Case Report

Skin Necrosis following Ilizarov Application in an Adolescent with Radial Club Hand

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Abstract

Congenital radial club hand is a well-recognized congenital malformation of the upper extremity. It is characterized by radial deviation of the hand and shortening of the forearm. Soft-tissue distraction with an Ilizarov fixator followed by ulnar centralization in a staged approach for the Type IV radial club hand is well described in the literature. In this report, we describe a unique complication following Ilizarov application for soft-tissue distraction in a patient with Type IV radial club hand. A longitudinal skin necrosis appeared at the radial side of the forearm and hand that healed spontaneously after the cessation of distraction.

Keywords: Club hand, complication, Ilizarov, necrosis, skin

Introduction

Congenital radial club hand or longitudinal radial deficiency is a well-recognized congenital malformation. It represents a "failure of formation of parts" anomaly of the upper extremity. It is a rare condition, occurring 1 in every 30,000–100,000 live births, with a slightly increased incidence in boys.[1] In 50% of the cases, it is bilateral and is frequently asymmetrical.[2] It is characterized by radial deviation of the hand along with significant shortening and bowing of the forearm. There is hypoplasia of bone and soft tissues including nerves and vasculature on the radial aspect of the forearm and hand. The thumb may be absent, and the ulna is typically 60% of the normal length.[2]

Conventionally, the radial club hand has been classified by Bayne and Klug according to the severity of one of the four types based on the length of radius present. Bayne and Klug Type IV, the most severe form, is clinically the most common type.[3]

The treatment of the radial club hand varies considerably with the extent of the deformity and the age of the patient.[1,2] Several authors described the concept of gradual soft-tissue distraction with an Ilizarov fixator followed by ulnar centralization in a staged manner for Bayne and Klug Type IV radial club hand.[4-11] In our report, we used the Ilizarov technique as a multistage procedure for soft-tissue stretching, centralization, and lengthening of the ulna. A thin longitudinal band-like painful ulceration-necrosis of the skin and the soft tissues appeared at the radial aspect of the forearm and the hand. To the best of our knowledge, this complication has never been reported before. We aim at presenting this rare complication encountered during the soft-tissue stretching in a case of Type IV radial club hand. The patient and the family gave an informed consent after we informed them that anonymous data and pictures concerning his case would be submitted for publication.

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Case Report

An 11-year-old boy has the right club hand with completely absent radius and thumb (Bayne and Klug Type IV). On examination, the wrist was stiff, in slight flexion and radially deviated with a minimal motion from the resting position and a limited forearm pronation/supination from a neutral position. The hand-forearm angle was +76 as measured on the

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AP forearm radiograph according to the method described by Manske *et al*.,[12] where the angle is formed by a line drawn through the third metacarpal bone intersecting the distal ulna bisector line and reflects the amount of radial deviation, indicated as a positive number, or ulnar deviation, indicated as a negative number, of the hand [Figure 1a].

Before surgery, the patient and his family were counseled regarding the commitment and stamina required to have this procedure. Ilizarov fixator was applied to the right forearm and hand to gain soft-tissue stretching and wrist centralization. The Ilizarov fixator consisted of two full rings. The transfixion wires were placed in the proximal ulna and distally in the metacarpals. The ring was fixed to the proximal ulna by two 1.5-mm Ilizarov smooth wires and two 3-mm olive wires. Distally, the ring was fixed to the metacarpals by one 1.5-mm Ilizarov smooth wires and two 3-mm olive wires in a radial to ulnar direction. The two rings were connected by two threaded rods with the hinge on the convex side at the level of the apex of the angular deformity [Figure 1b]. No bony shortening or carpal resection was performed.

On the 2nd postoperative day, the distraction was started at the usual rate of 1 mm daily (quarter-turn four times). At the 5th postoperative day, we noticed a painful band-like necrosis of skin along the radial aspect of the forearm [Figure 2]. Therefore, the distraction was halted for 2 weeks until the lesion spontaneously healed and then resumed at a slower pace (half-millimeter daily). Before discharge, the patient and his family were instructed to clean the wire sites daily with alcohol swabs, and any pin tract infection was treated accordingly. The patient was followed in the clinic at 2-week intervals, looking for clinical and radiological progress, and for any complication. The Ilizarov fixator was removed after 2 months, and the forearm was temporarily splinted for 3 months.

At the most recent 3-year follow-up, the patient had a good hand function with no recurrence of the deformity and radiographs revealed a hand-forearm angle of +6° [Figure 3].

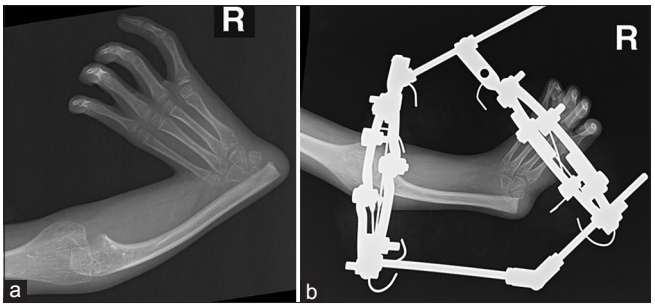

Figure 1: Plain radiographs of the right forearm and the hand showing (a) the preoperative radial club hand deformity (hand-forearm angle: +76°) and (b) immediate postoperative radiographs following application of Ilizarov fixator

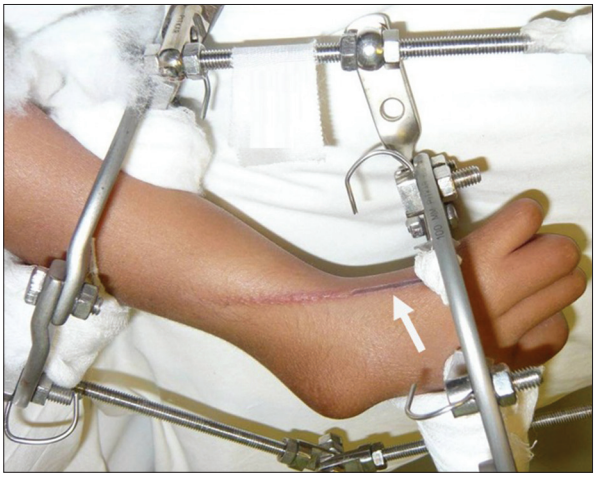

Figure 2: Clinical photograph showing the ulceration-necrosis at the radial side of the forearm and hand with Ilizarov fixator *in situ* at 5th postoperative day (white arrow)

Figure 3: Plain radiographs of the right forearm and hand at the most recent 3-year follow-up (hand-forearm angle: +6°)

Discussion

The introduction of the Ilizarov method has allowed the possibility of gradual distraction with neohistogenesis. Initial reports have described, the use of the Ilizarov fixator for only ulnar lengthening in the radial club hand.[5,6] Several authors later have described the use of the Ilizarov fixator for soft-tissue distraction preceding centralization of the ulna.[3,4,7,8] The authors reported excellent results using the Ilizarov fixator in a staged approach for Bayne and Klug Type IV radial club hand. They cited significant correction of the deformity and better functional position of the hand while minimizing the overall complications.

Several soft-tissue procedures have been described for the treatment of the radial club hand. These include z-plasty, dorsal-rotation flap, Evans bilobed flap, and microvascular joint transfer.[13-19] Although these single-stage procedures subject the patient to a single operation, it is associated with a higher rate of ulnar growth plate injuries, radial neurovascular injuries, ulnar shortening, and wound complications.[2] In a late-presenting patient with severe deformity as in the

presented case, an Ilizarov technique is more effective than a soft-tissue procedure.[10,11]

Complications, including nocturnal pain, early or delayed consolidation, joint contracture or stiffness, nerve palsy, pin track infection, and skin inversion, have been reported in the literature using the Ilizarov technique.[20] In our patient, we noticed a unique complication in the form of a longitudinal necrotic band of skin at the radial aspect of the forearm and the hand. It appeared in the early phase of distraction and healed spontaneously when the distraction was paused. We do not know whether distraction at a slower pace from the outset could have been successfully prevented this complication. In our view, we propose that stretching of the severely contracted soft tissues, combined with reduced blood supply of the inherently underdeveloped vasculature on the hypoplastic radial side of the club hand have contributed to the pathogenesis of this lesion.

In conclusion, we recommend that distraction using Ilizarov fixator should be closely monitored and preferably carried out at a slow pace in Type IV congenital radial club hand.

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Conflicts of interest
There are no conflicts of interest.

Author contributions
ADA edited the initial manuscript, wrote both the revised and the final manuscript and was the corresponding author, MA collected the data and wrote the initial manuscript, and the senior author SA provided the case material and supervised the whole work. All authors have critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript.

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