

Original Research Article

Functional outcome of small and long bone lengthening by distraction osteogenesis

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ABSTRACT

Background: Upper limb lengthening has limited indications because of the predominant non weight bearing nature. However, lengthening does have specific indications where the functional outcomes improve in selected patients. The aim of the study was to compare the clinical and radiological outcomes of upper limb long and short bone lengthening by distraction osteogenesis.

Methods: A total of twenty four lengthenings were done in 21 patients, three cases being bilateral. The study group included cases of club hand, symbrachydactyly, multiple hereditary osteocondromatosis, traumatic amputations and congenital defects. Operative technique involved gradual mechanical distraction by orthofix external fixator after osteotomy. Clinical and radiological evaluation was done on follow up. The functional outcome for daily activities was assessed. The Healing index, Percentage healing index and Consolidation time were calculated.

Results: The mean age of patients was 11.6 years. The percentage healing index of long bones was higher compared to short bones but the consolidation time was almost similar. Complications were seen in 35% patients. The satisfaction rates were higher in small bone lengthening and the complication rates were almost same as for long bones. Grasp, pinch strength and range of motion improved in cases of short bone lengthening.

Conclusions: Distraction osteogenesis is reliable and predictable method of correction of osseous defects with vascular bone without sacrificing any adjacent digits or toes. It also gains soft tissue that is sensate with good blood supply.

Keywords: Bone lengthening, Complications, Congenital, Distraction osteogenesis, Hand deformities

INTRODUCTION

Limbs shorten due to trauma, birth defects, osteomyelitis, and tumors.¹ Bone length reduction causes functional and aesthetic problems to the patient that can be resolved by restoration of normal bone length using modern techniques. Limb lengthening techniques were originally described for the lower extremity. The indications for limb lengthening in the upper extremity have been more

limited. In humans, the upper extremities are not weight-bearing and a moderate discrepancy in their lengths does not produce a significant functional deficit.²

Aim of the study

To compare the clinical and radiological outcomes of upper limb long and short bone lengthening by distraction osteogenesis.

METHODS

This was a retrospective and prospective study carried out in the department of Orthopaedics, Kasturba Hospital, Manipal. The case records and radiographs of 24 patients who underwent lengthening of the upper limb bones were reviewed over a period of five years.

Clinical and radiological evaluation of patients was done at their last follow up between the study periods of May 2010 to October 2012 to document the data.

The study group consisted of twenty four lengthenings in 21 patients including three bilateral cases. There were 8 cases of club hand, 7 of brachydactyly, 2 of multiple osteochondromatosis, 5 of traumatic amputations and two of congenital transverse deficiency at the level of upper forearm.

Data was analysed by SPSS (statistical package for social sciences) version 16.

Inclusion criteria

Inclusion criteria were cases requiring lengthening to improve functional and cosmetic status, club hand for functional improvement of forearm; excised tumor of distal ulna for multiple osteochondromatosis; to fill the bone defect; brachydactyly for functional improvement of digits; traumatic amputation of digits.

Exclusion criteria

Exclusion criteria were radial club hand undergoing distraction to improve the hand - forearm angle; so as to correct radial deviation were excluded; lengthening of digits by bone grafting only; lengthening of digits by pollicisation.

Preoperative work up

All patients/parents were counselled thoroughly regarding the treatment options, advantages and disadvantages, duration and possible complications of the procedure.

Operative technique involved gradual mechanical distraction applied through an external fixator.

Surgical technique

Under general anesthesia, a straight skin incision was made and bone was exposed. The extensor tendon and periosteum were dissected longitudinally, and an osteotomy was made horizontally at the center of the bone. Osteotomy, to accommodate the fixator pin was done at diaphysis and metaphysis for small and long bones respectively. An intramedullary Kirschner wire was used to maintain alignment. The periosteum and the extensor tendon were sutured during closure.

In cases of radial club hand, on the radial side of the forearm, soft tissue, bone and neurovascular bundle was either absent or fibrosed, so lengthening procedure was done only bringing after bringing the wrist to neutral by centralization or radicalization. To prevent recurrence of radial deviation during lengthening, cross K-wires were passed at the wrist and were left in place till consolidation.

Separate incisions were used for osteotomy and pin fixation to reduce infection rate. Intra operatively, the distraction with external applicator at the osteotomy site was confirmed under image guidance before closing the wound.

K wire fixation for reducing angular deformity

K wire was passed in 9 cases to reduce angular deformity (5- long bones and 4 - small bones). In some cases K wire could not be passed due to technical difficulties

Post-operative period

Distraction was started on day 5, with one quarterly turn every 6 hours a day and was confirmed under radiography (bone gap) on day 7.

The interval between surgery and the first appearance of radio opaque callus ranged from 2-4 weeks with a mean of 23 days.

Patients were reviewed at weekly intervals until sufficient length was achieved, and later were reviewed for consolidation twice a month.

Clinical evaluation at follow up was done to assess pain, itching, discharge from the wound, pin loosening, external fixator alignment, distal sensation and circulation and range of motion of joints

Radiological evaluation

Radiographs were taken at weekly intervals for callus formation, quality, length and width and alignment of regenerated bone, subluxation/ dislocation of joint and osteolysis around the pin.

Functional assessment in small and long bones

Pervical score and AHA (assisting hand assessment) scale was used for small and long bones respectively.

Pin tract infection was specifically looked for. Treatment consisted of wound care and oral antibiotics. Orthotic or physical rehabilitation were organized.

Distraction was stopped after gaining sufficient length. External fixator was used for twice as long as the distraction time. The distraction device was easily removed after achieving the desired length and width of

bone with adequate consolidation of at least three cortical surfaces as seen radiographically.

Follow up time

The average follow up period was 3 years 2 months. It was 4.2 years and 3.6 years for small and long bones respectively.

Follow up calculations

1. Healing index (Time taken to increase 1 cm of bone, Total number of days of treatment/ Total gain in length (cm)).

2. Percentage healing index (Total number of days of treatment /Total percentage gain in length, time taken to increase 1% of respective bone).
3. Consolidation time (Time taken for consolidation after distraction was stopped).

RESULTS

In Table 1, there were a total of 21 patients with 24 lengthening procedures, including 3 bilateral cases having 12 male and 9 female patients. The congenital and traumatic cases were 19 and 5 respectively. In congenital cases 12 were long and 9 were small bone lengthenings and in traumatic cases all were small bone lengthenings.

Table 1: Type of bone and indication for lengthening.

Long bone lengthening			Short bone lengthening		
Diagnosis	Bone lengthened	No. of cases	Diagnosis	Bone lengthened	No. of cases
Radial club hand	Ulna	5	Congenital brachydactyly	1st metacarpal	2
	Radius	2		5th metacarpal	2
Ulnar club hand	Radius	1		2nd metacarpal	3
Congenital transverse deficiency at the level of upper forearm	Ulna	2	Traumatic amputation	1st metacarpal	2
Multiple osteochondromatosis	Ulna	2		3 rd Proximal phalanx	3
Total number		12			12

Of the 24 upper limbs lengthening four were 1st metacarpal lengthening of which two were traumatic and two were congenital cases

Mean Age

In Table 2, overall mean age of patients was 11.5 years (range 3 - 32 years) with standard deviation (SD) of 8.7 years.

For long and short bones the mean age was 10 years (3 - 15 years) with SD of 5.6 years and 13 years (5 - 32 years) with SD of 10.6 years respectively.

In Table 3, the healing index (HI) of long bones was less compared to short bones, i.e. to increase 1 cm of long bone it took less time compared to small bones but this was statistically insignificant (p value 0.127).

Table 2: Percentage of lengthened bone compared to original length.

Percentage of bone lengthened	No. of small bones	No. of long bones	Total number of bones (%)
>5%	12	12	24 (100)
>20%	11	9	20 (83.3)
>30%	11	8	19 (79.9)
>40%	9	5	14 (58.9)
>50%	9	3	12 (50)
>60%	7	3	10 (41.6)
>70%	6	2	8 (33.3)
>80%	2	2	4 (16.7)
>100%	2	1	3 (12.5)

The% HI of long bones was higher compared to short bones, i.e. to increase 1% of long bone it took four times longer time. This value was found to be statistically significant (p value 0.021).

Consolidation time of long bones was greater but was statistically insignificant (p value 0.622).

There is no negative or positive correlation between age and healing index in this study.

The lengthening was a single stage procedure well-tolerated by all and all had well preserved sensation before and after distraction.

Table 3: Comparison of healing index and time of distraction for long and small bones.

Comparing criteria	Over all		Long bones		Small bones	
	Mean	SD	Mean	SD	Mean	SD
Initial length(cm)	6.42	5.26	10.27	4.90	2.57	1.26
Final length(cm)	9.12	6.49	14.10	5.55	4.13	1.78
Centimeter gain	2.65	1.99	3.73	2.22	1.57	.87
Millimeter gain	26.50	19.87	37.33	22.24	15.67	8.66
Percentage gain	55.68	35.25	43.00	30.54	68.33	36.11
Duration of distraction (days)	47.50	37.25	70.17	39.62	24.83	14.48
Duration of external fixator in situ (days)	99.29	48.41	124.83	40.17	73.75	43.18
Healing index	49.27	30.04	39.84	17.11	58.70	37.40
Consolidation time (days)	51.50	32.88	54.92	36.69	48.08	29.83
Percentage healing index	2.93	3.30	4.44	4.06	1.43	1.18

p value <0.05 was considered statistically significant; S: Significant, NS: Not significant.

Complications

Complication was considered major when it required surgery and as minor when it was treated non-operatively. Stiffness was the most common complaint in small bone lengthening initially which responded well to physiotherapy. Angular deformity in 3 cases and poor quality of regenerate bone, non-union and implant failure

in 2 cases each were seen. There was one case each of callus fracture, subluxation of joints, flexion contracture and paresthesia. Three cases had infection.

Postoperative evaluation

One month post external fixator removal patients were assessed for improvement in hand function.

Table 4: Functional outcome through Percival score in hand function in small bone lengthening.

Diagnosis	Bone lengthened	Percival score		Dominant hand	Follow up (Months)
		Pre-op	Post-op		
Brachydactyly	1 st MC	Poor	Fair	Right	30
Brachydactyly	2 nd MC	Poor	Fair	Right	20
Brachydactyly	2 nd MC	Poor	Fair	Right	32
Brachydactyly	1 st MC	Poor	Good	Right	15
Brachydactyly	5 th MC	Poor	Good	Right	12
Brachydactyly	1 st MC	Poor	Good	Right	36
Brachydactyly	2 nd MC	Poor	Fair	Right	15
Trauma	3 rd PP	Good	Good	Right	12
Trauma	1 st MCP	Poor	Good	Right	15

Table 5: Functional outcome through AHA score for long bone lengthening.

Diagnosis	Bone lengthened	AHA score		Dominant hand	Follow up (Months)
		Pre-op	Post-op		
Bilateral RCH	L ulna	46	52	Right	16
Left RCH	L ulna	48	60	Right	8
Bilateral RCH	R ulna	42	52	Right	20
Bilateral RCH	R radius	46	52	Right	23
Right RCH	R ulna	46	60	Left	26
Bilateral RCH	R ulna	42	48	Right	24
Bilateral RCH	L ulna	42	48	Right	20
Right UCH	R radius	46	52	Left	9
Phocomelia	L ulna	38	44	Right	27
Phocomelia	L ulna	40	48	Right	24
MOC	R ulna	66	66	Right	36
MOC	L ulna	66	66	Right	6
Mean	-	47.3	54	-	19.9

RCH: Radial club hand, UCH: Ulnar club hand, MOC: Multiple osteochondromatosis.

Grip and pinch strength improved in all cases especially in patients with radial club hand who demonstrated improved skill in activities of daily living but range of motion was restricted one month post external fixator removal which improved eventually with physiotherapy and anti-edema measures.

Functional Outcome in small bone lengthening

In Table 4, it was recorded by Percival Score preoperatively and postoperatively in 9 cases. In 4 cases it was poor to good, and in 4 cases improvement was from poor to fair and in one case it was from good to good.

Functional Outcome in long bone lengthening

In Table 5, it was recorded by AHA in 12 cases. The mean improvement noted was 6 points increase in raw score from 47.3 to 54.2.

This method effectively increased forearm length in unilateral deficiency and contributed to improved independence with bowel and bladder care for a child with severe bilateral shortening.

DISCUSSION

Bone lengthening by distraction osteogenesis was primarily used in lower extremities until 1970. Matev then presented successful thumb lengthening in three patients with amputation treated by callus distraction. Later this procedure was used to lengthen hypoplastic digits, traumatic defects and congenital anomalies. For upper limb bone lengthening the results and complications differ for long and small bones.³

Matev's original technique consisted of a subperiosteal osteotomy, gradual distraction, use of plaster, prolonged immobilization and subsequently filling the defect by a bone graft.³ Kessler et al recommended bone graft in all instances, to decrease the period of immobilization and complications due to prolonged external fixation.⁴ There are no definitive data which support the use of bone graft as a substitute to the process of osteogenesis. However, elimination of bone auto grafting also eliminates donor morbidity.^{5,6}

Matev felt distraction osteogenesis in the hand would be more successful in children especially above eight years rather than in older patients.³ But we used it successfully in nine patients under eight years. It has since been recommended that patients aged 25 years or older with gaps of 3 cm or more should receive bone graft. Some authors follow these recommendations.^{4,6,7} Other workers use distraction osteogenesis without bone graft.⁸ In the present study, 3 of 24 patients needed bone grafting after acute distraction.

Mean lengthening

In our study over all mean lengthening was 2.65 cm (range 0.6-9 cm) which was 55.67% (range 15%-130%) of original bone length. More than 40% of lengthening was achieved in 14 cases.

In long bone it was 3.73 cm (range 1.7-9 cm) which is 43% of original bone length, Raimondo et al has reported an increased forearm length on average 6.0 cm or 54% of the original length similar to our results.⁹

In short bones, mean lengthening was 1.57cm (range 0.6-3.1 mm). The minimum mean length of 1.7 cm was achieved by Ogino et al and Toh et al.^{7,10} The maximum mean length of 3 cm was achieved by Finsen et al.⁸

The mean lengthening achieved in small bones in our study was less than that of long bone lengthening but the percentage lengthening was found to be higher in small bones. Though the amount of bone lengthened in the small bones was lesser than that in the long bones, the percentage of gain in the small bones was far more than in the long bones, which explains the better functional outcome of small bones.

Mean consolidation time

In our study it was 52 days (range 15-130 days), which is similar to observations of Dhalla et al (33 days), Miyawaki et al (46.7 days) and Pensler et al (44 days).¹¹⁻¹³

The consolidation time for long and small bones was similar.

Healing index (HI)

Over all Mean healing index was 49.3 days/cm (range 13.3-75 days/cm)

For long bone lengthening it was 39.8 days/cm, i.e. 5.6 weeks/cm which is similar to observation by Matsuno et al (6.8 weeks).¹⁴

Mean HI for short bone lengthening was 58.7 days/cm i.e. 8.3 weeks/cm. Pickford et al reported a mean lengthening index of metacarpals of 3.8 weeks. The reduced time in their study could be the reason for the higher callus fracture and delayed union rate (4 of 8 lengthenings).¹⁵ In our study none of the small bones had callus fractures.

The mean HI in small bones was higher compared to long bones. The muscles surrounding the bone facilitate bone healing. In this respect, the hands have less favorable conditions.

The percentage healing index was found to be 4 times higher in long bones than in small bones (1.4 in small bones and 4.4 in long bones)

There was no positive correlation between age and healing index either in small or long bones.

Rate of distraction

The rate of distraction recommended by Matev was 1.5 mm/day, by Miyawaki was 0.6 mm/day and by Kato et al 0.5 mm/day in patients with congenital anomalies of the hand.^{3,12,16}

In our study distraction rate was 1mm /day (0.25 turn four times a day).

Complications

Pin-tract infections, early removal of the frame owing to pain, callus fracture, palmar bowing, premature consolidation, non-union and delayed union are some of the reported complications.^{5-8,17}

In our study, in small bone lengthening the complications seen were subluxation, stiffness, pin tract infection and poor quality of regenerate bone and in long bone lengthening were bowing, callus fracture, implant failure and nonunion.

There was no positive correlation between percentage of bone lengthening and complications.

35% of our patients had major complications requiring secondary surgery. Lengthening of more than 40% always required greater healing index and consolidation time.

Erdem et al observed metacarpo-phalangeal joint angulations, stiffness, dislocation, arthritis and delayed union or non-union in small bone lengthening. In their study, complications were more when the mean lengthening percentages were more than 40% and the lengthening rhythms were greater than 0.5 mm/day.¹⁸

If the periosteum is protected, distraction rates less than 2×0.25 mm/day and lengthening not more than 40% of original length is used then the complications can be reduced.

Rate of complications for both small and long bones was similar but major complications were more in long bone lengthenings. The most common complication encountered initially was angular deformity which was reduced by using K wire. Even after using K wire, angular deformity was found in cases where lengthening was achieved beyond the K wire. In small bones, in few cases K wire fixation could not be done because of technical difficulties like smaller diameter of the canal and using very thin K wires would not have served the purpose.

More than 40% of the original length was achieved in 58.9% of patients (14 cases), of these six patients had

complications of which four were angular deformity. The use of K wire can reduce this complication.

Pensler et al in his study of distraction osteogenesis on nine patients, reported that 17% of the digits had an angular deformity that necessitated reoperation.¹³ Miyawaki et al prevented this complication with the use of intramedullary Kirschner-wire support. This step facilitated longer and safer elongation, thereby making the procedure more time and cost-effective.¹²

We used K wire in long bone lengthening to reduce the angular deformity (5 cases). In few cases of radial club hand it could not be passed due to curved ulna. In five patients inspite of K wire use, angular deformity could not be prevented as the lengthened bone crossed the K wire, when lengthening achieved was more than 30-40% of original bone length. However, the deformity was far less compared to those cases where K wire had not been used.

The other complications in long bones were flexion contracture at the wrist in club hand as the radial aspect had hypoplastic soft tissue. In these cases if distraction rate was reduced it resulted in early bone consolidation. In one case of club hand normal distraction rate had neurovascular limitation necessitating strict vigilance.

In case of multiple enchondromatosis after excision of the tumor, the soft tissue space which is left intact may collapse or develop adhesions resulting in gross angular deformity of the regenerate leading to callus fracture if distraction is continued. As distraction is at a slower rate, patient doesn't complain of any pain even with callus fracture. Implant related problems observed were angular deformity leading to sliding of the rail over fixator pins during distraction.

Other complications encountered in small bones were poor quality of bone formation (smaller width) due to rapid distraction of more than 0.25 turn four times a day, in 2 cases to achieve desired length faster. In these cases the consolidation time was increased but the expected width of the bone could not be achieved. In one case of traumatic amputation of 5th PP joint, subluxation was seen due to angular deformity at the osteotomy which healed in flexion. In a case of traumatic amputation, infection occurred as the open wound was severely contaminated. These complications were not seen in long bone lengthening.

Infection

Infections as reported by various authors were encountered only in three patients (traumatic amputation, open wounds) in this series.¹⁹ In our study, none of the patients had pin tract infection as separate incisions were made for osteotomy and pin tract as suggested by Heo et al.²⁰

Separate incisions reduce the chance of infection.

Non union

The non-union rate observed by Raimondo et al was 1 in 4 and that by Pickford et al was 4 in 8. In all cases, the rate of nonunion requiring surgery was at least 10%. In some series, there was a trend toward higher nonunion rates with greater amounts of lengthening.^{9,15} In our series, two cases had non-union, one in type 2 RCH (osteotomy was done at distal metaphysis) and other in the 2nd metacarpal in symbrachydactyly.

Staged lengthening rather than a single sitting lengthening may decrease the chance of nonunion.

Site of osteotomy

The bone marrow has to be protected during the surgical procedures and hence, a limited corticotomy is advisable.²¹ However, a study by Kojimoto et al demonstrated that intact periosteum is essential rather than the endosteum or bone marrow and the clinical experience with leg lengthening by callotaxis has supported this experimental conclusion.²² Aronson et al stated that the formation of new bone and mineralisation in the experimental healing of distraction osteogenesis was better and quicker in the metaphyseal than in the diaphyseal bone.²³ Similarly Toh et al also advocated the metaphyseal site for osteotomy during distraction osteogenesis.¹⁰

In our series, an osteotomy was performed at diaphysis and metaphysis for small and long bones respectively. Careful suturing back of the periosteum was done in all patients.

We did not find any poor quality regenerate in these patients except in two cases where excess distraction was done by the patients to achieve more length who were not in regular follow up. During lengthening we observed non-union in two cases of which one was type 2 radial club hand (RCH), (osteotomy was done at distal metaphysis) and other in the 2nd metacarpal in symbrachydactyly.

Patient satisfaction

In this series, all patients except the patients with Club hand and Multiple Osteochondromatosis, were satisfied with the results, and would undergo the procedure again. This could be due to the extensive preoperative counselling. In Club hand patients, the dissatisfaction could be because of multiple surgeries and associated abnormalities like thumb hypoplasia.

In multiple osteochondromatosis, it could be because only a mild functional improvement was seen as only bone was lengthened after tumor excision and soft tissue was left intact.

Also success depends on the level of patient and parent motivation and intellect. However, compliance can be

maximized with detailed education and careful patient selection.

Functional improvement scoring

Percival scoring in small bones and assisted hand assessment in long bones was used. Assessment was not as expected in case of long bone lengthening because of the associated abnormalities like thumb hypoplasia in case of radial club hand.

Though the amount of bone lengthened in the small bones were lesser than that in the long bones, the percentage of gain in the small bones was far more than in the long bones, which can explain the better functional outcome for lengthening of small bones.

In this series we did not observe any disturbance in the growth rate of the elongated fingers at follow-up.

Lengthening in radial club hand

The management of patients with radial longitudinal deficiency is complex and requires a multidisciplinary approach.

For people with unilateral radial longitudinal deficiency, the difference in forearm length is very noticeable; for those with bilateral deficiency, short forearms may impair performance of activities of daily living. Both problems can be alleviated to some extent with successful distraction osteogenesis.

According to Peterson et al significant increase in ulnar length was possible by Ilizarov method of distraction osteogenesis in patients with radial longitudinal deficiency.²⁴ In our experience not a significant amount of length could be achieved. Maximum length achieved being 3.30 cm, as the process is arduous and frequently complicated by deformity, infection and/or patient noncompliance.

Comparisons of lengthening in radial club hand and other cases of forearm lengthening

Lengthening in radial club hand (8) and other bones of forearm, the following differences were observed,

The mean length achieved and percentage gain in case of club hand (32 mm) was lesser than other forearm lengthening (49 mm). The healing index was similar in both. The complications observed in radial club hand (5) were higher than that of other forearm lengthening (2). Of these 7 complications four of them needed surgical management (three in club hand and one in other forearm lengthening). The complications observed in club hands were flexion contracture and neurovascular limitation. Both the complications can be attributed to hypoplasia of radial side soft tissue and one was non-union (RCH Type

2) at osteotomy site (distal metaphyseal even this could be attributed to hypoplasia)

In case of other bone lengthenings, one was angular deformity, which lead to implant failure and callus fracture (soft tissue adhesions or collapse). In two cases of congenital transverse deficiency of upper forearm (CTDF) no major complications were observed. Club hand lengthening is more difficult due to the hypoplastic soft tissue on radial side.

Comparison of first metacarpal and other small bone lengthening

Results of thumb lengthening were better than other small bone lengthening.

The mean amount of length achieved, percentage of gain in length and healing index were similar. The consolidation time and complications in thumb lengthening were less compared to other small bone lengthenings. This could be because of the increased mobility of carpometacarpal (CMC) joint which compensates for angular deformity of the thumb and less disability even with reduced motion range.

CONCLUSION

Distraction osteogenesis is useful for reliable and predictable correction of osseous defects and provides vascular bone with preservation of adjacent digits or toes. The consolidation time is similar in small and long bones. There is no positive correlation between age and healing index either in small or long bones. Separate incisions for osteotomy and fixator pin reduce the risk of infection.

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