided at the distal end of the A2 pulley, proximal to the chiasm, to have enough length for tendon transfer into the radial lateral band, if required (Figure 4). As we advocate 'early' intervention, any fibrous adhesions are usually minor and respond to gentle passive manipulation intra-operatively; this is undertaken by exerting sustained gentle extension of the PIP and DIP joints while holding the MCP joint in flexion (Figure 5) to achieve further contracture release by breaking up residual adhesions (Figure 6). If left for a few years, the fibrous adhesions thicken and will require surgical release, as may the volar plate and other affected structures. The skin is sutured with 5-0 Vicryl Rapide (polyglactin 910).

Following full release of the camptodactyly contracture, it is important to assess central slip attenuation using the central slip tenodesis test, as previously described by the senior author; with the wrist and MCPJs fully flexed, observation of a passively correctable PIPJ extensor lag indicates central slip attenuation.[9] If the central slip attenuation is severe, as indicated by *a* >60° extensor lag, a requirement for an extended post-operative splinting period should be anticipated for simple cases. However, in complex cases, FDS transfer to the radial lateral band at the site of the lumbrical insertion and may be added to actively correct extension at the PIPJ; tenorrhaphy is achieved using a side to side and figure of eight 4-0/5-0 PDS (polydioxanone, Ethicon) suture technique (Figure 4).

While not routinely undertaken, Kirschner wire immobilisation across the PIPJ is recommended for 4 weeks in cases where post-operative splintage compliance is likely to be sub-optimal; this was the case in only 25% (3/12) of patients in this series. Mepitel, gauze, wool, plaster of Paris, and crepe are applied, fashioning a volar cast in the position of safety and ensuring full extension is maintained across the PIPJ for 4 weeks; dressings are reduced and changed at 2 weeks by the hand therapy team. If there is no evidence of central slip attenuation at 4 weeks, active flexion is encouraged from 4 weeks, with nighttime splintage utilised for 6 months post-operatively. If there is evidence of central slip attenuation at 4 weeks however, extended splintage is continued during the daytime with 5 min of active flexion allowed out of the splint every waking hour. This continues until the central slip has tightened back to normal and

Figure 2 Flexion contractures of the left little, ring, and middle fingers.

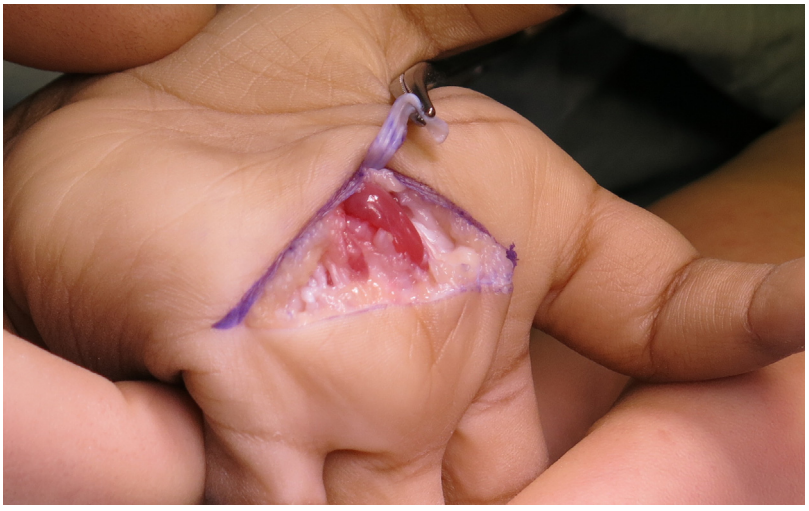

Figure 3 Transverse incision is made at the level of the distal palmar crease, and the FDS is released as distally as possible by flexing the digit; the effect of the release is illustrated here.

full active extension is possible. In general, it would not be uncommon for patients to be assessed on a 2 weekly basis post-operatively by both the hand therapist and hand surgeon in order to ensure progress. Clearly, there needs to be fairly intense attention to detail with regards to hand therapy during the first 3 months post-operatively; ideally, the more severe the camptodactyly is, the more frequent these sessions should be.

Results

There were 12 patients (3 males, 9 females) who underwent 15 operations for 24 involved digits. Three patients had camptodactyly associated with Jacobson's, Stuve-Wiedemann, and Tetrasomy 18p syndromes, respectively, and 3 cases were bilateral. Details of patient camptodactyly history, digits that were surgically released, operative techniques used, and pre- vs. post-operative contracture at follow-up are presented (Tables 1-4). Of note, the mainstay of surgery in 100% (24/24) of released digits in-

Table 1 Patient camptodactyly history. This indicates that median deterioration occurred within the first 3 years of life in the aggressive and progressive cases referred and included within the study cohort.

Camptodactyly history	Median (range)
Age at onset (years)	0 (0–10)
Age at progression (years)	2.5 (0.5–10)
Age at referral (years)	2.75 (2–12)
Age at surgery (years)	3 (1.5–12)

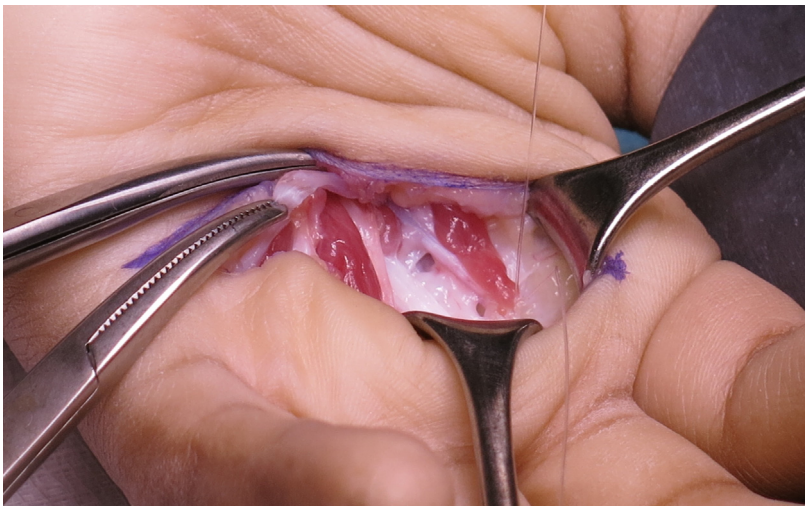

Figure 4 FDS is transferred to the radial lateral band of the lumbrical insertion, if required.

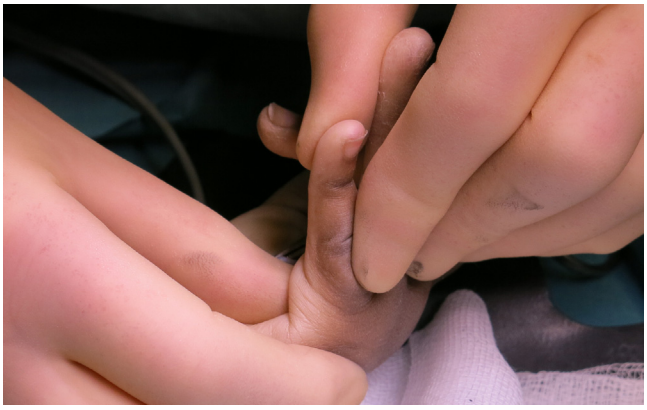

Figure 5 Gentle passive manipulation is applied.

volved FDS release and gentle passive manipulation, with 0% (0/24) of digits requiring formal joint contracture release; 50% (12/24) of digits had developed adhesions around the lateral bands or lumbricals which were therefore also released, and 33% (8/24) of digits with severe central slip attenuation required FDS to RLB transfers in order to improve cascade and passive tenodesis 'on-table', with the aim to ultimately improve active extension (Table 3). It is important to differentiate between the procedures required to release the joint contracture and those required to ensure that there was a dynamic dorsal extensor mecha-

Table 2 Number of digits surgically released.

Right hand digits		Left hand digits	
Middle	4	Middle	4
Ring	6	Ring	3
Little	4	Little	3
Sub-total	*14*	*Sub-total*	*10*
		Total digits released	24

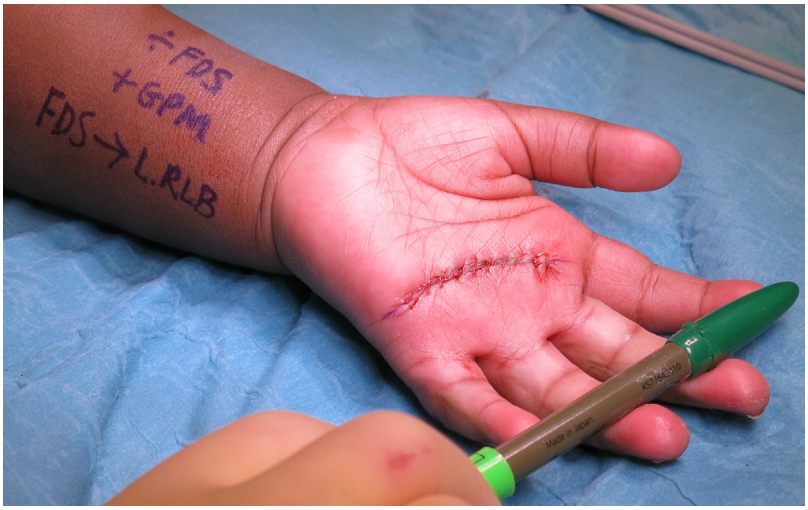

Figure 6 Final intra-operative result.

Table 3 Surgical techniques used. The mainstay of surgery in 100% (24/24) of released digits involved FDS release and gentle passive manipulation, with 0% (0/24) of digits requiring formal joint contracture release; 50% (12/24) of digits had developed adhesions around the lateral bands or lumbricals which were therefore also released, and 33% (8/24) of digits required FDS to RLB transfers in order to improve cascade and passive tenodesis 'on-table', with the aim to ultimately improve active extension. It is important to differentiate between the procedures required to release the joint contracture and those required to ensure that there was a dynamic dorsal extensor mechanism capable of producing extension at the PIPJ; the latter is achieved by releasing the component structures of the extensor apparatus and performing an FDS to RLB transfer when indicated. Local flaps or full-thickness skin grafts were required to address skin shortage post contracture release in 25% (6/24) of digits, with Kirschner wire stabilisation used only as a means of post-operative splintage in 42% (10/24) of released digits in cases where compliance was likely to be poor. FDS = flexor digitorum superficialis; GPM = gentle passive manipulation; LBs = both lateral bands; RLB = radial lateral band; ACL = accessory collateral ligament; FTSG = full-thickness skin graft; K-wire = Kirschner wire (inserted across proximal interphalangeal joint and removed at 4 weeks post-operatively).

Surgical techniques used		Number ofdigits
Contracture release	*Additional Procedures*, e.g., *active extension restoration, skin replacement, splinting*	
FDS Division, GPM	-	10
FDS Division, GPM	Freeing of LBs/Lumbrical, ACL Division	2
FDS Division, GPM	Freeing of LBs/Lumbrical, FDS→RLB Transfer, Local Flaps, FTSG	2
FDS Division, GPM	Local Flaps, FTSG, K-wire	2
FDS Division, GPM	Freeing of LBs/Lumbrical, Local Flaps, K-wire	2
FDS Division, GPM	Freeing of LBs/Lumbrical, FDS→RLB Transfer, K-wire	6

Table 4 Pre- vs. post-operative PIPJ contracture and range of motion arc at follow-up.

Patient presentation details	Median (range)	Median improvement (range)	p-value
Pre-operative contracture (°)	90 (30-90)	90 (20-95)	$p<0.001$
Post-operative contracture at follow-up (°)	0 (0-45)		
Pre-operative active range of motion arc (°)	0 (0-60)	90 (20-95)	$P<0.001$
Post-operative active range of motion arc (°)	90 (50-95)		

nism capable of producing extension at the PIPJ; the latter is achieved by releasing the component structures of the extensor apparatus and performing an FDS to RLB transfer when indicated. Local flaps or full-thickness skin grafts were required to address skin shortage post contracture release in 25% (6/24) of digits, with Kirschner wire stabilisation used only as a means of post-operative splintage in 42% (10/24) of released digits in cases where compliance was likely to be poor (Table 3). Of further relevance, patients had surgery by 3 months (median) post-referral as they were established cases of failed non-operative treatment, and there was a significant improvement in PIPJ contracture (90°(30°−90°) vs. 0°(0°−45°); $p<0.001$) and active range of motion arc (0°(0°−60°) vs. 90°(50°−95°); $p<0.001$), at a median (range) post-operative follow-up of 2.5(1-8) years (Tables 1 & 4). According to the Siegert grade, 87.5% (21/24) of digits had excellent or good post-operative outcomes, and 12.5% (3/24) had fair outcomes and no digits had a poor outcome (Figures 7 & 8); we acknowledge that the assessment of outcomes in children is difficult and have chosen the Seigert grade as it remains the commonly used camptodactyly-specific assessment tool in publications.[1,7,10-12]

Of the 18/24 (75%) digits that achieved full correction immediately post-release, 2 digits in 1 syndromic patient subsequently recurred to 45° and 1 digit in another syndromic patient subsequently recurred to 30°, both cases of which were associated with difficulties in post-operative compliance; a further 3 digits in 3 non-syndromic patients recurred to 5°, 5°, and 10° of contracture, respectively. Of the 6/24 digits that did not achieve full correction immediately post-release, 1 digit in 1 non-syndromic patient progressed from 20° to 40°, 2 digits in 2 patients remained the same at 20° and 28°, respectively, and 3 digits in 1 syndromic patient each subsequently corrected fully from 10°. Of the 2 patients who remained the same at 20° and 28°, respectively, the patient in whom 20° of on-table right ring finger correction was achieved, had previously undergone 2 operations at other UK centres for 4th web syndactyly and attempted camptodactyly release; the patient in whom 28° of on-table left middle finger correction was achieved, displayed vascular compromise despite full correction being possible, hence was splinted in this safe position post-operatively.

Discussion

Camptodactyly affects approximately 1% of the general population; although cases may occur sporadically, an autosomal dominant pattern of inheritance with variable penetrance and phenotypic expression may also be seen.[1,3,6,13-15] Parkes Weber initially proposed 2 forms of camptodactyly; congenital types were described as being present from early life and affecting males and females equally, with adolescent or acquired types gradually appearing in early teens and more commonly seen female patients.[16] In an analysis of 66 patients with 110 hands affected by camptodactyly, Flatt's group presented findings that were in keeping with

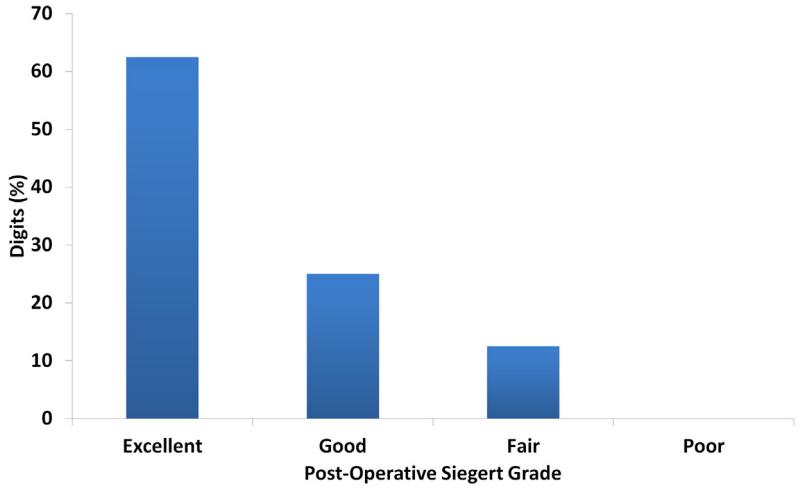

Figure 7 Percentage of digits classified according to the post-operative Siegert grade.[7] There were 15/24 (62.5%) excellent, 6/24 (25%) good, 3/24 (12.5%) fair, and 0/24 (0%) poor outcomes.

this hypothesis; age of onset within the first year occurred in 84% (the congenital group) of patients, 13% developed after 10 years (the adolescent or acquired group), and only 2 patients developed camptodactyly between 1 and 10 years of age.[17] Benson differentiated syndrome-associated camptodactyly, but in keeping with Barinka's view, we feel that it is likely that all presentations are exactly the same condition; severe (early) cases, however, presenting soon after birth and with rapid deterioration in the initial growth spurt, and milder (delayed) cases presenting in the adolescent growth spurt.[15,18]

Historically, general advice has been to avoid surgery, treat non-operatively with serial splintage and hand therapy, and to only operate in the presence of a fixed PIPJ contracture in excess of 60°; recent systematic review data reflects this continuing approach, with a trend towards a lower contracture threshold of >30°.[1,4] The exact definition and pathogenesis of camptodactyly and structural involvement remain a topic for debate; however, in the unifying theory of camptodactyly, where Smith and Grobbelaar reported that 83% of cases had excellent or good post-operative Siegert grades, virtually all structures crossing the PIPJ were demonstrated to be involved.[1,3,4,11,19,20] They therefore advocated using a combination of techniques, adopting some which had previously been established into their surgical approach; these included skin lengthening or replacement, release of the retinaculum cutis, FDS lengthening or division, release of the lateral bands from the proximal phalanx, repositioning of any abnormal lumbrical, possible division of the flexor tendon sheath, volar plate, and even the accessory collateral ligaments if after full release they appeared tight and 'flicked' over the condyles at the head of the proximal phalanx.[1,6,7,17,21-23] Central slip attenuation was addressed by appropriate post-operative splintage or K-wiring the PIPJ for 4 weeks.[1] Local flap cover could be taken from the side of the finger where there is always an excess of skin, using a transposition flap. This is a significant surgery and is not considered to be suitable for smaller fingers, rather it should be reserved only for adolescent hands with established contractures that are firmly fixed due to the development of dense fibrous adhesions.

The question, therefore, arises as to what the most appropriate course of action is for a young child with aggressive camptodactyly. Delaying surgical release until a 60° flexion contracture was observed and had become fixed as per accepted dogma may have contributed to more modest post-operative outcomes as reported in the literature; 80% of these patients will have had central slip attenuation.[5,10,24] The senior author, therefore, proposed a paradigm shift to the surgical group towards early release in all cases of rapid progression; early, meaning both at an early age and early in the progression of the deformity.

The data presented in this study reflect that patients presented with rapidly aggressive and progressive camptodactyly (age at progression = 2.5(0.5-10) years, age at referral = 2.75(2-12) years), with early surgery subsequently being performed by 3 months (median) post-referral (Table 1). The degree of camptodactyly severity is further reflected by the intentional inclusion of data from 8 digits released in 3 syndromic patients. When considering excellent and good post-operative Siegert grades together, the data presented in this study (87.5%) are similar to those previously published by Smith and Grobbelaar (83%); however, sub-analysis indicates a trend towards a higher proportion of digits with excellent post-operative outcomes compared to previous (62.5%(15/25) vs. 33.3%(6/18)); furthermore, the data presented in this study indicate a significantly higher pre-operative PIPJ contracture (90°(30°-90°) vs. 75°(35°-90°); $p<0.01$) and younger patient age at surgical release (3(1.5-12) years vs. 5(3-16) years; $p<0.05$) compared to those previously published by Smith and Grobbelaar (Tables 1 & 4, Figure 7).[1,7]

In view of the fact that all patients in this series had failed non-operative treatment, it would be difficult to justify randomising some into a treatment group in which they had already failed. It is important to appreciate that when a child puts their wrist into a functional position of neutral/extension, the presence of a 60°-90° PIPJ flexion deformity (even if the contracture is less), severely compro-

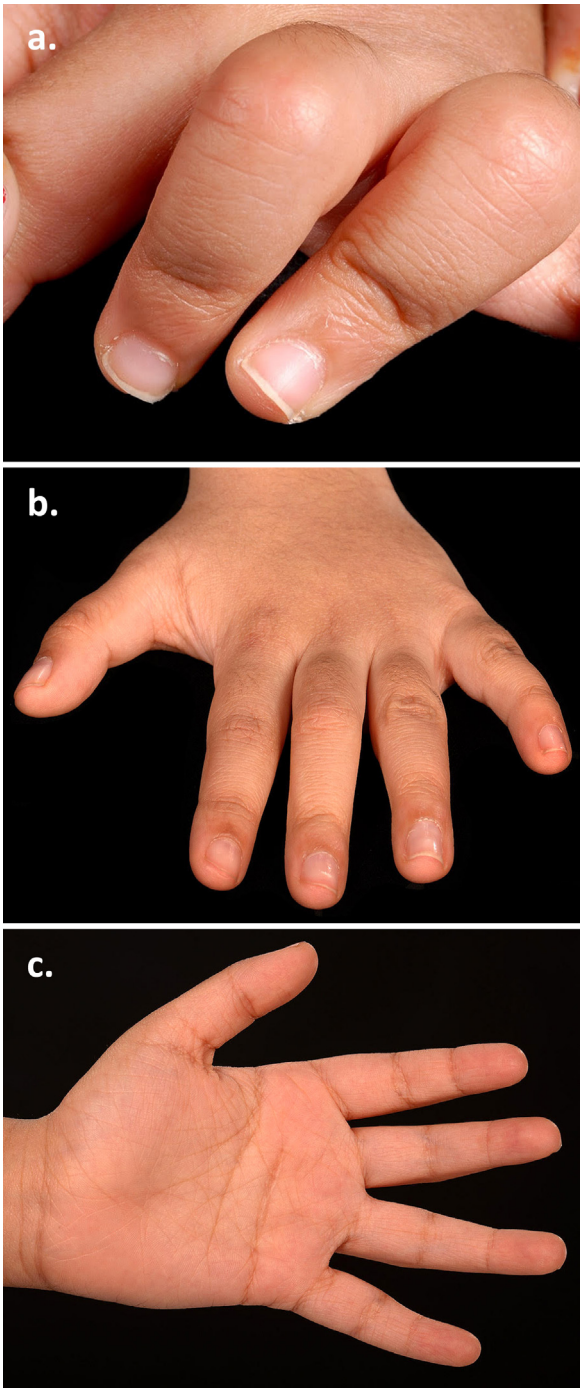

Figure 8 Pre- and post-operative series for left middle and ring finger camptodactyly. Pre-operative flexion contractures of the left middle and ringers (a) and post-operative images following release (b & c) are demonstrated.

mises function. This procedure addresses that issue. All the children had improved active range of motion post-operatively and none were made worse. The data presented in this multi-centre case series show uniformity of outcomes, with an improvement in PIPJ contracture (90°(30°-90°) vs. 0°(0°-45°); $p<0.001$) and active range of motion arc (0°(0°-60°) vs. 90°(50°-95°); $p<0.001$), and excellent or good post-operative Siegert grades in 87.5% of released digits, at 2.5(1-8) years follow-up.

Our aim was to ascertain whether a simpler operation involving early FDS release and GPM to break down any fragile adhesions could correct the deformity and avoid the need for more complicated surgery. Using this approach achieved a full correction at the time of surgery in the majority of cases (75%, 18/24). Gentle passive manipulation corrected the fragile adhesions in 100% (24/24) of cases and no joints required formal release of the volar plate (Table 3). Additional measures were required in some patients to achieve a dynamic dorsal apparatus and active extension. Crucial to the outcome is the role of post-operative splintage and hand therapy in which patient compliance is essential. Immobilisation in all patients was similar, but the method used differed as K-wires were used in patients judged likely to be non-compliant. Additional procedures may be required in older patients, those with multiple digit involvement (whether syndromic or not) as well as in patients where previous surgery has failed.

It may surprise some that the numbers in this series are small, but this reflects the current prevailing approach to camptodactyly (i.e., to avoid surgery); hence, referrals for consideration of surgery are limited. We recognise the need for a large series, followed until skeletal maturity, to establish whether early intervention may prevent later deterioration. Unfortunately, most surgeons are reluctant to operate on camptodactyly, and the described aggressive and progressive presentation is rarely encountered, achieving this will therefore take a long time. This should be regarded as a preliminary report which will achieve its aim if it challenges surgeons to question existing dogma. We know from Flatt's series that only 2/66 patients showed progression between 1 and 10 years following release.[17] It is therefore reasonable to assume that the children in our series will also remain contracture free until 10 years of age, with the associated improved function that this will deliver; however, it remains to be seen whether this early correction is maintained during the adolescent growth spurt. Our early results are promising, due to the functional improvement achieved post-operatively, and have encouraged us to operate earlier when deterioration progresses quickly and not wait to follow conventional advice on when to intervene. If maintained during the adolescent growth spurt, this will further strengthen the validity of the concept of early intervention in rapidly developing camptodactyly contractures in the young child, prior to the development of firmly fixed contractures by dense fibrous adhesions, and all the associated secondary changes that follow.

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Ethical approval

A retrospective, multi-centre case series was undertaken; clinical governance board approval was granted (Chelsea & Westminster: PCD977).

Declaration of Competing Interest

None.

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