Analysis of segmental residual growth after progressive bone lengthening in congenital lower limb deformity

D. Popkov\textsuperscript{a}, P. Journeau\textsuperscript{b,*}, A. Popkov\textsuperscript{c}, B. Pedeutour\textsuperscript{b}, T. Haumont\textsuperscript{b}, P. Lascombes\textsuperscript{d}

\textsuperscript{a} Scientific and Clinical Laboratory for Deformity Correction and Limb Lengthening. Federal Russian State-Financed Ilizarov Scientific Center for Restorative Traumatology and Orthopaedics, 6, M.-Ulyanova Street, 640014 Kurgan, Russia
\textsuperscript{b} Pediatric Orthopedic Surgery Dept, Nancy University Hospital Center, Children's Hospital, 11, allée du Morvan, 54511 Vandœuvre, France
\textsuperscript{c} Inter-Regional Orthopedic Center of the Federal Medico-Biologic Agency, 5 Plekhanov Street, 634000, Tomsk, Russia
\textsuperscript{d} Pediatric Orthopedic Surgery Dept, Geneva University Hospitals, Children's Hospital, 6, rue Willy-Donzé, Geneva 14, 1211 Switzerland

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KEYWORDS
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Congenital length discrepancy;
Lower limb;
Residual limb;
Children;
Limb length discrepancy

Summary

Introduction: The issue of prognosis in limb length discrepancy in children affected by congenital abnormality remains a subject of concern. Therapeutic strategy must take length prediction into account, to adapt equalization techniques and the timing of treatment. Initial prognosis, however, may need revising after completion of one or several surgical interventions on the pathologic limb. The aim of this study was to determine the different types of growth response that a bone segment can present after progressive lengthening in case of congenital limb length discrepancy.

Materials and methods: A series of 114 bone lengthenings with external fixator, performed in 36 girls and 50 boys with congenital lower limb length discrepancy, was retrospectively analyzed. Bone segment growth rates were measured before lengthening, during the first year after frame removal and finally over long-term follow-up, calculating the ratios of radiological bone length to the number of months between two measurements. Mean follow-up was 4.54 ± 0.2 years. Results: Changes in short- and long-term growth rate distinguished five patterns of bone behavior after lengthening, ranging from growth acceleration to total inhibition.

* Corresponding author. Tel.: +33 3 83 15 47 07; fax: +33 3 83 15 46 91.
E-mail address: p.journeau@chu-nancy.fr (P. Journeau).

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Discussion: These five residual growth patterns depended on certain factors causing acceleration or, on the contrary, slowing down of growth: age at the lengthening operation, percentage lengthening, and minimal period between two lengthenings. These criteria help optimize conditions for resumed growth after progressive segmental lengthening, avoiding conditions liable to induce slowing down or inhibition, and providing a planning aid in multi-step lengthening programs.

Level of evidence: Level IV. Retrospective study.

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Introduction

Residual growth after progressive lengthening has been the focus of several studies [1–9], but the evolution of congenital lower-limb length discrepancy in children undergoing progressive lengthening remains unforeseeable and the factors relevant to bone segment activity in these cases remain undetermined. Both slowing and boosted growth patterns have been reported, independently of the lengthening technique (Judet, Wagner, Callotasis or Cauchois) employed [2–6], other authors finding no change in growth rates [7–9].

The present retrospective study sought to identify factors affecting residual growth after progressive lengthening: age at lengthening, percentage lengthening, and single- or multistep strategy.

Material and methods

Residual bone segment growth after progressive lengthening was studied on a retrospective design. The series comprised 36 girls and 50 boys, with a mean bone age of 8.5 ± 0.23 years (range, 4.5 to 15 yrs) at first lengthening operation. The series was homogeneous for etiology and surgical procedure. It comprised 114 consecutive single-segment lengthening procedures using the Ilizarov technique (circular fixation, percutaneous osteotomy, and lengthening initiated as of postoperative day 5), and involved 59 femurs and 55 tibias with exclusively congenital etiology [10,11]. Eighty-seven procedures were primary and 24 iterative.

Mean femoral lengthening was 4.8 ± 0.16 cm (range, 2 to 6 cm; i.e., 19.7 ± 1.02%), and mean tibial lengthening 4.9 ± 0.26 cm (1.5 to 10 cm; i.e., 21.2 ± 0.62%). Mean consolidation index [12] was 26.3 ± 1.22 days/cm for the femur and 27.3 ± 1.94 days/cm for the tibia.

Any associated axial deformities were corrected during lengthening, to ensure a normal biomechanical lower-limb axis in all cases. The knee joint was bridged in case of clinical joint instability, with distraction maintained throughout the lengthening process.

The external fixator (EF) was removed as soon as consolidation was confirmed (homogenous callous formation, complete disappearance of regenerate bone area, and appearance of at least three cortices) on X-ray. Mean follow-up after fixator ablation was 4.54 ± 0.2 years.

Segmental growth was monitored 6-monthly or yearly from 1 year before to at least 2 years after lengthening, by comparative whole lower-limb X-ray of the lengthened and contralateral healthy segments. Bone age according to Greulich and Pyle was determined once a year.

Radiologic length (L), growth rate (R) and change in growth rate (C) were measured as follows:

Radiologic length (L) of each healthy and lengthened segment (Fig. 1) was measured:

- L0: 1 year before surgery;
- L1: immediately before surgery (L1);
- L2: 4 to 6 weeks after EF removal;
- L3: 7 to 12 months after EF removal;
- L4: greater or equal to 2 years after EF removal.

Growth rate (R, in mm/month) was determined in terms of differential radiologic length for a given segment:

- R0 = L1 − L0/month: growth rate before lengthening;
- R1 = L3 − L2/month: growth rate during the first year after EF removal (or "short-term growth rate");
- R2 = L4 − L3/month, "long-term growth rate", following the first year after EF removal.

Figure 1 Diagrammatic presentation of residual growth gain (RGG) calculation, taking account of length gain (LG), predicted final spontaneous segmental discrepancy (PFSSD) in case of no lengthening (mm), final residual segmental discrepancy (FRSD) (mm) obtained by subtracting healthy segment length from lengthened segment length, and final lengthened segment length (FLSL).

Following the criteria defined by Suva et al. [5], a length difference of 2 mm was counted as a change in growth for an observation period of at least 3 months.

Change in growth rate (Δ) was calculated from the following ratios:

\[
\Delta_1 = \frac{R_{t1} - R_{t0}}{R_{t0}} \quad \text{Short-term growth-rate change, during the first year after EF removal}
\]

\[
\Delta_2 = \frac{R_{t2} - R_{t1}}{R_{t1}} \quad \text{Long-term growth-rate change, following the first year after EF removal}
\]

Hechard-Carlioz charts [13] were used for monitoring femoral and tibial longitudinal growth. Preoperatively, predicted limb-length discrepancy and final segment length were assessed using the multiplier method [14] taking into account that, without treatment, the natural growth of a malformed segment is linear, with a constant rate, so that change in percentage discrepancy will likewise be linear [15].

Residual growth was studied by grouping the 114 lengthening procedures according to whether the growth phase was ongoing (group 1: n = 71) or bone maturity had been reached (group 2: n = 43), and further distinguishing between those studied over both the short and the long term (both during and after the first year after external fixator removal: n = 99) and those studied only over the long term (at least 1 year after external fixator removal: n = 15). The latter growth-rate analysis provided as precise an analysis as possible of the behavior of the lengthened segment, both as of fixator ablation and over the long term, regardless of pubertal status.

To investigate possible growth-rate change after lengthening, whether stimulation, transitory slowing of growth during the first year or definitive cessation of growth, the following index was calculated at end of growth phase:

\[
\text{RGG} (%) = \frac{PFSSD - (LG + FRSD)}{FLSL} \times 100
\]

where: RGG = residual growth gain; PFSSD = predicted final spontaneous segmental discrepancy (mm); LG = length gain achieved by external fixation (mm); FRS = final residual segmental discrepancy (mm); healthy segment length minus lengthened segment length; and FLSL = final lengthened segment length (mm) (Fig. 1).

This index represents the change in spontaneous growth after progressive lengthening with respect to the final length of the lengthened segment; it may be positive (growth stimulation), neutral (no change) or negative (slowing of spontaneous growth).

**Statistical methodology**

Change in growth rate was first assessed over the short and long terms (1 year after the end of the lengthening procedure and at end of follow-up: n = 99), and then in terms of bone maturity (group 1, before full maturity, n = 71; and group 2, after full maturity, n = 43). Factors potentially affecting growth rate were then assessed: age at procedure, percentage lengthening and single versus multi-step lengthening.

Analysis used the Student t test for independent samples, on StatPlus® Professional (2008) software, with the significance threshold set at P < 0.05.

**Results**

Short- and long-term growth rates both showed 3 distinct behavior patterns: acceleration, no change, and slowing. Procedures were therefore grouped first according to their associated short-term growth rate pattern: group A, with no slowing down of growth (i.e., either transitory acceleration or no change), and group B with slowed growth:

- group A showed no short-term slowing of growth, followed by 1 of 3 long-term patterns: acceleration (I), no change (II) or slowing (III);
- group B, which showed slowing of growth over the short term, then showed either resumption (I) or definitive cessation (II) of growth over the long term.
Figure 2  Representation of the five types of residual segmental growth following progressive bone lengthening for congenital length discrepancy.

Five patterns of growth rate change could thus be distinguished (Table 1 and Fig. 2). Types AI and AII are associated with good prognosis for final residual growth, and types AIII, BI and BII with poor prognosis:

- type AI: no change in or transitory acceleration of initial growth rate during the first year after EF ablation, with acceleration over the long term;
- type AII: no change in or transitory acceleration of initial growth rate during the first year after EF ablation, with growth progressively returning to its pre-procedural rate over the long term;
- type AIII: no change in or transitory acceleration of initial growth rate during the first year after EF ablation, followed over the long term by a reduced growth rate, falling below the initial pre-procedural rate;
- type BI: slowing or transitory cessation of during the first year after EF ablation, with long-term growth recovering or even exceeding the initial pre-procedural rate;
- type BII: slowing (or cessation) of growth during the first year after EF ablation, followed over the long term by continued slowing of growth until definitive cessation.

The differences between these five types were statistically significant; types AI and AII showed positive RGG and types AIII, BI and BII showed negative RGG.

Factors impacting growth rate

Certain factors could be identified as affecting the various residual growth patterns in the lengthened segments.

Age at initiation of lengthening
Age at initiation of lengthening directly impacted growth rates.

Figs. 3 and 4 show percentage residual growth gain according to age at initiation of 1-step lengthening in male and female children. The results show that initiation before the age of 12 years in boys and 9 years in girls was associated with long-term stimulation of spontaneous growth (types AI and AII, corresponding to positive RGG). Initiation immediately after the onset of the pubertal growth boost, in contrast, was associated with slowing of growth and thus with a negative RGG. In this subgroup of 1-step lengthening, the greatest reductions in RGG were found in case of initiation at 9–12 years bone age in girls and 12–14 years in boys. This accounts for the slowed growth associated with first lengthening procedure in the oldest children (types AII and BII). We suspect that the negative effect is greatest when the lengthening program is performed during the lower-limb pubertal growth boost, although this boost was not explicitly assessed in the present study; therefore defined a bone age interval as a factor meant to approximate the pubertal phase, within which lengthening procedures should be avoided.

These results were also found on analysis of groups 2 and 1 (having reached cessation of growth or not, respectively, at the time of study): growth was significantly slowed in children who were older at the time of lengthening, in both groups 1 and 2 (Tables 2 and 3); i.e., slowed growth of type...
Table 2  Distribution of results according to long-term change in growth rate in patients before the end of their growth phase (group 1: n = 71).

<table>
<thead>
<tr>
<th>Parameters</th>
<th>Acceleration</th>
<th>No change</th>
<th>Slowing</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number: 71 lengthenings</td>
<td>34</td>
<td>22</td>
<td>15</td>
</tr>
<tr>
<td>Age</td>
<td>7.1 ± 0.28</td>
<td>7.2 ± 0.41</td>
<td>8.4 ± 0.40</td>
</tr>
<tr>
<td>Length gain (cm)</td>
<td>5.0 ± 0.24</td>
<td>4.6 ± 0.35</td>
<td>5.1 ± 0.48</td>
</tr>
<tr>
<td>% length gain</td>
<td>21.7 ± 1.57</td>
<td>19.0 ± 2.37</td>
<td>22.2 ± 3.11</td>
</tr>
<tr>
<td>First lengthening</td>
<td>34</td>
<td>21</td>
<td>9</td>
</tr>
<tr>
<td>Iterative lengthening</td>
<td>0</td>
<td>1</td>
<td>6</td>
</tr>
<tr>
<td>Femoral HI</td>
<td>25.9 ± 2.06</td>
<td>24.8 ± 1.85</td>
<td>27.0 ± 3.01</td>
</tr>
<tr>
<td>Tibial HI</td>
<td>23.5 ± 1.37</td>
<td>21.8 ± 2.96</td>
<td>20.2 ± 2.05</td>
</tr>
<tr>
<td>Knee bridging</td>
<td>14</td>
<td>5</td>
<td>10</td>
</tr>
<tr>
<td>Δ₂</td>
<td>0.38 ± 0.05</td>
<td>0.02 ± 0.01</td>
<td>−0.43 ± 0.07</td>
</tr>
<tr>
<td>FU</td>
<td>4.2 ± 0.43</td>
<td>4.5 ± 0.51</td>
<td>4.4 ± 0.54</td>
</tr>
</tbody>
</table>

* All, All — Significant difference between groups (P < 0.05).

Table 3  Distribution of results according to long-term change in growth rate in patients after the end of their growth phase (group 2: n = 43).

<table>
<thead>
<tr>
<th>Parameters</th>
<th>Acceleration</th>
<th>Acceleration</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number: 43 lengthenings</td>
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<td>14</td>
</tr>
<tr>
<td>Age</td>
<td>7.8 ± 0.57</td>
<td>10 ± 0.4</td>
</tr>
<tr>
<td>Length gain (cm)</td>
<td>4.8 ± 0.34</td>
<td>4.7 ± 0.58</td>
</tr>
<tr>
<td>% length gain</td>
<td>25.9 ± 4.22</td>
<td>18.3 ± 3.32</td>
</tr>
<tr>
<td>First lengthening</td>
<td>7</td>
<td>12</td>
</tr>
<tr>
<td>Iterative lengthening</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Femoral HI</td>
<td>24.9 ± 2.31</td>
<td>22.8 ± 1.99</td>
</tr>
<tr>
<td>Tibial HI</td>
<td>23.8 ± 4.82</td>
<td>34.4 ± 6.18</td>
</tr>
<tr>
<td>% RGG</td>
<td>2.5 ± 0.35</td>
<td>0.8 ± 0.39</td>
</tr>
<tr>
<td>Knee bridging</td>
<td>5</td>
<td>4</td>
</tr>
<tr>
<td>Δ₂</td>
<td>0.36 ± 0.08</td>
<td>0.03 ± 0.01</td>
</tr>
<tr>
<td>FU</td>
<td>6.3 ± 0.44</td>
<td>5.3 ± 0.24</td>
</tr>
</tbody>
</table>

* All, All — Significant difference between groups (P < 0.05).

BI or BII did not depend on the child having reached the end of the growth phase.

Number of lengthening procedures per segment
Stimulation or resumption of normal growth (types I or II) was observed in most cases of first lengthening: 57 out of 60 (95%).

In contrast, in both femur and tibia, iterative lengthening severely impaired residual growth, even when performed outside of the pubertal growth boost period. Significant slowing followed by arrest (type BII) was observed in 17 of the 24 cases of iterative lengthening (70.8%), independently of end of growth: in both groups 1 and 2, slowing of growth was mainly observed in cases of iterative lengthening (Tables 2 and 3).

Interval between lengthening procedures
Growth disorder was mainly associated with iterative lengthening of the same segment. RGG at end of growth was negative (−1.8% to −12.8%) when the interval between procedures lay between 1 and 3 years, but was positive when the second lengthening procedure was performed before the pubertal growth boost and more than 3 years after the first procedure. Obviously, no slowing effect occurred when the second lengthening procedure was performed toward the end of the growth phase (Fig. 5).

Percentage lengthening
When the percentage lengthening in a first procedure exceeded 30% of initial segment length, transitory slowing of growth was observed during the first year after the procedure, followed by a return to the preoperative growth rate (type BII): i.e., negative RGG at end of growth. Percentage lengthening of less than 30%, however, showed no significant impact, and other factors must therefore be implicated.

No significant effects on residual growth rates were associated with external osteosynthesis duration or knee bridging.

Discussion

In congenital lower-limb length discrepancy, lengthening procedures often need to be performed before the end of spontaneous growth, due to the functional and psychological impact of the discrepancy on child and family. Prognosis for the lengthened segments should be assessed as rigorously as possible, so as to adapt the lengthening strategy.

The present series exclusively included congenital pathology, making it homogeneous, with a large enough sample in terms of statistical power to analyze the results. McCarthy’s et al. results were less clear-cut, but his series of discrepancies involved a variety of etiologies, which may account for the difference in results [9]. As congenital discrepancies show a percentage length difference that remains stable over growth in absence of treatment, we accepted the hypothesis that any observed change in growth pattern would be identical across all congenital etiologies, independently of segment location.

Growth stimulation following a first Ilizarov lengthening procedure was systematic when bone age was less than 12 years in boys or 9 years in girls, and could reach 1.1 to 4.7% of final lengthened segment length.

This mechanism may be explained by stimulation due to increased vascularization during lengthening, or by a mechanical cause [5]. This growth-stimulation effect was described in fracture, but lasted less than 2 years [16,17]. Some reports stressed the importance of having a correct biomechanical axis and of functional loading to minimize disturbance of conjunctive cartilage growth over periods exceeding 2 years [18,19]. Experimental studies showed that cell number per column in the growth area depends on the forces acting on the cartilage, increasing with increased loading. The greater the number of cells, the more active the cartilage [20]. Antonov demonstrated the growth-stimulating effect of functional forces under dynamic loading [18].

External fixation allows all deformities to be progressively corrected, avoiding impairment of joint range of motion during lengthening and ensuring a perfect biomechanical axis in three dimensions [21–24]. Malformed limbs are often misaligned, and the correction site is often epiphyseal, metaphyseal or meta-diaphyseal. Joint bridging avoids inducing subluxation and hyperpressure threatening the conjunctive cartilage. External fixation thus ensures optimal conditions for resumed growth, correcting conjunctive cartilage obliquity and producing a regular distribution of force, thereby improving lower-limb function in stance and gait.

Oostenbroek et al. recommended bridging the knee joint and maintaining joint distraction force throughout the period of external osteosynthesis in order to obtain subsequent growth stimulation [25]; they did not specify, however, whether distraction was applied throughout the lengthening process in their series, nor did they measure the distraction force between rings. Penneco et al. [2] reported a significant impact on the growth plate after progressive bone lengthening by Judet distractor. The harmful effects of hyperpressure on the conjunctive cartilage, impaired vascularization and the resultant shaft ischemia may account for subsequent slowing of growth. However, the 1–2 mm initial joint-line distraction reported by Pennecot et al. was probably insufficient to prevent contact between the joint surfaces and avoid shaft compression [24]. In the present series, there was no significant difference in residual growth rate associated with bridging of the knee, and no conclusion can thus be drawn as to whether bridging, performed basically to avoid joint impairment, also protects the tibial shaft.

The reduction in growth observed, however, was not necessarily related to the hyperpressure effects described by the above authors. Obtaining and maintaining a normal axis during the lengthening procedure seems to us to be a necessary but not sufficient condition for growth to be conserved; other factors are involved [23,24].

Percentage lengthenings emerged as one of the main factors impacting residual growth. An experimental study, however, reported that a lengthening of 30% did not affect longitudinal tibial growth [26]; these conclusions were drawn from morphological results at only 5 weeks’ follow-up. Clinical data, in contrast, demonstrate the negative impact of such a length gain. Viehweger et al. [6], in series of 34 children, reported a clear impact on residual growth following lengthening exceeding 30% using a unilateral external fixator. In the present series, this result corresponds to residual growth types BI and BII, where excessive percentage lengthening induces slowed growth over the first year following the procedure. It can reasonably be concluded that single-step lengthening of more than 30% of initial segment length is to be avoided in congenital lower-limb length discrepancy, as it may impair resumption of growth. In the present series, percentage lengthenings less than 30% had very little impact if any on growth. In groups BI and BII, other factors may be considered to have had much more harmful effect (Tables 1–3).

Osseous age at time of lengthening is a factor to be taken into account in treatment planning in congenital lower-limb abnormality. Pouliquen et al. [3] reported that lengthening performed before the age of 11 years induced growth disorder; this series, however, included only six children with congenital discrepancy. The present results, on the contrary, showed greater growth stimulation gain in children aged less than 8 years at time of procedure. These findings are in agreement with those of Suva et al. [5], who reported accelerated growth of a mean 2.5% in all children treated at less than 6 years of age.

Analysis of segmental residual growth after progressive bone lengthening

Other authors have shown that lower-limb lengthening performed just after the start of the pubertal growth boost inevitably slows growth in the operated segment [27]. This is confirmed by the present findings: lengthening performed after the start of the pubertal growth boost (here taken as after 12 years’ bone age in boys and 10 in girls, due to the impossibility of having a reliable physiological estimate of pubertal maturity) led to a negative RGG and all the more so in the early phases of puberty.

The exact mechanism of slowed growth can only be guessed at. During the intense pubertal spontaneous growth phase, the soft tissue of the lengthened segment cannot adapt to the change in length, which may result in hyperpressure on the growth plate. Suva et al. [5] suggest a phenomenon of growth capital exhaustion to account for slowing.

It also emerged that iterative lengthening of the affected segment, even when performed before the pubertal growth boost and remaining less than 30% of initial segment length, induced negative RGG when conducted at close intervals (of less than 3 years). These findings may guide planning length equalization when the discrepancy prognosis requires iterative lengthening [28]: iterative lengthening of a segment should be undertaken before the beginning of the pubertal growth boost but more than 3 years after the first procedure. The alternative is to perform the second lengthening around the end of spontaneous growth. This program can thus be applied to manage severe femoral or tibial hypoplasia.

In the light of these results, and on condition that the principles of concomitant correction of associated deformities are respected while protecting the joint by bridging if necessary to avoid or limit growth plate hyperpressure, certain favorable factors emerge for resumed growth in the lengthened segment:

- the upper bone age limit for initiating lengthening is 12 years in boys and 9 years in girls;
- lengthening should be systematically associated to restoring the mechanical axis of the lower limb;
- any second lengthening of the same segment should be performed before the beginning of the pubertal growth boost but more than 3 years after the first, or else around the end of spontaneous growth.

Factors liable to induce slow growth are:

- iterative lengthening less than 3 years’ interval;
- lengthening performed just after the beginning of or during the pubertal lower-limb growth boost;
- length gain exceeding 30% of initial segment length.

Conclusion

It is fundamental to the treatment strategy for severe congenital deformity and length discrepancy to respect or optimize subsequent resumption of growth in the lengthened segment. “Therapeutic” growth stimulation, as found in trauma cases, is here an illusion. The equalization program should take account of the factors detailed above as affecting spontaneous growth in the lengthened limb in case of congenital discrepancy; these factors may be somewhat different in the case of acquired limb-length discrepancy or of the upper limbs.

Disclosure of interest

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

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