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Case Report

Congenital Bilateral Hypoplasia of First Metacarpal in a 24 Year Old Male: A Rare Case Report

Arjun Ballal[1], Sanath Kumar Shetty[2], Anoop Hegde[1]

[1]Registrar, ARS Hospital, Tirupur.
[2]Assistant Professor, K.S.Hegde Medical Academy, Deralakatte, Mangalore-575018.

Corresponding Author: Arjun Ballal

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ABSTRACT

Hypoplasia of the thumb refers to a spectrum of clinical abnormalities ranging from a slightly small digit to complete absence (or aplasia) of the thumb unit. As a component of radial dysplasia, thumb hypoplasia can be either an isolated entity or seen in conjunction with other elements of radial longitudinal deficiency. Treatment of this condition initially involves identifying and addressing co-morbid anomalies in other body systems. The severity of thumb hypoplasia is then graded so that appropriate treatment methods can be instituted.
A 24 year old gentleman presented to us with an alleged history of trauma and he was diagnosed with an abrasion over the left palm. Incidentally, he was noted to have hypoplastic thumb in both hands. The radiographs showed bilateral hypoplastic first metacarpal with normal phalanges. There was an associated bilateral elongated ring fingers. There was no abnormality in the functions of the thumb and other digits. There were no other systemic abnormalities.
We would like to report this in our case report as this is a rare presentation.

Key Words: Congenital bilateral hypoplasia, first metacarpal.

INTRODUCTION

Hypoplasia of the thumb is included in the group 5 or undergrowth category of upper extremity malformations. [1] Included in this category are malformations that range from those in which the hypoplasia is difficult to recognize to a pouce flottant to complete aplasia of the thumb. Thumb hypoplasia has been reported to comprise 3.5% of congenital upper extremity malformations. [2] It may occur as an isolated malformation along with a more global radial hypoplasia.

The phalanges, trapezium, and scaphoid may be small, in addition to a small metacarpal. There have been case reports of all of the median nerve innervated thenar muscles being absent, or, in other cases, only a hypoplasia of the opponens pollicis and abductor pollicis are noted. [3] The extrinsic thumb flexor, the flexor pollicis longus (FPL), or extensor pollicis longus (EPL), may also be absent or hypoplastic. The FPL may also have an aberrant origin, such as from the index flexor digitorum profundus, or an aberrant insertion. [4] The ulnar collateral ligament is always hypoplastic and unstable. The first web space may also be narrowed.

A number of syndromes are associated with thumb hypoplasia or a more global radial hypoplasia, including Fanconi's anemia, thrombocytopenia, VATER association (vertebral anomalies, anal atresia, tracheoesophageal fistula, esophageal atresia, and renal defects), Holt-Oram syndrome, Cornelia de Lange's

syndrome, diastrophic dwarfism, and a 13q deletion. [5]

CASE REPORT

A 24 year old gentleman presented to us with an alleged history of fall and a wound over the left palm. On radiography no bony injuries were noted. But a hypoplastic first metacarpal with normal phalanges and an elongated ring finger was noted (Figure 1). Radiographs of the right hand also showed a hypoplastic first metacarpal with normal phalanges and an elongated ring finger. Clinical examination revealed no visible deformity of the hands (Figures 2 and 3)

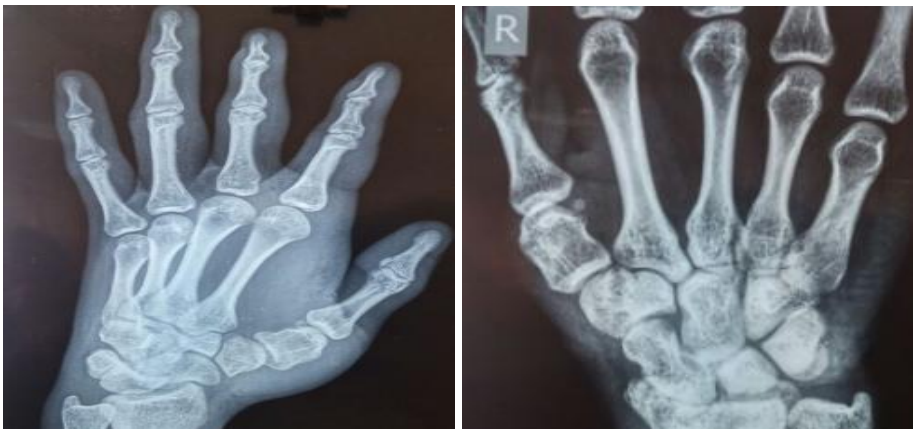

Figure 1 A Figure 1 B

Figures 1A and 1B: Plain radiographs of left and right hands showing bilateral hypoplastic first metacarpal.

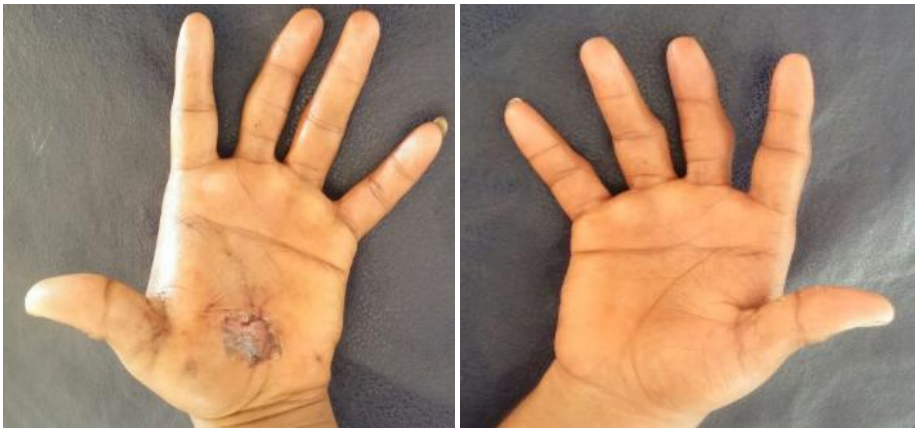

Figure 2 A **Figures 2B**

Figures 2A and 2B: Clinical images showing volar aspect of both hands. Abrasion noted in the left palm after the injury

On further clinical evaluation he was noted to have no deformities or abnormalities in the functions of the hands. He was doing his day to day activities without any difficulties. He had normal pinch, grasp and grip. He had no associated medical complaints or had no other symptoms or signs suggestive of any other systemic illness or deformities.

There was no surgical intervention performed or planned as the patient had no complaints.

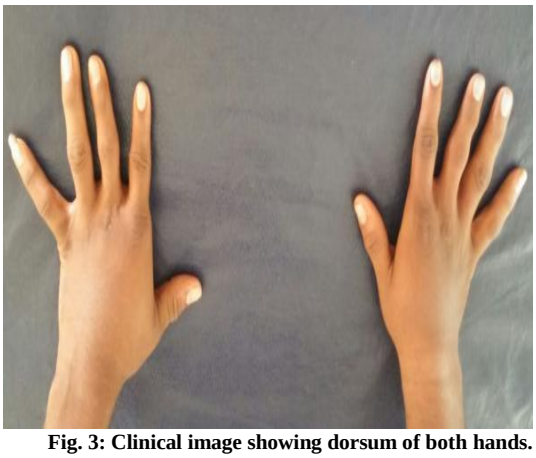

Fig. 3: Clinical image showing dorsum of both hands.

DISCUSSION

James et al. [6] found 63% to have bilateral involvement, 59% to have radial dysplasia, and 86% to have other anomalies.

Buck-Gramcko [7,8] has modified the Blauth classification of thumb hypoplasia into five types:

Type 1: All structures are minimally shortened and narrowed

Type 2: Mild underdevelopment of all structures; short bones; smaller diameter; mild hypoplasia of thenars; unstable thumb metacarpophalangeal joint; narrow first web space

Type 3A: Stable carpometacarpal joint; significant decrease in the size of the thumb; severe hypoplasia of the intrinsic and extrinsic musculature; unstable meta-carpophalangeal joint; narrow first web space.

Type 3B: Type 3A with an unstable carpo-metacarpal joint

Type 4: Pouce flottant; rudimentary phalanges

Type 5: Complete aplasia of the thumb

The role of nonoperative treatment in a child with a hypoplastic thumb is limited. In a child with a type 1 or minimally hypoplastic thumb with good pinch and grasp, surgery is not indicated. There may be a select child in this group who benefits from therapy for thenar strengthening or for activities of daily living, but few require this. In children with more advanced thumb hypoplasia, splinting or therapy, including thenar strengthening, is not helpful in improving the function. Splinting may be helpful for stability in a child who, for medical reasons, is not a surgical candidate.

The child's potential for grip and pinch plays a major role in the decision to operate on a child with a hypoplastic thumb. The thumb is an essential component for prehensile grasp. The inability to do prehensile activity because of poor thumb function is an indication for treatment of the hypoplastic thumb. One has to remember that these children are different from those who have had a traumatic injury or amputation of the thumb because of the lack of normal cerebrocortical representation. [9]

Most children develop prehensile patterns of hand use at approximately 6 to 12 months of age. [10] It is reasonable to offer reconstructive thumb surgery at 1 to 2 years of age. The children are usually large enough by 1 year of age that there is minimal risk with general anesthesia, and the structures around the thumb are large enough to make the surgery technically easier. If the surgery is delayed until the child is 3 or 4 years of age, the patterns of hand usage may be so well developed that retraining of the child is more difficult postoperatively. Also, the child is closer to school age, and it may take time to become facile with writing. If the child undergoing a pollicization is also undergoing surgery for radial aplasia, the pollicization is usually performed 6 months after the wrist surgery. [9]

Our case can be classified as type I. He did not undergo any surgical or medical intervention. As it is a rare case we would like to report it as a rare case report.

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