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Accurate Osteotomy for the Treatment of a Rare Case of Postaxial Polydactyly of the Foot That Originated From a Deformed Calcaneus Using a 3D-Printed Guiding Plate

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ABSTRACT

Polydactyly is a common congenital deformity of the foot that can be categorized as preaxial, central, or postaxial. Current treatments involve resecting the supernumerary toe(s) and repairing the normal toe(s) and soft tissue. Here, we present the first published report describing a very rare case of polydactyly of the foot, in which the supernumerary toe originated from a deformed calcaneus, which formed an abnormal bony bump. Preoperatively, 3-dimensional (3D) computed tomography reconstruction images revealed the morphology of the deformed toe and calcaneus, and gait analysis showed an abnormal weightbearing zone in the left foot. The 3D printing technology and a specially designed 3D-printed guiding plate were used for osteotomy. Postoperatively, x-ray showed that the calcaneus had a normal shape and surface, whereas gait analysis showed that the left foot was uniformly loaded and the area of pain was eliminated. Our findings should raise awareness among clinicians that a 3D-printed guiding plate is useful in the treatment of such an unusual deformity.

Polydactyly is one of the most common congenital diseases of the foot (1,2), with an estimated incidence of 0.37 to 1.2 in 1000 live births (3). Polydactyly of the foot can be classified into 3 types according to the position of the supernumerary toe(s): preaxial, central, or postaxial (4). Among these, postaxial polydactyly of the foot is the most common (4). Here, we describe a very rare case of postaxial polydactyly of the foot in which the supernumerary toe originated from the calcaneus, which was deformed, appearing as an abnormal bony bump. To the authors' knowledge, this is the first published report describing such a deformity. Current treatments for polydactyly of the foot involve resecting the supernumerary toe(s) and repairing the normal toe(s) and soft tissue, and are individualized according to the deformity (5–7). Surgery aims to improve the appearance of the foot while maximizing the patient's foot function postoperatively.

Three-dimensional (3D) printing technology has gained increasing popularity in medicine in recent years and has achieved good outcomes in the treatment of serious orthopedic diseases (8,9). In the current case, because the deformity of the calcaneus was complex, we determined that conventional surgery could not provide an osteotomy path that would restore the normal anatomical shape of the calcaneus; therefore, we used 3D printing technology and designed a suitable 3D-printed guiding plate to achieve an accurate osteotomy and satisfactory postoperative outcomes.

Case Report

A 56-year-old male presented to our clinic with an isolated extra toe in the lateral aspect of the left foot and associated pain in the lateral heel (Fig. 1). The patient had no other medical illness or congenital deformity and no family history of polydactyly. The supernumerary toe did not move autonomously, which made it difficult for the patient to wear shoes without cutaneous irritation; the other joints of this foot functioned normally. The patient was suffering from a painful callus on the supernumerary toe and heel. The American Orthopaedic Foot and Ankle Society (AOFAS) score, which has been widely used to evaluate foot and ankle function, was 71. The visual analog scale (VAS) pain score was 4. Radiographs of the patient's left foot and the supernumerary sixth toe showed separate distal, medial, and proximal phalanges, which originated laterally from a deformed calcaneus (Fig. 2). Gait analysis indicated an abnormal weightbearing zone located on the area of

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Fig. 1. Preoperative clinical appearance of the left foot. (*A*) Posteroanterior. (*B*) Planta.

the supernumerary toe and the lateral bump of the calcaneus, which was consistent with the patient's chief complaint.

The patient requested surgical treatment because he was experiencing significant pain. There were no contraindications to elective surgery. Polydactyly surgery usually involves resection of the extra toe and reconstruction of the soft tissue to restore normal anatomy and a functional foot; however, this patient's supernumerary toe originated laterally from the calcaneus, and there was an abnormal weightbearing zone on the lateral calcaneus. As a result, preoperative planning considered resection of the extra toe, reconstruction of the soft tissue, and repair of the bony structure of the abnormal calcaneus to provide a normal functional calcaneus without pain on weightbearing.

We treated this patient using our clinical experience of 3D printing technology based on the successful use of this approach for complex fractures, deformity, joint replacement, and bone tumor resection at our hospital. We obtained a data set for digital 3D modeling of the calcaneus and subsequent printing from the computed tomography scan. We designed a surgical guiding plate adjusted to the surface of the calcaneus. An osteotomy groove was designed on a suitable area of the guiding plate and adjusted to obtain an optimum path for osteotomy (Fig. 3).

The patient received lumbar anesthesia, and a lower limb pneumatic tourniquet was used to reduce bleeding. First, the supernumerary toe was resected; no tendon was found near the extra phalanges. Then, the calcaneus was exposed and the guiding plate was affixed to it with 3 K-wires. Another K-wire served as a guide for drilling a groove into the bone. An osteotome was used to resect the abnormal bone. After the bone was resected, the calcaneus was trimmed using a bone file and bone wax sealing material was brushed on the surface (Fig. 4). Postoperative x-ray showed that the calcaneus had a normal shape and a regular surface (Fig. 4). Postoperatively, the patient was non-weightbearing for 6 weeks, during which time he was placed in a cast to ensure that the bone was fully repaired. At the 6-month postoperative follow-up, the AOFAS score was 92 and the VAS pain score was

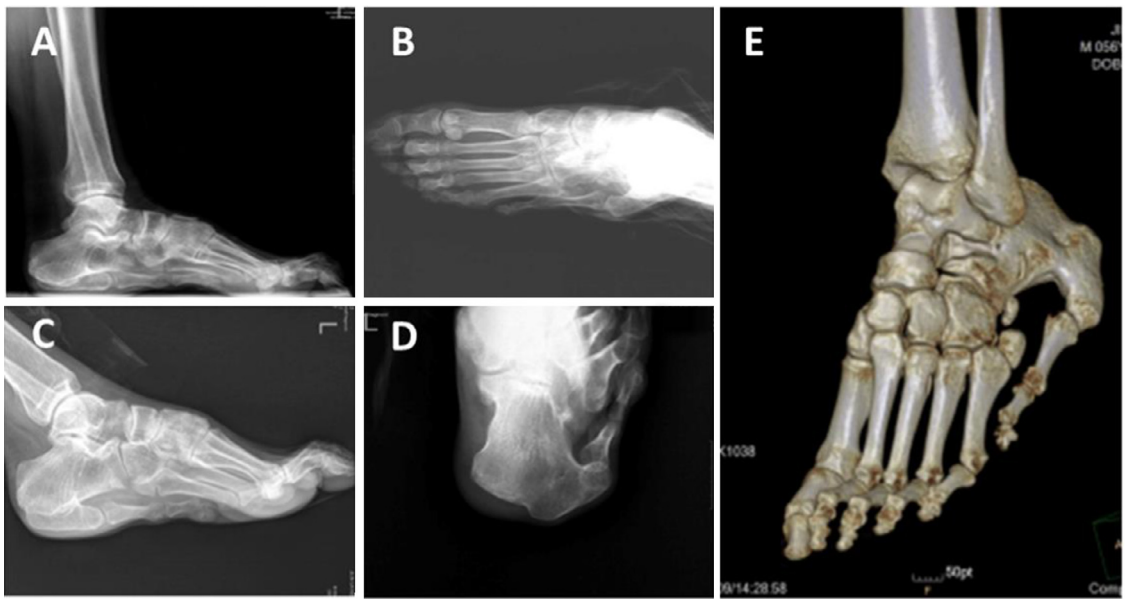

Fig. 2. Preoperative images of the left foot. (*A*) X-ray: weightbearing lateral view. (*B*) X-ray: weightbearing frontal view. (*C*) X-ray: lateral view. (*D*) X-ray: calcaneal axial view. (*E*) Three-dimensional computed tomography.

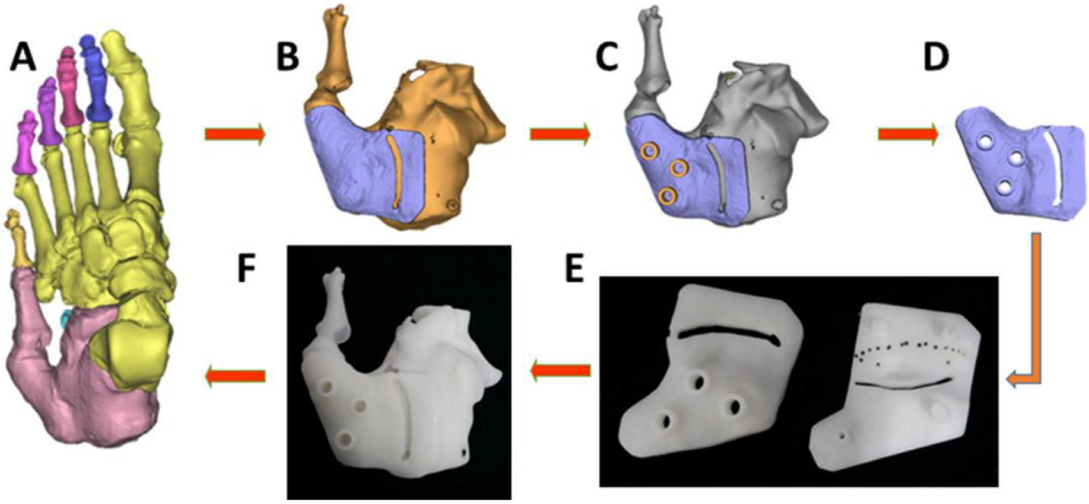

Fig. 3. Design of the three-dimensional (3D)-printed guiding plate. (*A*) 3D reconstruction of left foot. (*B*) Confirming the shape of osteotomy guiding plate. (*C*) Confirming the location of osteotomy groove and fixing holes. (*D*) 3D imaging construction of guiding plate. (*E*) 3D-printed guiding plate. (*F*) Matching evaluation.

Fig. 4. Postoperative appearance of the left foot. (*A*) X-ray: frontal postoperative view. (*B*) X-ray: oblique view. (*C*) Clinical appearance: front. (*D*) Clinical appearance: lateral.

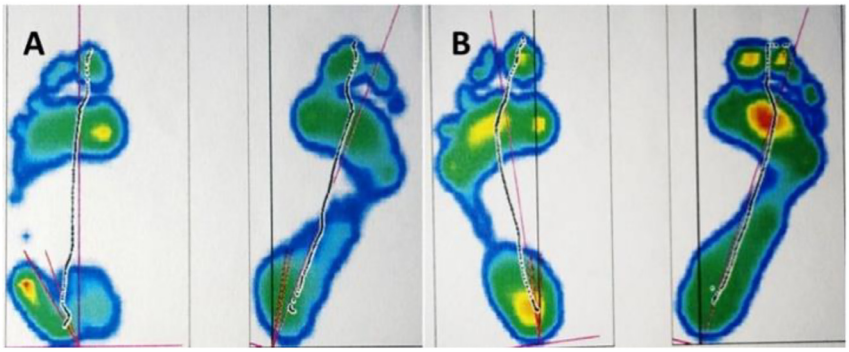

Fig. 5. Gait analysis. (*A*) Preoperative. (*B*) Postoperative.

0. Gait analysis showed that the left foot was uniformly loaded and the area of pain had been eliminated (Fig. 5). One year after the operation, the AOFAS score was 98 and there was no obvious discomfort (Table 1).

Discussion

Polydactyly is a common congenital deformity of the foot, occurring with an incidence that varies among racial groups from 0.37 to 1.2 in 1000 live births (3) but with varying frequency among different populations. Polydactyly of the foot is characterized by the presence of ≥6 toes, with or without duplication of the corresponding metatarsals or phalanges; tarsal deformity may be found in some severe cases (4).

Clinical complaints in patients with polydactyly of the foot include pain, discomfort when walking, unsatisfactory appearance, and psychological issues (10). Surgery is recommended in early childhood, before a child learns to walk, because the bone structure of children in this age group is adaptable and able to compensate for surgical intervention (11). In China, because of local economic and technical limitations, many patients in rural or remote areas are able to seek medical advice only when they are adults and the skeleton has matured. Preoperative decisionmaking is complex in these patients because surgeons need to improve the appearance of the foot while maximizing the patient's foot function postoperatively.

Technology surrounding 3D printing has gained increasing popularity in medicine and has been used successfully for tissue repair, arthroplasty, tumor therapy, and efficient drug delivery (8,9). In orthopedics, 3D printing can assist in preoperative planning for the surgical treatment of deformities. In these patients, 3D printing software can be used to design and print a model of the target tissue, which is then used to develop a patient-specific template for guiding surgery. The 3D-printed guiding plate has many advantages, including improving the accuracy of osteotomy, shortening operation time, and reducing surgical trauma compared with traditional approaches (9,12). In a cadaveric study, Chen et al (13) evaluated the accuracy and feasibility of an individualized thoracic pedicle screw guiding plate produced by 3D laser printing. They found that the guiding plate was accurate and convenient, concluding that this represented a new method for accurate placement of thoracic pedicle screws (13). Fu et al (14) found that guiding plates produced by various 3D printing techniques were useful for resection of bone tumors and reconstruction. Other studies have reported the application of 3D printing in craniofacial plastic surgery and achieved good results (15,16).

In the present case, there was a weightbearing zone located on the supernumerary toe and the lateral part of the calcaneus (Fig. 5). Here, we could see the formation of the corpus. Preoperative imaging indicated deformity of the calcaneus. Because the shape of calcaneus was complex and there was an issue with weightbearing, we determined that conventional surgery could not provide an osteotomy path that would restore the normal anatomical shape of the calcaneus and provide a functional heel without pain caused by abnormal weightbearing. Thus, we designed a model of the patient's left foot. The initial osteotomy guiding plate was designed and printed according to the anatomical characteristics of the supernumerary toe and calcaneus. We constantly adjusted the guiding plate. The most suitable plate was printed (Fig. 4) and used to perform a successful osteotomy. The calcaneus was repaired to its normal shape to balance loading of the foot and avoid the discomfort caused by abnormal weightbearing. The patient recovered well, and postoperative x-rays showed that the shape of the calcaneus was restored (Fig. 4). Six months after surgery, the VAS pain score had decreased to 0 from a preoperative value of 4, and the AOFAS score had increased to 92 from preoperative value of 71. One year after surgery, the AOFAS was 98 and there was no obvious discomfort.

In conclusion, this is a rare case of polydactyly of the foot originating from a deformed calcaneus. We used 3D printing technology to design a guiding plate and carried out an accurate osteotomy. Postoperative gait analysis indicated that weightbearing was balanced. After more than 1 year of follow-up, the patient did not show significant pain in the foot. Satisfactory results were obtained in this patient. Our findings should raise awareness among clinicians that a 3D-printed guiding plate is useful in the treatment of such an unusual deformity.

Table
American Orthopaedic Foot and Ankle Society score and visual analog scale pain score

	AOFAS	VAS
Preoperation	71	4
6 months postoperation	92	0
1 y postoperation	98	0

Abbreviations: AOFAS, American Orthopaedic Foot and Ankle Society; VAS, visual analog scale.

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