ORIGINAL STUDY

acta orthopaedica belgica, 2023, 89, 177-182

doi.org/10.52628/89.2.9675

Results of Ilizarov external fixator lengthening compared to lengthening and then plating in management of femoral shortening in children

Wael EL-ADLY, Kamal EL-GAFARY, Ahmed KHASHABA, Mohamed KHALED

From the Orthopedic Surgery Department, Assiut University, Assiut, Egypt.

Correspondence at: : Wael El-Adly, Orthopedic Surgery Department, Assiut University Hospital, Assiut University, University Street, 71515 Assiut, Egypt, Tel: 02 01224265850, Fax: +20 88 2333327. Email: eladlyw@aun.edu.eg

Femoral shortening in children is a challenging condition with complex functional and psychological implications. We study the results of Ilizarov external fixator (IEF) lengthening compared to lengthening and then plating (LAP) in the management of femoral shortening in children. Forty patients were included in the study and equally divided randomly into 2 groups, in group I LAP was used and in group II lengthening by IEF only was done. The two groups were analyzed for postoperative variables to adjudge the surgical outcomes. The mean follow up time was 24.05 ± 2.99 months, The gained length was 5.60 ± 0.60 cm in group 1 and 5.48 ± 0.64 cm in group II, group I had a shorter external fixator period (3.96 ± 0.22) months, better healing index (24.6 ±2.76) days/cm, earlier complete weight-bearing (5.55 ± 0.78) months than group II. The period of hospitalization for group I was longer more than group II. The complications were less in group I (n=7, 35%) than in group II (n=11, 55%). There was no significant correlation between the healing index with age, also no significant difference was detected between the healing index and gender. There was a significant correlation between the gained length and complete weight-bearing. This study efficiently demonstrates that LAP may be better than lengthening with IEF alone in the management of femoral shortening in children.

Keywords: Ilizarov External Fixator, lengthening, lengthening and then plating, femoral shortening.

INTRODUCTION

Femoral shortening in children is a challenging condition with complex functional and psychological implications. This reduction in bone length, if neglected during the growing phase, may lead to other orthopedic deformities and problems like scoliosis and joint degeneration[1]. In contrast, early intervention and monitoring to manage the shortening by multiple phased surgeries ensuring better results and prevent complications arising from a progression of the discrepancy[2]. While the most common causes for femoral shortening may be congenital, post-traumatic, or post-infectious, other less frequent factors like vascular deficiency, neuromuscular disorders, and tumor resection may also result in shortening[3]. Correction of shortening is done by limb lengthening procedures, devices for which have evolved in the last years[4]. With Ilizarov's revolutionary concepts, the principle of using a circular fixator was employed worldwide despite their known complications[5,6]. Bone lengthening and then minimally-invasive sub-muscular plating were developed that were more suitable for children and eventually the incidence of fracture in the regenerated bone after removal of external fixation was reduced[7]. Therefore, this study presents a comparison of Ilizarov external fixator (IEF) lengthening with lengthening and then plating (LAP) in the management of femoral shortening in children.

PATIENTS AND METHODS

This study was prospective comparative randomization included 40 children who were recruited from the orthopedic department and outpatient clinics of our university hospital between 2017 and 2019. The patients aged 5 to 17 years with a shortening >3.5 cm up to 10 cm, had type 1 congenital femoral shortening, post-infectious or post-traumatic shortening were included. Those beyond the age limit specified above, shortening of <3 cm and >10 cm, and femoral shortening due to other causes were excluded. Most cases were referred to our hospital complaining of chronic limping, Trendelenberg gait, or scoliosis. An initial assessment of the child was done by a thorough history, complete physical examination, and rule out apparent shortening, relevant investigations, and radiological evaluation by a scanogram. The children were randomly assigned to

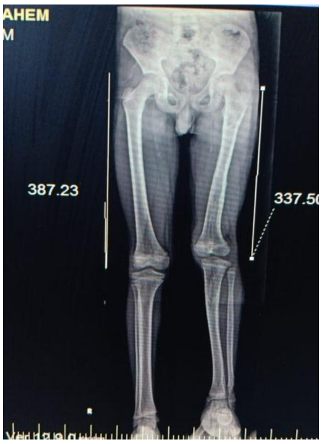

Fig. 1a — Radiograph of 11years old male with 5 cm left femoral shortening was treated by LAP.

Fig. 1b — After 4 months with complete lengthening by IEF.

Fig. 1c — After 2 months with complete union on submuscular locked plate.

Fig. 1d — After metal removal with equal femoral lengths.

Fig. 2a — Radiograph of 9 years old female with 5 cm left femoral shortening.

Fig. 2b — Radiograph after 2 months with complete lengthening by IEF.

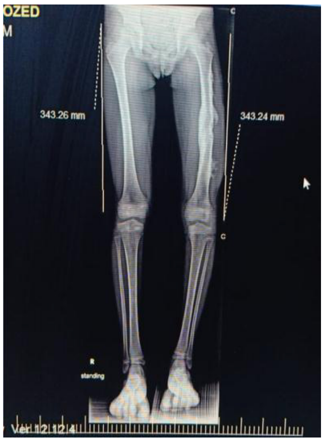

Fig. 2c — Radiograph after metal removal 3 months later with equal femoral lengths.

two groups, maintaining the blinding of the subject to the allotment. In group I, the distraction osteogenesis by IEF was done (as described later) until reaching the target lengthening followed by removal of the frame and insertion of minimally-invasive submuscular locked titanium plate under fluoroscopy guidance as described by Uysal et al.[8]. The plate length selected could accommodate three screws in each section, proximal and distal, to the distracted segment (Fig.1), Patients were allowed partial weight-bearing as tolerated from the start of frame application and continued after the plate insertion until three of four cortices of bridging callus of the regenerate were observed in radiographs. Loading of the limb was progressively increased until full weight-bearing was achieved. Whereas, the patients in group II were treated with distraction osteogenesis by IEF only. An Ilizarov construct of the appropriate size was assembled preoperatively. The frame consisted of 2 distal rings and 2 proximal femoral arches or one ring and one arch. Corticotomy was carried out by low energy technique using multiple drilling by a 3.2mm drill bit and completed by osteotomy. After a latency period of 10 to 14 days, distraction was started at the rate of one-quarter turn (0.25mm) four times a day. The patients were advised to bear weight early as tolerated from the start of frame application and progressively increased up to full weight-bearing. At the time of reaching the targeted length, IEF was kept stabilized. When the patients were able to weight-bear pain- free with radiological evidence of healing, the frames were dynamized. Finally, the frame was removed after full bridging of the lengthened area. (Fig. 2) Patients were followed regularly every 2 weeks in the outpatient clinic with clinical and radiographic evaluations, the two groups were analyzed for the following postoperative variables to assess the surgical outcomes, external fixator period, healing index, knee range of motion (ROM), the bony alignment, time of full weight-bearing, hospitalization period, exposure to secondary anesthesia, and presence of complications. The study has been approved by the institutional review board of the authors' affiliated institution and was conducted following the ethical principles of the Declaration of Helsinki. Parents of the patients provided informed written consent for participation in the study.

All data were analyzed using SPSS version 22 (Statistical Package for Social Science). Descriptive statistics, i.e., frequency and percentage, mean and standard deviation were computed according to the data. Qualitative variables were compared using the Chi-square ($\chi 2$) and Fisher's exact tests, while Student's independent-samples t-test was used for comparing quantitative variables. Also, Pearson's correlation was calculated to measure the correlation between quantitative variables. P< 0.05 was considered for statistical significance.

RESULTS

The mean ± SD of age at the time of surgery was 11.25 ± 3.21 in group I and10.05 ± 3.02 in group II. Twenty-one patients were males and 19 were females. Congenital femoral shortening type 1was the most common cause of femoral shortening (Table I). The mean± SD of follow-up time was 24.05 ± 2.99 months. The mean ± SD of the amount of shortening in group 1 was 6.60 ± 1.67 cm while in group II was 5.78 ± 1.59 cm. The gained length mean± SD was 5.60 ± 0.60 cm in group 1 and 5.48 ± 0.64 cm in group II (P=0.527). Moreover, incomplete compensation of shortening (residual shortening) was presented in 45% and 5%of patients in group I and group II respectively with a significant difference (P=0.003). On comparing the external fixator period, group I had a shorter period than group II with a significant difference (P<0.001). Group I had a better healing index than group II with a significant difference (P<0.001). Likewise, patients in group I achieved full weight-bearing earlier than in group II with a significant difference (P= 0.001). Eighteen patients (90 %) in group I attaining full weight-bearing by 8 months after the targeted lengthening, and the remaining 2 patients (10%) by about 12 months. Whereas in group II, only 8 patients (40%) were enabled of full weight-bearing by 10 months post-lengthening, and the remaining 12 patients (60%) achieved full weight-bearing at about 12 months. The period of hospitalization for group I was longer more than group II as 19 patients (95 %) in group I were admitted for more than 2 weeks and only one patient required less than 2 weeks admission. While in group II 17 patients (85%) were admitted for less than 2 weeks, and only 3 patients (15%) required > 2 weeks admission. On comparing the readmission, all the patients in group I were readmitted (thrice or more) to the hospital during the study, while only 3 patients in group II had two readmissions and the rest 17 had

Table I. — Demographic data of the studied patients.

Item	Group I (n= 20)	Group II (n= 20)	P-value
Age: (years) Mean ± SD	11.25 ± 3.21	10.05 ± 3.02	0.231
Sex: n/%			0.752
Male	10(50%)	11(55%)	
Female	10(50%)	9(45%)	
Etiology: n/%			0.702
Congenital	12(60%)	13(65%)	
Post-traumatic	7(35%)	5(25%)	
Post-infection	1(5%)	2(10%)	
Side: n/%			0.752
Right	11(55%)	10(50%)	
Left	9(45%)	10(50%)	

SD: standard deviation, n: number.

Table II. — Comparison of Ilizarov external fixator lengthening and lengthening and then plating.

Item	Group I (n= 20)	Group II (n= 20)	P-value
Shortening: (cm) Mean ± SD	6.60 ± 1.67	5.78 ± 1.59	0.119
Lengthening: (cm) Mean ± SD	5.60 ± 0.60	5.48 ± 0.64	0.527
Residual shortening: n/%	9(45%)	1(5%)	0.003*
External fixator period: (months) Mean ± SD	3.96 ± 0.22	5.98±1.08	<0.001*
Distraction index: (days) Mean ± SD	56.00 ± 5.98	54.75 ± 6.38	0.527
Healing index: (days/cm) Mean ± SD	24.6 ± 2.76	36 ± 3.34	<0.001*
Full weight-bearing after lengthening: (months) Mean ± SD	5.55 ± 0.78	7.03 ± 0.82	<0.001*
Hospitalization: (days) Mean ± SD	19.40 ± 1.90	15.65 ± 1.87	<0.001*
Readmission: n/%	20(100%)	3(15%)	<0.001*

*Significant difference.
SD: standard deviation, n: number, cm: centimeter.

Table III. — Complications.

Complication : n	Group I (n= 20)	Group II (n= 20)
pin tract infection	1	11
Limited ROM	3	0
Stiff knee	1	8
Nonunion	1	0
Malalignment	2	4
Re-fracture	1	3
knee subluxation	1	1
Weak regenerate	2	4
Premature consolidation	0	1

ROM: range of motion,n: number.

Table IV. — Correlation of healing index with age and lengthening, and of lengthening with full weight-bearing.

	Healing index (m) r-value	**P-value**
Age (year)	0.081	0.617
Lengthening (cm)	0.342	0.031*
	Lengthening (cm) r- value	**P-value**
Full weight-bearing (m)	0.397	0.011*

*Significant difference.
m: month, cm: centimeter.

a single one with a significant difference (P<0.001) (Table II). Finally, the complications were less in group I than group II, 7 cases (35%) had complications in group I, while 11cases (55%) had complications in group II. In group I one patient had limited last 30° knee flexion, and 2 patients had limited knee extension (15°, and 20°).While one patient had a completely stiff knee at 30°. Contrarily, 8 patients in group II developed a transient stiff knee. When comparing the alignment after the targeted length was achieved by scanogram and measurement of the lateral distal femoral angle (LDFA), it was seen that 2 patients in group I acquired a 6-8°valgus malalignment. However, 4 patients in group II progressed to a 6-8 varus malalignment (Table III). The correlational analysis showed that there was no statistically significant correlation between the healing index and age. There was a significant positive correlation between the length gained and both the healing index and full weight-bearing (Table IV). Also, no significant difference was detected between the healing index and gender (P=0.502).

DISCUSSION

Distraction osteogenesis with IEF is a predictable technique for bone lengthening; however, it is associated with a prolonged external fixation period. In turn, this leads to an increased risk of pin tract infection, hardware breakage, re-fracture, and deformity after frame removal[5,9]. During bone lengthening, once the desired length is achieved, the consolidation phase begins. At this time, conversion to internal fixation using a submuscular plate can be performed to shorten the duration of external fixation and decrease the incidence of previously mentioned complications. This method provides mechanical stability to support the newly formed bone structure after lengthening and can preserve periosteal blood supply[10]. In the comparison of IEF lengthening with LAP, different important parameters must be considered. The external fixator period is an important parameter to evaluate the prognosis of management. Patients in group I had a shorter period than group II, A longer application of the fixator is often associated with an increased incidence of pin tract infections, as evident from our results, it was observed in one and 11 patients in group I and group II respectively, all were treated by increasing the frequency of pin site cleaning, topical, and parenteral antibiotics in the outpatient clinic except 2 patients in group II who were treated by debridement and readjustment of Schanz screws in the operative theater. This agrees with the results of previous researches that reported a higher incidence of pin tract infection when they used an external fixator only in lengthening due to a long period of external fixator[11-13]. On the other hand, other authors reported a lower incidence of pin tract infection by using the LAP procedure[10,14]. There was a potential risk of infection when plating was done at the moment of removal of external fixator with pin tract infection, in the current study, one patient in group I devolved pin tract infection. We waited for the healing of the pin tract before inserting the plate with daily shower, washing the pin sites with antibacterial soap, topical and parenteral antibiotics, these regimens succeeded in avoiding the plate infection. In the current study the mean healing index in group I was shorter than that in group II. Munajat et al. employed a similar methodology of LAP to improve the healing index and achieved comparable results[15]. Lie et al. described the comparison between lengthening by IEF only, and LAP, the healing index in the LAP was better and shorter than the lengthening without plating[16]. Another parameter to be considered after the femoral lengthening procedure is the knee ROM both during and after the procedure. Restricted knee ROM is a common complication after lengthening, poor postoperative rehabilitation; long periods of an external fixator and pin tract infection often contribute to this. Endo et al. recorded one case that had knee contracture when used the LAP technique and had complete resolution with regular physiotherapy[17]. Similarly, Horn et al. have reported 6 cases of a stiff knee at 30-40°due to pin tract infection and prolonged application of an external fixator[13]. Munajat et al. have described stiff knee as a complication with the LAP as 3 of their patients had transient knee stiffness, and recovered well with physiotherapy, while one patient underwent quadricepsplasty to improve knee flexion[15]. Our results were comparable to others regarding knee ROM. All patients with limited ROM recovered by physiotherapy except one patient who had a completely stiff knee, where quadricepsplasty was done to improve the knee flexion. Knee subluxation is a major complication that was seen in two patients, one in each group. The patient in group I had type I fibular hemimelia, while that in group II had congenital hypoplasia of the lateral condylar of the femur, and both were gradually corrected by IEF and full extension was achieved, however, knee flexion after removal of the frame was limited to 90° flexion in one patient, and 100° in the other. Jones et al. performed femoral lengthening using the Wagner technique and reported a 33% of patients in their study had varying degrees of knee subluxation, while critical assessment demonstrated that each of these was associated with a hypoplastic femur. It was postulated that posterior displacement of the tibia on the femur occurred as a result of the distal femoral and intra-articular bony deficiencies[18]. In the study of El-Rosasy et al., lengthening was done by IEF only and they observed that hypoplastic lateral femoral condyle, anterior and posterior cruciate ligaments deficiency, and tight posterolateral structures were common findings and knee dislocation was present in six cases that were treated by reduction and reconstruction of ligaments deficiency[19]. Bony alignment is another important factor to ascertain the success of the two procedures. Patients with malalignment in our study may be attributed to soft-tissue contractures associated with a congenitally short femur. After the targeted length was achieved during the consolidation phase scanogram was done to assess the bone alignment. Four patients in group II had 6-8 varus malalignment that occurred during lengthening. After removal of the frame, no malalignment was recorded. Munajat et al. used LAP and there were no malalignment complications in their study[15], however, Iobst et al. used LAP and had two cases with procurvatum[14]. In another study, 36.8% of patients had malalignment when lengthening by IEF only was done[20]. Likewise, re-fracture was less in group I than group II in our study. This was probably due to a short period of external fixator and rigid support by plating which provided more rigidity and stiffness to the bone. In group, I, only one case had re-fractured due to falling from a bicycle after 4 months of plate removal and was treated by plating. Contrarily in group II, 3 cases had a re-fracture due to weak regenerate. Danziger et al. reported 9 regenerate fractures in their series of 18 femoral lengthenings by IEF[21]. In the study of Iobst et al. who used the LAP technique, there was one case that had re-fracture due to falling on the ground[14]. Weak regenerate is a serious problem during limb lengthening and results from many systemic or local causes[1]. Once delayed regeneration has been diagnosed, alternate cycles of compression distraction can solve the problem[22]. Our results revealed that group I had no case of weak regenerate probably due to the submuscular plate which gives more bony stability and promotes healing of the regenerate. Comparably, in group II, 4 patients had a weak regenerate, and 2 of them had a history of osteomyelitis femur. Song et al. had 2 cases of weak regenerate and delayed consolidation due to local wound infection at the osteotomy site[12]. We detected that nonunion occurred in one case in group I, it was treated by refreshing the edges and autogenous bone graft. Premature consolidation of the regenerated bone has been reported due to irregular distraction of the osteotomy, especially in children[1], it was seen in our patients only in one case in group II. This was due to weak distraction because of pin tract infection and loosening the pins, it was treated by re-osteotomy. We measured the period for attaining complete weight-bearing for both groups. LAP ensured early weight-bearing in group I that was attained by better healing using a minimally-invasive submuscular locked titanium plate. Full weight-bearing was not achieved at an early stage in group II despite the patients being advised to bear weight early from the start of frame application, this may be due to the complications that occurred as transient stiff knee, weak regenerate, and re-fracture that increased the mean± SD time to full weight-bearing. Consequently, the hospitalization period was longer in group I than group II as the longer admission was necessary for lengthening, plate insertion, metal removal, and management of complications. Whereas in group II, just one admission is required for putting up the IEF, and frame removal can be done in outpatient clinics. However, 3 cases in group II needed readmission for the management of complications, 2 had a pin tract infection, treated by debridement and readjustment of Schanz screws, while the third had a knee subluxation necessitating readmission for reduction by IEF. Similarly, the re-exposure to anesthesia was seen for all patients in group I compared to only 3 cases or re-exposure in group II.

Limitations of this study include a relatively small number of patients, a short follow-up period, and also we did not consider the expectation programs in our study as Multiplayer and bone Ninja programs. Therefore, more studies are needed to further reaffirm our results.

CONCLUSION

This study efficiently demonstrates that LAP may be better than lengthening with IEF alone in the management of femoral shortening in children. A reduction of external fixator period, early weight-bearing, better healing index, and lesser complications favor the use of the LAP technique. However, this comes at the cost of a longer hospital stay and more exposure to anesthesia.

REFERENCES

1. Hosny G. Limb lengthening history, evolution, complications and current concepts. J Orthop Traumatol. 2020 Mar; 21(1):3.
2. Ilizarov GA. The tension-stress effect on the genesis and growth of tissues: Part II. The influence of the rate and frequency of distraction. Clin Orthop Relat Res. 1989 Feb; (239):263-285.
3. Hvid I, Horn J, Huhnstock S, Steen H. The biology of bone lengthening. J Child Orthop. 2016 Dec; 10(6):487-492.
4. Hasler CC, Krieg AH. Current concepts of leg lengthening. J Child Orthop. 2012 Jun; 6(2):89-104.
5. Paley D. Problems, obstacles, and complications of limb lengthening by the Ilizarov technique. Clin Orthop Relat Res. 1990 Jan; (250):81-104.
6. Paley D, Herzenberg JE, Paremain G, Bhave A. Femoral lengthening over an intramedullary nail. A matched-case comparison with Ilizarov femoral lengthening. J Bone Joint Surg. Am. 1997 Oct; 79(10):1464-1480.
7. Fadel M, Hosny G. The Taylor spatial frame for deformity correction in the lower limbs. Int Orthop. 2005 Apr; 29(2):125-129.
8. Uysal M, Akpinar S, Cesur N, Hersekli MA, Tandoğan RN. Plating after lengthening (PAL): technical notes and preliminary clinical experiences. Arch Orthop Trauma Surg. 2007 Dec; 127(10): 889-893.
9. Dahl MT, Gulli B, Berg T. Complications of limb lengthening. A learning curve. Clin Orthop Relat Res. 1994 Apr; (301):10-18.
10. Oh CW, Shetty GM, Song HR, Kyung HS, Oh JK, Min WK, et al. Submuscular plating after distraction osteogenesis in children. J Pediatr Orthop. B. 2008 Sep; 17(5): 265-269.
11. Ko KR, Shim JS, Chung CH, Kim JH. Surgical Results of Limb Lengthening at the Femur, Tibia, and Humerus in Patients with Achondroplasia. Clin Orthop Surg. 2019 Jun; 11(2): 226-232.
12. Song HR, Myrboh V, Oh CW, Lee ST, Lee SH. Tibial lengthening And concomitant foot deformity correction in 14 patients With permanent deformity after poliomyelitis. Acta Orthop. 2005 Apr; 76 (2):261-269.
13. Horn A, Sipilä M. Femoral lengthening in children. South African Orthopaedic Journal. 2020; 19(1):12-17.
14. Iobst CA, Dahl MT. Limb lengthening with submuscular plate Stabilization: a case series and description of the technique. J Pediatr Orthop. 2007 Aug; 27(5):504-509.
15. Munajat I, Sulaiman AR, Mohd EF, Zawawi MSF. Submuscular Plate Stabilisation After Lengthening: Standard and Modified Techniques. Malays Orthop J. 2020 Mar; 14(1):49-54.
16. Lie CW, Chow W. Limb lengthening in short-stature patients using monolateral and circular external fixators. Hong Kong Med J. 2009 Aug; 15(4):280-284.
17. Endo H, Asaumi K, Mitani S, Noda T, Minagawa H, Tetsunaga, et al. The minimally invasive plate osteosynthesis (MIPO) Technique with a locking compression plate for femoral lengthening. Acta Med Okayama. 2008 Oct; 62 (5):333-339.
18. Jones D C, Moseley C F. Subluxation of the knee as a complication of femoral lengthening by the Wagner technique. J Bone Joint Surg. B. 1985 Jan; 67(1):33-35.
19. El-Rosasy MA, Ayoub M. Salvage of dislocated hip and knee during limb lengthening for congenital femoral deficiency. The Egyptian Orthopaedic Journal. 2017; 52(2) 100-108.
20. Reitenbach E, Rödl R, Georg Gosheger G, Vogt B, Schiedel F. Deformity correction and extremity lengthening in the lower leg: comparison of clinical outcomes with two external surgical procedures. Springerplus. 2016 Nov; 24;5(1):2003.
21. Danziger MB, Kumar A, DeWeese J. Fractures after femoral lengthening using Ilizarov method. J Paediatr Orthop. 1995 Mar-Apr; 15(2):220-223.
22. Makhdom AM, Cartaleanu AS, Rendon JS, Villemure I, Hamdy RC. The accordion maneuver: a noninvasive strategy for absent or delayed callus formation in cases of limb lengthening. Adv Orthop. 2015 Oct; 2015: 912790. https://doi.org/10.1155/2015/912790.