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A Neglected Rare Case of Thumb Duplication: A Case Report

İhmal Edilmiş Nadir Bir Başparmak Duplikasyonu Olgusu: Bir Olgu Sunumu

Yavuz ÖNEL

İzmir City Hospital, Department of Hand Surgery

Abstract

Aim

Preaxial polydactyly (PP) is one of the most common congenital anomalies of the upper extremity and exhibits a wide spectrum of clinical presentations. Surgical management aims to achieve a stable, well-aligned thumb with preserved function and acceptable aesthetic appearance. This case report presents a rare and neglected case of thumb duplication and discusses surgical outcomes in light of existing classification and scoring syste.

Case

A nine-year-old male patient presented with preaxial polydactyly of the right hand and functional impairment. Physical examination revealed absence of interphalangeal joint motion in the radial thumb, whereas motion was preserved in the ulnar thumb. Radiographic evaluation demonstrated a triphalangeal radial thumb and an epiphyseal bony connection between the duplicated digits. Surgical treatment involved excision of the radial thumb, preservation and reconstruction of collateral ligament and abductor pollicis brevis insertion, and stabilization using Kirschner wires. Postoperative follow-up showed complete metacarpophalangeal joint motion, minimal interphalangeal extension deficit (5°), and mild radial deviation (15°). Opposition and pinch strength were comparable to the contralateral thumb, and the patient and family reported high satisfaction with functional and aesthetic outcomes. No complications related to Kirschner wire fixation were observed.

Conclusion

PP exhibits wide morphological variability, and traditional classification systems may not fully encompass all clinical presentations. Individualized surgical planning and the combination of appropriate reconstructive techniques are essential for achieving optimal outcomes. In this case, collateral ligament reconstruction and temporary Kirschner wire stabilization provided satisfactory functional and aesthetic results. The outcome aligns with established scoring systems, supporting the effectiveness of tailored surgical strategies in managing complex PP cases.

Keywords: polydactyly; congenital abnormalities; thumb; reconstructive surgical procedures

Öz

Amaç

Preaksiyal polidaktili (PP), üst ekstremitenin yaygın görülen konjenital anomalilerinden biri olup belirgin klinik varyasyon göstermektedir. Cerrahi tedavinin amacı; stabil, aksiyel dizilimi düzgün, fonksiyonu korunmuş ve estetik açıdan kabul edilebilir bir başparmak elde etmektir. Bu olgu sunumunda, başparmak duplikasyonu bulunan nadir ve ihmal edilmiş bir olgu ele alınmış ve cerrahi sonuçlar mevcut sınıflama ve skorlama sistemleri ışığında değerlendirilmiştir.

Olgu

Dokuz yaşında erkek hasta, sağ elde preaksiyal polidaktili ve fonksiyonel kısıtlılık ile başvurdu. Fizik muayenede radial başparmakta interfalangeal eklem hareketinin olmadığı, ulnar başparmakta ise hareketin korunduğu saptandı. Radyolojik değerlendirmede radial başparmağın trifalangeal yapıda olduğu ve duplike parmaklar arasında epifizyal düzeyde kemiksel bağlantı bulunduğu belirlendi. Cerrahi tedavi kapsamında radial başparmak eksize edildi; kollateral ligament ve abductor pollicis brevis insersiyosu korunarak rekonstrüksiyon uygulandı ve Kirschner telleri ile stabilizasyon sağlandı. Postoperatif takipte metakarpofalangeal eklem hareket açıklığının tam olduğu, interfalangeal eklemde yalnızca 5° ekstansiyon kaybı bulunduğu ve yaklaşık 15° radial deviasyon mevcut olduğu görüldü. Opozisyon ve pinch kuvveti karşı taraf ile benzerdi; hasta ve ailesi fonksiyonel ile estetik sonuçlardan yüksek düzeyde memnuniyet bildirdi. Takip süresince Kirschner teli ile ilişkili komplikasyon izlenmedi.

Sonuç

PP geniş morfolojik varyasyonlar gösterdiğinden, geleneksel sınıflama sistemleri tüm klinik prezentasyonları kapsamada yetersiz kalabilmektedir. Optimal sonuçlara ulaşılabilmesi için bireyselleştirilmiş cerrahi planlama ve uygun rekonstrüktif tekniklerin kombinasyonu önemlidir. Bu olguda kollateral ligament rekonstrüksiyonu ve geçici Kirschner teli stabilizasyonu ile tatmin edici fonksiyonel ve estetik sonuçlar elde edilmiştir. Sonuçlar, mevcut skorlama sistemleri ile uyumlu olup, kompleks PP olgularında hasta bazlı cerrahi stratejilerin etkinliğini desteklemektedir.

Anahtar kelimeler: polidaktili; konjenital anomaliler; başparmak; rekonstrüktif cerrahi prosedürler

Corresponding Author: Yavuz ÖNEL, MD
İzmir City Hospital, Department of Hand Surgery
yavuzonel48@gmail.com
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Introduction

Polydactyly is one of the most common congenital anomalies of the upper extremity anomalies. It arises from abnormal differentiation of the limb bud during the sixth to eighth weeks of embryogenesis. Its reported incidence ranges from approximately 0.08 to 1 per 1,000 live births (1). Preaxial polydactyly (PP) is a congenital anomaly characterized by considerable clinical variability. The Wassel–Flatt classification system, which categorizes duplication based on the skeletal level from distal to proximal, is the most widely used method for distinguishing different clinical variants (2). Wassel–Flatt Type IV is the most common variant, accounting for approximately 40% of cases. PP is typically unilateral and does not demonstrate a clear familial inheritance pattern. However, particularly in Wassel–Flatt Type VII cases, associations with systemic conditions such as Holt–Oram syndrome or Fanconi anemia have been reported (3).

Case

A nine-year-old male patient presented to our outpatient clinic with complaints of preaxial polydactyly (PP) of the right hand. According to the history obtained from the patient's mother, difficulties in writing and psychosocial distress due to being stigmatized by peers were reported. Physical examination revealed absence of active and passive motion at the interphalangeal joint (IPJ) of the radial-sided duplicated thumb. In contrast, the ulnar-sided thumb demonstrated flexion and extension movements at the IPJ (Figure 1). Sensory examination revealed no deficits. Vascular assessment using the Allen test was negative. Direct radiographic evaluation demonstrated that the radial-sided thumb had a triphalangeal structure. A bony connection at the epiphyseal level between the radial and ulnar thumbs was observed. Additionally, the ulnar-sided thumb exhibited ulnar deviation of the proximal phalanx and radial deviation of the distal phalanx (Figure 2). Genetic analysis performed on the patient revealed no abnormalities.

Figure 1. Preaxial polydactyly of the right thumb.

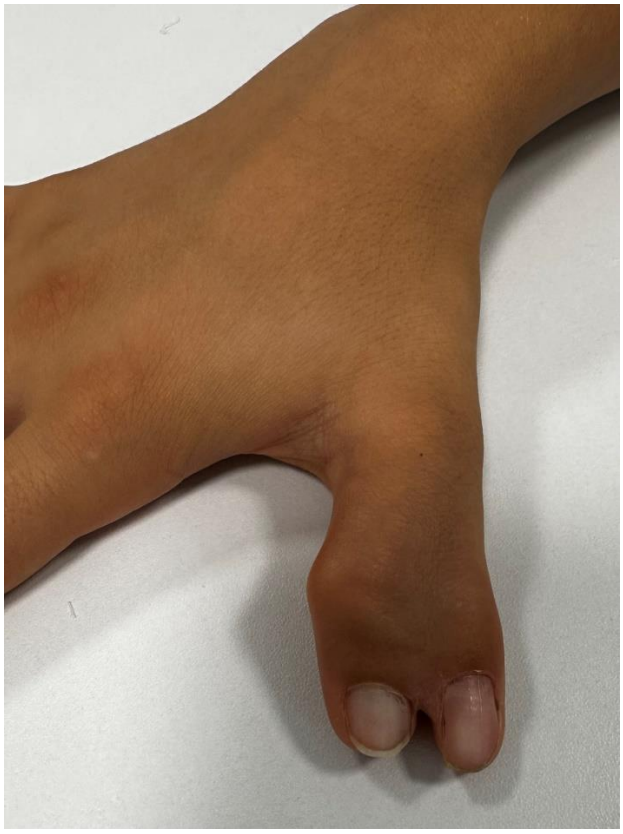
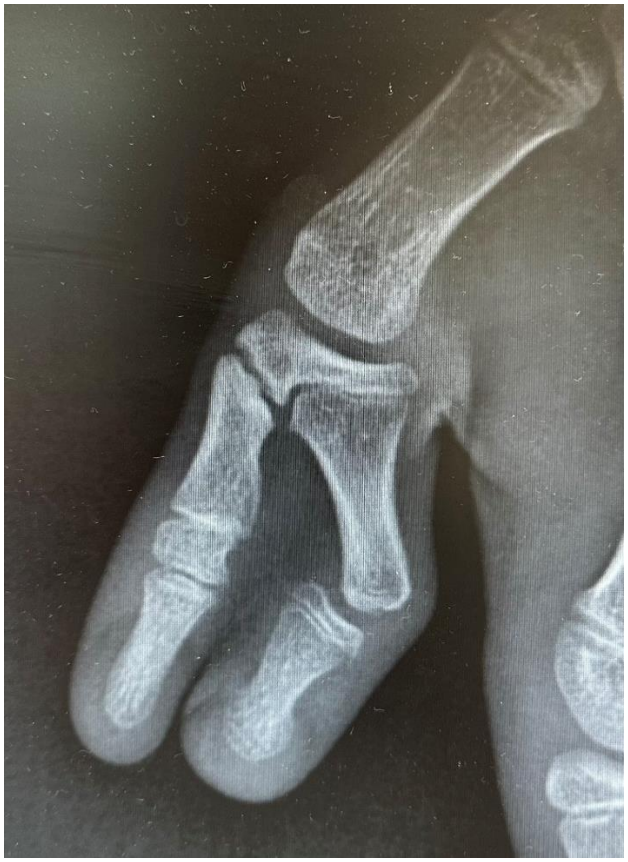


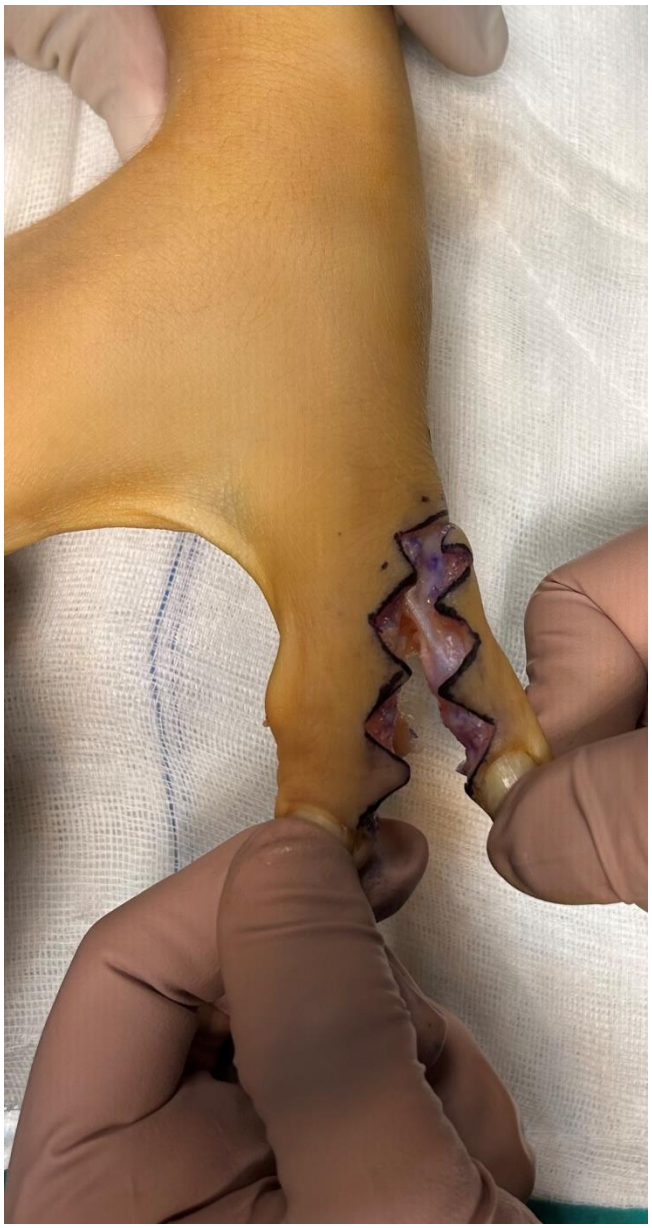
Figure 2:.Radiographic image.



The patient's family was provided with detailed information regarding the planned surgical intervention, alternative treatment options, and potential complications; following the acquisition of informed consent, surgical treatment was scheduled.

During the surgical procedure, multiple incisions were made on the dorsal and volar aspects of the duplicated thumbs to create appropriate skin flaps. The digital arteries and nerves of the ulnar-sided thumb were carefully dissected and preserved. Similarly, the extensor pollicis longus and flexor pollicis longus tendons on the ulnar side were observed to be well developed and were maintained intact (Figure 3).

Figure 3. Intraoperative image.



The abductor pollicis brevis muscle and the bony insertion of the radial collateral ligament belonging to the radial-sided thumb were preserved and removed together with a bone fragment (Figure 4). Subsequently, the ulnar-sided thumb was separated from the radial aspect in accordance with markings made under fluoroscopic guidance. The abductor pollicis brevis and radial collateral ligament, whose bony insertion had been preserved, were reattached at an appropriate anatomical location with sufficient tension and secured using two Kirschner wires (K-wires). Repair of the ulnar collateral ligament of the interphalangeal joint (IPJ) was performed. The joint was reduced into an appropriate position and stabilized with a K-wire. After confirming adequate circulation and hemostasis, the created skin flaps were adapted to their new anatomical positions, and the surgical procedure was completed (Figure 5).

Figure 4. Preservation of the bony insertion of the radial collateral ligament (black arrow).

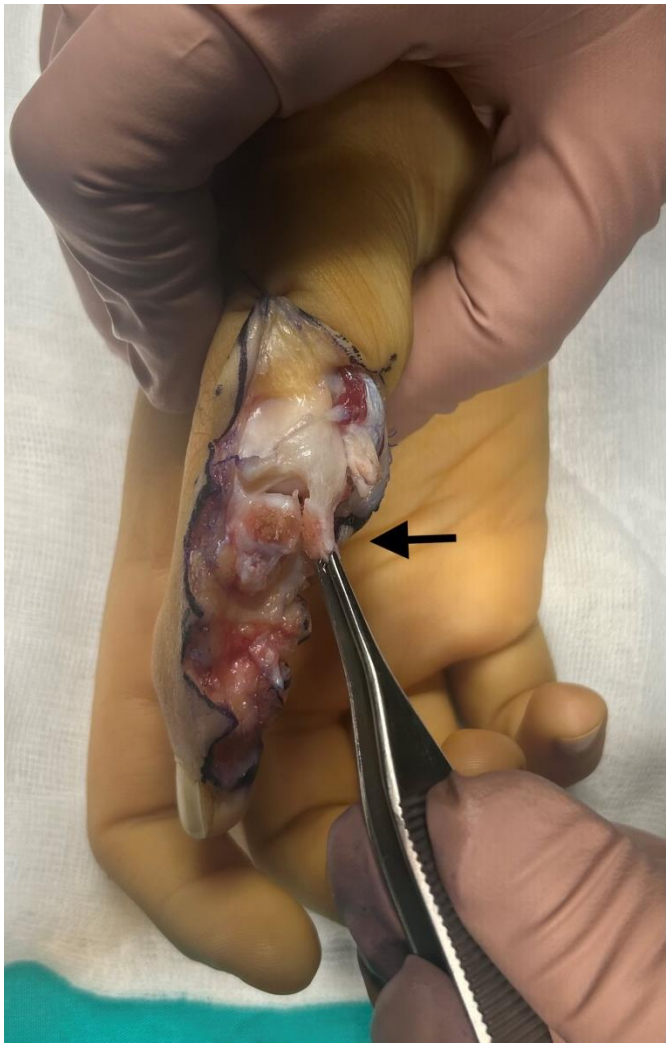
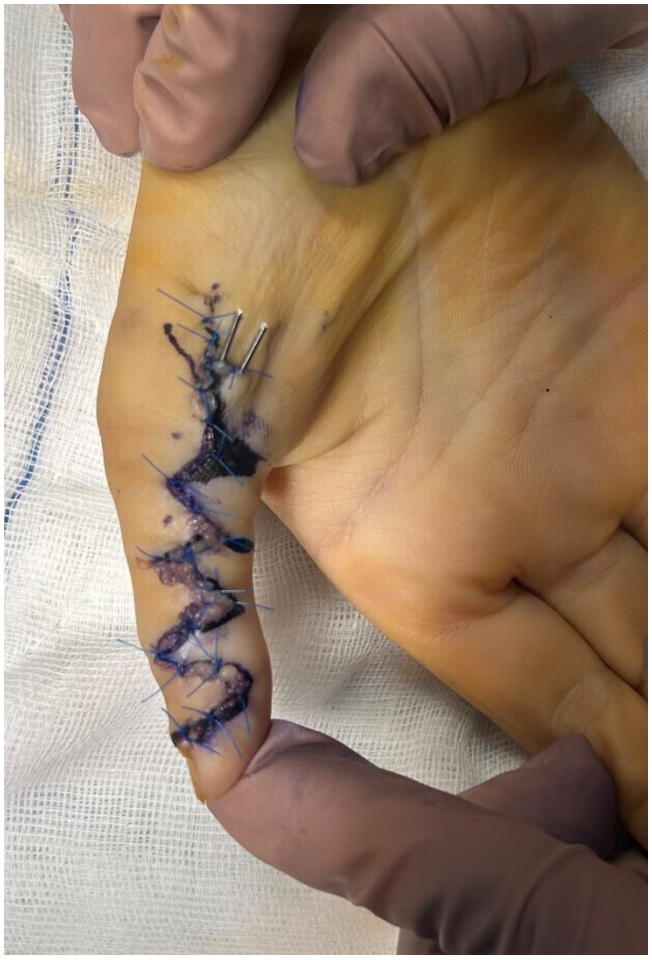


Figure 5. The radial collateral ligament was fixed with two K-wires, and the skin was sutured.



Postoperatively, the patient was followed with weekly outpatient clinic visits during the first month; at the end of this period, the Kirschner wires were removed and a rehabilitation program was initiated. Follow-up was subsequently maintained through regular monthly outpatient evaluations. At the one-year control, metacarpophalangeal (MCP) joint flexion and extension were found to be complete, with no joint instability detected (Figure 6 and 7). IPJ flexion was also full, while a 5-degree extension deficit was observed. Additionally, approximately 15 degrees of radial deviation was present (Figure 8). Opposition and pinch strength were comparable to those of the contralateral thumb. The patient and family reported a high level of satisfaction with both functional and aesthetic outcomes.

Figure 6. The aesthetically acceptable appearance of the hand at the one-year follow-up.

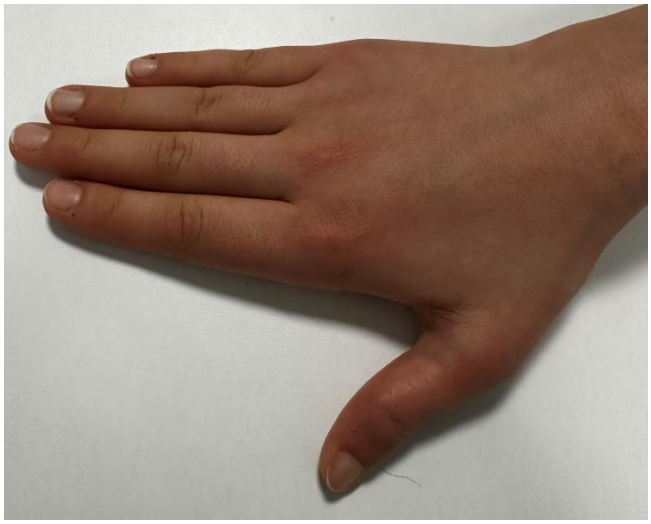
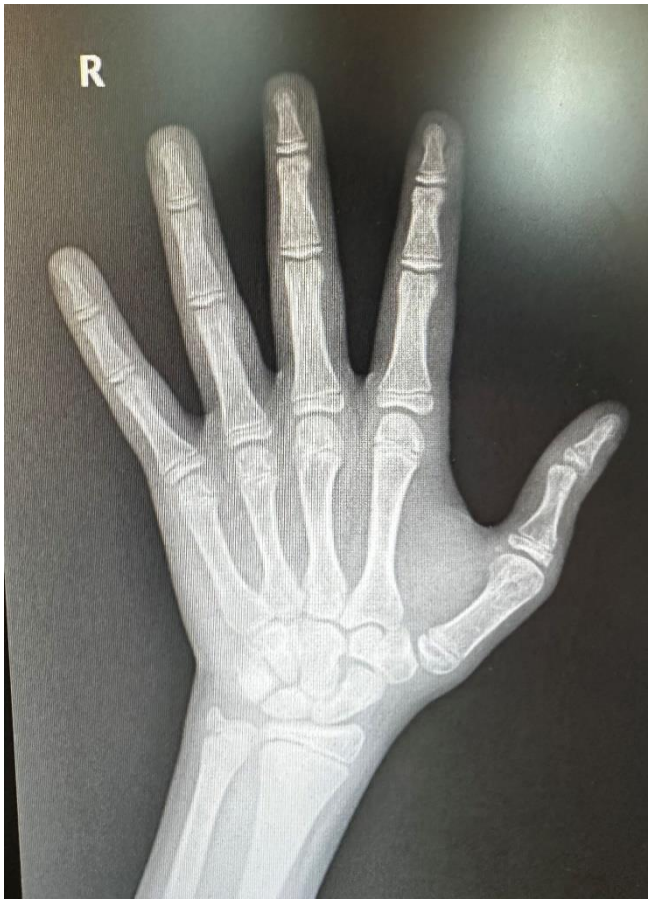


Figure 7. Normal functional use.

Figure 8. One-year follow-up control radiograph.



Discussion

In PP, the timing of surgical intervention is regarded as a determining prognostic factor influencing functional and aesthetic outcomes. It has been reported in the literature that the optimal time window for surgical treatment is between 12 and 18 months of age, given that functional pinch grasp has not yet fully developed and aesthetic awareness remains relatively limited (4,5). However, in the present case, due to sociocultural and economic constraints, surgical intervention could only be performed at the age of 9 years. This delay resulted in difficulties with fine motor tasks, particularly writing activities during the school period, and subjected the patient to peer-related psychosocial distress.

PP may exhibit a slight male predominance in the literature, although it can occur in both sexes. The majority of cases are unilateral, whereas bilateral involvement is observed less frequently. From a genetic perspective, PP may present as sporadic isolated cases; however, in some instances, it may follow an autosomal dominant inheritance pattern. Additionally, syndromic forms have been described, and therefore genetic evaluation is recommended, particularly in cases with bilateral involvement or associated anomalies (6). The present case, characterized by male sex, unilateral involvement, and the absence of hereditary transmission on genetic evaluation, is largely consistent with the epidemiological and genetic features reported in the literature.

PP classification is of considerable importance for prognostic assessment, determination of the appropriate surgical approach, evaluation of potential intraoperative complications, and standardized reporting of postoperative outcomes (7,8). For this purpose, the most widely used system is the Wassel–Flatt classification. However, PP exhibits a broad clinical spectrum, and this heterogeneity may render the Wassel–Flatt system insufficient for categorizing certain atypical morphological variants. In the present case, although the dominant and nondominant thumbs demonstrated a duplicated appearance as two separate digits, the presence of an epiphyseal bony connection does not fully correspond to the classical definition of duplication. Furthermore, the observed morphology does not conform to a bifid configuration. Consequently, the Wassel–Flatt classification is inadequate for categorizing the morphological characteristics of this case. In this context, considering the subclassification system proposed by Hori and colleagues specifically for Wassel type IV cases (9), it is deemed more appropriate to classify the present case as subtype A.

The primary goal of thumb reconstruction is to obtain a thumb that is stable, preserves joint range of motion, has appropriate size and shape, demonstrates correct axial alignment, provides acceptable aesthetic appearance, and offers functionality as close to normal as possible (10). The surgical approach varies depending on the type and morphological characteristics of polydactyly. In cases of accessory polydactyly, simple excision may be sufficient. In more complex cases, excision of the hypoplastic nondominant thumb and reconstruction of the remaining thumb are generally preferred. Within this scope, collateral ligament reconstruction, corrective osteotomies in the presence of malalignment, and tendon reconstructions when necessary may be performed. Alternatively, the modified Bilhaut-Cloquet procedure, which aims to combine the symmetrical and functional components of both thumbs into a single reconstructed thumb, also represents a viable surgical option (11,12).

Wang and colleagues proposed excision of the radial digit, metaphyseal osteotomy of the ulnar side, and

correction of MCP joint deviation using Kirschner wires as the treatment approach for Type A cases within the WF Type IV subclassification. The authors further reported that axial alignment at the IPJ level could be achieved through ulnar collateral ligament plication or various corrective osteotomies (13). In the present case, deformity correction was pursued without performing osteotomy; instead, collateral ligament reconstruction was carried out and stabilization was achieved using temporary Kirschner wires. Alignment at the IPJ level was addressed in accordance with the authors' recommendations through collateral ligament plication.

In the literature, studies have reported that either Kirschner wires or polydioxanone sutures may be utilized for bony fixation in collateral ligament reconstruction (14). Although both techniques have been shown to yield comparable clinical outcomes, Kirschner wire fixation is suggested to carry a greater predisposition to complications such as infection, wire migration, and skin maceration. Nevertheless, in the present case, stabilization was achieved using Kirschner wire fixation, and none of the aforementioned complications were encountered during the follow-up period.

The Tada scoring system, described by Tada K. and colleagues, is the most widely used method for evaluating surgical outcomes in radial polydactyly. This system assesses thumb length, range of motion of the MCP and IPJ, axial deviation, and stability on a scale of 0–8 points; scores of 6–8 are considered "good" outcomes (15). In the present case, given the complete and stable range of motion at the MCP joint, a mild 5° extension deficit at the IP joint, and approximately 15° of radial deviation despite preserved functional capacity, the surgical outcome was evaluated as consistent with an "excellent" result, corresponding to a score of 7 points according to the Tada scoring system.

In conclusion, PP may not always be classified precisely within traditional classification systems due to its wide range of clinical variations. Although various classification systems have been described in the literature to address this need, many of them may not provide sufficient guidance for surgical decision-making, operative planning, or the assessment of potential requirements for reoperation. On the other hand, numerous surgical techniques have been described for the treatment of preaxial polydactyly. In this context, evaluating each case individually according to its unique anatomical and functional characteristics, planning the surgical approach on a patient-specific basis, and possessing the surgical expertise to combine different techniques when necessary are considered a more rational approach for achieving optimal functional and aesthetic outcomes.

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