Archives of Clinical & Experimental Surgery

Case Report
Arch Clin Exp Surg 2017;6:45-48
doi:10.5455/aces.20150516031930

A seven-toed central polydactyl in an adult: A neglected, unclassifiable case

Kenan Koca[1], Bahtiyar Demiralp[1], Yuksel Yurttas[1], Inanc Guven[2], Ercan Koseoglu[3], Serkan Akpancar[1], Mustafa Basbozkurt[1]

ABSTRACT

The aim of this study was to present a neglected, unclassifiable case that involved a central type polydactyl adult with 7 toes and metatarsals, 4 cuneiforms and 1 cuboid. A 22 year-old male soldier with a right polydactyl was referred to our hospital. He suffered from the need of excessively wide-shoes and occasional shoe irritation. He was evaluated with plain radiography and 3D tomography. The patient had central-type polydactyl with 7 toes and metatarsals, and 4 cuneiforms and 1 cuboid. Ankles and hind feet were completely normal. All toes were capable of tendon flexion and extension. His medical and family history was unremarkable. We planned to excise the excessive toes and metatarsals, but the patient denied the surgery. We present a very rare case with a central polydactyl having 7 toes and metatarsals, 4 cuneiforms and 1 cuboid. The striking point in our case was that he was a neglected, unclassifiable case.

Key words: *Polydactyl, central, classification*

Introduction

Polydactyl is one of the most common types of congenital deformities involving the foot. It is defined as the presence of a supernumery digit or metatarsal. The excessive digit or metatarsal may be partially or completely developed. It may exist as an isolated condition or part of a congenital syndrome [1, 2]. According to the classification system of Temtamy and McKusick, polydactylies are classified as pre-axial or tibial (hallux side), postaxial or fibular (side of the little toe), or central (middle toes) [3, 4]. In addition, polydactyl can be of a mixed type, exhibiting pre-axial, postaxial and central characteristics. Most polydactyl foot deformities are postaxial; the pre-axial and the central types are rare. The central ray polydactyl has been found in 3 to 6% of all cases of pedal polydactyl [5].

The hexadigit is the most common form of polydactyl of the foot [6]. Eight-toed polydactyls have been reported numerous times in the literature [7-10]. Two seven-toed polydactyl cases of a seven-year-old girl and a one-month-old girl were reported by Kapetanos [11] and Haber [12]. These cases were of a mixed-type polydactyl. We present an extremely rare case that involved a central-type polydactyl with 7 toes and metatarsals, 4 cuneiforms and 1 cuboid in an adult. He had received no treatment until now.

Author affiliations : *[1]Department of Orthopedic Surgery, [2]Department of Radiology, Gulhane Military Medical Academy, Ankara, Turkey [3]Department of Orthopedic Surgery, Eskisehir Military Hospital, Eskisehir, Turkey*
Correspondence : *Serkan Akpancar, MD, Department of Orthopedic Surgery, Gulhane Military Medical Academy, Ankara, Turkey*
e-mail: drserkanakpancar@gmail.com
Received / Accepted : *December 18, 2014 / February 10, 2015*

Case Report

A 22-year-old man with polydactyl was referred to our hospital. He suffered from the need of excessively wide-shoes and occasional shoe irritation for many years. His medical and family history was unremarkable. Physical examination revealed that he had 7 toes, with one foot that was wider than the other foot. Supernumerary toes included the second and third toes. All toes were capable of active and passive plantar flexion and dorsiflexion. The radiography and 3D tomography showed that the foot had 7 toes with 7 metatarsals, 4 cuneiforms and 1 cuboid. The hind foot and ankle were completely normal. In addition, his left foot was completely normal by physical examination and radiography. He had a normal karyotype. Although we offered to excise the excessive central 2 toes together with the metatarsals and cuneiforms, the patient rejected surgery. Clinical appearance, X ray and 3D CT images of the case are presented in Figure 1A, 1B and 1C.

Discussion

Polydactyl is a common congenital foot deformity that has various clinical presentations. The frequency of polydactyl varies widely among populations. Its incidence is greater than 5.1 cases per 10,000 newborns [13].

The classification of polydactyl of the foot was usually based on the number, location, and morphological difference of the supernumeraries [14]. Venn-Watson delineated classification of the polydactyl according to anatomic and morphologic configurations, specifically duplication of the metatarsals [15]. They reported four major types of polymetatarsals, including Y-shaped, T-shaped, wide metatarsal head, and full duplication in postaxial polydactyl; pre-axial duplication depicted the metatarsal head and they were further classified as either of the wide or short block type. Masada et al. improved the classification of Venn-Watson by focusing on pre-axial polydactyl [16]. However, they did not mention central-type polydactyl. Watanabe et al. classified the morphologic alterations of polydactyl in 265 cases, indicating the type of ray involvement and the duplication level of the metatarsal [13]. According to Watanabe et al., central ray polydactyly is classified into 4 types; metatarsal type, proximal phalangeal type, middle phalangeal type, and distal phalangeal type.

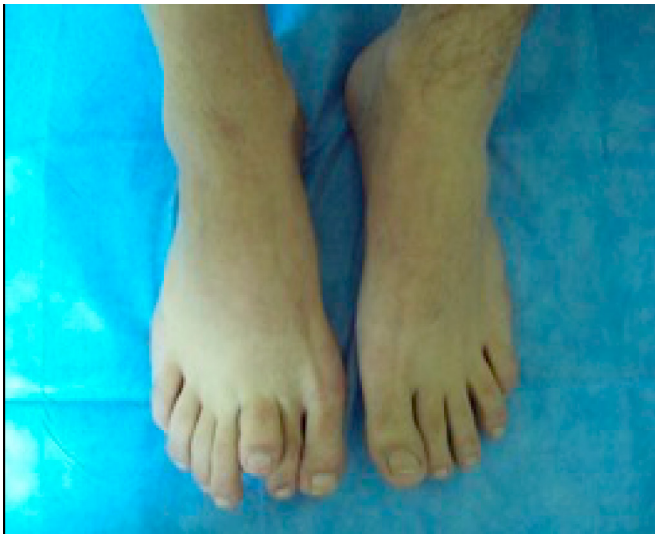

Figure 1A. Clinical appearance of the case. The left foot was completely normal, while 7 toes were defined on the right foot.

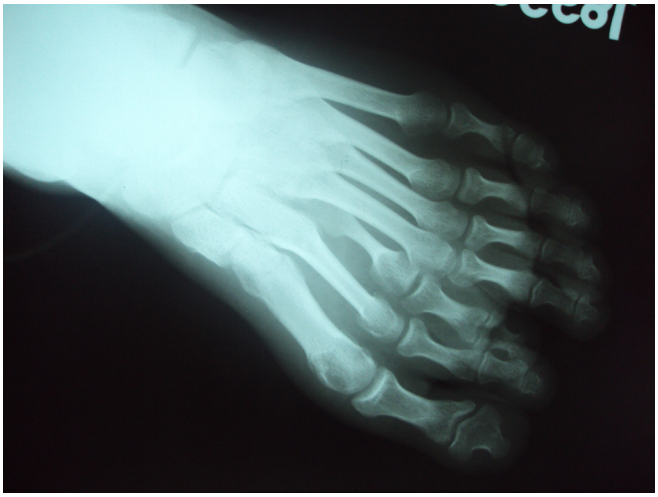

Figure 1B. X-ray images of the right foot showing 7 toes with 7 metatarsals.

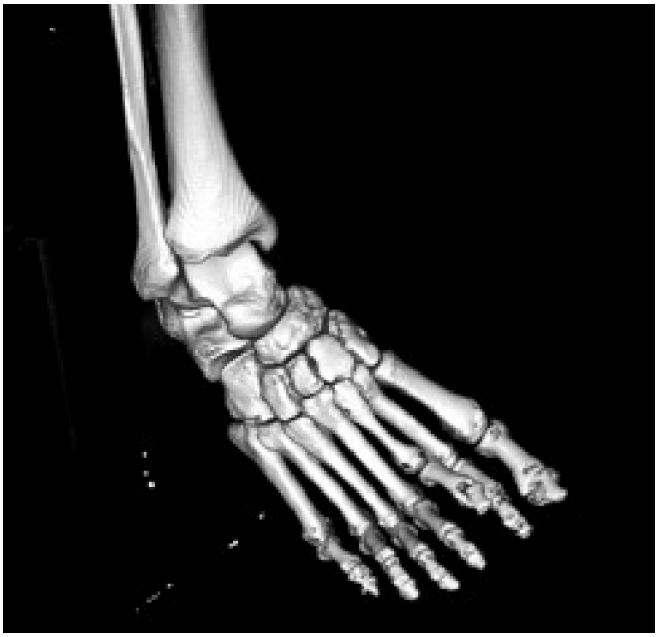

Figure 1C. 3D CT images of the foot showing 7 toes with 7 metatarsals, 4 cuneiforms and 1 cuboid.

Proximal phalangeal and metatarsal types are rarely seen [13]. In the central type of polydactyl, supernumerary digital rays occur between the original first and second rays. Our case involved a 7-toed polydactyl with 7 metatarsals, 4 cuneiforms and 1 cuboid in an adult, with the supernumeraries located between the first and second digits. The present polydactyl did not fit into the Venn-Watson or Masada and Watanabe classification systems because the patient had a poly-cuneiform together with metatarsals and toes. According to the literature, poly-cuneiforms are not associated with polydactyly. However, there was a report of a poly-cuneiform in a mirror foot [17]. Kapetanos [11] and Haber [12] each reported a case with seven toes. However, these two cases involved crossed and mixed polydactylies. The term crossed polydactyly refers to postaxial polydactyly of the hands combined with pre-axial polydactyly of the feet (Type I), or pre-axial polydactyly of the hands combined with postaxial polydactyly of the feet (Type II). The present case involved no cross polydactyly, just central polydactyly.

Polydactyl of the foot can be found as an isolated case or can occur as a part of congenital syndromes that may follow an autosomal-dominant or recessive inheritance pattern [18]. Syndromic polydactylies include Tryzomi 13, Ellis-Van Creveld syndrome, Orofacial-dygital syndrome, Meckel Gruber syndrome, and Rubinstein Taybi syndrome. Patients with syndrome polydactyly have severe medical conditions and more complex polydactyly compared to cases of isolated polydactyly. In addition, syndromic polydactylies are grouped together with syndactylies. In the present case, there was no hereditary history and additional pathology of the other organ systems and the karyotype were normal. Therefore, we classified the present case as an isolated polydactyly.

Patients with polydactyl suffer from cosmetic problems and require large-sized shoes and a different number of shoes. These problems are especially distressful to the family. Our case involved an adult who suffered no psychological trauma from his polydactyl condition. He just required abnormal, extra-wide shoes when he was admitted to our clinic. He reported no pain and walked normally, as indicated by observing his gait.

The aim of surgery for polydactyl is to achieve a cosmetically pleasing foot and allow the patient to wear well-fitting shoes. The optimal time of surgery is not known, but many authors delay the surgery until 1 year of age to avoid problems of anesthesia and to allow additional development of the osseous and soft tissue structures [19]. Some authors reported that they achieved good results performing excision of polydactyl in children [20, 21]. Although resection of the extra toes in central polydactyl results in a wide gap, it can be closed by repair of the transverse metatarsal ligament and by using compressive K-Wire [22, 23]. The present case, however, involved an adult who had not received any treatment. There is limited knowledge in the English literature regarding surgery of the polydactyl toe of adults. Even though Galois et al. reported that surgery was successful in adults with polydactyl [24], their cases were not as complex as our case. Although it may have been difficult to close the wide gap that occurred after the resection of excessive toes, we planned to excise the second and third toes, second and third metatarsals, as well as the cuneiforms next to the second and third metatarsals. Then we could have fused the first and second cuneiforms or implanted soft tissue in the gap to remove the cuneiform. However, the patient rejected the proposed surgery.

In conclusion, we presented a rare central polydactyl with seven metatarsals, seven toes, four cuneiforms and one cuboid. The polydactyl in our case involved an adult who was neglected and unclassifiable.

Conflict of interest statement

The authors have no conflicts of interest to declare.

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