Lengthening of short bones by distraction osteogenesis—results and complications

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Abstract We performed bone lengthening surgery on 12 metacarpals and 14 metatarsals of 15 patients. The mean age for metacarpal and metatarsal lengthening was 14.5 (10–21) and 17.5 (10–25) years, respectively. We used a unilateral or a circular external fixator. The mean healing index of the metacarpals and metatarsals was 1.6 (1.1–2.3) and 1.6 (1.0–2.0) months/cm, respectively. The mean increase in metacarpal and metatarsal length was 17.6 (13–26) and 24.3 (20–30) mm, respectively. The functional scores of the metatarso-phalangeal (MTP) joint of lengthened metatarsals for the lesser toe were excellent in 12 and good in two cases based on the American Orthopaedic Foot and Ankle Society (AOFAS) scoring system. Complications were seen in six of the metatarsal lengthening cases including four angulations, one subluxation and one non-union. We conclude that the periosteum must be protected with percutaneous osteotomy and lengthening should be performed at a rate of 0.25 mm twice a day and should not exceed 40% of the original bone length (or >20 mm).

Résumé Nous avons réalisé une chirurgie d'allongement sur 12 métacarpiens et 14 métatarsiens chez 15 patients.

L'âge moyen était respectivement de 14.5 ans pour les métacarpiens (10 à 21) et 17.5 ans pour les métatarsiens (10 à 25). Nous avons utilisé un fixateur unilatéral ou circulaire. L'index moyen de consolidation a été de 1.6 mois/cm d'allongement, de 1.1 à 2.3 pour les métacarpiens et de 1.0 à 2.0 pour les métatarsiens. L'allongement moyen a été de 17.6 mm (13 à 26) pour les métacarpiens et de 23 mm (20 à 30) pour les métatarsiens. Le score fonctionnel de l'articulation métatarso-phalangienne a été excellent chez 12 patients, et bon chez 2 patients (score de l'AOFAS). Les complications ont été constituées par 4 angulations, 1 subluxation et 1 pseudarthrose parmi 6 allongements métatarsiens. Nous pouvons conclure que le périoste doit être protégé lors de l'ostéotomie percutanée mais la vitesse d'allongement doit être de 0.25 mm deux fois par jour. L'allongement ne doit pas excéder 40% de la longueur originale de l'os (plus de 20 mm).

Introduction

The incidence of congenitally short metacarpals and metatarsals is less than 1/1,000. Urano and Kobayashi [18] reported the incidence of congenitally short metatarsals to be approximately 0.022%. The occurrence rate for females is five times greater than for males. Bilateral involvement has been observed in 72% of the patients [18]. The fourth ray is the most frequently involved digit. It has been widely recognised that early closure of the epiphyseal plate is the primary aetiological factor contributing to congenitally short metacarpals and metatarsals [4, 7, 9]. The presence of short metacarpals does not impair hand function and surgical intervention is performed primarily for cosmetic reasons [4, 20]. However, patients with short metatarsals often suffer from unacceptable cosmesis,

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plantar pain, callosities on the heads of the metatarsals, difficulty in wearing shoes and in walking due to the cock-up deformity [2, 17]. Mansoor [8] was the first to introduce a technique that allows for metacarpal lengthening. Eleven years later, Matev [10] was able to report on his vast experience using this procedure. Further, there have been numerous other practitioners who have reported the specifics of methods that they have employed, either as a one-stage procedure or gradual procedures that lengthen short metatarsals [2, 5, 16].

This study summarises our experience in the lengthening of congenital short metacarpals and metatarsals by the callotasis method.

Patients and methods

Between October 1998 and September 2005, 15 (13 females, two males) patients with congenital short metacarpals and metatarsals underwent a lengthening procedure using the callus distraction method. We examined and performed bone lengthening surgery in 13 female and two male patients. Of these, procedures were conducted on 12 metatarsals of eight patients, four metacarpals and one metatarsal of a single patient, one metacarpal and one metatarsal of a single patient and seven metacarpals of five patients. Bilateral involvement was detected in six patients (short metacarpals, $n=3$, and short metatarsals, $n=3$).

The mean age of the patients who underwent lengthening of their metacarpals, at the time of the operation, was 14.5 (10–21) years. Of the 12 metacarpals included in this study, the fourth ray in ten patients and the fifth ray in two patients were affected. Lengthening was performed using an Ilizarov type circular external fixator for two metacarpals and a unilateral external fixator for ten metacarpals. The correction of short metacarpals was only performed for cosmetic reasons.

The mean age of the patients who underwent the lengthening of metatarsals was 17.5 (10–25) years at the time of operation. The fourth ($n=12$), second ($n=1$) and third rays ($n=1$) were affected in these patients. Lengthening was performed using an Ilizarov type circular external fixator for eight metatarsals and a unilateral external fixator for the remaining six metatarsals. Indications for the treatment of short metatarsals were plantar pain, callosities on the head of the metatarsi, painful cock-up toe, functional metatarsalgia and unacceptable cosmesis. The American Orthopaedic Foot and Ankle Society (AOFAS) scoring system [6] for the lesser toe was used for the clinical evaluation of metatarsal lengthening (excellent, >85 ; good, 71–85; fair, 56–70; poor, <56). Preoperative good and excellent AOFAS scores were obtained in five and nine cases, respectively.

Technique

The lengthening was performed using either a unilateral external fixator (Mini-DAF, Orthofix Srl, Verona, Italy) with self-drilling, self-tapping screws with 3-mm-diameter shafts and two proximal and two distal screws placed on the dorsal aspect for one group or a circular external fixator (Tasarimmed, Turkey) for the other group. A half-ring was mounted to the proximal and distal end of the short bone segment to be lengthened. Two 3-mm self-tapping screws or one self-tapping screw and a 1.5-mm-long K wire passing through both below and above these half-rings was used to fix these half-rings to the bone. The screws, or screws and K wires, were fixed at 60°–90° to each other and perpendicular to the bone segment to be lengthened. Then, the screws and K wires were fixed to the half-rings with special rod holders.

A 1-cm straight incision was made to dissect the extensor tendon. Osteotomy was performed with minimal damage to the periosteum using a percutaneous drilling method [14]. Following a latency period of 7 days, distraction was started at the rate of 2×0.25 mm/day. Determining the appropriate amount of lengthening of a short bone is challenging in a bilateral involvement. We used a formula presented by Aydinlioglu et al. [1] to determine the amount of metacarpal lengthening. This formula indicates the mathematical relations between the lengths of metacarpal bones and phalanges as follows:

- First metacarpal length = $0.67 \times$ second metacarpal length
- Second metacarpal length = $0.71 \times$ third metacarpal length
- Third metacarpal length = $0.78 \times$ fourth metacarpal length
- Fourth metacarpal length = $0.84 \times$ fifth metacarpal length

We determined the final length of metatarsals involved in the lengthening procedure using the formula outlined by Magnan et al. [7] ($1=2>3>4>5$). Partial weight bearing was permitted with crutches on the second postoperative day and full weight bearing was allowed after the removal of the fixator. All of the technical details are summarised in Tables 1 and 2.

Results

The mean follow-up was 57.5 (12–96) months for short metacarpals. All patients who underwent metacarpal lengthening reported that they were satisfied and could perform their daily living activities with good functional results and cosmetic appearance. None of the patients demonstrated a decrease in the range of metacarpo-

Table 1 Results of metacarpal lengthening by the callotasis method

Case	Age	Sex	Fixator	Lengthened metacarpals	Amount of lengthening (mm)	Ratio of lengthened segment (%)	Healing period (months)	Healing index (months/cm)	External fixator period (months)	Daily living activities	Cosmetic results	ROM of MCP postop./preop. (%)	Patient's satisfaction	Complications
1	21	F	C	MC (R4)	15	40	3	2	3	full	G	80	yes	Pin-tract infection
			C	MC (L4)	13	27	3	2.3	3	full	E	100	yes	
			U	MC (R5)	14	40	3	2.1	3	sufficient	G	80	yes	
			U	MC (L5)	14	29	2.5	1.8	2.8	full	E	100	yes	
2	14	F	U	MC (R4)	15	32	2.8	1.9	3	full	E	100	yes	
			U	MC (L4)	15	32	2.7	1.8	3	full	E	90	yes	
3	10	F	U	MC (R4)	22	35	2.6	1.2	2.6	full	E	100	yes	Pin-tract infection
			U	MC (L4)	20	36	2.5	1.3	2.7	full	E	90	yes	
4	10	F	U	MC (L4)	17	30	2	1.2	2.2	full	E	100	yes	
5	17	M	U	MC (R4)	26	35	2.9	1.1	3	full	E	100	yes	
6	14	F	U	MC (R4)	23	40	2.7	1.2	3	full	E	100	yes	Pin-tract infection
7	16	M	U	MC(R4)	18	36	3	1.7	4	full	E	100	yes	

ROM=range of motion; MCP=metacarpo-phalangeal joint

C=circular fixator; Tasarimmed, Turkey

U=unilateral fixator; Mini-DAF, Orthofix srl, Verona, Italy

E=excellent; G=good

phalangeal (MCP) joint motion of more than 20% when compared to preoperative values (Fig. 1a–c). The mean amount of lengthening was 17.6 (13–26) mm, which corresponded to 36.8% of the preoperative bone length. The average time to union was 2.7 (2.0–3.0) months, the average healing index was 1.6 (1.1–2.3) months/cm and the mean length of time that the external fixator was in place was 2.9 (2.2–4.0) months for the metacarpal procedures (Table 1).

The mean follow-up was 48.3 (12–72) months for metatarsals. None of the patients demonstrated a decreased range of metatarso-phalangeal (MTP) joint motion of more than 20% when compared to the preoperative values. Based on the AOFAS score for the lesser toe, the functional results of the MTP joint of the lengthened short metatarsals were excellent in 12 and good in two cases (Fig. 2a–c). The mean amount of lengthening was 24.3 (20–30) mm, the mean lengthening compared to the original bone length was 36.9%, the average time to union was 3.8 (2.2–6.0) months, the average healing index was 1.6 (1.0–2.0) months/cm and the mean length of time that the external fixator was in place was 3.8 (2.5–5.2) months for the short metatarsals that underwent the procedure (Table 2).

Complications

In none of the lengthening procedures for short metacarpals did the lengthened segment increase to 40% of the total original preoperative length of the bone. Therefore, stiffness of the MTC joint of more than 20%, subluxation, dislocation, refracture, non-union, arthritis or neurovascular injury were not observed. However, three patients had pin-tract infections, which were controlled with oral antibiotics and local wound care.

Four metatarsals of three patients [cases 3 (right and left), 5 and 7] had angulation of not less than 10° at the end of metatarsal lengthening. In these cases, a unilateral external fixator was used for lengthening and segments were lengthened to more than 40% of the preoperative bone lengths. These patients did not undergo surgery for this angulation because they had no functional problems. Another patient (case 1) in whom a unilateral external fixator was used resulted in subluxation of the MTP joint. We performed capsular release and included the distal phalanx into the frame. All lengthened metatarsal segments healed except one. One patient (case 2) had non-union three months after the index operation. In this patient, we lengthened the metatarsal by 30 mm and used a unilateral external fixator. This gap was grafted with autogenous fibula strut graft and union occurred within three months. No complications were observed in three instances where metatarsal lengthening of more than 20 mm (or >40%) was

Table 2 Results of metatarsal lengthening by the callotasis method

Case	Age	Sex	Fixator	Lengthened metatarsals	Amount of lengthening (mm)	Ratio of lengthened segment (%)	Healing period (months)	Healing index (months/cm)	External fixator period (months)	ROM of MTP postop./preop.(%)	AOFAS score	Cosmesis	Patient's satisfaction	Complications
1	21	F	U	MT (L4)	26	48	4	1.5	4.2	80	G	E	yes	Subluxation of MTP joint
2	15	F	U	MT(R4)	30	48	6	2	3	90	E	E	yes	Non-union, autogenous fibular graft
3	13	F	C	MT (L4)	30	47	3.9	1.3	4	100	E	E	yes	Angulation
			U	MT (R3)	22	49	3.2	1.5	3.5	80	G	E	yes	Angulation
			U	MT (R4)	22	43	3.4	1.5	3.7	90	E	E	yes	Angulation
4	23	F	C	MT (R4)	20	33	3.5	1.8	3.7	100	E	G	yes	Angulation
5	21	F	U	MT (R4)	23	43	3.6	1.6	3.7	100	E	G	yes	Angulation
6	10	F	C	MT (R4)	22	29	2.2	1	2.5	100	E	E	yes	Angulation
7	18	F	U	MT (L2)	22	45	3.7	1.7	4	100	E	E	yes	Angulation
8	13	F	C	MT (R4)	23	38	2.3	1	2.5	100	E	E	yes	Pin-tract infection
			C	MT (L4)	20	37	3.3	1.7	3.5	100	E	E	yes	Pin-tract infection
9	25	F	C	MT (L4)	20	37	3.7	1.9	4	100	E	E	yes	Pin-tract infection
10	16	M	C	MT (R4)	30	48	5	1.7	5.2	100	E	E	yes	Pin-tract infection
			C	MT (L4)	30	48	5	1.7	5.2	100	E	E	yes	

C=circular fixator; Tasarimmed, Turkey

U=unilateral fixator; Mini-DAF, Orthofix srl, Verona, Italy

ROM=range of motion; MTP=metatarso-phalangeal joint

AOFAS score; the American Orthopaedic Foot and Ankle Society score of unaffected toes was used for clinical evaluation (excellent, >85; good, 71–85; fair, 56–70; poor, <56)

E=excellent; G=good

Fig. 1 A 10-year-old female with left fourth short metacarp. **a** Preoperative X-ray. **b** X-ray taken during distraction osteogenesis using a unilateral fixator. **c** X-ray after the removal of the frame

achieved using the circular external fixator. In addition, three patients had pin-tract infections, which were controlled with oral antibiotics and local wound care.

Discussion

Several techniques have been described to lengthen congenitally short metacarpals and metatarsals. The most frequently used technique is either one-stage lengthening with an intercalary bone graft or gradual lengthening by callotasis [2, 5, 18]. One-stage lengthening of metacarpals has several advantages, including a relatively short union period with less scar tissue formation [2, 3, 5]. However, there are some disadvantages of one-stage lengthening, such as a small gain in length, morbidity of the donor site, neurovascular impairment caused by rapid stretching, graft problems related to multiple lengthening and the limitation of range of motion due to cast immobilisation [2, 3].

Distraction osteogenesis is currently more popular for bone lengthening [15, 16, 17]. The advantages of this method is the achievement of greater final bone length, avoidance of bone grafting, no morbidity at the donor site, suitability for multiple lengthening procedures, shorter delay after metatarsal lengthening before the patient can put weight on the affected limb and a lower incidence of neurovascular damage [4, 16, 19]. However, when the distraction osteogenesis method is used, some morbidities, such as pin-tract infection, limitation of range of motion, stiffness, subluxation or dislocation of the MTP–MCP joint, angulation and non-union, have been reported [9, 17]. We prefer the callotasis method as it can achieve a greater degree of lengthening (average gains: 17.6 mm for metacarpals and 24.3 mm for metatarsals) and also avoid post-operative complications.

Many authors reported that a healing index of less than 1.5 months/cm could be achieved by applying an extension rhythm of 0.25 mm twice a day [4, 12, 17]. A healing index

Fig. 2 A 13-year-old female with bilateral fourth short metatarsal and right metatarsal lengthening. **a** Preoperative X-ray. **b** X-ray taken during distraction osteogenesis using a unilateral fixator. **c** X-ray taken after the removal of the fixator

of more than 2.0 months/cm can be achieved by applying an extension rhythm of 0.75–1 mm/day [11, 16]. Many factors have been identified to support a longer consolidation period after distraction osteogenesis. This includes damage to the periosteal tissue, which can be prevented with percutaneous osteotomy [14], and a distraction rhythm of greater than 2×0.25 mm/day in older patients who undergo lengthening of more than 40% (or 20 mm) of the original bone length. In our study, the healing index was 1.6 months/cm for both metacarpal and metatarsal lengthening. Our healing index for metatarsal lengthening is

greater than the healing index reported by other authors [16, 19]. We feel that these results are due to a greater amount of lengthening (average lengthening of 24.3 mm) achieved in our study compared with others (average lengthening not less than 17.6 mm) [4, 12, 17].

MTP or MCP joint angulation, stiffness, subluxation or dislocation, arthritis and delayed union or non-union are the most frequent complications encountered in the lengthening of metatarsals and have been observed in instances in which the mean lengthening percentages were more than 40% and the lengthening rhythms were greater than

0.5 mm/day [13, 16, 17]. Unfortunately, we also observed similar complications. In six cases, the rate of metatarsal lengthening was greater than 40% (20 mm) when unilateral fixators were used. We also encountered angulation ($n=4$), MTP joint subluxation ($n=1$) and non-union ($n=1$). Fortunately, we were able to change our strategy and use a circular fixator, especially when the metatarsal lengthening exceeded 20 mm. In order to prevent subluxation (in some instances), the phalanges and MCP or MTP joints were temporarily fixed with an axial K-wire or included into the frame.

In conclusion, the distraction osteogenesis method is successful and reliable for the lengthening of short metacarpals and metatarsals. However, severe complications such as stiffness, angulation, subluxation of the MCP or MTP joint and delayed union or non-union are associated with this procedure. To avoid these complications, we suggest protection of the periosteum, refraining from distraction rates of more than 2×0.25 mm/day and, if possible, avoid lengthening a bone by more than 40% (or not more than 20 mm of the preoperative bone length). If the anticipated amount of lengthening is more than these predefined values, then we suggest the usage of a circular fixator for the temporary fixation of the MTP or MCP joint with inclusion of the proximal phalanx in the frame.

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