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CASE REPORT

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A 23-year-old patient with bilateral hypoplastic thumbs and toes: A case report

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Key Clinical Message

However, it is noteworthy that certain patients with minor functional limitations in their hand may have experienced undiagnosed thumb hypoplasia during their childhood years. These individuals may have successfully adapted to their condition without seeking medical intervention and may express a preference for nonintervention (as in this case).

Abstract

Thumb hypoplasia is a congenital underdevelopment of the thumb, accounting for 5%–15% of congenital hand disorders. It occurs equally among both genders and can affect both thumbs. The condition is categorized using Blauth's classification with Type I being the mildest form. We report a 23-year-old Syrian male presented with a bilateral restriction in opposition movement when using a pen or razor. Clinical examination and x-ray imaging revealed a bilateral Type I hypoplastic thumb with bilateral minimal hypoplasia of the toes. Despite the surgical treatment options available, the patient opted not to undergo surgery due to his adaptation to his condition. Hypoplastic thumb Type I is a congenital condition characterized by underdevelopment of the thumb. Bilateral thumb hypoplasia with toes hypoplasia is extremely rare. A full systemic evaluation should be done due to its associations with other syndromic manifestations and treatment options are discussed concerning the best functional outcomes and patient preferences.

KEYWORDS

congenital, hypoplasia, thumb, toes

1 | INTRODUCTION

Thumb hypoplasia is a condition of congenital underdevelopment of the thumb. It accounts for approximately 5%–15% of congenital hand disorders.[1]

It occurs equally among both genders. In over half of the cases, both thumbs are affected, while among one-sided hypoplastic patients the right thumb is reported to be more commonly involved.[2]

Blauth's classification is the most used categorization. The principle of hypoplastic thumb reconstruction is to address each abnormal clinical element.[3]

Type I thumbs have minimal shortening and narrowing. Type II thumbs have thumb-index web space

narrowing, hypoplasia intrinsic thenar muscles, and metacarpal joint instability. Type IIIA thumbs have Type II features plus extrinsic tendon abnormalities and hypoplastic metacarpal with a stable carpometacarpal (CMC) joint. Type IIIB thumbs have Type IIIA features plus partial metacarpal aplasia with an unstable CMC joint. Type IV thumbs are floating thumbs, and Type V thumbs are absent thumbs.[4]

2 | CASE HISTORY

A 23-year-old male patient comes to the rheumatology clinic for a bilateral restriction in opposition movement especially when using a pen or razor. The clinical examination revealed a bilateral inability to use the interphalangeal joint of the thumb and toes with a mild weakness in grip strength.

3 | DIFFERENTIAL DIAGNOSIS, INVESTIGATIONS, AND TREATMENT

X-rays imaging of the hands and feet, along with a physical examination, demonstrated a bilateral Type I hypoplastic thumb with bilateral minimal hypoplasia of the toes (Figures 1 and 2).

After presenting the surgical treatment options to the patient and their limited benefit toward his condition and the fact that he is adapted to his condition, the patient did not want to undergo any surgical intervention.

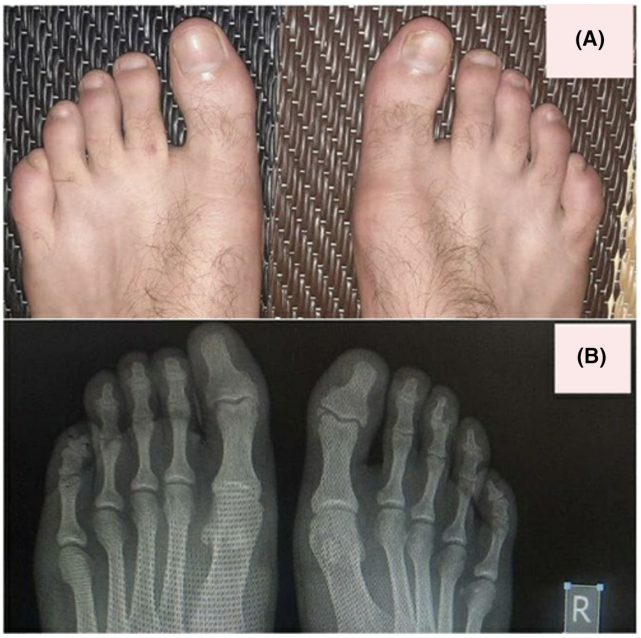

FIGURE 2 (A) Dorsal view of both feet demonstrated minor hypoplasia of the both halluxes and absence of dorsal wrinkles are seen. (B) X-ray of the both feet demonstrated both-side hypoplastic hallux with an avulsion fracture in the right hallux because of previous trauma.

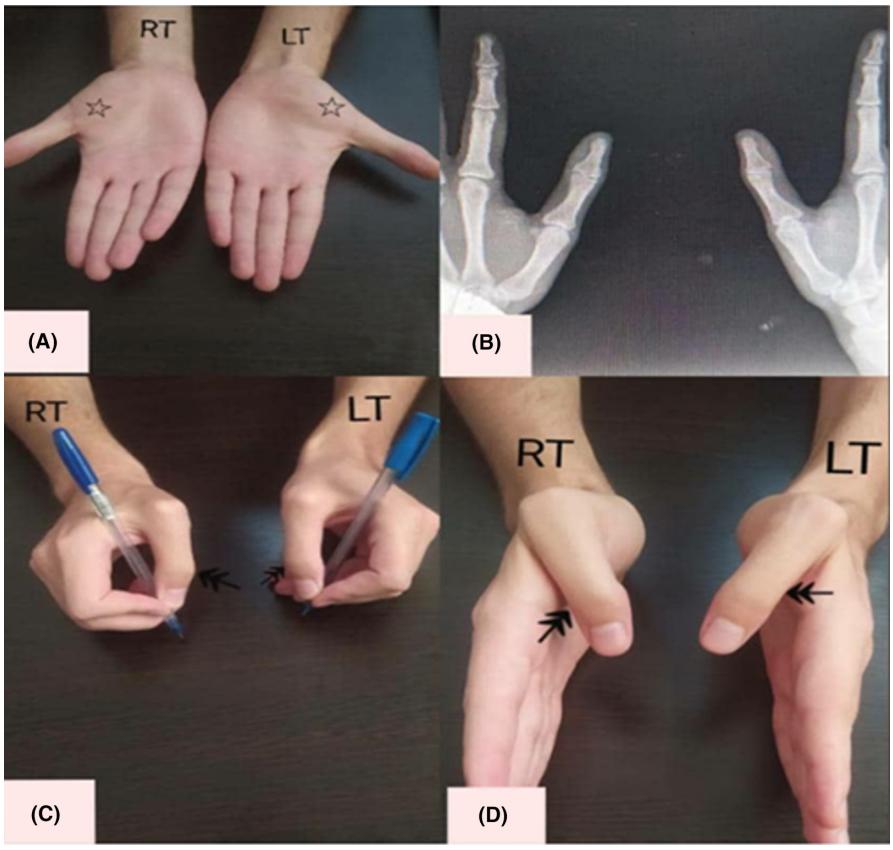

FIGURE 1 (A) Palmar surface of the hands shows absent flexion creases (arrows) of bilateral thumbs. Thenar eminence atrophy is absent (asterisk). (B) X-ray of the hands demonstrated both-side hypoplastic thumb Type I. Radiograph of both hands shows mild skeletal and soft tissue hypoplasia of bilateral thumbs. (C) Functional limitation in holding a pen; note the weakness of active flexion of the interphalangeal joints of both the thumbs. (D) Dorsal view of both thumbs. Minor hypoplasia of both the thumbs and absence of dorsal wrinkles are seen (arrows).

4 | OUTCOME AND FOLLOW-UP

The patient's mobility improved well on physical therapy. After follow-up for 2 years, the patient's condition was very good.

5 | DISCUSSION

Thumb hypoplasia is distinguished by extrinsic tendon dysplasia, thenar muscle hypoplasia, metacarpal adduction, and instability of the metacarpophalangeal (MCP) joint. In the most severe cases, thumb aplasia or MCP joint instability is present, and thumb removal surgery is advised as treatment.[5]

Thumb hypoplasia affects one in 100,000 live births. On the other side, 50% of these kids will also have a comparable impairment, however, the degree may vary.[6,7]

Both sexes are equally affected by the condition.[8,9]

Many classifications have graded congenital thumb hypoplasia and theses classifications aid in surgical decision-making. It was initially categorized by Müller, but Blauth's classification (and BuckGramcko's revision) is the one that is most frequently used today.[10]

Blauth's classification is based on different abnormalities of the bones, MCP joint, CMC joint, and thenar muscles of the thumb.[8]

Clinical presentation ranges from minimal underdevelopment of the thumb (Grade I) to a completely absent thumb (Grade V) (Table 1).[9]

Type I thumb hypoplasia in both hands is extremely rare, with an estimated incidence of less than 1 in a million births. The occurrence of Type I hypoplasia in both hands and feet is even rarer, with only a few reported cases in the medical literature.

Congenital hypoplastic thumb can be a part of the radial longitudinal deficiency spectrum or an isolated deformity.[11]

It can be also associated with syndromic manifestations such as thrombocytopenia-absent radius syndrome, Holt–Oram syndrome, Fanconi syndrome, and VACTERL sequences. Thus, a systematic evaluation of the musculoskeletal system, cardiovascular system, and complete blood count should be done for diagnosing such comorbidities. The initial management involves identifying and managing associated extra-skeletal abnormalities for better outcomes.[6]

Treatment options vary based on the severity of the thumb deformity and the functional impairment of the hand. Patients with hypoplastic thumb Type I often have a well-functioning thumb and may require no further intervention, while patients with Type II and IIIA may undergo surgical thumb reconstruction that involves releasing the tight web space between the thumb and index finger using a skin graft and stabilizing the MCP joint through ligament reconstruction. Patients with Type IIIB, IV, and V have a completely absent or severe malfunctioning thumb and require pollicization; a surgical procedure that creates a functional thumb by transferring another finger (usually the index) to the thumb position.[12]

However, it is noteworthy that certain patients with minor functional limitations in their hand may have

TABLE 1 Modified Blauth classification.

Grade	Clinical manifestations
I	Narrow, small (near normal) Minimal underdevelopment Stable Mild intrinsic hypoplasia
IIA	Narrow, small Adduction contracture Uniaxial ulnar collateral ligament (UCL) metacarpophalangeal (MCP) joint instability, poorly developed thenar muscles
IIB	2A plus biaxial/global MP joint instability and/or extrinsic anomalies
IIC	2B plus inadequate carpometacarpal (CMC) joint
IIIA	Aplasia proximal 1/3 thumb MC (absent CMC joint) Severe underdevelopment Unstable Extensive intrinsic and extrinsic musculotendinous deficiencies
IIIB	3A with aplasia proximal 2/3 thumb Metacarpal (absent CMC joint)
IV	Absent metacarpal (pouce flottant)
V	Absent thumb

experienced undiagnosed thumb hypoplasia during their childhood years. These individuals may have successfully adapted to their condition without seeking medical intervention and may express a preference for nonintervention (as in this case). Hypoplastic thumb Type I is a congenital condition characterized by underdevelopment of the thumb. It is the mildest form among the different types of hypoplastic thumbs. Bilateral thumb hypoplasia with toes hypoplasia is extremely rare. Despite its small size and limited functionality, individuals with Type I hypoplastic thumbs can still perform most daily activities. However, specialized medical evaluation and potentially surgical intervention may be required to address functional limitations and improve overall hand function.

AUTHOR CONTRIBUTIONS

Suaad Hamsho: Writing – original draft. **Mouhammed Sleiay:** Writing – review and editing. **Mohammed Alaswad:** Formal analysis; resources; writing – original draft. **Hasan Alsmodi:** Writing – original draft. **Khaled Al-Hakeem:** Resources; writing – original draft.

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The authors have no conflict of interest to declare.

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Not applicable.

ETHICS STATEMENT

Not applicable because all data belong to the authors of this article.

CONSENT

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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REFERENCES

1. Van Wyhe RD, Trost JG, Koshy JC, Pederson WC. The duplicated thumb: a review. *Semin Plast Surg*. 2016;30(4):181-188.
2. Abdel-Ghani H, Amro S. Characteristics of patients with hypoplastic thumb: a prospective study of 51 patients with the results of surgical treatment. *J Pediatr Orthop B*. 2004;13(2):127-138.
3. Spinner M. Fashioned transpositional flap for soft tissue adduction contracture of the thumb. *Plast Reconstr Surg*. 1969;44(4):345-348.
4. Ozols D, Butnere MM, Petersons A. Methods for congenital thumb hypoplasia reconstruction. A review of the outcomes for ten years of surgical treatment. 2019;55(10):610.
5. Riley SA, Burgess RC. Thumb hypoplasia. *J Hand Surg Am*. 2009;34(8):1564-1573.
6. Bhat AK, Acharya AM. Current concepts in the management radial longitudinal deficiency. *J Clin Orthop Trauma*. 2020;11(4):597-605.
7. Basat S, Orman C, Kayadibi T, Scetin I, Bozkurt M. Hypoplastic thumb type IIIB: an alternative method for surgical repair. *Hand Microsurg*. 2014;3(2):59-61.
8. Mende K, Suurmeijer JA, Tonkin MA. Surgical techniques for reconstruction of the hypoplastic thumb. *J Hand Surg Eur*. 2019;44(1):15-24.
9. Nakada M, Tada K, Nakajima T, Matsuta M, Tsuchiya H. A case of a 5-year-old boy with a Blauth type IIIB hypoplastic thumb reconstructed with a nonvascularized, hemilongitudinal metatarsal transfer. *Case Rep Orthop*. 2018;2018:8205285.
10. Little KJ, Cornwall R. Congenital anomalies of the hand—principles of management. *Orthop Clin North Am*. 2016;47(1):153-168.
11. Tay SC, Moran SL, Shin AY, Cooney WP 3rd. The hypoplastic thumb. *J Am Acad Orthop Surg*. 2006;14(6):354-366.
12. Zargarbashi R, Panjavi B, Bozorgmanesh M. Congenital hypoplastic thumbs treated by staged nonvascularized MTP joint transfer for absent MCP joints and abductor digiti minimi tendon transfer for opposition: a case series study. *BMC Musculoskelet Disord*. 2023;24(1):179.

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