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DUPLICATION OF FOOT*

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SUMMARY

A rare case of partial duplication of foot is presented. A few reports are scattered in the literature and this case is presented with the hope that a larger collection, with morphological details, may help in the understanding of the embryogenesis and a possible classification. The literature describing its occurrence has been briefly reviewed.

Duplication of the foot (Diplopodia) is a rare developmental anomaly. The incidence of all congenital anomalies of both the upper and lower extremities including club foot is estimated at 0.3% (Rogola et al., 1974). According to Kelikian (1974), about 60 cases of mirror hands have been reported. The instances of foot duplication are scarcer. The first authentic documented case was described by Jackson in 1853.

Case Report

A ten year old girl with eight toes in her right foot was brought because of the abnormal appearance and difficulty in wearing shoes. (Fig. 1 and 2). She used to wear sandals by gripping the strap between the two great toes. The family history was negative for consanguinity or other congenital anomalies. There was no history of intragestational drug ingestion or illness. The mother had a full term normal delivery. The right foot of the patient was broader than the left with a digital formula 5, 4, 3, 2, 1, 1, 2, 3 and there was also partial syndactyly between the supernumerary second and third toes. There was normal range of movements at all the toe joints.

Skiagrams of the right foot showed fusion of both the medial cuneiform bones with partial bifurcation which were also fused to the bases of both the first metatarsals and the supernumerary intermediate cuneiform. It was also fused with the cuboid and the lateral cuneiform (Fig. 3). The first metatarsal of the apparently normal side was short with its base pushed laterally. The other tarsal bones and leg bones were normal.

The extra medial three toes were amputated through a racquet shaped incision and though the necessity of needing extra skin was kept in mind, a resultant raw area measuring 12 cm × 5 cm had to be covered by a sheet of split skin graft. It would have been preferable to reflect skin flaps by an incision along the medial border of the foot to give proper shape to the foot (Fig. 4). The patient has been provided with properly fitting boots and is able to walk comfortably. Dissection of the amputated segment did not reveal any abnormal neurovascular or tendinous structures.

The unusual features about this patient were :

1. Only one foot was affected, when usually the condition is described as being bilaterally symmetrical.
2. The leg bones were normal, when usually there is associated tibial hemimelia or hypoplasia.

Discussion

Development of the zygote which weighs only 0.005 mg into a highly differentiated newborn of more than 3000 g requires about 44

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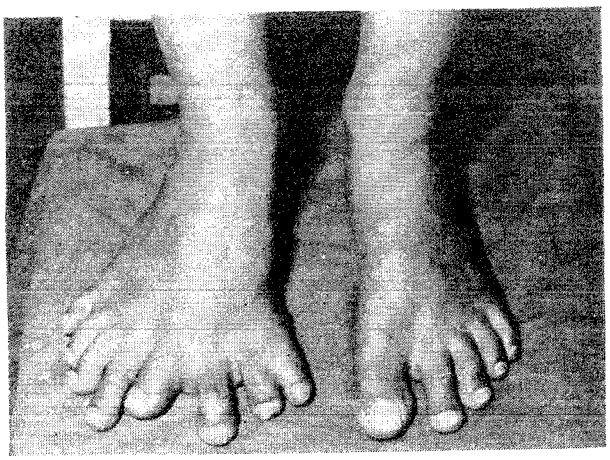

Fig. 1. Dorsum of both feet showing duplication (Right Foot).

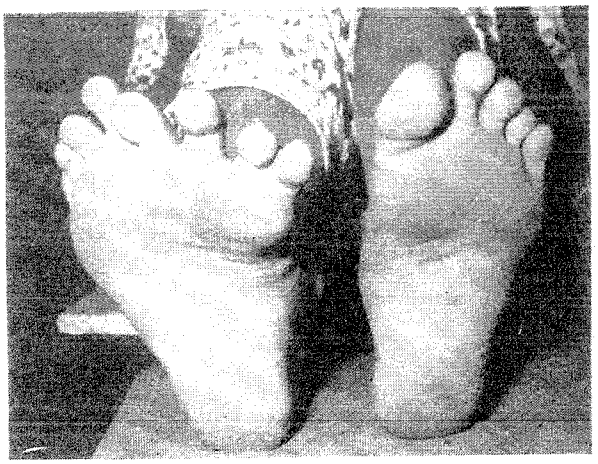

Fig. 2. Showing the appearance of the soles in the patient.

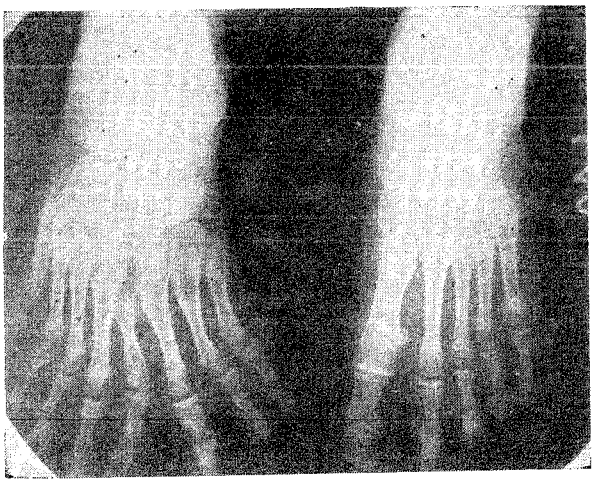

Fig. 3. Pre-operative skiagrams.

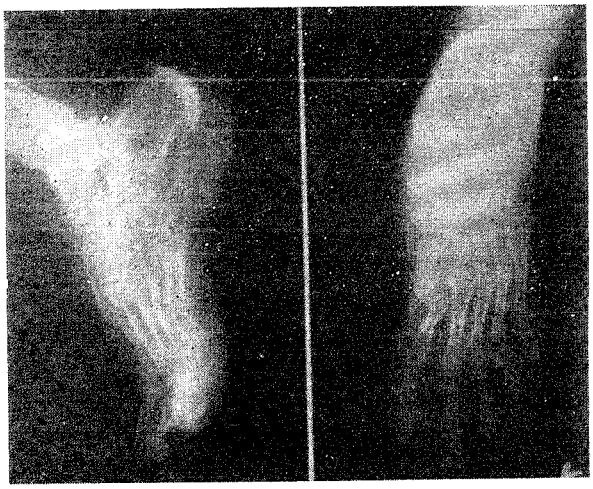

Fig. 4. Post-operative skiagrams.

successive cell divisions that are directed by the genes contained in the zygote and influenced by the intrauterine and extrauterine environments. Limb buds are first seen when the embryo is about 4 mm in length and about 4 weeks in age. Almost as soon as the limb bud is visible, an ectodermal thickening is formed on its ventral aspect. Within a few days the thickening gives rise to an apical ectodermal ridge which has been the object of numerous experimental investigations.

It is difficult even in experimental work to ascertain the time of origin to limb defects but it is possible to ascertain termination periods that sometimes permits exclusion of an event as the cause of the deformity. Malformations of excess, such as polydactyly or duplication, date back at least to the embryonic period, when they arise as mesodermal structures. Thus in a 23 mm (8 weeks old) embryo, the digits are well formed and their number is determined (Warkany, 1971).

The skeletal elements of the limb bud are first seen as mesenchymal condensations which chondrify in the fifth week in a definite sequence and ossification follows (Swanson, Barsky

and Entin, 1968). Tarsal fusions may occur between almost any two elements and probably manifests as an absence of joint cavitation and chondrification of the embryonic 'interzone' (O'Rahilly, 1953). Hence, anomalies in which the number of skeletal elements is increased originate under the influence of factors that act before the seventh post ovulatory week. After the seventh post ovulatory week the remainder of intrauterine life is termed the foetal period. Experimental work in mouse on "Cytochemical predifferentiation" i.e. the histochemical detection and localization of certain substances prior to morphologic differentiation has indicated that the perichondrium, the muscles and the tendons arise by a centripetal migration from the superficial mesenchyme, while the skeletal blastemata appear in the depth of the limb (Milaire, 1967).

Extreme cases of polydactylia are rare. Gould and Pyle (1901) quoted Voight, who recorded a case with 13 fingers on each hand and 12 toes on each foot and Saviard who saw an infant with 40 digits, ten on each limb. Stein and Bettman (1940) reported a patient with 15 fingers and 13 toes. Extreme cases of this type often involve duplication of wrist and forearm. Incomplete diplopodia or diplocheria with high degree of polydactyly usually are associated with hypoplasia or absence of tibia and radius. O'Rahilly (1951) considered duplication of ulna and the distal components of the ulnar ray as the most constant feature of mirror hands, conversely, the components of the radial ray are absent.

O'Rahilly (1953) noted extensive tarsal anomalies in limb duplication. Bilateral duplication of the ulna and fibula was reported by Lewin et al. (1964). The polydactyly in the hands was concealed by marked syndactyly, but in each foot ten toes could be identified. According to Warkany (1971) polydactylies of more than six digits are usually associated with serious or even lethal malformations; but polydactyly of high degree can be compatible with life. Gruber (1958) described a woman shown in country fairs in Germany whose legs were so short and deformed that she had to walk on all four extremities. She was named 'Lionella'; her hands showed merely duplication of the thumbs, on the left foot she had eight digits of the formula 4, 3, 2, 1, 2, 3, 4, and on the right foot seven digits with the formula 4, 3, 2, 2, 3, 4, 5. Potter and Craig (1975) described a case with seven digits on all four extremities and observed that excessive division of the terminal part of the limb buds is usually bilaterally symmetrical, in contrast to reduction in numbers, which is often unilateral. According to Birchjensen (1949) all bilaterally symmetrical abnormalities and those in which the abnormalities consist of an excessive number of parts are endogenous.

Neel (1961) has estimated that not more than 20 percent of congenital defects in man result from simple Mendelian dominant or recessive inheritance and a further 10 percent from extrinsic teratogens such as viruses, drugs, radiation etc. About 10 percent may be the result of major chromosomal abnormalities. In about 60 percent of congenital developmental abnormalities no known or demonstrable cause can be found. Their aetiology is presumably multifactorial and the result of combined genetic and extrinsic factors.

Apart from these abnormalities of the growth process, congenital abnormalities may result from abnormalities of cellular degeneration during ontogenesis (Saxen and Rapola, 1969). Several theories of embryogenesis have been suggested:

1. The apex of the developing limb bud retains an intrinsic capacity to form distal portions of the limb as it progressively lays down developing portions of the limb in an orderly proximodistal sequence (Saunders, 1948). Division of the ectodermal cap at any time during embryogenesis can cause duplication defects ranging from complete duplication of digits to simple polydactyly.
2. Gabriel (1946) suggested on the basis

of experimental work on polydactylous fowls that accessory digits may represent the partial development of duplicated limb fields which is inhibited in its differentiation by the dominance of the primary, normal limb field.

3. It has been hypothesized that the human foot has been derived from the 15 odd tarsal bones, attributed to the earliest labyrinthodant amphibian e.g. the lobefin of the barramunda, an Australian lung fish (Giannestras, 1973). The concept of phylogenetic polydactylism, based on the principle that 'Ontogeny recapitulates phylogeny' is an example according to which the normal five fingered hand of man is a derivative of an ancestral limb possessing six or more digits. A hand with supernumerary fingers represents an atavism (Kelikian and Doumanean, 1957).
4. O'Rahilly (1951) described supernumerary digits as a reversal of symmetry by a second growth centre.

REFERENCES

1. Birch-Jensen, A. : Congenital deformities of the upper extremities. Odense Denmark, 1949.
2. Gabriel, M. L. : The effect of local applications of colchicine on leghorn polydactylous chick embryos. J. of Experimental Zoology. 1946; 101 : 339-350
3. Giannestras, Nicholas, C.: Foot disorders. Second Edition. Philadelphia, Lea and Febiger, 1973.
4. Gould, C. and Pyle, W. L.: Anomalies and curiosities in Medicine. Philadelphia, W. B. Saunders Company (1901) quoted by Warkany, 1971.
5. Gruber, G. B. (1958): quoted by Warkany Josef, 1971.
6. Kelikian, H. : Congenital deformities of the hand and forearm. Philadelphia, London, Toronto, W. B. Saunders Company, 1974.
7. Kelikian, H. and Doumanean, A.: Congenital anomalies of the hand. Part-I. Journal of Bone and Joint Surgery, 1957; 39(1) : 1002-1015.
8. Lewin, G. A., Favrian, J. C. and Labella, P.: Bilateral absence of radius and tibia with bilateral reduplication of ulna and fibula—A case report. Journal of Bone and Joint Surgery, 1964; 46 (A): 137-142.
9. Milaire, J. (1967): quoted by Giannestras, 1971.
10. Neel, R. (1961): quoted by Saxen and Rapola, 1969.
11. O'Rahilly, R.: Morphological patterns in limb deficiencies and duplications. American Journal of Anatomy 1951; 89 : 135-191.
12. O'Rahilly, R.: Survey of carpal and tarsal anomalies. Journal of Bone and Joint Surgery, 1953; 35(A): 626-639.
13. Potter, Edith Louise and Craig, John, M.: Pathology of the fetus and Infant. Third Edition. Chicago Year Book Medical Publishers, 1975.
14. Rogola, E. J., Wynne-Davis, R., Littlejohn, A. and Gormley: Congenital limb anomalies: frequency and actiological factors; data from the Edinburgh register of the new born (1964-68) Genetics 1974; 11: 222-230.
15. Saunders, J. W. G.: The proximo-distal sequence of origin of parts of the chick wing and the role of ectoderm. Journal of Experimental Zoology, 1948; 108: 363-404.
16. Saxen, E. and Rapola, J. : Congenital defects. Developmental Biology Series. New York. Holt Rinehart and Winston, 1969.
17. Stein, H. C. and Bettman, E. H.: Rare malformation of the arm, double humerus with three hands and fifteen fingers. American Journal of Surgery, 1964; 50: 336-340.
18. Swanson, Alfred, B., Barsky, Arthur, J. and Entin, Martin, C.: Classification of limb malformations on the basis of embryological failures. Surgical Clinics of North America, 1968; 48: 5-15.
19. Warkany, Josef: Congenital Malformations. Chicago Year Book Medical Publishers, 1971.

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