ORIGINAl ARTICLE

Fracture following lower limb lengthening in children: A series of 58 patients

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KEYWORDS
Lengthening; lower limb; fracture; children; limb length inequality

Summary

Introduction: Fracture is one of the main complications following external fixator removal used in cases of progressive lower limb lengthening; rates as high as 50% are found in the literature. The aim of this study was to determine the factors influencing this complication.

Materials and methods: One hundred and eleven cases of lower limb lengthening were performed in 58 patients (40 femurs and 71 tibias). The mean age at surgery was 10.1 years old. Lengthening was performed in all cases with an external fixator alone, associated in 39.6% of cases with intramedullary nailing. The patients were divided into three groups according to disease etiology (congenital, achondroplasia and other). The fractures were classified according to the Simpson classification.

Results: Twenty fractures were recorded (18%). Sixteen fractures were found in patients with congenital disease, four with achondroplasia and none in the group of other etiologies. The fracture was more often in the femur (27.5%) than in the tibia (12.7%).

Discussion: The rate of fracture is influenced by different factors depending on the etiology of disease. In congenital diseases, the fracture rate is higher when there is lengthening of more than 15% of the initial length and a delay between surgery and the beginning of lengthening of less than 7 days. In patients with achondroplasia, the influence of a relative percentage of lengthening is less important than in those with congenital disease. However, to avoid fractures, lengthening should not be started in children under the age of nine. Moreover, lengthening should begin at least 7 days after the fixator has been placed.

Type of study: Retrospective.

Level of evidence: Level IV.

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Introduction

Limb lengthening is associated with numerous complications: infections [1,2], stiff joints [2,3], pseudarthrosis [2−4], early union [3,4] and neurological sequella [3,4]. Fractures also occur after the fixator is removed in 3−50% of the lengthened segments [1−5]. Although the principles of distraction are applied and the callotasis technique has been popular since the 1950s, this rate remains high [6−8]. These principles are essential to obtain optimal union of the distraction site, and to limit the risk of fracture after removal of the external fixator. Bone fixation must be stable [6−8], while preserving the peristeme as much as possible during the osteotomy [6−8], and a low energy osteotomy should be performed [6−8], with a rhythm of lengthening of 0.5−2 mm/d [6−8] while the delay between the osteotomy and the start of lengthening should be 4−15 days [6−8].

Several non-invasive procedures may be used to evaluate bone union and determine when the external fixator should be removed, such as ultrasound, Dual Energy X-ray Absorptiometry (DEXA) osteodensitometry which provides quantitative and qualitative evaluation of bone, as well as digital X-ray and CT scan [9−16]. The fracture rate after removal of the external fixator is reduced to a rate of 3.6% with the use of osteodensitometry [9,13]. However, in most centers, the most common technique is standard AP X-ray because it is simple to use, inexpensive and accessible. Thus, based on the principles of Fischgrund, the fixator is removed when three continuous cortices at least 2 mm thick are present [17].

The aim of our study was to analyze whether other factors influence the risk of these fractures based on a retrospective series of leg lengthening.

Materials and methods

Between 2000 and 2010, at the Timone Children’s Hospital in Marseille, 111 limb lengthenings were performed in 58 patients. The femur was lengthened in 40 cases (36%) and the tibia in 71 (64%). All clinical files and X-rays were retrospectively evaluated by an observer who was different from the practicing surgeon. Patient data was obtained from the clinical file (date of birth, sex, type of disease, limb operated on), the chronology of events in relation to the procedure (date of surgery, date of removal of fixator, date of fracture), as well as the different complications which occurred during distraction and their management. X-rays were analyzed to measure the initial length of the lengthened limb, the length of regenerated bone at the end of distraction, the diameter of the lengthened bone at the osteotomy site, the diameter of the regenerated bone at the end of distraction as well as to classify the fractures. Measurements were performed with a centimeter ruler in X-rays before 2008 and on the computer (PACS) in X-rays performed after 2008.

The mean age of patients at surgery was 10.1 years old (range 2.1−20.3). Patients were divided into three groups according to the etiology of disease:

- Group 1: congenital lower limb length discrepancies, such as fibular hemimelia or congenital short fibula (58 lengthenings in 35 patients);
- Group 2: patients with achondroplasia or a similar disease operated on for their short stature (34 lengthenings in eight patients);
- Group 3: post-traumatic, post-infectious or post-tumoral lower limb length discrepancies (disease on healthy bone) (19 lengthenings in 15 patients).

Lengthening was obtained with an external fixator in all cases (Fig. 1): monolateral fixator in 38 lengthenings (34.2%), circular in 68 lengthenings (61.3%), and hybrid in five lengthenings (4.5%). A monolateral fixator was used in all patients in group 2 (achondroplasia and associated diseases), bilateral in all cases and crossed lengthening was never performed.

Lengthening was associated with perioperative intramedullary nailing (Küchler wires of different diameters, and Metaizeau nails (Fig. 2)) in 44 segments. The osteotomy was performed with a drill and an osteoma by the “postal stamp” technique (low energy osteotomy) in 60 cases (54%) and with a Gigli saw (high energy osteotomy) in 51 other cases (46%). The level of the osteotomy was the diaphysis in 92 segments (82.9%) and the metaphysis in 19 segments (17.1%). The peristeme was opened and the entire circumference was rasped to perform the osteotomy in all cases.

After a mean delay of 7.2 days (2−14 days) between the date of surgery and the beginning of lengthening, the rhythm of lengthening was 0.5−1 mm/d. The rigidity of the callus was evaluated with standard AP and profile X-rays based on Fischgrund criteria (3/4 continuous cortices at least 2 mm thick), to determine when to remove the external fixator.

After removal of the external fixator, 56 segments (50.4%) were immobilized and 22 (19.8%) were allowed immediate
partial or total postoperative weight-bearing; the remaining cases only applied weight later. At the end of distraction, the diameters and lengths were calculated and recorded according to the following methods:

- recording of diameters: by comparing the smallest diameter of the callus to the diameter of the former osteotomy site on standard AP and/or profile X-rays (Fig. 3);
- recording of lengths: expressing the rate of lengthening by comparing the length of regenerated bone to the initial length of the lengthened limb (Fig. 4).

All of the fractures were classified according to the Simpson classification [1] (Figs. 5–7).

Statistical analysis was performed using the Pearson Chi² test. When the theoretical participants were fewer than five, the Fisher test was used. The Student-t test was used for means. The differences were considered to be statistically significant when the P value was less than 0.05.

Results

Absolute lengthening obtained was 5.3 cm (range 2–8.6 cm). Relative lengthening in relation to the initial length of the operated bone segment was 23% (range 6–57%). Lengthening lasted a mean 65.7 days (28–152 days) and mean fixation was 228.5 days (98–448 days). The Healing Index was 45.1 days per centimeter (21–102 d/cm).

There were 20 fractures/111 lengthenings (18%). The distribution of fractures in relation to the lengthened bone segments is found in Table 1. The mean delay between removal of the external fixator and the fracture was 63.2 days (0–547 days). Seventy-five percent of these fractures (15/20) occurred within the first month. The distribution of fractures in relation to the Simpson classification is found in Table 2. Placement of intramedullary nails reduced the number of fractures after removal of the external fixator, but this was not significant (Table 3). Nevertheless, it should be noted that the rate of infection at the site of external fixation when intramedullary nails were used (25 infections/ 44 lengthenings or 56.8%) was slightly higher than the rate of infection without intramedullary nails (34 infections for 67 lengthenings or 50.7%). Once again the difference was not statistically significant.

We found a greater number of femoral fractures in patients in group 1 (congenital disease) (Table 4). We also
Figure 5  Simpson classification: Type 1A: deformity without any real continuity of the callus; Type 1B: fracture of the center of the regenerated bone; Type 2: fracture of the bone/regenerate interface; Type 3: fracture at a distance from the callus in the same bone segment; Type 4: fracture of another bone segment.

Table 1  Distribution of fractures in relation to the lengthened bone segment and the etiology of disease.

<table>
<thead>
<tr>
<th></th>
<th>Number of lengthenings</th>
<th>Number of fractures</th>
<th>Percentage</th>
<th>( P )</th>
</tr>
</thead>
<tbody>
<tr>
<td>Femur</td>
<td>40</td>
<td>11</td>
<td>27.5</td>
<td>S</td>
</tr>
<tr>
<td>Tibia</td>
<td>71</td>
<td>9</td>
<td>12.7</td>
<td>( P = 0.05 )</td>
</tr>
<tr>
<td>Group 1</td>
<td>58</td>
<td>16</td>
<td>27.6</td>
<td>S between G1 and G3</td>
</tr>
<tr>
<td>Group 2</td>
<td>34</td>
<td>4</td>
<td>11.8</td>
<td>( P = 0.01 )</td>
</tr>
<tr>
<td>Group 3</td>
<td>19</td>
<td>0</td>
<td>0.0</td>
<td></td>
</tr>
</tbody>
</table>

S: significant; NS: non-significant; G1: Group 1; G2: Group 2.

Figure 6  Type 1B femoral fracture.

Figure 7  Type 2 femoral fracture.
noted that the number of fractures increased with the relative lengthening was more than 15% of the initial length of the bone and when the delay between surgery and the beginning of lengthening was more than 7 days (Table 4). By combining a rate of lengthening of more than 15% and a delay before beginning lengthening of no more than 7 days, we identified 100% of the femoral fractures in group 1 (5 fractures in 5 femurs). The difference was significant in relation to the other femurs in group 1 ($P = 0.01$).

We also found more femoral fractures in the patients in group 2 (achondroplasia) (Table 5). We noted that the rate of fracture was higher in patients under the age of nine and when the delay between surgery and the beginning of lengthening was less than 7 days (Table 5). By combining age less than nine and a delay between surgery and the beginning of lengthening of less than 7 days, half of the segments were fractured (4 fractures/8 segments including 1/2 tibias and 3/6 femurs). The difference was significant ($P = 0.0001$) in relation to the other segments in group 2.

The presence of an infection during the lengthening program increased the number of fractures after removal of the external fixator (Table 6). However, the presence or absence of radiological signs of deep infection was not a predictive factor of the incidence of fracture (Table 6).

### Discussion

#### Lengthening and intramedullary nailing

In the past few years, the efficacy of stable elastic intramedullary nailing has been reported in the treatment of fractures in children [18,19]. Other authors have evaluated the association of external fixation and rigid internal fixation with rigid intramedullary nails [20–22]. Although this association has reduced the duration of fixation and the Healing Index, several major complications were observed [20,21], such as osteomyelitis, failure of the intramedullary nail or the incarceration of the intramedullary nail.

The goal of flexible intramedullary nails is to improve fixation of the bone callus and limit medullary destruction. This was observed during lengthening of the upper limbs in particular the forearm, in the study by Launay et al. [23], with better union of the bone callus, shorter fixation, fewer deformities and no infection.

In the studies by Lascombes et al. [24–26], the Healing Index was reduced in the group with intramedullary nailing, with a reduction in fixation time and in the rate of fractures. These results were significant in particular in patients with congenital diseases of the femur and the forearm. No fractures occurred in their clinical series in 1994 [24], after lengthening in 14 segments (2 upper limbs and 12 lower limbs). The rate of fracture in their comparative series [25] was lower in the group with intramedullary nailing (1 type 1A fracture/92 cases in the group with nailing and 21 fractures/194 cases in the group without). These results included lengthening of lower and upper limbs.

In the series by Launay et al. [27], no fractures occurred after removal of the external fixator for lengthening of seven congenital femurs associating Flexible Intramedullary Nailing (FIN) and a monolateral fixator.

In the present series, the Healing Index was reduced in group 2 when intramedullary nailing was used, whatever the bone lengthened. The Healing Index of the femur was also reduced in group 1 although the difference was not significant. Fractures occurred in 6/44 bone segments (13.6%) in the population with intramedullary nailing. Fractures occurred in 14/67 segments (20.9%) in the population without intramedullary nailing. Although the fracture rate was reduced with intramedullary nailing, the difference was not significant. This confirms the results described above.

#### Lengthening in patients with achondroplasia

In patients with achondroplasia and associated diseases (group 2), relative lengthening can be as high as 50% of the lengthened segment without any increase in the fracture rate. This may be explained by the elasticity of the ligament and tendon in this disease [28–30]. In our series, we did not observe any relationship between the rate of lengthening and the fracture rate. On the other hand, we did observe that the fracture rate was significantly higher in patients who were under the age of nine (33.3%), compared to patients who were older than nine (0%), $P = 0.003$. We did not find any difference when we varied the age limit. In their work on progressive lengthening in achondroplasia, Aldegheri et al. suggest that lengthening should be performed later, between 12 and 16 years old [8,28,29]. Ganel et al. suggest that lengthening can be performed in boys at 8 years old in boys and at 15 in girls because union of the callus is poorer in the latter [30]. The fractures in our series were found in male patients, but there was no significant difference found in relation to female gender. In the literature, the delay between the osteotomy and beginning lengthening is 4–15 days [6–8,27–29,31]. In the series by Aldegheri et al. [29], a delay of 5 days was proposed in patients with achondroplasia. We observed an increased risk of fracture when the delay was less than 7 days (28.6%), while no fractures were observed when the delay was more than 7 days ($P = 0.01$). Moreover, the association of lengthening before the age of nine and less than 7 days delay before beginning lengthening resulted in a higher risk of fracture (1/2 segments were broken) and the statistical difference with the rest of the population (age older than 9 and delay of less than 7 days) was even greater $P = 0.0001$. We did not find any association between the two parameters (age of the patient and delay before lengthening) and the rate of fracture in the literature.
Fracture after lower limb lengthening in children

<table>
<thead>
<tr>
<th>Table 3</th>
<th>Distribution of fractures in relation to the presence of Flexible Intramedullary Nailing (FIN) (or stable elastic intramedullary nailing).</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Number of lengthenings</td>
</tr>
<tr>
<td>With FIN</td>
<td>44</td>
</tr>
<tr>
<td>Without FIN</td>
<td>67</td>
</tr>
<tr>
<td>NS: non-significant.</td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Table 4</th>
<th>Distribution of fractures in group 1 (congenital diseases).</th>
</tr>
</thead>
<tbody>
<tr>
<td>Congenital</td>
<td>Number of lengthenings</td>
</tr>
<tr>
<td>Femur</td>
<td>15</td>
</tr>
<tr>
<td>Tibia</td>
<td>43</td>
</tr>
<tr>
<td>Lengthening ≤15%</td>
<td>13</td>
</tr>
<tr>
<td>Lengthening &gt;15%</td>
<td>45</td>
</tr>
<tr>
<td>Delay ≤7 days</td>
<td>21</td>
</tr>
<tr>
<td>Delay &gt;7 days</td>
<td>37</td>
</tr>
<tr>
<td>S: significant; NS: non-significant.</td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Table 5</th>
<th>Distribution of fractures in group 2.</th>
</tr>
</thead>
<tbody>
<tr>
<td>Achondroplasia</td>
<td>Number of lengthenings</td>
</tr>
<tr>
<td>Femur</td>
<td>18</td>
</tr>
<tr>
<td>Tibia</td>
<td>16</td>
</tr>
<tr>
<td>Patients &lt;9 years old</td>
<td>12</td>
</tr>
<tr>
<td>Patients &gt;9 years old</td>
<td>22</td>
</tr>
<tr>
<td>Delay ≤7 days</td>
<td>14</td>
</tr>
<tr>
<td>Delay &gt;7 days</td>
<td>20</td>
</tr>
<tr>
<td>S: significant; NS: non-significant.</td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Table 6</th>
<th>Distribution of fractures depending on the presence or not of infection.</th>
</tr>
</thead>
<tbody>
<tr>
<td>Infections</td>
<td>Number of lengthenings</td>
</tr>
<tr>
<td>Infected segments</td>
<td>59</td>
</tr>
<tr>
<td>Non-infected segments</td>
<td>52</td>
</tr>
<tr>
<td>Infection w/ radiological sign</td>
<td>23</td>
</tr>
<tr>
<td>Infection w/o radiological sign</td>
<td>36</td>
</tr>
<tr>
<td>S: significant; NS: non-significant.</td>
<td></td>
</tr>
</tbody>
</table>

Lengthening in patients with congenital diseases

For Dahl et al. [32], the suggested rate of lengthening for the fewest number of complications (all complications combined, including fractures) should not be more than 15%. For Maffulli et al. [33], it should be less than 18%, and 25% for Karger et al. [34] and Antoci et al. [3,35]. The series by Kamlesh et al. [36] found that all fractures occurred in cases with lengthening of more than 30%. These results apply, in particular to congenital diseases (fibular hemimelia and congenital short femur) because in achondroplasia a rate of lengthening as high as 50% or more can be applied [27–29].

In our series, we did not find a relationship between the rate of fractures and the rate of lengthening in the achondroplasia and associated group, while fracture rate of 33.3% (15/45) was found in the congenital disease group when lengthening was more than 15%. On the other hand, when lengthening was 15% or less, the fracture rate was 7.7% (1/13), although the difference was not significant (P=0.06).

We chose a threshold of 7 days for the delay between surgery and lengthening in patients with congenital disease. Nine fractures/21 segments (42.9%) were observed when the delay was less than 7 days and 7/37 fractures when the delay was 7 days or more. The difference was not significant. On
the other hand, by associating a rate of lengthening of more than 15% and a delay of less than 7 days, we found fractures in all femurs, or 5 fractures/5 femurs. The difference was significant with the rest of the femurs in the congenital disease group. $P = 0.01$. It should be noted that none of these femurs had associated intramedullary nailing.

**Conclusion**

The rate of fracture after removal of external fixation in a lengthening program is still high in particular in patients with congenital length discrepancies of the lower limb. There is no single rule to avoid fracture in all cases. However, certain elements such as relative lengthening, delay between osteotomy and beginning lengthening or placement of intramedullary nails can markedly reduce their occurrence.

**Disclosure of interest**

The authors declare that they have no conflicts of interest concerning this article.

**References**

Fracture after lower limb lengthening in children


