

Case Report

Polydactyly of the Foot Diagnosed from a Minor Nail Problem

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Polydactyly is one of the most common foot congenital anomalies. It is often detected immediately after birth, but the diagnosis can sometimes be delayed if the symptoms are less evident. A 2-year-old girl with a complaint of recurrent bleeding from the right toenail was diagnosed with foot polydactyly. She underwent corrective surgery, and her family was satisfied with the outcome. Although the diagnosis of polydactyly may be difficult in cases with minor nail problems, further radiographic evaluation will be needed for the improvement of the patient's quality of life.

Key words: polydactyly, hyperdactyly, hexadactyly

Polydactyly is one of the most common congenital anomalies of the limbs and is often noticed at birth. Diagnosis is relatively easy in patients with apparent cases, but slight polydactyly that appears normal at a glance, such as in the present case, may be overlooked. These patients should not be ignored, since they may have other syndromic diseases, or their daily lives may be affected by superficial problems such as needing different two shoe sizes or bleeding and pain at the duplicated toe.

In the present case, the patient had only one symptom, *i.e.*, recurrent bleeding from a toenail. Although the diagnosis of polydactyly was delayed until 2 years of age, the patient and her family were relieved of the problem after corrective surgery.

Case Report

A 2-year-old girl presented to the hospital with a complaint of occasional bleeding from the right toenail.

Although she had visited other hospitals several times for the same symptom, further follow-up was not completed. She was born healthy and had no apparent past medical or family histories. Her mother reported that her right toenail occasionally bled after a minor injury since she began walking at approximately one year of age. Her toe appeared normal except that the second toenail of the right foot was wider than the third to fifth toenails and was separated in the middle (Fig. 1A). No active bleeding was noted. Neither joint instability nor functional impairment of the foot was detected. She had no other congenital anomalies upon physical examination, and blood tests and a chest plain radiograph were normal. A radiograph of the right foot revealed a duplicated second distal phalanx (Fig. 2). Genetic tests were not performed.

The patient was referred to a plastic surgeon and diagnosed with polydactyly of the foot on the first visit. The surgical plan was to preserve the medial side of the second distal phalanx because it was larger than the lateral side and located in good alignment with the

middle phalanx. Corrective surgery was then performed, in which the middle phalanx was found to be bifurcated. Its lateral side was resected, and the medial side was fixed to a medial distal phalanx with a Kirschner-wire. The clinical course after surgery was satisfactory (Fig. 1B,C). The wire was removed on postoperative day 13, and the patient was discharged on postoperative day 15.

Discussion

Polydactyly, also known as hyperdactyly or hexadactyly, is the most common congenital anomaly of the limb and is often observed at birth. Its overall incidence is 0.3-3.6/1,000 live births [1], and it is more common in African-Americans. The incidence of foot polydactyly in Japan is 0.4-1.0/1,000 [2,3], but precise data for other ethnic groups are not available. This might be because in Western countries there is less time spent without shoes, or because there is no indication for surgical treatment for slight cases.

The phenotype of polydactyly varies from a minor cutaneous protuberance on the lateral aspects of a digit to the doubling of a single phalanx. Polydactyly is classified into three groups: preaxial, central, and postaxial (the “big” toe, the second to fourth, and the fifth digits). The postaxial type is the most common in polydactyly of the foot, whereas the central is the rarest, as in our patient’s case. Polydactyly is significant because of other occasionally coexistent congenital anomalies and their effects on the quality of life. Most unilateral

polydactylies occur sporadically.

The detection rate of gene mutations in isolated preaxial cases is low [4], and that in central cases is uncertain. The familial types are mostly bilateral and inherited in an autosomal dominant manner. At least 10 genes causing non-syndromic polydactyly have been identified, including *ZNF141*, *GLI3*, *MIPOL1*, *IQCE*, *PITX1*, and *GLI1* [5]. Polydactyly may occur in the context of syndromes such as the Ellis-Van Creveld syndrome, trisomy 18, and trisomy 21 [6].

Plain radiographs should be obtained for the diag-



Fig. 2 Radiograph of the right foot showing the duplicated second phalanx. The medial bone is larger than the lateral bone.



Fig. 1 A, Separated second nail on the right foot of the patient, a 2-year-old girl; B, Removal of the lateral side of the duplicated phalanx; C, Postoperative view (2 weeks after the operation) with a good cosmetic result.

nosis of polydactyly to understand the accurate anatomy and to decide on the surgical plan. Surgery is usually planned around 1 year of age because of the lower risk of anesthesia and the lower effects on development and walking ability at this age. Nevertheless, it can be performed at any age with good outcomes for functionality optimization, pain relief, shoe fit improvement, and cosmetic enhancement [7]. The surgical approach includes the ablation of the duplicated digit and reconstruction. Complications include hallux valgus, residual angular deformity, and a widened foot, particularly in central polydactyly.

In the present case, polydactyly was diagnosed at 2 years of age because the patient had no other remarkable symptoms except for a separated toenail. Nail bleeding has not recurred since the operation, and she and her family are satisfied with the outcome. Patients with polydactyly occasionally suffer from other congenital anomalies. Patients may be encouraged to improve their development and quality of life through surgery. Although it may be difficult to diagnose polydactyly in patients with minor nail problems, further radio-

graphic evaluation is needed for these patients.

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