A Rare Case: Co-Existence of Simultaneous Anterograde and Retrograde Ulnar Polydactyly

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Dear Editor,

Ulnar polydactyly has been described as one of the most common congenital anomalies of the upper extremity. It manifests itself as a supernumerary digit that is rudimentary and is represented by a small appendage, loosely attached to the ulnar border of the hand mostly. Several classification systems exist for ulnar polydactyly. Here, we report a rare case of postaxial polydactyly, in which well-formed digits were oriented in opposite directions.

A 5-year-old child was brought to the plastic surgery clinic for the evaluation of bilateral ulnar polydactyly. The patient had no family history of bilateral ulnar polydactyly. On physical examination, the patient was noted to have a mobile supernumerary ulnar digit orientated from proximal to distal on the left and a similar extra digit orientated in the opposite direction on the right (Figure 1). At operation, the abductor digiti minimi insertion was observed at the base of the proximal phalanx on the right side, but not on the left side. Supernumerary digits were removed and the neuro-muscular bundles were ligated. The abductor digiti minimi tendon was separated from the origin and transferred to the ulnar side of the fifth digit on the right side. Also, collateral ligaments were reconstructed.

Extend of ulnar polydactyly varies from a completely formed digit to a single phalanx or skin tag. Ulnar polydactyly can be classified on the basis of genetic, morphologic, and clinical implications. The most commonly used classification system belongs to Temtamy and Mc Kusick1, which defines type A as a well developed digit with skeletal articulation and type B as a small, poorly developed digit that represents as a skin tag. However, many surgeons have considered this classification simplistic and prefer using other classification systems such as Stelling2, Rayan and Frey3, modified Rayan classification4, and our proposed classification system5. A single case of ulnar polydactyly with retrograde development and fusion has been reported previously in English literature.6 The duplication of on the right side in our case most closely approximates type IVb according to our classification system and Rayan–Frey classification type III, but a triphalangeal digit and greater than 90° angle of development. We assume that the insertion of the abductor digiti minimi may be the cause of a greater than 90° angle extension on the right side.

Our case represents an unusual form of ulnar polydactyly that was treated using similar surgical principles of ulnar polydactyly. Currently, five genetic loci for isolated ulnar polydactyly in humans have been identified. The genetic loci are PAP-A1, PAP-A2, PAP-A3, PAP-A4, and ZNF 141. However, the genetic and molecular bases for the embryologic development of ulnar polydactyly are just beginning to be understood.

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REFERENCES