

Case Report

Surgical correction of club hand by radialization of ulna and leaving distal epiphysis insitu: a case study

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ABSTRACT

Radial club hand is a very rare congenital anomaly which gives deformity as well as functional disability. A 2-year-old female child presented with a radially deviated wrist and an absent thumb on the left side. She was unable to use her left hand. We performed the surgery by radialization of the ulna, leaving the distal epiphysis of the ulna in situ. The leftover distal epiphysis of the ulna started growing proximally and joined the radialized ulna, which provided additional stability to the wrist. The outcome of the procedure was successful, with improved functionality and cosmesis. The procedure was deemed safe with no recurrence in the short term for one year. Further research and long-term follow-up are needed to assess any potential complications associated with this procedure. This case is reported to enlighten the readers about the new technique for treating a rare disorder.

Keywords: Radialization, Distal epiphysis, Club hand, Apical ectodermal ridge, Sonic hedge hog gene.

INTRODUCTION

Radial club hand is a rare disease with an incidence of 1 in 100,000 population.¹ This syndrome has a range of severity, ranging from minor hypoplasia of the thumb to the total loss of the radius. A careful understanding of embryology is required to understand the pathology in the radial club hand. During the third to seventh week of gestation, the upper limb buds form and the radial component arises from the apical ectodermal ridge (AER). The Sonic Hedgehog gene plays a pivotal role in limb patterning and disruptions in SHH signaling result in a radial component deficiency. The primary objective of surgical care is to attain functional alignment and ensure stability. The techniques employed in this context encompass centralization (specifically tendon transfers),

distraction with radialization, and microvascular epiphysis transfer.²⁻⁴ The task of achieving a balance between function and cosmesis remains daunting.

There are very few studies that were done in the radial club hand. Still, further research has to be conducted and more cases have to be reported as of the rarity to the disease. Surgeries are also very difficult to perform and long follow-ups are required for understanding the complications of the disease. Follow-up should be continued till the child grows, because recurrence is very common. Just correction is not the end. Skeletal survey and also screening of the entire body (USG abdomen). Sometimes, a geographical survey has to be done. Although it is rare, it's an anomaly to be corrected and it

should not be neglected as the limb is very useful in daily activities and balance.

CASE REPORT

A 2-year-old female child presented with deformity of the left hand, wrist and forearm with no history of trauma and infection. As per history, the child was born with a deformity. There was no history of difficult labour and the child was born after the completion of 40 weeks. History of consanguinity present. On examination, the left forearm looks short and thin compared to the right forearm. Absence of thumb with radial deviation deformity at the wrist was noted. No radial pulsations could be palpated.

The child was unable to perform active wrist movements. Finger movements were present, but she was unable to use her hand. Passive supination and pronation were restricted; ulnar deviation was impossible. Wrist flexion and extension were grossly restricted. A color Doppler test of both upper limbs was done and it was found that the radial artery was absent on the left side. The radiograph of the left upper limb shows the absence of the radius and 1st metacarpal ray along with phalanges (thumb) with club hand deformity.



Figure 1 (A-C): Pre operative radiograph of left upper limb with absence of radius and I metacarpal and phalanges (thumb) along with radial deviation of hand.

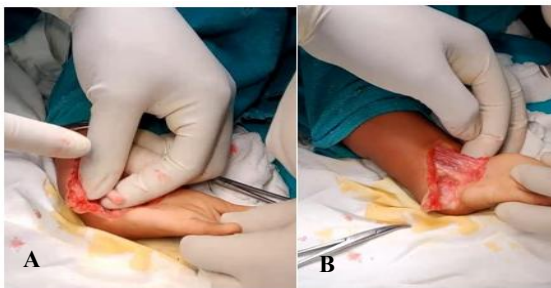


Figure 2 (A and B): An intraoperative picture of the ulna that is being radialized.

The child was thoroughly assessed clinically and radiologically. Then the child was posted for surgery under general anaesthesia and tourniquet control in the supine position. The incision is made on the ventral aspect from the middle of the forearm up to the proximal 1/3rd of the palm. The lower half of the ulna is exposed by separating from the surrounding muscles and osteotomized just

proximal to distal epiphysis of the ulna and shifted towards the radial side at the base of the scaphoid (radialization).

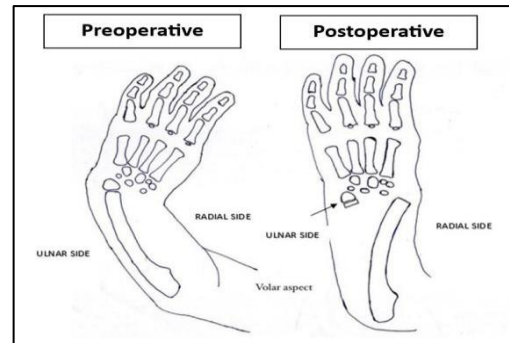


Figure 3: Radialization of the ulna, leaving a distal end along with the epiphysis. Black arrow showing the epiphysis left over on the medial side

Later intra-medullary fixation was done with a K-wire connecting the 2nd metacarpal and ulna. The distal part of the ulna with the epiphysis was left in-situ with an anticipation for it to grow proximally and join the radialized ulna, which gives additional stability to the wrist. The wound closed in layers after securing homeostasis and dressed.



Figure 4: Post-operative radiograph with k wire insitu.



Figure 5: A 2-month follow-up clinical picture with an above elbow cast in-situ.

Then, the limb is immobilized with a plaster of paris (POP) slab and bandage, keeping the elbow in flexion and forearm in supination and wrist in neutral position. After two weeks, the wound was inspected and sutures were removed. The wound healed well. Again, the limb was immobilized in the above elbow POP casing. After two months the POP was removed, and the K-wire was also removed. The wrist splint was given to maintain the correction. The mother was advised to do passive movements of the child's forearm, wrist and fingers intermittently. The mother was also advised to encourage the child to do active finger and wrist movements.

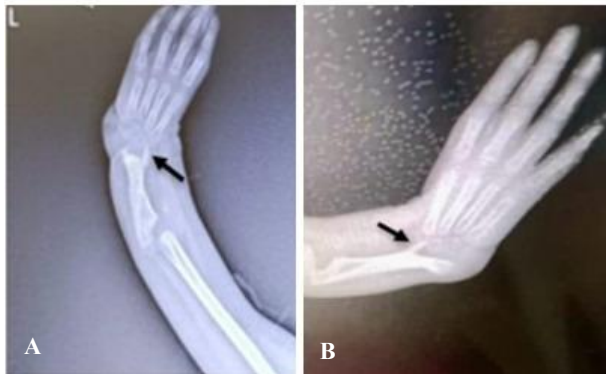


Figure 6 (A and B): 6 months and 1 year follow-up post-operative radiograph of left upper limb showing growth of epiphysis towards Radialized ulna.



Figure 7: 6 months follow up clinical picture.



Figure 8: 1 year follow up clinical picture.

The child was followed for one year and the patient had a better outcome compared to pre operative period in terms of improved functionality and cosmesis. As anticipated, the leftover distal epiphysis of the ulna has grown proximally and joined the radialized ulna, which is acting as a support to the wrist and helping to improve its stability and functionality of the wrist. The child is able to hold objects like a pen.

DISCUSSION

Radial club hand is a very rare congenital anomaly. It varies from the mere absence of distal epiphysis to the complete absence of radius in total, along with the absence of thumb. As per the Bayne and Klug classification, this case falls under type 4.⁵ There are many procedures for correcting the deformity and improving the functionality of the hand and wrist. The current procedure aims to maintain cosmesis and functionality at the wrist in a child as his or her functionality of the hand would affect his or her daily activities as the child grows. In Bayne and Klug type 4, commonly performed procedures are radialization with tendon transfers, centralization and micro epiphysis transfers and gradual correction with Ilizarov external fixator. This technique is a novel procedure and further modification of radialization, where we are leaving the distal epiphysis with the distal part of the ulna insitu and shifting the remaining part of the ulna bone towards the radial side, the process left the distal epiphysis insitu anticipating that it will grow proximally and join the medial surface of distal 1/3rd of the ulna which gives stability to the wrist and also prevents ulnar deviation of wrist. To the surprise it took place and the distal ulna looks like an inverted Y and also, it helped us in preventing transplant of epiphysis with metatarsal from foot. On follow-up for two years, correction is good and the child is able to use the hand and hold small objects like a pen. Growth of ulna will be continued with the help of proximal epiphysis and as the forearm depends on only the ulna there will not be any chance of deformity. At the most the length of the left forearm may be short when compared to the opposite normal right forearm. The purpose of surgery is good cosmesis and functionality, that is already achieved.

This procedure requires skills and expertise as the affected limb has no radial artery and depends on only one artery, i.e., the ulnar artery.

A long period of follow-up until child has attained skeletal maturity is needed. The literature is very sparse and limited in accessibility.

Management of Bayne and Klug type 4 radial longitudinal deficiency remains one of the more challenging problems in congenital hand surgery, and reported outcomes vary considerably with the technique used. In their original description of the classification, Bayne and Klug followed 53 patients for an average of 8.6 years after centralization and reported good or satisfactory results, defined as a

hand-forearm angle under 30 degrees, in 81% of hands, which established centralization as the historical benchmark against which newer techniques have since been measured.⁵ However, later long-term series have shown a substantial rate of recurrent radial deviation after this procedure. Damore et al followed 19 clubhands for a mean of 6.5 years and found that the mean total angulation, corrected from 83 degrees preoperatively to 25 degrees immediately after surgery, drifted back to a mean of 63 degrees by final follow-up, an average loss of 38 degrees.⁶ Shariatzadeh et al, in a series of 11 hands followed for a mean of 90 months, similarly reported 66% correction at surgery but a 54% loss of that correction over time.⁷ Set against this experience, our patient has shown no recurrence to date, though we recognise that recurrence in these comparative series generally became evident only several years after surgery, well beyond our current one-year follow-up.

A major contributor to recurrence and stiffness after centralization is injury to the distal ulnar physis during seating and pinning of the carpus. Buck-Gramcko³ introduced radialization as an alternative that translocates the carpus to the ulnar side of the ulnar head rather than impaling it onto the ulna, and reported a lower recurrence rate of 7.5%, though growth arrest of the distal ulnar epiphysis from avascular necrosis of the growth plate still occurred in 11% of hands. Sestero et al, studying ulnar growth patterns in radial longitudinal deficiency more broadly, confirmed that the ulna in these children typically reaches only half to three-quarters of the length of the unaffected side, and that surgical handling of its distal end can further compromise this already limited growth potential.⁸ Our technique was conceived to sidestep this specific problem: by osteotomising the ulna proximal to its distal epiphysis and translocating only the shaft, while leaving the epiphyseal fragment undisturbed in its native soft-tissue bed, avoided direct instrumentation of the physis altogether. The subsequent proximal migration of this leftover epiphysis to fuse with the radialized shaft, producing the inverted-Y configuration seen on our follow-up radiographs, does not appear to have been previously described, and if reproducible in further cases would represent a way of gaining epiphyseal apposition and wrist stability without the physeal handling inherent to standard centralization or radialization.

Preservation of a growth centre on the radial side of the wrist has also been the rationale behind microvascular epiphyseal transfer. Vilkki originally described transferring the second metatarsophalangeal joint to reconstruct a mobile, growing radial support, and in a later long-term review with de Jong and Moran, ulnar length at final follow-up averaged 67% of the contralateral side with a mean wrist range of motion of 83 degrees, figures that compare favourably with the 48-58% relative ulnar length reported after conventional centralization.^{4,9} This lends support to the general principle that protecting a growth plate on the radial side of the wrist translates into better preserved forearm length over time, the same principle

underlying our own technique. Because our method uses the patient's own distal ulnar epiphysis rather than a free vascularized joint transfer, it avoids the donor-site morbidity, longer operating time, and microsurgical expertise that limit the wider use of the Vilkki procedure, which may be a practical advantage in resource-limited settings such as in this case.

More recent modifications have pursued the same goal of stability without recurrence or physeal injury through other means. Paley described ulnarization, performed through a volar approach with flexor carpi ulnaris transfer and acute ulnar shortening, and reported no recurrent deformity or growth arrest at follow-up, although the technique requires a period in an external fixator and a level of deformity-correction expertise that is not universally available.¹⁰ Bhat et al, working at an Indian centre, described a metaphyseal ulnar cuff osteotomy performed during radialization that is designed to correct the deformity while sparing the epiphysis and carpus; at a medium-term follow-up averaging 66 months, their patients maintained ulnar growth to 62% of the contralateral side with a low rate of recurrence.¹¹ This technique shares the same underlying philosophy of epiphyseal preservation as this series, though it differs technically: rather than an osteotomy confined to the metaphysis, left the entire distal epiphyseal fragment in situ and allowed it to unite spontaneously with the radialized shaft, an approach that, if the growth response we observed proves consistent in further cases, may achieve a comparable result without an additional cuff-osteotomy step.

Taken together, these comparative series point to a consistent theme: techniques that protect the distal ulnar physis, whether by microvascular transfer, volar ulnarization, or metaphyseal-sparing osteotomy, report lower recurrence and better preserved forearm growth than standard centralization^{4-7, 9-11}. This case adds to the body of current evidence by describing a further physis-preserving modification of radialization that requires neither microsurgical transfer nor a specialised fixator system, which may make it more readily applicable in general paediatric orthopaedic practice, a gap also highlighted in recent reviews of the management of radial longitudinal deficiency.¹²

Table 1: Bayne & Klug classification.

Bayne and Klug classification of the Radial Deficiency	
Type 1	Deficient distal radial epiphysis
Type 2	Deficient distal and proximal radial epiphysis
Type3	Present proximally (partial aplasia)
Type 4	Completely absent (total aplasia – most common)

Our conclusions remain limited to a single case with one year of clinical and radiographic follow-up; recurrence in

the comparative series discussed above became apparent only after several years, and growth of both the radialized ulna and the migrated epiphyseal fragment will need to be followed at least until skeletal maturity before this technique can be recommended more widely.^{6,7}

CONCLUSION

This is a novel procedure in which we leave the epiphysis with the distal part of the ulna at its position and shift the remaining part of the ulna bone towards the radial side along with the muscles. This procedure requires a steep learning curve and experience as is usually the case with this type of club hand, with the radial artery absent and the ulnar artery is the only artery available for vascularity.

Recommendations

Regular follow-up of the child until skeletally matured. The mother was advised to observe for any deformity, instability, or disability.

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Ethical approval: Not required

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