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ARTICLE

Long-term outcomes after phalangeal distraction lengthening in patients with constriction band syndrome

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ABSTRACT

We investigated the long-term post-operative outcomes, more than 10 years after surgery, of distraction lengthening of phalanges in patients with congenital constriction band syndrome. Five shortened digits of two patients with congenital constriction band syndrome underwent distraction lengthening of the proximal phalanges using external fixator for the reconstruction of grasp and pinch at age of 53 and 41 months. The post-operative followed-up was 126 and 124 months. Subjective evaluation of hand function was performed using the tape measure method and Functional Dexterity Test pre-operatively and at 1 year after surgery. The parameters for proximal phalangeal distraction and the changes in the length ratio of the phalanges were evaluated. Grasp and pinch function was markedly improved after phalangeal lengthening. Regarding the radiographic findings, the length of distraction was 9.7 mm, % increase of lengthening: 52%, duration of distraction: 28 days, duration of fixation: 83 days, and healing index: 90 days. At the latest follow-up, the relative length of the proximal phalanx did not change significantly (−6%) during the 10 years after distraction lengthening. Patients with constriction band syndrome obtained favorable hand function and preserved those functions for more than 10 years after distraction lengthening of short phalanges using an external fixator.

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Introduction

Distraction lengthening of the digital rays using an external fixator is widely regarded as a useful treatment for shortened digits in patients with congenital hand anomalies. A number of studies demonstrated that distraction osteogenesis of phalanges and metacarpals is an effective technique for the establishment of pinch and grasp function [1–3]. Those studies obtained excellent results after the lengthening, however, most of the studies did not demonstrate long-term changes to the hands of the children as they grew after surgery, which is of importance as bone growth could differ between the lengthened and un-lengthened digits. In addition, external fixation is used less frequently in the phalanges [4,5] than in the metacarpals [1–3,6], with the major reason appearing to be the lack of a suitable external fixator for the small bones.

This report describes post-operative outcomes, more than 10 years after surgery, for distraction lengthening of phalanges in our cases with congenital constriction band syndrome using an external fixator.

Methods

Patients

Bone lengthening procedures were performed from December 2009 to June 2010 at our hospital for five proximal phalanges in two boys with shortened digits associated with congenital constriction band syndrome. The phalanges consisted of one index finger, one middle finger, two ring fingers, and one little finger. Associated anomalies in the contralateral hand and the right foot were observed in 1 patient. Neither of the patients had any generalized abnormalities, syndrome or other congenital anomalies of the limbs. The patients underwent distraction lengthening procedures for the phalanges at age of 53 and 41 months, respectively. Pre-operative problems included impairment of pinch and grasp function due to their short fingers. The post-operative follow-up period was 126 and 124 months, respectively.

Surgical technique

The distraction length of each phalanx was preoperatively decided by measurement of the finger length to enable the patient to achieve grasp and pinch function. The operation was performed according to the previous method [2]. Briefly, a slightly curved skin incision was made on the dorsal side of the affected fingers and the extensor tendon was retracted. The periosteum was incised longitudinally and retracted carefully at the intended osteotomy site. A unilateral external fixator (Hoffman II Micro lengthener, Stryker, Switzerland) and four half-pins with a diameter of 1.65 mm were used as the lengthening device. Under fluoroscopic control using an external fixator frame as a guide, two half-pins were inserted into the proximal phalanx, and the other two half-pins were inserted into the metacarpal. Those half-pin was placed 3–4 mm away from the epiphyseal plate to avoid distraction damage. After all 4 pins were inserted, transverse osteotomy was performed using an osteotome at the proximal of the proximal phalanx where was 5 mm proximal from the half-pin. The external fixator was adjusted and all clamps were firmly

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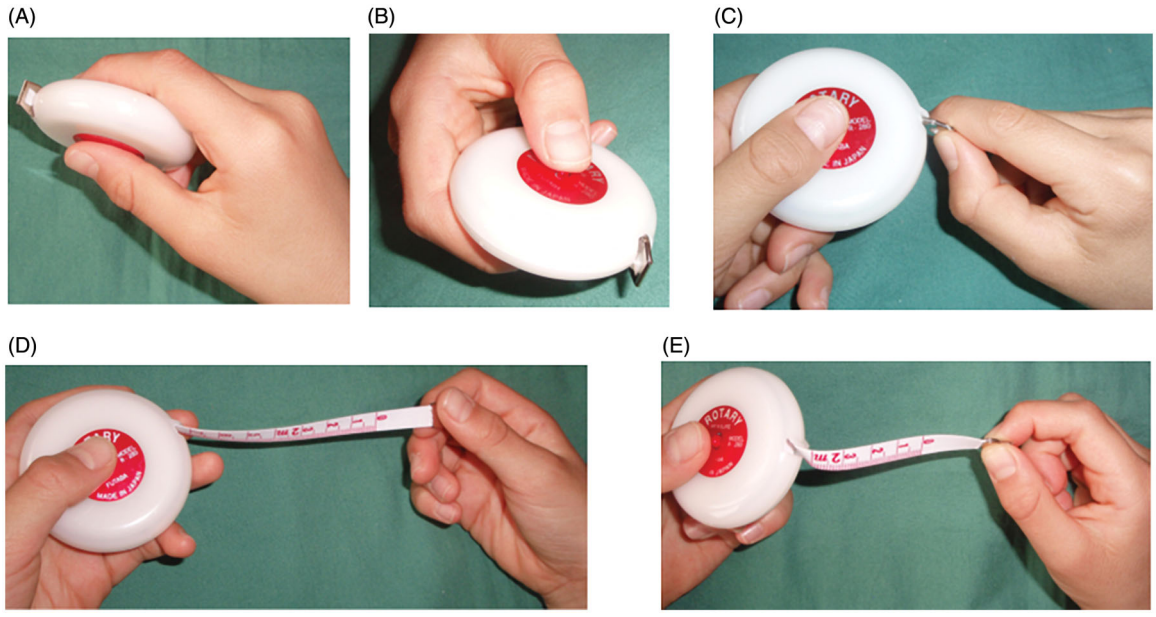

Figure 1. Evaluations of the impairment of pinch function using a tape measure. The scoring system using a tape measure were as follows: 1 point, grasp = 1 (A); push button = 2 (B); pinch the end of the measuring tape = 3 (C); pull out tape by side pinch = 4 (D); and pull out tape by pulp pinch = 5 (E).

secured across the metacarpophalangeal (MP) joints. The periosteum was sutured and the skin was closed. Lengthening was started 5 days after the operation. The patients were discharged for home recovery and resumed going to kindergarten as well as before being hospitalized. The patients' parents conducted lengthening at a rate of 0.25–0.5 mm twice a day. The radiograph findings of bone distraction were monitored once a week. Based on the callus formation, the rate of distraction was changed from 0.25 mm to 1.0 mm per day. When the desired length was achieved, the distraction was concluded. At the completion of the consolidation of the callus, the external fixator device and the pins were removed [2].

Assessment

Objective evaluation of hand function was performed using the tape measure method [7] and Functional Dexterity Test (FDT) [8] method pre-operatively and at 1 year after phalangeal lengthening of the fingers. Briefly, the impairment of grasp and pinch function was evaluated using a tape measure (Rotary Measure, Japan). The criteria were scored as follows: 1 point, grasp = 1; push button = 2; pinch the end of the measuring tape = 3; pull out tape by side pinch = 4; and pull out tape by pulp pinch = 5 (Figure 1). The highest score is 5 point and the lowest one is 1 point for hand function in each patient. Before starting the assessment, we pulled out the tape by pulp pinch and wound the tape by push button as a demonstration. As the movement for winding tape was tricky and attractive to the children, they took an active interest in the performance and desired to pull out the tape or push the button by themselves as we had done [7]. The FDT enables to the evaluation of not only grasp and release but also measures tripod pinch and in-hand manipulation functions even in young children. The FDT consists of a 16-hole wooden pegboard arranged in four rows of four, with each peg hole 3 cm deep, 2.5 cm in diameter, and located 2 cm from the adjacent peg holes. Each patient was asked to sit on a chair with the FDT pegboard on a table in front of them. The proximal edge of the pegboard was placed 10 cm from the edge of the table. The examiners were hand therapists, and they explained to each patient that the purpose of the test was to turn all the pegs over in the air as quickly as possible. The examiner showed each patient how to perform the task by turning over four pegs, then asked the patients to practice by turning over all the pegs on the board one time. The test was then performed, and the examiner recorded the time taken to turn over all the pegs [8–10]. Both tests are suitable for the evaluation of hand function in young children.

The bone length was measured on posteroanterior radiographs of the hand, and several parameters related to proximal phalangeal distraction, including length of distraction, percent (%) increase of lengthening, duration of distraction, duration of fixation and healing index [days required for 1 cm of lengthening; duration of fixation (days)/length of distraction (cm)], were measured based on the radiographic findings. In addition, the ratio of the proximal phalanx length between the lengthened finger and non-lengthened thumb [(proximal phalanx length of the finger/proximal phalanx length of the thumb) × 100] was measured immediately after and 10 years after the external fixator device was removed. We evaluated the changes in the length ratio of the phalanges over the 10 years after surgery. Post-operative complications that occurred during the first 6 months after surgery were noted.

Results

Both patients achieved grasp and pinch function within 6 months after phalangeal bone lengthening. The post-operative score (4.5 point) evaluated by the tape measure method, which reflected grasp and pinch function, was improved in comparison with the pre-operative score (1.5 point). The post-operative FDT score (32.4 s), which reflected the grasp and release, tripod pinch, and in-hand manipulation functions, was also improved in comparison with the pre-operative score (64.0 s). Regarding the radiographic findings, the mean value of the parameters for the 5 fingers was 9.7 mm (8–14 mm) for length of distraction, 52% (34–78%) for % increase of lengthening, 28 days (22–35 days) for duration of distraction, 83 days (56–88 days) for duration of fixation and 90 days (58–123 days) for healing index. The ratio of proximal phalanx

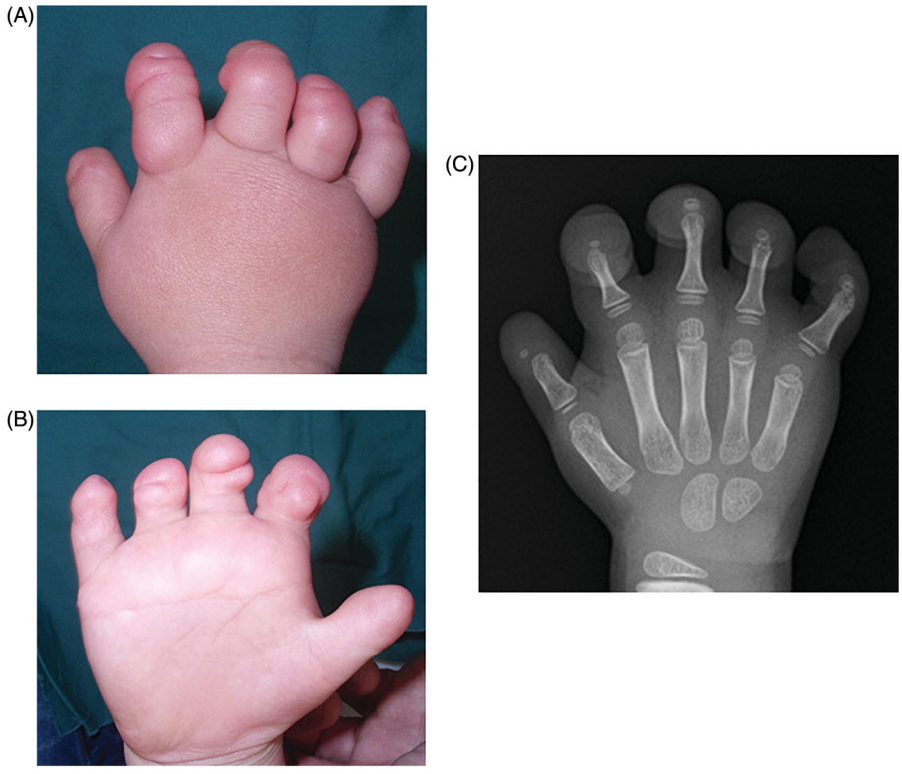

Figure 2. A 4-year old child with constriction band syndrome of the right hand. The index, middle, ring and little fingers are too short to allow pinch function (A,B) and show hypoplastic proximal phalanges without distal and middle phalanges (C).

length between the lengthened finger and the thumb did not change significantly [−6% (−10∼+1%)] over the 10 years after distraction lengthening.

Mild superficial infection around the half pin was found during external fixation after bone lengthening as a complication in one finger. It was resolved by treatment with oral antibiotics without removal of the fixation device. At the latest follow-up, neither patient had pain nor impairment in grasp and pinch function, and neither showed any restriction in the range of motion of the MP joint.

Case report

A 4-year old child presented with constriction band syndrome of the right hand. The index, middle, ring and little fingers were too short to allow pinching with the thumb and consisted of a floppy soft-tissue envelope with hypoplastic proximal phalanges without any distal or middle phalanges (Figure 2). There were no associated anomalies in the left hand and the lower limbs. Regarding the impairment of grasp and pinch function, the tape measure method score was 1 point, indicating that the patient could not pinch. First, the proximal phalanges of the middle and ring fingers were lengthened simultaneously by 8 mm and 10 mm respectively, to reconstruct pinch function (Figure 3(A,B)). After proximal phalangeal lengthening, side pinch became possible, with a score of 4 points (Figure 3(C)). The external fixator device and the pins were removed at 88 days after the surgery. At 6 months after the first operation, the proximal phalanges of the index and little fingers were lengthened simultaneously by 11 mm and 14 mm respectively to reconstruct stable grasp and improved hand function (Figure 3(D)). The external fixator device and the pins were removed at 81 days after the surgery. Within 6 months after the second operation, the patient achieved satisfactory pinch and grasp function due to the lengthened fingers (Figure 3(E)) and improvement in hand function based on the evaluation by FDT method, the scores for which improved from 63 s before to 38 s after bone lengthening. At the latest evaluation, the hand function including pinch was preserved, and the lengthened phalanges had not developed any significant relative shortening based on the radiograph findings (Figure 4). The post-operative level of the MP joint and hand shape were preserved in comparison with the pre-operative findings.

Discussion

A previous study demonstrated that distraction lengthening of the metacarpals in patients with brachymetacarpia provided good post-operative outcomes. On the other hand, the study recommended that patients with brachymetacarpia should undergo bone lengthening when they are 10–15 years of age, with one of the reasons given being that as the epiphyseal plate is almost closed, the lengthening of the final shortening of the metacarpal can be appropriately estimated [2]. However, regarding impairment of pinch and grasp function due to short digits in congenital hand anomalies such as symbrachydactyly and constriction band syndrome, it is important for a patient to have function reconstructed as a young child. In this report, the patients with constriction band syndrome underwent phalangeal lengthening at 3 and 4 years of age, respectively, and adequate lengthening of the phalanges was obtained to allow pinch and grasp function, which was preserved more than 10 years. Thus, we believe that phalangeal lengthening should be performed as soon as possible when small digital bones grow to a suitable size for external fixation.

The patients could pinch immediately after metacarpal lengthening but several patients could not pinch at the follow-up evaluation, because the different growth rates among the metacarpals caused the hand shape to change undesirably [4]. The effect of

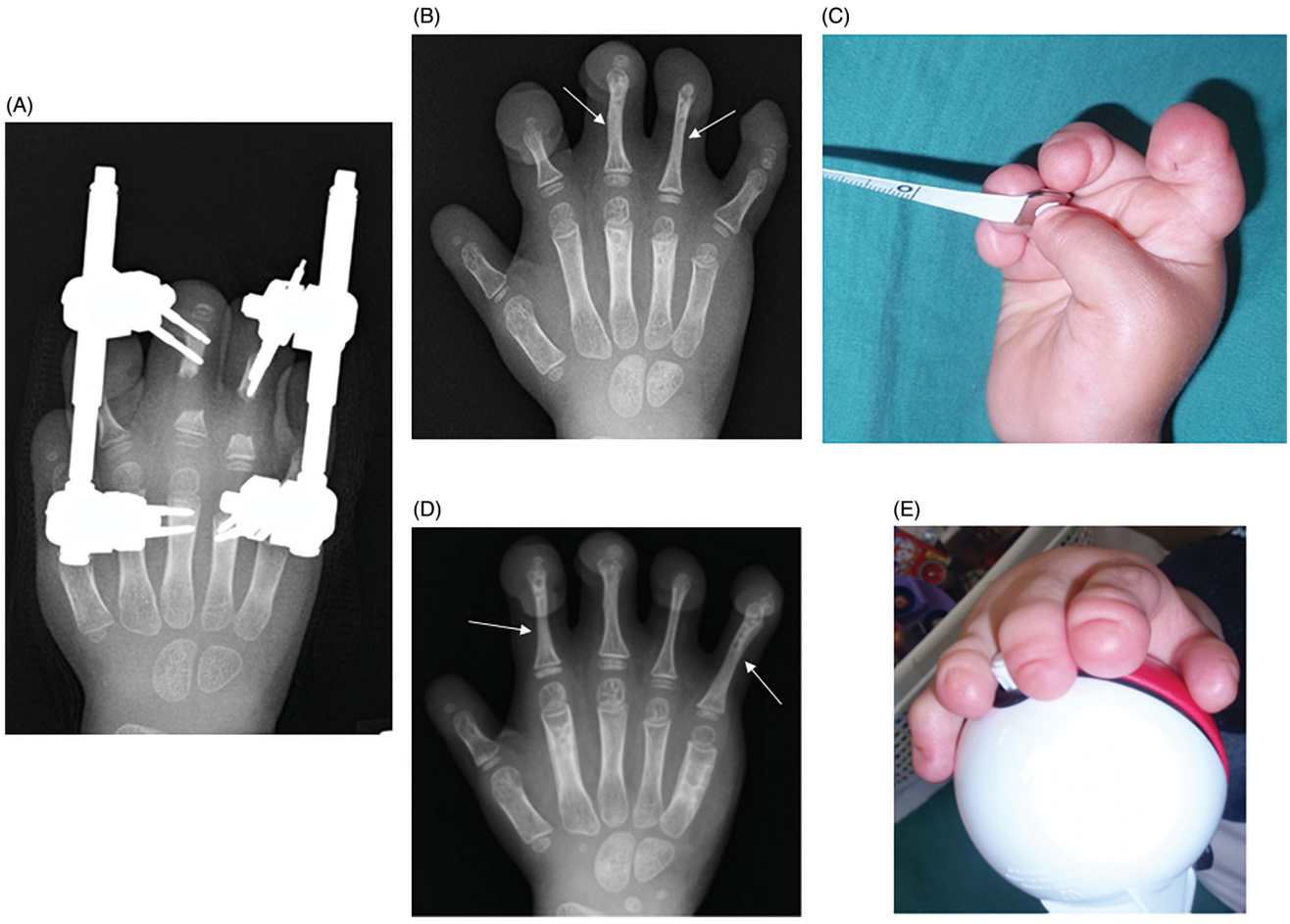

Figure 3. Distraction lengthening of the fingers to reconstruct grasp and pinch function. The proximal phalanx lengthening of the middle and ring fingers (white arrow) (A,B) enabled to pinch (C). Additional lengthening of the proximal phalanges of the index and little fingers (white arrow) improved grasp and hand function (D,E).

lengthening on growth cartilage has not yet been determined. Some studies have described the mechanical stresses on the growth cartilage, by which the lengthened metacarpal may be subjected to extra compressive forces [4,11,12]. In our cases, the lengthened phalanges in the patients with constriction band syndrome did not reveal any significant relative shortening in terms of bone growth. However, it is unknown that a lesion of growth plate is caused by the natural course of the disease or the mechanical stress. We, therefore, need further study to elucidate the relationship between lengthening and the growth potential of the growth plate.

Regarding the lengthening of small bones such as the phalanges, the fixation apparatus was placed across the MP joints due to the lack of a suitable external fixator for the phalanx [5]. It is unknown whether distraction force could affect joint components such as the ligaments or capsule during phalangeal lengthening. In the current cases, we did not find any damages to the MP joint components or growth plate, although further careful follow-up is needed. On the other hand, bone lengthening of the phalanges preserved the post-operative level of the MP joint in comparison with pre-operative level. Thus, we consider that phalangeal lengthening could preserve hand shape and function.

This study has several limitations. First, we acknowledge that the sample size was small although we focused on long-term post-operative follow-up more than 10 years. Second, assessment using the tape measure and FDT methods only reflects limited hand functions such as grasp and pinch, and those assessment methods have not been widely used. Third, there were no control cases without bone lengthening. Fourth, in one patient, the external fixator device and the pins in middle and ring fingers, or those in index and little fingers were removed at the same time, respectively.

In conclusion, we investigated post-operative outcomes after distraction lengthening of short phalanges using an external fixator in the patients with constriction band syndrome and followed-up these patients for more than 10 years. The patients obtained favorable hand functions including grasp and pinch and preserved those functions for more than 10 years after phalangeal lengthening.

Ethical approval

Informed consent was obtained from patients before enrollment in this study and the study was approved by the institutional review board of the university.

Authors' contributions

All the named authors were actively involved in the planning, enactment and writing up of the study.

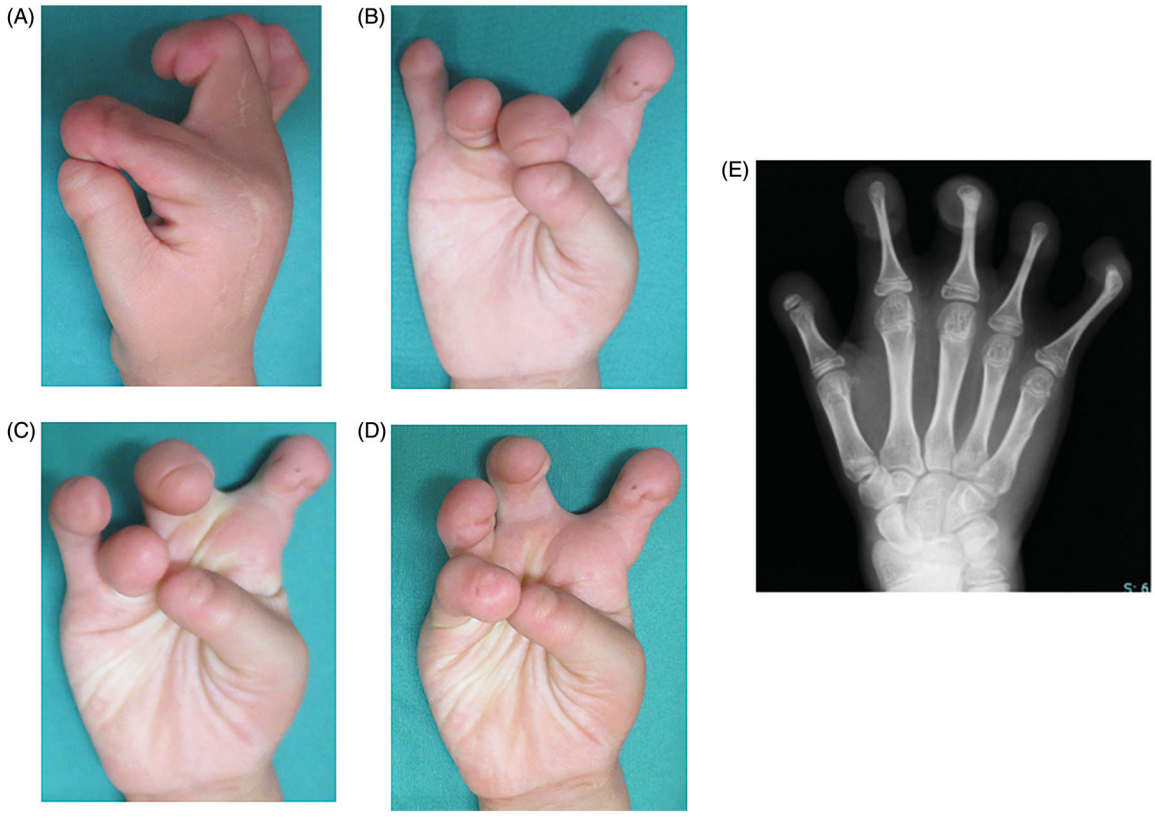

Figure 4. Postoperative f hand function and phalangeal length more than 10 years. At the latest evaluation, hand functions were preserved (A–D), and there was no significant relative shortening of the lengthened phalanges on the radiographic findings (E).

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Disclosure statement

The authors declare no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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