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Axial deformity correction in children via distraction osteogenesis

Received: 9 November 2005 / Revised: 17 November 2005 / Accepted: 18 November 2005 / Published online: 18 March 2006
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Abstract We performed a retrospective analysis of the results of 62 tibial and 54 femoral lengthenings in 88 consecutive patients. The patients mean age was 13.5 years and mean follow-up was four years. There was a significant difference between metaphyseal (27 ± 1.2 days/cm) and diaphyseal (39.4 ± 1.7 days/cm), tibial (34 ± 1.7 days/cm) and femoral (31 ± 1.4 days/cm) lengthening ($P < 0.05$), but no significant difference among the lengthening indexes when treating one-, two-, or three-dimensional deformities, congenital (34 ± 2.4 days/cm) and acquired (32 ± 1.0 days/cm) limb length discrepancy (LLD) ($P > 0.05$). The lengthening index was 33 ± 1.1 days/cm, distraction regenerate length 6 ± 0.4 cm, and lengthening percentage 21 ± 2.1 . The scatter plots of new regenerate length against time and the scatter plots of neurological complication, residual deformities, broken pins, joint contractures, and hypertension rate against lengthening percentage showed a positive linear relationship ($r = 0.8$). We found the correlations between quantitative and qualitative parameters that should help to predict the treatment outcomes. Lengthening index depends on the amount of length gained. Higher length of new bone regenerate leads to a decrease in lengthening index. Expected gain in bone length can aid in estimating the duration of treatment. The lengthening percentage correlates very well with the complication rate and can be used to predict the complication rate.

Résumé Nous avons pratiqué une analyse rétrospective du résultat de 62 allongements du tibia et de 54 allongements du fémur lors d'une série de 88 patients traités consécutivement. L'âge moyen était de 13 ans et demi, le suivi moyen

de 4 ans. Il y avait une différence significative entre les allongements métaphysaires (27 ± 1.2 jours/cm), diaphysaires (39 ± 1.4 jours/cm), tibiaux (34 ± 1.7 jours/cm) et fémoraux (31 ± 1.4 jours/cm) ($P < 0.05$), mais pas de différence significative de l'index de consolidation au cours du traitement des déformations congénitales de type 1, 2 ou 3 (34 ± 2.4 jours/cm) ou acquise (32 ± 1 jour/cm). L'inégalité de longueur était d'origine congénitale ou acquise (34 ou 32) ($P > 0.05$). La moyenne d'allongement était de 33 ± 1.1 jours/cm, l'allongement moyen était de 6 ± 0.4 cm et le pourcentage d'allongement de $21 \pm 2.1\%$. Ce travail a montré une relation significative entre le pourcentage d'allongement et le taux de complications neurologiques, de déformation résiduelle, de rupture de broches, de raideurs articulaires, d'hypertension et d'anomalies du régénérat osseux ($r = 0.8$). L'index de consolidation dépend du pourcentage d'allongement, avec une bonne régénération osseuse lorsque celui-ci est réduit. Le gain estimé d'allongement permet d'estimer la durée du traitement, le pourcentage d'allongement est tout à fait bien corrélé avec le taux de complications et permet de prédire celles-ci aux patients.

Introduction

Distraction osteogenesis is based on progressive gradual tension stress across an osteotomy or corticotomy site, which leads to new bone formation [13, 14]. Treatment time is divided into three successive periods: latency, distraction, and consolidation. The latency period is the time from the date of osteotomy until distraction begins. It allows early callus formation. For the period of distraction biological and mechanical conditions should be optimised to avoid delayed bone formation and non-union [1, 4, 25], decrease the rate of residual axial deformity [3, 5, 6, 22], avoid joint contractures and joint subluxations [2, 7], and increase fixation stability [5, 6, 21, 22]. During the consolidation period the new bone regenerate is allowed to consolidate by holding the fixator in a neutral phase before a subsequent dynamisation is performed to allow

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Fig. 1 **a** There was a significant difference between metaphyseal (27 ± 1.2 days/cm) and diaphyseal (39.4 ± 1.7 days/cm) lengthening ($P < 0.05$). **b** There was no significant difference among the lengthening indexes when treating one-, two-, or three-dimensional deformities ($P > 0.05$). **c** The lengthening indexes in congenital (34 ± 2.4 days/cm) and acquired (32 ± 1.0 days/cm) LLD were not significantly different ($P > 0.05$). **d** There was a significant difference ($P < 0.05$) when comparing tibial (34 ± 1.7 days/cm) vs femoral lengthening (31 ± 1.4 days/cm)

maturation of the regenerate [11]. The fixator is left in place until the new bone regenerate is strong enough to prevent collapsing, shortening, bending, buckling, or fracturing [1, 4, 10].

We reviewed the results of distraction osteogenesis for femoral and tibial lengthening in children and performed a statistical analysis to find out the effect of the corticotomy site, deformity complexity, new bone regenerate length, percent increase in bone length, and aetiology on the outcome as well as on the prevalence of complications and of additional procedures.

Materials and methods

We performed a retrospective analysis of the results of 62 tibial and 54 femoral lengthenings performed between January 1990 and December 2003 in 88 consecutive patients. The data included medical records, radiographs were reviewed to determine the diagnosis, the age at time of lengthening, the bone segment involved (femur, tibia), deformity (one-, two-, and three-dimensional), osteotomy type (subperiosteal corticotomy, osteotomy with multiple drill holes and an osteotome, osteotomy with an oscillating saw, and osteotomy with Gigli saw), level of distraction (diaphysis, metaphysis, bifocal), external fixation length (days), treatment periods (latency, distraction, consolidation), the rate of distraction, the length of distraction regenerate (cm), angular correction, lengthening percentage (percent increase in bone length), lengthening index (days/cm), external fixator brand, additional operations, and complications. When angular deformity accompanied the shortened bone, the lengthening was performed at the deformity site. Patients were encouraged to mobilize fully weight bearing as soon as possible, with appropriate

physiotherapy. Consolidation of the distraction regenerate was considered sufficient for fixator removal when the regenerate was corticalised on three of four sides as seen on anteroposterior and lateral radiographs [10]. Because the decision to remove an external fixation device based on radiographic assessment alone resulted in intra-observer and inter-observer variability [24], all limbs were tested for structural integrity by partially disassembling the frame and manually stressing the bone before removing the fixator.

Statistical analyses were conducted using SAS (SAS Institute, Cary, NC, USA). The *t*-tests two-sample assuming unequal variances and analysis of variance (ANOVA) followed by post hoc *t*-tests with an alpha level of $P < 0.05$ were applied. Continuous data were reported as the mean ± standard deviation. Scatter plots of complication rate (%) against lengthening percentage and of new regenerate length (cm) against time (days of external

Fig. 2 The scatter plots of new regenerate length (cm) against time (days of external fixation) showed a positive linear relationship with $r = 0.8$

Fig. 3 The scatter plots of neurological complication rate against lengthening percentage showed a positive linear relationship with $r=0.8$

fixation) were constructed, and linear regression was used to investigate the mathematical relationship between the variables.

Results

The aetiology was congenital in 46 lengthened segments and acquired in 70 lengthened segments. The patients' mean age was 13.5 years (range: 4–20 years) at the time of operation. Follow-up was from two to 12 (mean: four) years. Most of the lengthening was done by subperiosteal corticotomy (about 75%) and by osteotomy with multiple drill holes and an osteotome (about 15%). Only 10% of osteotomies were done using an oscillating saw or a Gigli saw. There were 50 metaphyseal (43%), 50 diaphyseal (43%), and 16 bifocal (14%) lengthenings. There was a significant difference between metaphyseal (27 ± 1.2 days/cm) and diaphyseal (39.4 ± 1.7 days/cm) lengthening ($P < 0.05$) (Fig. 1a). There was lengthening alone in about half of the cases (57 bones, 49.1%), two-dimensional deformity correction in 41 bones (35.3%), and three-dimensional deformity correction in 18 bones (15.5%). The lengthening index was not influenced by the deformity complexity. There was no significant difference among the lengthening indexes when treating one-, two-, or three-dimensional deformities ($P > 0.05$) (Fig. 1b). Lengthening was initiated on average 5.7 days after osteotomy at a rate of 0.25 mm four times a day at the distraction site. The distraction rate was adjusted according to the amount of new bone formation or to patients' tolerance. After fixator

Fig. 4 The scatter plots of residual deformities rate (a), broken pins rate (b), joint contractures rate (c), and hypertension rate (d) against lengthening percentage showed a positive linear relationship with $r=0.8$

Table 1 Complication rate

Complications	Per patient		Per segment	
	Absolute	%	Absolute	%
Pin tract infections	85	96.6	113	97.4
Half-pins	85	96.6	97	83.6
Wires	24	27.3	24	20.7
Osteomyelitis	5	5.7	5	4.3
Acute	2	2.3	2	1.7
Chronic	3	3.4	3	2.6
Bacteremia	1	1.1	1	0.9
Bacterial endocarditis	1	1.1	1	0.9
Toxic shock syndrome	1	1.1	1	0.9
Neurological	27	30.7	28	24.1
Peroneus	18	20.5	19	16.4
Ischiatic	3	3.4	3	2.6
Saphenous	3	3.4	3	2.6
Sural	3	3.4	3	2.6
Contractures	17	19.3	18	11.4
Ankle	5	5.7	5	4.3
Knee	9	10.2	10	8.6
Hip	3	3.4	3	2.6
Subluxations	4	4.5	4	3.4
Hip	2	2.3	2	1.7
Knee	2	2.3	2	1.7
Regenerate fracture	7	8.0	7	6.0
In external fixator	5	5.7	5	4.3
After fixator removal	2	2.3	2	1.7
Premature consolidation	21	23.9	21	18.1
Delayed union	6	6.8	8	6.9
Broken pin	16	18.2	16	13.8
Half-pins	14	15.9	14	12.1
Wires	5	5.7	5	4.3
Loose pin	1	1.1	1	0.9
Hypertension	12	13.6	17	14.7

removal, most limbs were further protected for 4–6 weeks in an external orthosis or cast. The lengthening index was 33 ± 1.1 days/cm. The lengthening indexes in congenital (34 ± 2.4 days/cm) and acquired (32 ± 1.0 days/cm) limb length discrepancy (LLD) were not significantly different

Table 2 Post lengthening deformity and complication residuals

Residuals	Per patient		Per segment	
	Absolute	%	Absolute	%
LLD	19	21.6	22	19.0
Axial deformity	25	28.4	28	24.1
Valgus	14	15.9	15	12.9
Varus	7	8.0	9	7.8
Flexion	1	1.1	1	0.9
Extension	3	3.4	3	2.6
Chronic osteomyelitis	3	3.4	3	2.6
Recurrent clubfoot	1	1.1	1	0.9
Knee contracture	1	1.1	1	0.9
Peroneus neuropraxia	2	2.3	2	1.7

($P > 0.05$) (Fig. 1c). There was a significant difference ($P < 0.05$) when comparing tibial (34 ± 1.7 days/cm) vs femoral lengthening (31 ± 1.4 days/cm) (Fig. 1d). The length of distraction regenerate was 6 ± 0.4 cm. The scatter plots of new regenerate length (cm) against time (days of external fixation) showed a positive linear relationship with $r = 0.8$ (Fig. 2). The lengthening percentage was 21 ± 2.1 . The scatter plots of neurologic complication rate (Fig. 3), residual deformities rate (Fig. 4a), broken pins rate (Fig. 4b), joint contractures rate (Fig. 4c), and hypertension rate (Fig. 4d) against lengthening percentage showed a positive linear relationship with $r = 0.8$. The complication rates, residual deformities, and additional operations are presented in Tables 1, 2, and 3, respectively.

Discussion

The results of limb lengthening in children via distraction osteogenesis are impressive. Although the gain in bone length is imposing, the outcomes should be evaluated in the context of extended time in external fixator and the fact that many complications occur and additional procedures are needed [1, 4, 9, 16]. The results of distraction osteogenesis are currently assessed using quantitative parameters such as the amount of length gained, lengthening index, and lengthening percentage and qualitative parameters such as complication rate and prevalence of additional procedures. Analysing our results, we made an effort to establish the correlations between these parameters that would help to predict the treatment outcomes.

Our results compare well with many previous studies on limb lengthening. Lengthening index depends on the amount of length gained [10, 18, 23]. Lengthening index was related to the amount of length gained (Fig. 2). Increased length was associated with decrease in lengthen-

Table 3 Additional operations

Operations	Absolute	Per patient	Per segment
Total	124	1.4	1.1
Frame adjustment	80	0.9	0.7
Additional corticotomy	8	0.09	0.07
Pin site release	4	0.05	0.03
Pin exchange	8	0.09	0.07
Peroneal nerve exploration	3	0.03	0.03
Sciatic nerve exploration	1	0.01	0.01
Fibular osteotomy	2	0.02	0.02
Pin removal	5	0.06	0.04
Bone marrow injection	3	0.03	0.03
Bone graft	4	0.05	0.03
Tendon Achilles Lengthening	2	0.02	0.02
Joint manipulation under anesthesia	1	0.01	0.01
Irrigation and debridement	1	0.01	0.01
Adductors release	1	0.01	0.01
Frame exchange	1	0.01	0.01

ing index (Fig. 2). The external fixation period had a direct relationship with length gain (Fig. 2) [23]. Paley [19] pointed out that distraction osteogenesis is four-dimensional and time is the fourth dimension. It needs to be adjusted carefully when planning the lengthening procedure. Metaphyseal sites have a propensity to be more favourable for osteogenesis (Fig. 1a) [8, 10]. The total time needed for the treatment of the tibia was longer than that needed for the femur (Fig. 1d) [10, 17, 18, 23]. Merloz [17] stated that the time for bone consolidation was on average 30% longer for the tibia than for the femur when comparing 75 tibial and femoral lengthenings. We agree with Noonan et al. [18] that in cases where both bones require lengthening, the tibia should be lengthened before the femur. Performing the tibial lengthening when the patient is younger requires less time for the osteotomy site to consolidate. Bone is uniformly produced regardless of aetiology (Fig. 1c) [23]. The lengthening index was not influenced by the complexity of the deformity in our patients (Fig. 1b). It has been shown that the periosteum of the bone is a potent site of osteogenesis during distraction and acute angular correction results in an insult to this layer [12, 13, 14]. We used a gradual correction of angular deformity, which did not disturb the periosteum and did not lengthen the consolidation time.

Distraction osteogenesis can be used to obtain the desired amount of length, but the prevalence of complications increases with the increase in lengthening percentage (Figs. 3 and 4). While new bone formation is the most identifiable and remarkable effect of limb lengthening, Putti's observations made in 1934 are still valid: That soft tissues around bone present [20] the greatest difficulty encountered in obtaining a successful result. Karger et al. [15] presented a statistically significantly higher complication rate in patients with lengthening of more than 25% of the original length. Maffulli and Fixsen [16] concluded that lengthening should be limited to 25% of the original length. Our study shows that lengthening the bone to 25% of the original length can lead to neurological complications and residual axial deformities in about one-third of patients (Figs. 3 and 4a), broken pins and joint contractures in about one-fourth of patients (Fig. 4b,c), and more than 10% of other complications. Practically all patients may be expected to develop at least one pin tract infection (Table 1) and need one additional unplanned operative procedure related to the lengthening (Table 3).

We found the correlations between quantitative and qualitative parameters that should help to predict the treatment outcomes. Lengthening index depends on the amount of length gained. Higher length of new bone regenerate leads to a decrease in lengthening index. Expected gain in bone length can aid in estimating the duration of treatment needed for every planned lengthening procedure. The lengthening percentage correlates very well with the complication rate. During planning the lengthening percentage should be a useful tool to predict the complication rate and to discuss the risks and benefits with patients and their families. Unplanned operative procedures related to the lengthening should be anticipated.

Knowledge about predictable complications should help prevent or detect them early.

We believe that distraction osteogenesis is an excellent method for extremity lengthening but complications and problems are common. We suggest that no surgeon contemplate limb lengthening without taking into account the expected significant complication rate and prolonged treatment time. Any surgeon undertaking lengthening should be comprehensively versed in the numerous complications that can occur and their prevention and treatment. The patient and family should be thoroughly counseled before undertaking the procedure.

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