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Case Report

Central hexadactylia of the foot: a rare case presentation

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ABSTRACT

Polydactyly is congenital duplication type of anomaly of fingers and toes. Its embryogenesis is associated with a disturbance of the anterior-posterior axial development of the limb. Classified in pre-axial, central and post-axial. Classification depends on location of duplication. Details of the case-3 years old, female. Examination findings were single supernumerary toe over metatarsal bone. Extra toe is inclined to transverse arch of foot. Bony structure of supernumerary digit with Y-shaped bifurcation of second metatarsal bone proximal to head of metatarsal. For right upper limb type IV radial club hand. For Left upper Limb-Wassel Type 5 Radial polydactyly. Foramen ovale L to R shunt was also found in 2D Echo. Central polydactyly is a very rare phenotypic presentation. Patient in our case was operated with excision of the supernumerary digit. On dissection, anomalous tendons were found with digital nerve and artery. Tendons were removed, digital nerve incised and digital arteries coagulated. This case is a rare variety. Extra toe dorsally oriented, metatarsal angulated to transverse arch of foot. No metatarsal widening. This case does not fit into the classifications described.

Keywords: Bone, Foot, Polydactyly

INTRODUCTION

Polydactyly is congenital duplication type of anomaly of fingers and toes. It can be syndromic or non-syndromic. Inheritance with an autosomal dominant trait with variable penetrance.[1] Its embryogenesis is associated with a disturbance of the anterior posterior axial development of the limb.[2] Classified into pre-axial, central and post-axial.

Classification depends on location of duplication. Belthur et al reported 80% post axial, 15% pre-axial and least common is central type.[3] Pre-axial polydactyly is defined as an extra digit on radial/tibial digits while post axial polydactyly involves ulnar/peroneal digits. Axial (central) polydactyly refers to duplication of three central hand/foot digits. Mirror image polydactyly and Hass-type polysyndactyly are rare and do not fit into these three categories.[4] Aetiology multifactorial genetic, family history, environmental factors.

Aim

To report a case of hexadactyly of left foot which is central polydactyly with type iv right radial club hand and left-hand radial polydactyly.

CASE REPORT

3 years old, female presented with the chief complaint of poor cosmesis of left foot, difficulty wearing footwear, absent right thumb and bowed right wrist and an extra digit in left hand. Third born baby by full term normal vaginal delivery to a non-consanguineous couple. Maternal History uneventful. She cried normally at birth, no cyanotic episodes. No positive family history for similar complaint. Examination findings were single supernumerary toe over metatarsal bone. Extra toe is inclined to transverse arch of foot. Rudimentary nail was present. Sensations were present Figure 1 and 2.

Figure 1: Central polydactyly of foot.

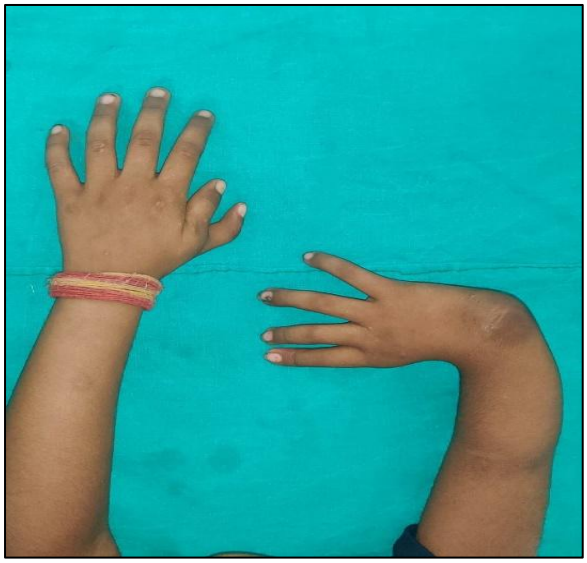

Figure 2: Left hand radial polydactyly and right radial club hand with absent thumb.

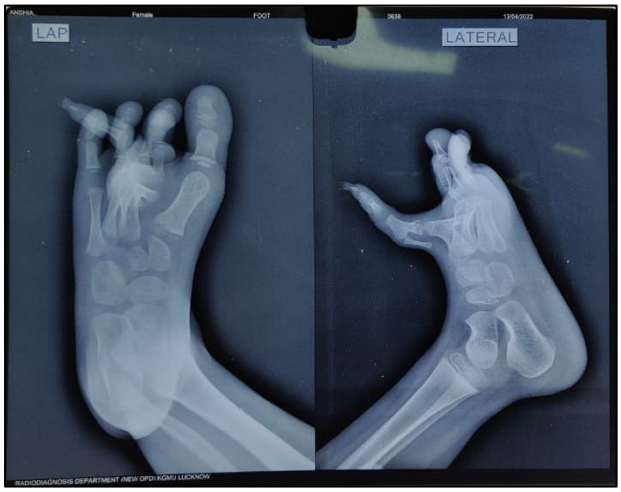

Figure 3: X-ray image of left foot.

Figure 4: Excised supernumerary digit.

Movements appreciated. Both transverse and longitudinal arches were maintained. X-ray image of left foot shows Bony structure of supernumerary digit with Y-shaped bifurcation of second metatarsal bone proximal to head of metatarsal. For Right upper limb- Type IV Radial club hand. For Left Upper Limb-Wassel Type 5 Radial polydactyly. Foramen Ovale L to R shunt was also found in 2D Echo.

DISCUSSION

Central polydactyly is a very rare phenotypic presentation. Presentation can be isolated feature or associated with other limb or cardiac anomalies. Duplication of one of the three middle digits of hand and foot is its characteristic feature. The most common manifestation of hand central polydactyly is duplication of fourth digit. Second toe duplication is the most common presentation of foot central polydactyly.[5] Polydactyly can present as a soft tissue remnant, bony remnant without a joint or a completely functional digit.[6] It can be diagnosed during antenatal period via USG done between 16-20 weeks of gestation.[7]

Polydactyly patients experience difficulty with using shoes, pain during walking, psychosocial issues and poor cosmesis.[3,7] Non-operative treatments include footwear modification which are not of much use. The goals of treatment /surgery include preservation of digits with the greatest axial alignment, resection of symptomatic digits, alignment correction of the remaining great toe, stabilisation of soft tissues and adequate soft tissue coverage.[8] Patient in this case was operated with excision of the supernumerary digit. On dissection, anomalous tendons were found with digital nerve and artery. Tendons were removed, digital nerve incised and digital arteries coagulated. Distractor was placed for Radial club hand. Minimal postoperative deformity was present. Polydactyly is the most common congenital anomaly of foot (45% of all).[9] Various methods of classification based on morphological presentation. Venn-Watson gave

classification based on anatomical configuration of metatarsals and duplicated bony parts. T shape, Y shape, wide fifth metatarsal head, short block first metatarsal head.[10] Classification according to the longitudinal level of bifurcation such as distal, middle or proximal phalangeal, metatarsal or tarsal, given by Blauth et al and Olason et al.[6] Classification by Seok HH-SAM, S indicates need for syndactyly release, need for SSG, A indicates axis of deviation, M indicates metatarsal duplication status.[11] In the literature, polydactyly of foot is usually treated during infancy after onset of walking. Many authors support delaying of surgery until ossification of affected rays has occurred. The reason given for delaying surgery after 1 year of age is that there is better definition of the bony anatomy and the duplicated structures on radiography. Second, at this age, the child starts to walk and we can better evaluate whether polydactyly is causing gait problems.[11]

To remember during surgery is to prevent any residual deformity. If there is widened metatarsal arch, may require repair of transverse metatarsal ligament.

CONCLUSION

Central polydactyly is rare. Metatarsal extension needs particular attention, to prevent post-op residual intermetatarsal widening. This case is a rare variety. Extra toe dorsally oriented, metatarsal angulated to transverse arch of foot. No metatarsal widening. Patient can also be regarded as a case of Holt–Oram Syndrome. This case does not fit into the classifications previously described.

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