

Case Report

Camptodactyly correction: A report of two cases

Surya Rao Rao Venkata Mahipathy*, Alagar Raja Durairaj, Narayanamurthy Sundaramurthy, Anand Prasath Jayachandiran, Suresh Rajendran

Department of Plastic and Reconstructive Surgery, Saveetha Medical College & Hospital, Thandalam, Kanchipuram Dist., Tamil Nadu, India

Received: 24 September 2021

Revised: 18 October 2021

Accepted: 20 October 2021

***Correspondence:**

Dr. Surya Rao Rao Venkata Mahipathy,
E-mail: surya_3@hotmail.com

Copyright: © the author(s), publisher and licensee Medip Academy. This is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

ABSTRACT

Camptodactyly is a condition where there is a permanent flexion contracture at the proximal interphalangeal joint mostly involving the little finger. This condition has a varied presentation and hence treatment is controversial, whether conservative management or surgical correction. Here, we present two cases of camptodactyly that were managed with surgery.

Keywords: Rare, Little finger, Flexion contracture, Non-operative, Surgery

INTRODUCTION

Camptodactyly is an uncommon hand anomaly involving varying degrees of congenital or acquired flexion contracture of the fingers at the proximal interphalangeal (PIP) joint, which can be unilateral or bilateral.^{1,2} This condition affects about 1-2% of the general population and most commonly involves the little finger.³ Many etiologies have been attributed which include abnormal lumbricals, short flexor digitorum superficialis (FDS), skin shortening, tight fascial bands, deficient extensor central slip and changes in the distal interphalangeal joint or metacarpophalangeal joint.⁴

The onset of this condition could be early or late, and it has been proven to show an autosomal dominant pattern of inheritance.⁵ Camptodactyly usually does not cause any functional impairment, but patients come for cosmetic corrections.

Many treatment modalities have been proposed from splinting or stretching exercises, release of tendons, fascial bands, transfer of muscles and tenotomy.⁵⁻⁷

CASE REPORT

A 17-year-old male presented to us with a deformity of the left mid finger since birth. The deformity was initially mild but worsened as the child grew. Now, he is having difficulty in grasping objects in the left hand. He was born to parents of a non-consanguineous marriage born by a full term normal vaginal delivery.

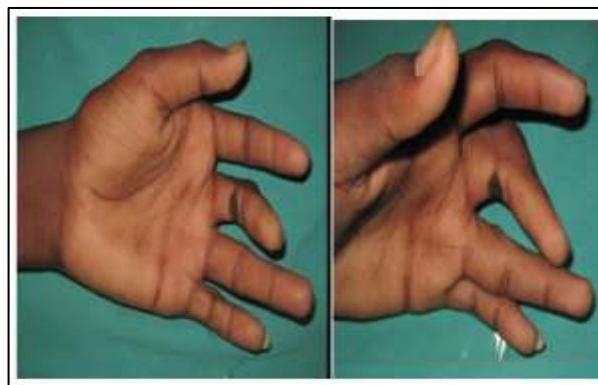


Figure 1: Camptodactyly of the left middle finger.



Figure 2: Contracted FDS tendon.



Figure 3: Defect after contracture release.



Figure 4: After cross finger flap cover.

The antenatal period was uneventful. On examination, there was a flexion deformity of the left middle finger at the PIP joint with inability to fully extend the joint passively. (Figure 1) X-ray was unremarkable with a narrowing of the PIP joint space and periarticular changes. A diagnosis of camptodactyly was made and we proceeded for exploration. Under axillary block and tourniquet control, an incision was made at the PIP joint level and slowly dissected. The skin and subcutaneous tissue were contracted with the FDS as a contracted band. (Figure 2) The FDP was intact and the neurovascular bundle was shifted anteriorly. The involved skin, soft tissue and the

FDS contracture were released and the position maintained with 1.25 mm K-wire. (Figure 3)



Figure 5: Late post-operative picture showing good result.



Figure 6: Camptodactyly of left little finger.



Figure 7: After release and cross finger flap cover.

The resulting raw area was covered with a cross finger flap from the ring finger. (Figure 4) Flap inset given with 4-0 nylon sutures and the secondary defect was resurfaced with a SSG. Post-operative was uneventful and the sutures and the K-wire were removed two weeks later. (Figure 5)

13 year old male presented to us with camptodactyly of the left little finger since birth. (Figure 6) He too was explored under anaesthesia. The contracture was gradually released and the position was stabilized with 1.25mm K wire. Post release, the defect of size 1×0.5 cm covered with cross finger flap from ring finger. (Figure 7) Post-operative was uneventful and the surgical outcome was satisfactory. (Figure 8)



Figure 8: Late post-operative picture showing good result.

DISCUSSION

Camptodactyly is a condition with progressive flexion contracture of the PIP joint unrelated to trauma, systemic disease, or neurologic abnormality.⁸⁻¹⁰ This condition, first described by Tamplin in 1846, may be present at birth (congenital type) or acquired later in childhood (acquired or adolescent type).^{3,8-10} It can be simple or complex when it is associated with other deformities. Benson et al. stated that while patients who had an early presentation have an equal sex distribution whereas late-onset patients are mostly females.¹¹ The little finger is the first to be affected and the incidence of isolated, atraumatic camptodactyly affects approximately 1-2% of the general population.⁸⁻¹⁰ Management is either conservative or surgical based on the clinical and radiological findings defining the extent of the deformity and joint flexibility.¹² This condition is usually not diagnosed until late in childhood or early teens, because of progression which is concurrent with normal childhood and adolescent growth spurts.¹³ Pain or swelling can occur but cosmetic appearance and functional deficits are most often the reasons for seeking medical attention.^{10,13} While the genetic occurrence of camptodactyly is generally sporadic, studies indicate camptodactyly may, at times, be an autosomal dominant trait with variable penetrance.^{3,9,13,14} The underlying abnormality in camptodactyly is an imbalance between the flexor and extensor mechanisms around the PIP joint.^{3,8-10,13,14} Although the DIP joint is rarely involved, the MP joint may show some compensatory hyperextension deformity, especially with more severe flexion deformities of the PIP joint.^{3,10,15} The following structures at the base of the finger have been implicated as a factor causing the

deformity: anomalous intrinsic muscles, lumbrical muscles, or flexor tendons as well as defects in the dorsal extensor apparatus, contracture of the volar skin, and congenital fibrous bands beneath the palmar skin.^{13,15,17,18} Smith et al concluded that all of these defects are involved to different degrees in camptodactyly.¹⁹ The conservative management uses various stretching exercises and splinting techniques and is usually done in mild contractures (less than 45 degrees of flexion). Although splinting techniques have been used to correct camptodactyly, the results are inconsistent. A study by Engber and Flatt on stretching and splinting showed that they were effective in 43% of patients.¹⁰ However, in 57% of the patients, there was progression of the contracture with a success rate of less than 20%. Dynamic splints used by Hori et al and Miura et al initially demonstrated improvements in both flexion and extension of the affected PIP joint for the majority of their patients.^{6,20} The use of both dynamic and static splints by Siegert et al resulted in good to excellent results in 66% of their patients, especially those with mild deformities (less than 30 degrees of contracture).²¹ To be classified as a good to excellent result, the contracture must be corrected to within 20 degrees of full PIP joint extension, or more than a 40 degree increase in PIP joint extension with less than a 30 degree loss of flexion.²¹ The surgical approach to treatment should be performed only if a trial of conservative measures fails to correct the defect.¹⁹ Surgical treatment has mostly been directed at rebalancing the extension and flexion forces about the PIP joint. The majority of the procedures have included tendon transfers and volar soft-tissue release.^{5,6,8,14,16,19,20} Surgical options include FDS release alone, FDS release and transfer to the extensors, FDS release with split skin graft or Z-plasty, FDS release along with release of fascial bands, and transfer of anomalous lumbrical insertion to the lateral band. Static splinting was continued for 2 to 3 weeks in cases of tendon transfer, after which patients were taught to engage in gradual mobilisation of the PIP joint for 6 weeks. Night splinting was continued for a prolonged time in all cases. Ogino and Kato performed surgery on patients with abnormalities of the flexor digitorum superficialis tendon, but most of them had recurrence.¹³ McFarlane et al did surgical repairs in patients with anomalous lumbrical muscle insertions and concluded that surgical intervention tends to be most successful in patients who are identified early in age, have contractures less than 50 degrees, and have passively correctable contractures.¹⁵ Siegert et al reported 66% of patients achieving good to excellent results with nonoperative treatment, whereas only 18% had good results from surgery like release of the flexor digitorum superficialis, with joint capsulotomy and skin grafts.²¹ Siegert et al divided the patients into three categories based on their preoperative degrees of contracture (mild <30, moderate 30-60, severe >60). Patients in the nonoperative group with mild deformities experienced complete correction, whereas surgery for this group caused an increase in their flexion contractures. Patients with moderate contractures had greater improvement with the

nonoperative method. But, patients with severe contractures clearly benefited more from surgical intervention than splinting. Smith and Grobbelaar studied the use of surgical treatment for patients with severe contractures who failed conservative management and using an extensive postoperative splinting protocol, they improved the range of movement for 83% of their patients achieving good to excellent results.²⁰ Hence, the above studies illustrates the unpredictable and often poor results of surgery for the correction of camptodactyly and therefore the decision for surgical intervention must be taken only after careful evaluation.

CONCLUSION

Camptodactyly is a rare condition with not much mentioned regarding the ideal treatment in the literature. A thorough clinical examination with radiological support would dictate the line of management. For mild contractures, non-operative method would be adequate and surgery for the severe cases. Moderate contractures may benefit from both methods. Recurrence is a common complication which must be emphasized to the patient.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

REFERENCES

1. Choi BR, Lim YH, Joo KB. Camptodactyly, arthropathy, coxa vara, pericarditis (CACP) syndrome: a case report. *J Korean Med Sci.* 2004;19:907-10.
2. Hamilton KL, Netscher DT. Multidigit camptodactyly of the hands and feet: a case study. *Hand (N Y).* 2013;8:324-9.
3. Welch JP, Temtamy SA: Hereditary contractures of the fingers (camptodactyly). *J Med Genet.* 1966;3:104-13.
4. Santosh R, Haobijam N, Barad AK. Absent flexor digitorum profundus (FDP): an unreported component of camptodactyly. *J Med Soc.* 2014;28:120-2.
5. McFarlane RM, Classen DA, Porte AM. The anatomy and treatment of camptodactyly of the small finger. *J Hand Surg Am.* 1992;17:35-44.
6. Miura T, Nakamura R, Tamura Y. Long-standing extended dynamic splintage and release of an abnormal restraining structure in camptodactyly. *J Hand Surg.* 1992;17:665-72.
7. Glicenstein J, Haddad R, Guero S. Surgical treatment of camptodactyly. *Ann Chir Main Memb Super.* 1995;14:264-71.
8. Smith RJ, Kaplan EB. Camptodactyly and similar atraumatic flexion deformities of the proximal interphalangeal joints of the fingers. A study of thirty-one cases. *J Bone Joint Surg Am.* 1968;10:1187-203.
9. Littman A, Yates JW, Treger A. Camptodactyly: A kindred study. *JAMA.* 1968;206:1565-7.
10. Engber WD, Flatt AE. Camptodactyly: An analysis of sixty-six patients and twenty-four operations. *J Hand Surg Am.* 1977;2:216-24.
11. Benson LS, Waters PM, Kamil NI. Camptodactyly: classification and results of nonoperative treatment. *J Pediatr Orthop.* 1994;14:814-9.
12. Foucher G, Lorea P, Khouri RK. Camptodactyly as a spectrum of congenital deficiencies: a treatment algorithm based on clinical examination. *Plast Reconstr Surg.* 2006;117:1897-905.
13. Ogino T, Kato H. Operative findings in camptodactyly of the little finger. *J Hand Surg Br.* 1992;17:661-4.
14. Koman LA, Toby EB, Poehling GG. Congenital flexion deformities of the proximal interphalangeal joint in children: A subgroup of camptodactyly. *J Hand Surg Am.* 1990;5:582-6.
15. McFarlane RM, Curry GI, Evans HB. Anomalies of the intrinsic muscles in camptodactyly. *J Hand Surg Am.* 1983;8:531-44.
16. Minami A, Sakai T. Camptodactyly caused by abnormal insertion and origin of lumbrical muscle. *J Hand Surg Br.* 1993;18:310-11.
17. Maeda M, Matsui T. Camptodactyly caused by an abnormal lumbrical muscle. *J Hand Surg Br.* 1985;10:95-6.
18. Inoue G, Tamura Y. Camptodactyly resulting from paradoxical action of an anomalous lumbrical muscle. *Scand J Plast Reconstr Surg Hand Surg.* 1994;28:309-11.
19. Smith PJ, Grobbelaar AO. Camptodactyly: A unifying theory and approach to surgical treatment. *J Hand Surg Am.* 1998;23:14-9.
20. Hori M, Nakamura R, Inoue G. Nonoperative treatment of camptodactyly. *J Hand Surg Am.* 1987;12:1061-5.
21. Siegert JJ, Cooney WP, Dobyns JH. Management of simple camptodactyly. *J Hand Surg Br.* 1990;15:181-9.

Cite this article as: Mahipathy SRRV, Durairaj AR, Sundaramurthy N, Jayachandiran AP, Rajendran S. Camptodactyly correction: a report of two cases. *Int J Res Med Sci* 2021;9:3460-3.