Surgery and Conservative Management of Camptodactyly in Pediatric Patients: A Systematic Review

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Annie M. Q. Wang¹, Min Kim², Emily S. Ho³, and Kristen M. Davidge^{2,3}

Abstract

Camptodactyly is a pediatric hand condition, the treatment of which remains controversial. The authors' aim was to improve patient care through clarifying the definition of camptodactyly and indications for surgical and/or conservative management, summarizing outcomes, and defining risks. A systematic review was conducted of articles in all languages on outcomes following surgical and/or conservative management of idiopathic camptodactyly in children using MEDLINE (Medical Literature Analysis and Retrieval System Online), PubMed, EMBASE (Excerpta Medica database), AMED (Allied and Complementary Medicine), and CINAHL (Cumulative Index of Nursing and Allied Health Literature) (until January 2017). The primary outcome was posttreatment flexion contracture, and the secondary outcomes were indications for surgery, complications, and patient satisfaction. Database searching generated 16 final articles, with 7 case series and 9 retrospective cohort studies. There was a lack of consistency on the definition of camptodactyly and in outcome reporting. All 16 studies received a "Weak" global rating and demonstrated low-quality evidence, suggesting that treatment of camptodactyly with operative or nonoperative measures reduces the degree of flexion contracture in most patients (from pretreatment averages of 20°–85° to posttreatment averages of 5°–37°). There was general agreement that surgery should be reserved for contracture >30° or failure to respond to conservative management. Surgery generally led to more complications compared with conservative management. Only one study reported on functional limitations, and another reported on patient-reported outcomes. Current evidence of the effectiveness of camptodactyly treatment in addressing both joint-specific deformity and patient-perceived function and appearance is insufficient to guide patient care. Future research may consider the development of decision aids to guide patients and families through selecting management strategies and to promote shared decision making.

Keywords: camptodactyly, hand surgery, hand therapy, pediatric hand, systematic review

Introduction

Camptodactyly is a pediatric hand condition estimated to affect 1% of the population.¹ Although commonly described as a flexion contracture of the proximal interphalangeal (PIP) joint in one or multiple digits, the exact definition and pathogenesis of camptodactyly remain a topic of debate.^{2–4} Camptodactyly can appear in isolation or be associated with a congenital syndrome, and it most frequently affects the little finger, followed by the fourth, the second, and then the third digit of the hand.⁵ Two-thirds of all patients with camptodactyly have bilateral involvement.⁶

Camptodactyly can present in infancy, affecting males and females equally, or it can develop during a rapid period of growth in adolescence, during which time it occurs more frequently in females.⁵ There is significant variation in patient presentation, with the most common complaints

being dissatisfaction with appearance followed by functional limitation.^{5,7,8} While some individuals have minimal functional impact, others have difficulty playing the piano, typing, wearing gloves, writing, or performing manual labor.^{7,9,10} Some patients also report associated pain, although this is rare.⁷

Both surgical and conservative management have been used to treat camptodactyly. Surgical approaches focus on

¹The University of British Columbia, Vancouver, Canada

²University of Toronto, ON, Canada

³The Hospital for Sick Children, Toronto, ON, Canada

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Corresponding Author:

Kristen M. Davidge, The Hospital for Sick Children, 555 University Avenue, Toronto, ON, Canada M5G 1X8.
Email: kristen.davidge@sickkids.ca

correcting the underlying structural anomalies and are widely varied, including capsular release, flexor digitorum superficialis (FDS) lengthening, tendon transfer, lumbrical resection, osteotomy, and arthrodesis.¹¹ Conservative interventions are similarly diverse and include one or more of the following interventions: passive range of motion stretching exercises, serial casting, dynamic splinting, and static splinting.¹² The treatment for camptodactyly remains controversial. Some prefer conservative approaches,^{2,7,13} whereas others advocate for surgical intervention to prevent PIP joint flexion contracture progression and metacarpophalangeal (MP) joint hyperextension over time.⁵ The current trend in the literature generally favors conservative treatment for most patients, only recommending surgery for severe deformity, functional limitation, pain, or cases refractory to nonoperative management.^{7,14,15}

Ambiguity in the literature regarding the pathogenesis of camptodactyly, high variability in both conservative and surgical treatments, and lack of consistency in outcomes evaluation lead to significant challenges when guiding patients and their families through selecting management strategies. The aim of this systematic review was to improve patient care and decision making in camptodactyly management, through: (1) clarifying the definition of camptodactyly and indications for treatment; (2) summarizing treatment outcomes for patients with camptodactyly; and (3) defining risks of treatment, including complications, pain, and burden of care. We hypothesize that for pediatric patients with isolated idiopathic camptodactyly in single or multiple fingers, conservative management leads to favorable outcomes for mild-to-moderate contractures but has a high burden of treatment. Furthermore, we hypothesize that surgical intervention yields modest improvements in range of motion for severe flexion contractures and that recurrence is common. The resultant evidence from this comprehensive summary will be used to propose recommendations to guide camptodactyly management and to identify areas needed for future research.

Materials and Methods

We performed a systematic review following guidance from the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) statement¹⁶ and registered details of the protocol with International Prospective Register of Systematic Reviews (PROSPERO 2016: CRD42016050075). The literature regarding surgical and conservative management of camptodactyly in pediatric patients was searched using MEDLINE (Medical Literature Analysis and Retrieval System Online), PubMed, EMBASE (Excerpta Medica database), AMED (Allied and Complementary Medicine), and CINAHL (Cumulative Index of Nursing and Allied Health Literature). A comprehensive electronic search strategy was developed for MEDLINE,

and the search terms were adapted for use with other bibliographic databases in combination with database-specific filters for controlled trials. The search was performed from the inception of each database until January 2017. The detailed search strategies are outlined in Supplemental Material.

We included randomized control trials, prospective and retrospective cohort studies, case-control studies, and case series and case reports on outcomes following the surgical and/or conservative management of idiopathic camptodactyly in children aged 0 to 18 years, with idiopathic camptodactyly defined as isolated camptodactyly not associated with another known syndrome or multisystemic disorder. Exclusion criteria were studies with fewer than 4 patients (this threshold has been suggested to constitute the number of patients needed for a case series vs a case report¹⁷), patients with a known diagnosis of multisystem disorder or syndrome, and studies with level 5 evidence. The titles and abstracts of all identified articles were independently reviewed by 2 investigators (AW and MK) for eligibility. When necessary, full-text articles were obtained to allow the assessment of inclusion and exclusion criteria. The included articles were then fully reviewed by the 2 investigators and senior author (KD) to ensure the criteria were met and to resolve any study inclusion disagreements. The reference lists of included studies and key articles encountered throughout the search were also manually screened for additional records and added if eligible.

A standardized data extraction form was created by the first author (AW) following recommendations from the Cochrane Data Extraction and Assessment Template,¹⁸ which included the following variables: study characteristics (authors, year, journal, language, and level of evidence), definition of camptodactyly, study population (sample size, patient demographics, and comorbidities), treatment modality (surgery, stretching, or splinting), and treatment outcomes (passive and active range of motion, functional ability, burden of care, adherence, imaging findings, and complications). It was pilot tested with the first English study and the first non-English study and modified based on discrepancies. The English and non-English studies were all reviewed by those fluent in the respective languages.

The quality of the studies was independently evaluated by 2 reviewers (AW and MK) and scored in accordance with the criteria published by the National Collaborating Center for Methods and Tools.¹⁹ Case series articles were additionally appraised using the criteria proposed by Chan and Bhandari.²⁰ When performing quality assessments, the 2 investigators were not blinded to study authors, institutions, or journal.

Results

The literature review process is presented in a flow diagram in Figure 1. Removal of duplicates generated 805 articles, and then title and abstract review eliminated additional

Figure 1. Flow of literature search.

Note. MEDLINE = Medical Literature Analysis and Retrieval System Online; EMBASE = Excerpta Medica database; AMED = Allied and Complementary Medicine; CINAHL = Cumulative Index of Nursing and Allied Health Literature.

articles that did not assess treatment or failed to quantitatively evaluate treatment outcomes. Full article review was carried out on the remaining 57 articles, with inclusion and exclusion criteria applied. This resulted in 16 articles included in this systematic review, with 7 case series and 9 retrospective cohort studies.

Study and Patient Characteristics

Study and patient characteristics are presented in Table 1, and clinical outcomes are summarized in Table 2. The most common definition of camptodactyly was a nontraumatic flexion deformity of the PIP joint, but classification schemes based on pathogenesis and timing of onset were varied (Table 2). Fourteen articles (88%) were written in English, with one in French and one in German. The median year of publication was 1993 (range, 1968-2015). Three studies included 7 adult patients, and one study included 22 patients who were not treated; these patients were excluded. Three

articles²¹⁻²³ likely studied overlapping patient populations because they were authored by the same research group with overlapping patient recruitment periods. We included all 3 articles in the qualitative analysis as each had a different aim, so their conclusions still uniquely contributed to the overall understanding of camptodactyly; however, we only included quantitative results from the most recent article of the three²³ when aggregating outcomes data.

This resulted in a total of 354 patients from 7 countries. Patients ranged in age from 0.1 to 18 years, with 53% of patients being females. The little finger was involved 67% of the time, and bilateral involvement was observed in 86% of patients. Follow-up was reported in 14 of 16 studies and ranged from 0.2 to 20.0 years. Study results are synthesized qualitatively, as the highly varied study outcomes did not allow for statistical assessments. The treatments performed (including both surgical and conservative approaches) were also highly heterogeneous, making direct comparisons difficult.

Table 1. Study and Patient Characteristics.

Author	Year	Level of evidence	Language	Study design	No. of digits	No. of patients	Gender (M,F)	Mean age (range), y
Smith and Kaplan ²⁴	1968	4	English	CS	14	9	6,3	NR (4.0-19.0)
Engber and Flatt ¹⁴	1977	3	English	RC	56	32	13,19	Improved: 5.0 (NR) Same/Worse: 10.2
Miura ²⁵	1983	3	English	RC	—	63	29,34	NR (1.0-18.0)
Hori et al ⁹	1987	4	English	CS	34	23	9,14	NR (0.3-10.0) ^a
Koman et al ²⁶	1990	4	English	CS	27	8	4,4	NR (1.0-8.5)
Siegert et al ⁷	1990	3	English	RC	79	31	20,37 ^b	Conservative: 14.1 (2.0-18.0) ^c Surgical: 13.3 (1.0-18.0) ^d
McFarlane et al ²⁷	1992	3	English	RC	—	53	22,31	15 (NR)
Ogino and Kato ²⁸	1992	4	English	CS	6	4	0,6 ^b	12.0 (9.0-14.0) ^e
Benson et al ¹³	1994	3	English	RC	59	22	—	4.1 (0.1-17.0)
Glicenstein et al ²⁹	1995	3	French	RC	95	48	27,32 ^b	NR (0-15.0)
Smith et al ³⁰	1998	4	English	CS	18	16	10,6	8.0 (3.0-16.0)
Hasselbacher et al ³¹	2002	3	German	RC	16	14	6,6	11.0 (NR)
Rhee et al ³	2010	3	English	RC	61	22	13,9	1.0 (0.3-3.0)
Hamilton and Netscher ²¹	2015	4	English	CS	18	12	8,5 ^b	8.0 (0.8-15.0)
Netscher et al ²²	2015	4	English	CS	18	9	6,3	11 (4.0-15.0)
Netscher et al ²³	2015	3	English	RC	18	12	8,5 ^b	8 (0.8-15.0)

Note. CS = case report; NR = not reported; RC = retrospective cohort.

^aDoes not include one 42-year-old adult patient.

^bIncludes patients who were not treated.

^cDoes not include one 30-year-old adult patient.

^dDoes not include 3 adult patients, aged 19 to 36.

^eDoes not include 2 adult patients, aged 22 and 41.

Study Quality

Study quality is presented in Tables 3 and 4. All 16 studies (7 case series and 9 retrospective cohort studies) received a “Weak” global rating as per the Quality Assessment Tool for Quantitative Studies,¹⁹ with selection bias, confounders, blinding, and data collection being the most common sources of bias identified (Table 3). The 7 case series were further assessed using the guidelines proposed by Chan and Bhandari.²⁰ Overall, there was a high likelihood of selection bias across all included case series, as consecutive patient enrollment, exclusion criteria, and total duration spent for participant recruitment were not met or explicitly stated for most studies.

Indications for Surgery

Of the 12 studies that assessed surgical outcomes, 9 outlined the indications for surgery (Table 2). Four studies utilized poor response to conservative management, whereas 3 studies defined flexion contracture of greater than 30° as indication for surgery. The remaining 2 studies stated severe contraction as an indication for surgery but did specify any threshold angles or values. One study also used PIP joint skeletal changes noted on radiograph as a surgical indication (the exact radiographic change was not described).²³

Posttreatment Flexion Contracture

A total of 161 patients (46%) in 8 studies underwent conservative treatment only, whereas 187 patients (54%) in 14 studies had at least one surgical operation, with at least 178 of these patients (95%) receiving a trial of nonsurgical treatment before surgery (Table 2). Of these 187 surgical patients, 70 (37%) were explicitly reported to have failed prior conservative management, although this number may be higher as many studies were unclear about the patients' indication for surgery.^{14,26,31}

Overall, both conservative and surgical approaches demonstrated a general tendency toward reduction in the flexion contracture of the affected joint (Table 2). All studies demonstrated a reduction in flexion contracture in most patients, with pretreatment flexion contracture averages ranging from 20° to 85° and posttreatment averages ranging 5° to 37°. Only 7 studies reported both pretreatment and posttreatment values for PIP joint flexion contracture for all patients. Of these, only Rhee et al³ statistically analyzed the impact of treatment on change in flexion deformity. They found that passive stretch exercises improved mean flexion contracture from 20° to 1° ($P < .001$) in the mild group ($<30^\circ$ initial flexion contracture), from 39° to 12° ($P < .001$) in the moderate group (30°-60° initial flexion contracture), and from 75° to 28° ($P < .001$) in the severe group ($>60^\circ$ initial

Table 2. Clinical Outcomes.

Author	Definition of camptodactyly	Conservative treatment				Surgical treatment						
		Total No. of patients	No. of patients before treatment	Flexion contracture before treatment	Flexion contracture after treatment	Mean follow-up (range), y	No. of patients	No. of patients who failed CT	Indications for surgery	Flexion contracture before treatment	Flexion contracture after treatment	Mean follow-up (range), y
Engber and Flatt ¹⁴	Slowly progressing painless deformity of the little finger beginning in childhood, lacking associated nodular swelling near the joints/tendons	32	14	NR	n = 6 improved, n = 8 same/worse	3.7 (0.7-1.4)	18	18	NR	57°	n = 11 improved, n = 7 same or worse	3.4 (0.2-14.0)
Miura ²⁵	Nontraumatic flexion deformity of the PIPJ of one or several fingers	63	44	(20°-80°)	(0°-30°)	NR	19	19	Poor response to >6 mo of CT before puberty	(40°-85°)	(0°-60°)	NR
Koman et al ²⁶	Flexion contracture of the PIPJ without limitation of flexion, articular swelling, or pain. MCPJ may show a dynamic hyperextension deformity. Palmar fascia is never thickened	8	2	NR	n = 1 improved	(1.0-4.5)	6	6	NR	85.4° (70°-90°)	8° (0°-20°); n = 4 improved	(1.0-4.5)
Siebert et al ⁷	—	31	13	37° (5°-85°)	15° (0°-70°)	6.5 (1.2-13.7)	18	NR	Severe contracture	51° (15°-95°)	37° (0°-95°)	6.8 (0.8-20.0)
Benson et al ¹³	Severe flexion contractures of the PIPJ present at birth	22	18	19° (5°-55°)	5° (0°-60°)	2.8 (0.6-9.4)	4	4	Poor response to CT or patient preference	51° (20°-90°)	21° (0°-65°)	2.8 (0.6-9.4)
Glicenstein et al ²⁹	Nontraumatic flexion contracture of the PIPJ, usually of the little finger. Involvement of DIPJ or MCPJ suggests traumatic cause and not camptodactyly	48	25	NR	n = 19 improved	NR	23	NR	Flexion contracture >30°	NR	n = 23 improved	NR
Smith and Kaplan ²⁴	Congenital intrinsic minus deformity of the small finger and occasionally the ring and middle fingers	9	0	—	—	—	9	NR	Severe contraction	(15°-90°)	n = 8 improved	NR (1.0-4.0)
McFarlane et al ²⁷	Flexion deformity of the PIPJ, often increasing during the growth spurt	53	0	—	—	—	53	NR	Flexion contracture >30°	49°	n = 44 improved	NR (1.0-NR)
Ogino and Kato ²⁸	Nontraumatic flexion deformity of the PIPJ, usually affecting one or both small fingers	4	0	—	—	—	4	4	Poor response to CT and/or patient preference	58° (30°-85°)	16° (0°-40°)	2.3 (1.3-3.3)
Smith and Grobbelaar ³⁰	Trouble of flexor-extensor balance at the level of the PIPJ	16	0	—	—	—	16	16	6 months of CT failed to stop contracture progression	69° (8°-90°)	11° (0°-50°)	2.8 (0.7-9.0)
Hasselbacher et al ³¹	Irreducible flexion contracture affecting the PIPJ, presents in early childhood or in adolescence, and may be static or progressive	14	0	—	—	—	14	NR	NR	58°	34°	NR (1.2-8.5)
Hamilton and Netscher ²¹ and Netscher et al ^{22,23}	—	12	0	—	—	—	12	12	Flexion contracture >30°-40° or when skeletal changes are seen on radiographs	57° (35°-75°)	3° (0°-25°)	0.9 (0.3-2.7)
Hori et al ⁹	Fixed flexion contracture of the PIPJ of at least 10°	23	23	40°	10° (1°-28°)	3.8 (0.8-8.8)	0	—	—	—	—	—
Rhee et al ³	Persistent flexion contracture of interphalangeal joints of the hand	22	22	(20°-75°)	(1°-28°)	2.2 (1.0-3.9)	0	—	—	—	—	—
	Nontraumatic flexion contracture of digit											

Note. CT = conservative treatment; NR = not reported; PIPJ = proximal interphalangeal joint; DIPJ = distal interphalangeal joint; MCPJ = metacarpophalangeal joint.

Table 3. Quality of All Included Studies.

Author	Study design	Selective bias	Study design	Confounders	Blinding	Data collection	Withdrawals and dropouts	Global rating
Smith and Kaplan ²⁴	Case series	W	W	W	M	W	—	W
Engber and Flatt ¹⁴	Retrospective cohort	W	M	W	W	W	W	W
Miura ²⁵	Retrospective cohort	W	M	W	W	W	W	W
Hori et al ⁹	Case Series	W	W	W	W	W	—	W
Koman et al ²⁶	Case series	W	W	S	W	W	—	W
Siebert et al ⁷	Retrospective cohort	W	M	W	W	W	S	W
McFarlane et al ²⁷	Retrospective cohort	M	M	W	W	W	M	W
Ogino and Kato ²⁸	Case series	W	W	W	M	W	—	W
Benson et al ¹³	Retrospective cohort	W	M	W	W	W	S	W
Glicenstein et al ²⁹	Retrospective cohort	W	M	W	W	W	S	W
Smith and Grobbelaar ³⁰	Case series	W	W	W	W	W	—	W
Hasselbacher et al ³¹	Retrospective cohort	W	M	W	W	W	S	W
Rhee et al ³	Retrospective cohort	W	M	S	W	S	W	W
Hamilton and Netscher ²¹	Case series	W	W	W	M	W	—	W
Netscher et al ²²	Case series	W	M	W	W	W	—	W
Netscher et al ²³	Retrospective cohort	W	M	W	W	W	S	W

Note. W = weak; M = moderate; S = strong.

Table 4. Quality of Included Case Series.

Author	Clear objectives stated prior to study	Well-defined study protocol	Consecutive participant enrollment	Inclusion criteria explicit	Exclusion criteria explicit	Total duration spent recruiting participants	Outcome measures clinically relevant?	Any use of health-related quality of life measures	Follow-up rate	Reasons for loss of follow-up stated?
Smith and Kaplan ²⁴	No	No	Unclear	No	No	Unclear	Yes	No	Unclear	NR
Hori et al ⁹	Yes	No	Unclear	Yes	No	Unclear	Yes	No	Unclear	NR
Koman et al ²⁶	Yes	Yes	Unclear	Unclear	No	Unclear	Yes	No	100%	No loss
Ogino and Kato ²⁸	Yes	Yes	Unclear	Yes	No	Unclear	Unclear	No	NR	NR
Smith and Grobbelaar ³⁰	Yes	Yes	Unclear	Yes	No	Unclear	Unclear	No	Unclear	NR
Hamilton and Netscher ²¹	Yes	Yes	Yes	Yes	Yes	Unclear	Yes	No	100%	NR
Netscher et al ²²	Yes	Yes	Yes	Yes	Yes	Unclear	Yes	No	No	NR

Note. NR = not reported.

flexion contracture).³ Given inconsistent reporting of results, quantitative synthesis of the studies could not be performed. In addition, 3 of the 14 studies did not report on pretreatment flexion contracture, demonstrating further deficiency of the literature.^{14,26,29}

Posttreatment Complications

Complications of conservative and/or surgical management are reported in 10 of 16 studies ($n = 216$ patients), with length of follow-up ranging from 0.2 to 20.0 years (Table 5). Surgical treatment generally led to more complications compared with conservative management and included the need for further surgery (14/130, 11%), loss of flexion (10/130, 8%), ankylosis (6/130, 5%), and

worsened contracture after treatment (2/130, 2%). Complications reported for conservative management included contracture recurrence (4/86, 5%), loss of flexion (2/86, 2%), and worsened contracture after treatment (4/86, 5%). Contracture recurrence was observed in the 2 studies with the longest duration of follow-up: Hori et al,⁹ with a follow-up duration of 3.8 (0.8-8.8) years, described 1 patient (out of 23) developing 35° of contracture recurrence after a 2-year interval without splinting; Siebert et al,⁷ with a follow-up duration of 6.5 (1.2-13.7) years, noted that 3 patients (out of 13 who underwent conservative management) developed contracture recurrence. Studies with higher reported incidences of complications tended to be those with a longer duration of follow-up.

Table 5. Complications of Conservative and Surgical Treatments.

Author	Conservative treatment			Surgical treatment			
	Total No. of patients	No. of patients	Mean follow-up (range), y	Complications	Surgical approach	Complications	Mean follow-up (range), y
Engber and Flatt ¹⁴	32	14	3.7 (0.7-14)	None	Release of skin and soft tissue ± release of PIP; release of FDS; PIP bony decompression; osteotomy; arthrodesis	n = 2: ankylosis	3.4 (0.2-14.0)
	63	44	NR	NR	Fibrous band release; soft tissue release; skin graft; ± FDS division; K-wire joint fixation	NR	NR
	8	2	(1.0-4.5)	NR	FDS lengthening/transfer ± release of palmar plate/check rein ligaments; extensor mechanism exploration; release of transverse retinacular ligament	n = 2: required further surgery	NR (1.0-4.5)
Koman et al ²⁶							
Siegert et al ⁷	31	13	6.5 (1.2-13.7)	n = 2: loss of flexion n = 3: contracture recurrence	Z-plasty/Bruner incision; FDS release ± palmar capsulotomy; collateral ligament release; PIP pinned in extension; skin grafts	n = 9: loss of flexion n = 4: ankylosis n = 5: skin slough n = 1: persistent paresthesia n = 8: required further surgery	6.8 (0.8-20.0)
Benson et al ¹³	22	18	2.8 (0.6-9.4)	NR	NR	NR	2.8 (0.6-9.4)
Glicenstein et al ²⁹	48	25	NR	n = 4: worsened contracture	Total anterior tenoarthrolysis with flexor apparatus recession; palmar skin rotation flap	NR	NR
Smith Grobbelaar ²⁸	9	0	—	—	Z-plasty ± tenotomy of FDS; anterior capsulorhaphy with K-wire fixation	n = 1: loss of flexion	NR (1.0-4.0)
McFarlane et al ²⁷	53	0	—	—	Z-plasty; dissect FDS V from FDS IV from vincula longa until carpal tunnel ± FDS V or IV passed dorsal to transverse metacarpal ligament and then sutured; Zancolli lasso procedure to the A2 pulley; soft tissue release at PIP	n = 4: required further surgery	NR (1.0-NR)
Ogino and Kato ²⁸	4	0	—	—	Release of subcutaneous ligaments; incision of flexor tendon sheath; release of check rein ligament and space between volar plate and proximal phalanx; K-wire; transverse palmar skin; skin graft	NR	2.3 (1.3-3.3)
Smith and Grobbelaar ³⁰	16	0	—	—	Z-plasty; release of retinaculum cutis, pretendinous bands, and Grayson ligaments; release of lateral bands and interosseous muscles	NR	2.8 (0.7-9.0)
					± Lengthening and transposition of FDS insertions at the chiasma level; release of the FDS; release of flexor tendon sheath/volar plate/accessory collateral ligaments		
Hasselbacher et al ³¹	14	0	—	—	Palmar arthrolysis of PIP	n = 2: worsened contracture	(1.2-8.5)
Hamilton and Netscher ²¹	12	0	—	—	Z-plasty; skin graft; release of underlying fascia and subcutaneous tissues; transverse FDS tenotomy at level of the Camper chiasm ± sliding volar plate release; FDS tendon transfer; Fowler distal extensor tenotomy	None	0.9 (0.3-2.7)
Hori et al ⁹	23	23	3.8 (0.8-8.8)	n = 1: contracture recurrence			—
Rhee et al ³	22	22	2.2 (1.0-3.9)	NR			—

Note. PIP = proximal interphalangeal joint; FDS = flexor digitorum superficialis; K-wire = Kirschner wire; NR = not reported.

Other Outcomes

Koman et al was the only study that reported on patient function, noting that in their case series of 8 camptodactyly patients, 4 of the 6 patients who had operative interventions obtained “functional grasp/release” (pretreatment function was not discussed).⁷ Along the same lines, only one study by Ogino and Kato included data on patient-reported outcomes, whereby patient satisfaction was evaluated using a simple yes/no question. In this study, 5 out of 6 patients expressed satisfaction with the results of surgery.²⁸ Finally, no studies explicitly reported on burden of care, adherence to conservative management, and pain or costs associated with treatment.

Discussion

Camptodactyly is a pediatric hand condition that can have both functional and aesthetic implications. Indeed, the average PIP joint flexion deformity in patients presenting for treatment in these studies ranged from 20° to 85°. Overall, the results of this systematic review suggest that treatment of camptodactyly with operative or nonoperative measures reduces the degree of flexion contracture in most patients.

This finding corroborates that of Lethbridge and Wolin¹² in their review of camptodactyly outcomes following conservative treatment protocols. Yet, studies of the treatment of camptodactyly were limited in number and quality. Only 16 studies met our inclusion criteria, and all were case series or retrospective cohort studies that received a “Weak” global rating and demonstrated low-quality evidence. Consistent measurement and reporting of the key treatment outcomes—PIP joint flexion deformity and range of motion—were found to be lacking in the current literature. Indeed, most studies (56.3%) failed to report both pretreatment and posttreatment values for PIP joint flexion deformity or range of motion, and only one study performed a statistical analysis of the change in flexion deformity following treatment.

Importantly, the broader impact of the treatment of camptodactyly on the patient has not been adequately assessed to date. No study evaluated aesthetic outcomes following treatment, despite the fact that dissatisfaction with the aesthetic appearance of the hand is the most common reason for patients with camptodactyly to present to a health care provider.^{5,7,8} Other relevant outcomes to this population, such as function and satisfaction with care, were each evaluated in a single study, but utilization of validated outcome measures was lacking. In summary, evidence supporting the effectiveness of available treatments in addressing both the joint-specific deformity and patient-perceived function and appearance is insufficient to guide patient care.

Compounding the inconsistency in outcome reporting in this review was the heterogeneity in treatment protocols both among and within studies. Surgical and nonsurgical patients were grouped collectively, rendering it difficult to interpret the effectiveness of each individual intervention. Conservative treatment approaches ranged from stretching and range of motion exercises to progressive or dynamic splinting and serial casting. Surgical treatment approaches ranged from skin and soft tissue release to FDS tendon lengthening, release or transfer, ligament release, osteotomy, and arthrodesis. This treatment heterogeneity reflects that “camptodactyly” is a catch-all term for a spectrum of congenital anomalies resulting in PIP joint flexion contracture. Classification schemes based on anatomy and clinical examination, such as that proposed by Foucher et al,² may be useful in framing future studies to better clarify the relative effectiveness of different treatment approaches.

There appears to be a general agreement that surgery should be considered for severe PIP joint flexion contracture >30° or failure to respond to conservative management. However, the factors leading to failure of conservative treatment have not been explored. Such factors include patient costs, burden of care, and patient adherence, which are important elements to developing a patient-centered treatment decision-making paradigm. As might be anticipated, surgical approaches were generally associated with more complications than conservative approaches, but overall complication rates in this review were low (2%-11%). It would be useful for future studies to better elaborate on the nature and duration of postoperative therapy, as well as the long-term risk of recurrence or worsening of the PIP joint flexion deformity following surgical or nonsurgical treatment, particularly during periods of growth.

The strength of this systematic review is limited by the strength of the included studies, and we were not able to quantitatively summarize data or perform meta-analyses on treatment effects as desired owing to small sample sizes, weak study designs, treatment heterogeneity, and inconsistency in measurement and reporting of outcomes. Importantly, however, this review highlights the paucity of good-quality evidence guiding our current management of pediatric patients with camptodactyly. The knowledge gaps identified in this review can serve as a platform for the design of future studies in this population to improve patient-centered and evidence-based care in camptodactyly. Specifically, future research may consider the development of decision aids to guide patients and families through selecting management strategies and to promote shared decision making.

Ethical Approval

This study was approved by our institutional review board.

Statement of Human and Animal Rights

This article does not contain any studies with human or animal subjects.

Statement of Informed Consent

No patients were recruited for the study and therefore informed consent was not required.

Declaration of Conflicting Interests

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