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Lengthening by distraction osteogenesis in congenital shortening of metacarpals

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Objective: The aim of this study was to present the results of seven cases of metacarpal lengthening by distraction osteogenesis and to discuss the ideal daily rate of distraction.

Methods: Metacarpal lengthening was performed by distraction osteogenesis in the seven metacarpals of four patients (3 females, 1 male; mean age: 14.9 years). A unilateral external fixator was used for lengthening. Lengthening was initiated with a distraction rate of 2×0.5 mm/day in the patient with bilateral involvement of the middle and ring metacarpals. On the tenth day of lengthening, distraction was discontinued due to pain and contracture. Then, distraction was continued with a rate of 2×0.25 mm/day. In all other cases, the distraction rate was 0.5 mm/day. Pre- and postoperative range of motion was measured with a goniometer. Patient satisfaction was evaluated with visual analog scale.

Results: The mean pre- and postoperative metacarpal lengths were 34.6 mm (range: 33 to 37) and 49.7 mm (range: 47 to 52), respectively. The mean lengthening achieved was 15.1 mm (range: 14 to 17), while the mean distraction rate was 0.55 mm/day (range: 0.48 to 0.63). No functional loss was observed in the fingers at the final check-up. The patients were happy with the functional and cosmetic results.

Conclusion: Distraction osteogenesis is a safe method providing acceptable cosmetic and functional results in patients with congenital metacarpal shortness. The length of metacarpals and muscles that will be affected from lengthening should be considered when determining the daily rate of distraction.

Key words: Congenitally brachymetacarpia; distraction osteogenesis; external fixator; lengthening.

Short metacarpals may be congenital or may occur secondary to damaging of the growth plate due to postnatal injuries and infections.[1] Although the reason of the congenital type is unknown, premature closure of the growth plate is thought to be the cause.[2] The condition may present itself as an isolated shortness of the metacarpal or as a part of various syndromes.[3,4] Cases where shortness of the first metacarpal is accompanied by hyperphalangism have also been reported in the literature.[5,6]

Short metacarpal is frequently seen at the 3rd, 4th and 5th metacarpals.[7-9] Other than firm gripping, hand functions are not much affected by this pathology. Surgery is mostly performed for cosmetic reasons.[3] Lengthening techniques for the metacarpal include lengthening in single session and fast distraction with bone grafting and distraction osteogenesis (callotasis).[3] Progressive distraction osteogenesis has been the most preferred technique in recent years. Despite lack of

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large series and published articles, successful results have been reported through this technique.[1,3,7,9-13]

In this study, we aimed to evaluate the results of 7 cases with congenital shortening of the metacarpals where lengthening was performed through distraction osteogenesis.

Patients and methods

Seven metacarpals of 4 patients (3 females, 1 male; mean age: 14.9, range: 13 to 18) who presented to our clinic between 2004 and 2007 and had complaints of deformity and loose grip were included in the study. One patient had bilateral involvement on the 3rd and 4th metacarpals; one had involvement on the 4th right metacarpal, one on the 4th left metacarpal, and one on the 5th left metacarpal (Table 1).

An institutional review board approval and written informed consent of all patients was obtained prior to the study. Length of short metacarpals was measured on plain radiographs. All cases were categorized as Type E (short metacarpal) according to Bell's classification.[14] None of the patients had an additional pathology or family history. Lengthening by distraction osteogenesis was performed on 7 metacarpals of 4 patients. To determine the length, Aydınlıoğlu et al.'s[15] technique was used on the 13-year-old case with bilateral involvement. In cases with unilateral involvement, length of the contralateral metacarpal was used as reference. Active finger exercises were recommended in the course of lengthening.

Surgeries were performed under general anesthesia and tourniquet application. An unilateral mini external fixator with four 1.5 mm Schanz screws was applied for distraction in all cases. Two Schanz screws were percutaneously inserted both on the proximal and distal ends with fluoroscopic control. Screws were applied at a radial angle of 15 degrees on the 3rd metacarpals and ulnar angle of 15 degrees on the 4th and 5th metacarpals to protect the extensor tendons. Then a longitudinal incision of 1.5 cm was made on the dorsolateral aspect of the metacarpal to be lengthened. Subcutaneous soft tissues, the extensor mechanism and periosteum were incised parallel to the skin incision, protecting vascular and nervous tissues. Transverse osteotomy was performed through the midline of the screws at the proximal and distal ends. The periosteum and soft tissues were repaired and the skin was closed. A single incision was used for osteotomy of the 3rd and 4th metacarpals in the patient with bilateral involvement.

Patients were discharged on the first postoperative day. No distraction was performed in the first week. On the one week follow-up, the patients were instructed on lengthening process and distraction was started. Lengthening was slowed down or paused considering pain in the hand, limited flexion of the fingers, and consolidation of the callus. Control radiographs were taken twice in the first two weeks and then once every week to evaluate the alignment metacarpal. Active finger exercises were introduced through the course of lengthening.

Following the seven-day latency period, simultaneous lengthening at a speed of 2×0.5 mm/day on both hands was initiated on the patient with bilateral shortness on the 3rd and 4th metacarpals. On the 10th day, lengthening was paused for 3 days due to pain and limited flexion in the hand. Bilateral lengthening was continued at 2×0.25 mm/day at the end of this period, following pain relief. Despite active finger exercises, slightly limited flexion on 4 fingers and swan neck deformity on the right 4th finger was observed at the end of the lengthening. Radiograph taken on the 70th postoperative day revealed sufficient consolidation and the fixator was removed. Limited flexion and swan neck deformity were observed to have decreased gradually in intermittent follow-ups, resulting in full recovery 3 months after removal of the device (Fig 1). In light of this experience, other patients received lengthening at a speed of 2×0.25 mm/day, following the latency period.

Table 1. Preoperative data of the patients with short metacarpals.

Patient No	Gender	Age	Affected metacarpal
1	F	13	Right-3
1	F	13	Right-4
1	F	13	Left-3
1	F	13	Left-4
2	F	17	Right-4
3	F	18	Left-5
4	M	17	Left-4

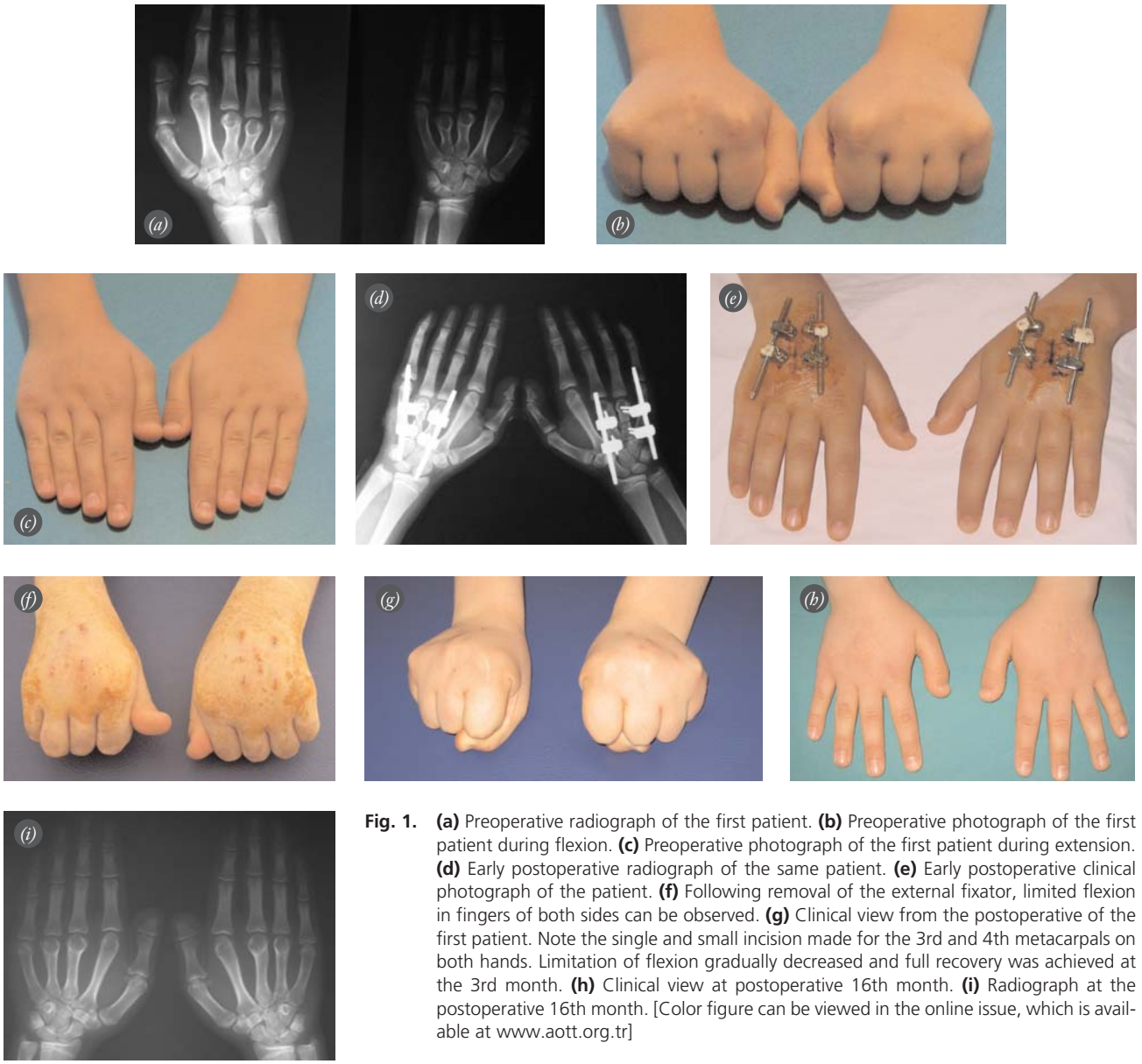

Fig. 1. **(a)** Preoperative radiograph of the first patient. **(b)** Preoperative photograph of the first patient during flexion. **(c)** Preoperative photograph of the first patient during extension. **(d)** Early postoperative radiograph of the same patient. **(e)** Early postoperative clinical photograph of the patient. **(f)** Following removal of the external fixator, limited flexion in fingers of both sides can be observed. **(g)** Clinical view from the postoperative of the first patient. Note the single and small incision made for the 3rd and 4th metacarpals on both hands. Limitation of flexion gradually decreased and full recovery was achieved at the 3rd month. **(h)** Clinical view at postoperative 16th month. **(i)** Radiograph at the postoperative 16th month. [Color figure can be viewed in the online issue, which is available at www.aott.org.tr]

Postoperative 32nd day plain radiograph of our case with shortness on the 4th metacarpal on the right hand revealed instability in the fixation system and malalignment. The external fixator was removed under general anesthesia and repositioned parallel to the metacarpal and stabilized. After a 5-day break, lengthening was resumed at a speed of 2×0.25 mm/day. The fixator was removed on the 99th day, after the radiographic evidence of consolidation (Fig. 2). No complication was observed in the other two patients treated similarly (Figs. 3 and 4).

Preoperative and postoperative 12th month metacarpophalangeal (MCP) joint range of motions (ROM) were measured with a goniometer and length of metacarpals with plain radiographs. Patient satisfaction level was evaluated using visual analog scale (VAS). Statistical analysis of pre- and postoperative length differences was performed utilizing Wilcoxon test. Significance level was set at $p \leq 0.05$.

Results

Mean follow-up time was 34.6 months (range: 26 to 37). Mean lengthening time was 27.7 days (range: 24 to 35) and external fixator application time 79.1 days (range: 70 to 99). Mean length of the metacarpals was 34.6 mm (range: 33 to 37) preoperatively and 49.7 mm (range: 47 to 52) postoperatively, with significant difference ($p<0.001$). Mean elongation amount achieved

Fig. 2. Postoperative radiograph of the second patient. Fixator was replaced due to malalignment and instability.

Fig. 3. **(a)** Postoperative radiograph of the 3rd patient from the course of lengthening. **(b)** Early postoperative clinical photograph of the 3rd patient. [Color figure can be viewed in the online issue, which is available at www.aott.org.tr]

Fig. 4. Postoperative radiographic image of the 4th patient.

was 15.1 mm (range: 14 to 17) and the percentage of lengthening was 44.2% (range: 41.6% to 51.5%). Mean recovery index was noted as 51.7 day/cm (range: 45 to 58) and daily elongation amount 0.55 mm/day (range: 0.48 to 0.63) (Table 2).

Mean preoperative extension/flexion of the MCP joints with short metacarpals was 19.3/64.3 degrees, and postoperative 12th month as 16.4/78.6 degrees, respectively. The increase in flexion was statistically significant ($p<0.05$). Patient satisfaction of cosmetic outcome was evaluated using VAS. Preoperative values were 45.7±3.4 and postoperative values 82.9±6.9, demonstrating a significant difference ($p<0.05$) (Table 3). Patients confirmed firmer gripping with their fingers compared to their preoperative status.

The first patient experienced finger contracture and pain during the initial lengthening period at 2×0.5 mm speed. The complaints decreased gradually and disappeared following reduction of the lengthening speed. The fixator had to be replaced in the second patient, due to malalignment and instability caused by the non-parallel placement of the fixator with the metacarpal. The third patient developed pin tract infection which resolved after wound care and antibiotics.

Table 2. Postoperative data of the patients.

Patient No	Lengthening time (days)	Consolidation time (CT) (days)	Duration of fixator application (days)	Recovery index (days/cm)	Lengthening percentage (%)	Follow-up time (months)	Distraction speed (mm/days)
1	24	56	70	45.4	41.6	37	0.63
1	24	56	70	50	42.4	37	0.58
1	24	56	70	45.4	41.6	37	0.63
1	24	56	70	50	42.4	37	0.58
2	35	64	99	58.2	51.5	35	0.48
3	31	54	85	56.6	44.1	33	0.48
4	32	58	90	56.2	45.7	26	0.50
Mean	27.71	57.1	79.14	51.68	44.18	34.57	0.55

Table 3. Pre- and postoperative metacarpal length, degree of the MCP joint extension/flexion, and VAS scores of the cases.

Patient No	Preoperative length (mm)	Amount of lengthening (mm)	Final length (mm)	Preoperative extension/flexion of the MCP joint (degrees)	Postoperative extension/flexion of the MCP joint (degrees)	Pre- and postoperative patient satisfaction (100 mm VAS)
1	37	15	52	15/70	15/80	45/80
1	33	14	47	20/60	15/75	45/80
1	37	15	52	15/70	15/80	45/80
1	33	14	47	20/60	15/75	45/80
2	33	17	50	25/60	20/75	50/75
3	34	15	49	20/65	20/85	50/95
4	35	16	51	20/65	15/80	40/90
Mean	**34.57**	**15.14**	**49.71**	**19.3/64.3**	**16.4/78.6**	**45.7/82.9**
Statistical analysis			p=0.017		p=0.046 (for ext.) p=0.016 (for flex.)	p=0.016

Visual Analog Scale: (0-25) full dissatisfaction, (26-50) mild dissatisfaction, (51-75) mild satisfaction, (76-100) full satisfaction

Complications, such as non-union, angulation, premature consolidation, refracture, necrosis, osteomyelitis, or neurovascular damage was not observed. There were no osteoarthritic changes in the radiographs.

Discussion

Congenital shortening of the metacarpals is a rare anomaly. Patients usually present to healthcare professionals for weakness in gripping function of the hand and cosmetic purposes. Distraction osteogenesis is a commonly preferred method in lengthening of the short bones and has proved successful outcomes in lengthening of the congenitally short metacarpals in recent years.[1,3,10,12,13,16] In their study, comprising one of the largest series in this regard, Smith et al.[13] treated 41 metacarpals of 24 patients and reported osteotomy and lengthening distraction osteogenesis as a perfect method in the management of short metacarpals. Prevalence of short metatarsals in women is 25 times of that in men, as reported by Davidson.[2] We did not come across with any information in the literature, regarding the prevalence ratio of short metacarpals in men and women. However, studies have shown that it is more frequently seen among women.[7,9,11] Three of our cases were female and one was male.

The 3rd, 4th and 5th metacarpals are the mostly involved ones in isolated shortening of the metacarpals without any genetic or biochemical anomalies.[7,9-11,17] Our cases had isolated shortening of the metacarpals and showed no sign of biochemical anomaly or genetic inheritance. The 4th metacarpal was affected in four, 3rd metacarpal in two and 5th metacarpal in one of our cases.

The optimum time for lengthening in congenital shortening of the metacarpals is proposed to be adolescence, and the possibility of repeat surgeries are emphasized for interventions before the growth of the bone is finalized; suggesting callotasis as the most practical method in lengthening procedures.[9,11] All of our patients were either adolescents or adults, thus, repeat surgeries were not required.

Distraction devices to be used in congenital shortening of the metacarpals are categorized in three groups, however, most authors suggest the use of unilateral external fixators with half pins due to their relatively sleek design and ability to allow for free movement of the finger.[9,11,12,15,18,19] We, too, used unilateral external fixators with half pins in our cases. One patient experienced a complication due to loosening of the fixator. The complication was caused by the non-parallel alignment of the fixator with the metacarpal.

Minguella et al.[11] reported that lengthening with callus distraction was the best choice for older children and achieved the maximum lengthening, with a mean of 17.2 mm, through this method. Kato et al.[9] suggested callotasis lengthening in children between ages of 10 to 15 and noted a mean lengthening of 15.2 mm using this technique. Belusa[12] and Messina,[16] applied distraction osteogenesis in their case reports and achieved satisfactory results in the metacarpals, lengthened by 20 and 22 mm respectively, with no complications. Our mean lengthening was 15.1 mm.

Recovery indices in various studies have been reported between 32 to 62.3 days/cm.[7,9,11,17] In their study emphasizing the protection of the periosteum,

Şen et al.[17] reported this index as 1.4 months/cm with a mean lengthening of 20 mm. Minguella et al.[11] had a mean recovery index of 47 days/cm, duration of fixator application of 82 days, and lengthening ratio of 53.4%. Bozan et al.,[7] on the other hand, noted the recovery index as 49.6 days/cm, the mean lengthening 54.6%, and duration of fixator application 122.3 days. In Kato et al.'s[9] study, mean recovery index was 62.3 days/cm, duration of fixator application 13.9 weeks, and lengthening ratio 43.8. We ensured the preservation of the periosteum and found out the mean recovery index as 51.7 days (range: 45.4 to 58.2), duration of fixator application 79 days (range: 70 to 99), and lengthening ratio 44.2% (range: 41.6% to 51.5%).

Masada et al.[20] and Takakura et al.[21] postulated that the total amount of lengthening should not exceed the size of the original bone by more than 40%, which otherwise would lead to limited ROM and contracture. Mean lengthening amount in our study was 44.2% of the original metacarpal length. Limited ROM and contractures in our study was observed in early days of the lengthening process when the achieved amount was still way below 40% of the original metacarpal. Thus, we believe the limited ROM and contractures seen in our cases was due to lengthening speed rather than our mean lengthening ratio of 44.2%.

In their study on 18 metacarpals of 8 patients, Bozan et al.[7] found a positive correlation between age and recovery index and argued that elder patients required more time for recovery and consolidation. Our series consisted of young patients of a close age group and did not experience any complication related with consolidation.

The mostly encountered complications with this method are mainly pin tract infection and less frequently hypertrophic scar on the dorsal tissue.[9,11] Another complication is the decreased ROM in the adjacent joint which in turn may lead to a slow down or cessation of the lengthening process.[20-22] We did not observe any serious infection other than a pin tract infection in one case, which responded to antibiotics and local wound care.

There is no consensus on the ideal speed of lengthening. Recommended lengthening amount varies between 0.3 mm to 1.5 mm/day.[8,9,23,24] From our series of 7 cases, we started with a lengthening speed of 2×0.5 mm, took a pause for 3 days on the 10th day due to pain and limited flexion in both hands, and then resumed the process at a speed of 2×0.25 mm in one patient with bilateral involvement. In all other cases, we performed lengthening at a speed of 2×0.5 mm throughout the whole process and observed no complications.

The size of the bone to be lengthened and the affected muscles should be taken into account when determining the daily lengthening speed. A lengthening of 1 mm per day will cause much more tension and pain in a muscle with a size of 5 to 6 cm when compared with a muscle with a size of 30 to 40 cm. Failure of the soft tissue in complying with the lengthening of the bone will cause contractures. Therefore, sizes of the lumbrical and interosseous muscles to be affected by the lengthening and possible excessive tension in the periosteum should be considered when lengthening of the metacarpals is intended.

In conclusion, it is possible to obtain cosmetically and functionally successful results without severe complications in distraction osteogenesis performed on the adolescents with congenital shortening of the metacarpals. However, the unilateral external fixator should be placed properly and stabilized; and daily lengthening speed should be well adjusted.

Conflicts of Interest: No conflicts declared.

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