

Regenerate bone fracture rate following femoral lengthening in paediatric patients

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Abstract

Purpose Femoral lengthening using a circular or mono-lateral frame is a commonly used technique. Fracture at the site of the regenerate bone is a major concern especially following removal of the external fixator. This aim of this study was to assess the rate of fracture of the regenerate bone in this single surgeon series of paediatric patients and determine potential risk factors.

Methods Retrospective review of all the femoral lengthening performed by the senior author was performed. The medical and physiotherapy notes were reviewed. The gender, age at time of surgery, disease aetiology, total days in the external fixator and length of the new regenerate bone were recorded. Patients who sustained a regenerate fracture were identified.

Results A total of 176 femoral lengthening procedures were performed on 108 patients. Eight regenerate fractures occurred in seven patients (4.5%). The mechanism of injury was a fall in five cases and during physiotherapy in three cases. The regenerate fracture occurred a median number of nine days following removal of frame. There was no significant difference between gender, age at time of surgery, total time in external fixator between those who sustained a regenerate fracture and those patients who did not. A significant difference was noted between the amount of lengthening between the 'regenerate fracture group' and the 'no fracture group' (50 mm vs 38 mm, respectively; $p = 0.029$). There was no association between disease aetiology and risk of regenerate fracture.

Conclusions Femoral lengthening of more than 50 mm increases the risk of a fracture at the regenerate site regardless of the disease aetiology. We recommend avoidance of aggressive physiotherapy for the initial four weeks following external fixator removal.

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Introduction

Distraction osteogenesis using an external frame fixator is now a well-established surgical procedure to lengthen the femur.¹⁻⁴ It allows for the gradual correction of multi-planar deformities of the lower limb as well if necessary. This method of limb length correction may be preferable to epiphysiodesis of the contralateral femur, which would result in an overall loss of height. This would lead to an unsatisfactory outcome for many paediatric patients with a congenital, short stature, post-infectious or post-traumatic cause.

Femoral lengthening with a circular or mono-lateral frame is associated with complications such as infection, joint contractures, neurological deficit, pseudoarthrosis and early union.^{2,5-7} Another major complication is fracture occurring at the site of the regenerate bone after the removal of the external fixator. Rates vary in studies between 3.6% and 50%.^{8,9} To prevent or minimise the risk of fracture it has been suggested that the external fixator should remain in place until the newly formed bone is 'radiologically mature', along with protection of the limb following removal of the frame with a support such as a cast.¹⁰

To assess the maturity of the callus is challenging for an orthopaedic surgeon. Fischgrund et al recommends three continuous cortices, at least 2 mm thick on plain radiographs.¹¹ Other methods such as ultrasound, DEXA scan and CT scan are also used to assess the quality of new bone formation.^{8,12,13} The aim of this study was to assess the fracture rate of the regenerate bone following removal of the circular frame in this large single surgeon series

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and identify any potential risk factors in these paediatric patients.

Patients and methods

Institutional review board approval was granted for this study. A retrospective review of paediatric patients who had undergone a femoral lengthening procedure by the senior author was performed. Between 1993 and 2013, 176 femoral lengthening procedures were performed on 108 patients. These patients were identified from a prospective database. Patients over the age of 17 years or with removal of the external fixator after October 2013 were excluded from the study. The medical and physiotherapy notes were reviewed for each patient. The age at time of surgery, gender, disease aetiology, total number of days in the circular or mono-lateral frame (inclusive) and length of the regenerate bone for each patient were recorded. All patients were included in the retrospective review.

The patients who sustained a fracture at the site of regenerate were identified. The number of days following removal to frame to the fracture was obtained, as well as the mechanism of injury. The radiographs of these patients were reviewed with recording of the original femoral length, the length of the regenerate following distraction with the external fixator, the percentage increase in length compared with the original femur and the callus width at the time of frame removal. The bone healing index was calculated as the number of days of external fixation per centimetre of lengthening. The fracture of the regenerate bone was classified according to Simpson et al.²

For comparison purposes, the patients were initially divided into the regenerate fracture group and the no fracture group. These were then further categorised into four groups according to the disease aetiology, which resulted in the patient requiring the femoral lengthening. Group 1 comprised patients with congenital aetiology; group 2 consisted of short stature patients (including achondroplasia); group 3 comprised patients with a post-traumatic or post-infectious cause; and group 4 included patients requiring femoral lengthening as a result of tumour excision.

Descriptive statistics are reported for as number of people for categorical variables, and median (range) are used for continuous variables. The data collected did not follow a normal distribution for the 'regenerate fracture group' and the 'no fracture group', or for the four categories of the disease aetiology. The median scores are discussed, and the non-parametric Mann-Whitney U test was used to evaluate the significance of the relationship between the age, length of time in external fixator (inclusive days) and amount of femoral lengthening obtained (mm) of the regenerate fracture group compared with those which did not fracture. The Kruskal-Wallis test was used to assess

these variables in the four different aetiology groups. The regenerate fracture rate was assessed at five-year intervals. The p value was considered statistically significant if less than 0.05.

Surgical technique

All surgical procedures were performed by the senior author (DM). A three or four ring circular frame construct (Ilizarov, Smith & Nephew, London, UK; Taylor Spatial Frame, Smith & Nephew) is applied depending on whether a unifocal or bifocal osteotomy is required. Fixation is obtained distally with tensioned olive-tipped fine wires and proximally with half pins. Minimal circumferential periosteal stripping is performed at the osteotomy site. The multiple 'drill-hole technique' is used, with completion of the corrective osteotomy with an osteotome. This is usually performed at the distal femoral metaphysis or at the level of the centre of rotation of angulation (CORA) in those with a more complex deformity. Chlorhexidine-soaked dressings are applied around the pin sites and held in place with rubber bung.

Lengthening was commenced at day 6 post-surgery, at a rate of 1 mm per day over four increments. Patients are allowed to mobilise full weight-bearing with crutches and commence knee, hip and ankle mobilisation exercises prior to discharge by the physiotherapy team. Each patient is counselled by the limb reconstruction clinical nurse specialist prior to each procedure on the risk and benefits of the lengthening operation and provided with a video explaining the procedure and the post-operative regime. This information is again reinforced prior to discharge. All patients are followed up regularly at the outpatient clinic, and the distraction rate is adjusted according to the radiological evidence of bone formation. Once the desired length is achieved, consolidation of the regenerate bone commences. The senior author applies the Fischgrund principles regarding removal of the frame. After removal, the patient can mobilise full weight-bearing with crutches to assist with balance and discontinue when confident to mobilise independently. A mono-lateral fixator (Orthofix, TX, USA) is used occasionally for femoral lengthening. This was usually for patients undergoing bilateral simultaneous lower-limb lengthening. A similar low-energy osteotomy is performed, and the post-operative lengthening and physiotherapy regime remain unchanged.

Results

A total of 176 femoral lengthening procedures were performed in 108 patients over a 20-year period. A frame system was used to treat 165 and 11 were treated with a mono-lateral external fixator. There was a total of eight fractures in seven patients (four females, three male) at

the regenerate site following removal of the external fixator (4.5%) over 20 years. From the eight cases of regenerate fracture, six cases were treated with a circular frame system while two cases were treated with a mono-lateral external fixator. In the first five years, 61 femoral lengthening procedures were carried out with a regenerate fracture rate of 9.8%. The regenerate fracture rate for the following five-year period declined to 4.3% in 46 femoral lengthening procedures. There were no regenerate fractures in the third (49 procedures) and fourth (20 procedures) five-year interval periods. The median number of days the regenerate fracture occurred after removal of the external fixator was nine. All regenerate fractures occurred within four weeks of frame removal. The characteristics of the overall study sample are outlined in Table 1.

The descriptive statistics for the regenerate fracture group are shown in Table 2. There was no significance

Table 1. Overall characteristics of the patients which underwent femoral lengthening

| Demographics | |
|---|----------------------|
| Patients (n) | 108 |
| Lengthening | 176 femurs |
| Gender | 61 males, 47 females |
| Mean age at time of surgery (years) | 11.2 (2.8-16.9) |
| Total time in external fixator (months) | 6.42 (2.3-45.3) |
| Median lengthening of the femur (mm) | 42 (10-220) |
| Median Bone Healing Index (days/cm) | 47.2 |

Table 2. Descriptive statistics of the regenerate fracture group (n = 8)

| Descriptive statistics | Median value (interquartile range) |
|--|------------------------------------|
| Age at time of surgery (years) | 10.0 (9.4-13.9) |
| Median time in external frame (days) | 184 (155-218) |
| Median amount of femoral lengthening (mm) | 50 (45.5-67.5) |
| Median percentage lengthening (%) | 18.7 (15.5-27.9) |
| Median time to fracture after removal of the frame (days) | 9 (3.5-15.5) |
| Bone Healing Index (days/cm) | 33.5 (26.0-43.9) |
| Median callus width of regenerate at time of removal of frame (mm) | 17.8 (15.0-18.5) |

Table 3. Descriptive statistics for the four different disease aetiology groups

| | Femoral lengthening (n) (n = 176) | Median age at time of surgery (years) | Median time in external frame (days) | Median amount of lengthening (mm) | Median Bone Healing Index (days/cm) | Regenerate fractures (n) |
|--|-----------------------------------|---------------------------------------|--------------------------------------|-----------------------------------|-------------------------------------|--------------------------|
| Congenital | 73 | 10.5 (5.7-14.7) | 162 (128-218) | 40 (30-50) | 43.6 (34.8-62.7) | 5 |
| Short stature (including achondroplasia) | 19 | 13.0 (11.0-14.7) | 154 (127-210) | 23 (20-43) | 58.6 (35.3-84.7) | 2 |
| Post trauma / post infection | 56 | 11.0 (7.7-15.0) | 175 (137-250) | 40 (29-50) | 44.7 (34.3-61.1) | 1 |
| Tumour | 28 | 12.4 (8.9-14.1) | 173 (116-249) | 37 (20-56) | 48.3 (34.0-73.6) | 0 |

Value (interquartile range)

difference in age ($p = 0.667$), gender ($p = 0.541$) and total time in external fixator ($p = 0.506$) when comparing the regenerate fracture group ($n = 8$) with those patients which did not fracture ($n = 168$). There was a statistical difference in length of the regenerate bone between the two groups with the regenerate fracture group lengthened significantly more (50 mm vs 38 mm; $p = 0.029$). The percentage of femoral lengthening compared with the original femoral length was 18.7%. All regenerate fractures were Type 1B according to the Simpson classification (i.e. through the regenerate bone).

The disease aetiology of patients who underwent femoral lengthening is outlined in Table 3. There was no significance between age at time of surgery, amount of lengthening or bone healing index between the four groups of differing disease aetiology. The mechanism of injury in five cases was a traumatic fall. Two patients sustained a fracture following physiotherapy, which involved a concomitant bilateral femoral fracture in a patient with achondroplasia (Fig. 1a-d). One patient had replacement of a circular frame to stabilise the regenerate fracture site and the rest were treated in a long leg cast (Fig. 1e). All the regenerate fractures went on to heal and had no further complications.

Discussion

This is the largest known published series for femoral lengthening using an external fixator such as a circular frame or mono-lateral frame in paediatric patients by a single surgeon in the English literature. The rate of regenerate fracture was 4.5% in this series and this is comparable with the other studies with a regenerate fracture rate of between 3.6% and 5% following removal of the external fixator.^{2,3,8,14} The initial five years demonstrate an elevated regenerate fracture rate of 9.8% in the first 61 femoral lengthening procedures, which decreased gradually to 4.3% and to zero in the subsequent ten years, demonstrating a learning curve in femoral lengthening.

A low-energy osteotomy improves the quality of the new bone formation, commencing distraction of the bone ends at around day 6 or 7, and also by a slow steady daily distraction rate (usually 1 mm per day).^{1,15-17} It has been

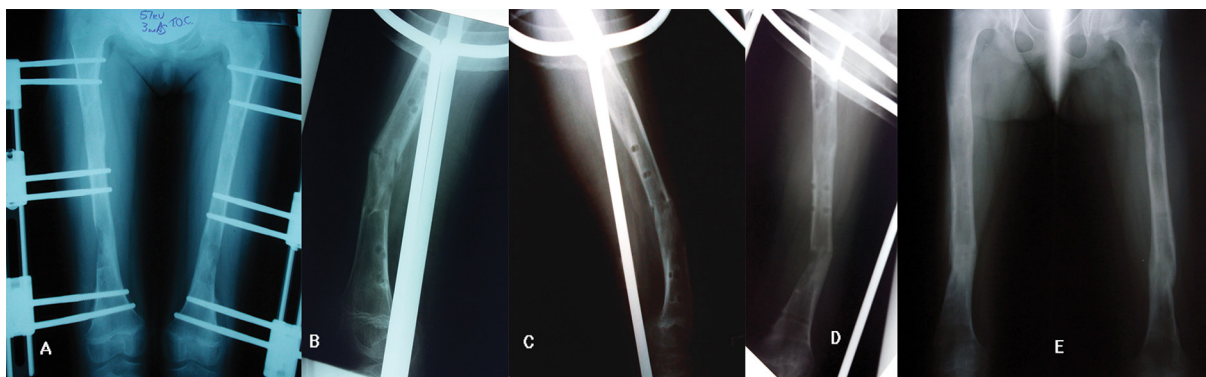


Fig. 1 (a) Anteroposterior (AP) radiographs during femoral lengthening, (b-d) AP and lateral femoral radiographs four days post treatment and physiotherapy, (e) AP radiographs after the regenerate fracture has healed.

shown that when the regenerate bone is of poor quality the fracture will occur within the regenerate, while if the bone quality is good the fracture will occur at the junction of the regenerate and original bone.⁴ All fractures in this series occurred within the regenerate bone suggesting the bone quality was poor, and all occurred within three weeks (three to 18 days) of removal of the frame. Poor quality or immature bone is a major factor in a regenerate fracture occurrence. Five of the cases were associated with a traumatic fall and three cases occurred during physiotherapy soon after removal of the frame. The stresses applied to this immature or poor quality bone was too great especially during the mobilisation exercises. In view of this, we recommend a gentle physiotherapy regime during the first four weeks following removal of the external fixator.

In our study, a nine-year-old girl with achondroplasia underwent bilateral femoral lengthening with mono-lateral frames, and sustained bilateral femoral fractures during a physiotherapy session three days after removal of the frame. In such cases, Ganel et al suggested that lengthening in girls should be at the age of 15 years and in boys at eight years as inadequate union of callus occurs if commenced before these ages.¹⁸ Aldehegri recommended lengthening should be performed between the ages of 12 and 16 years.⁴ Launay et al observed that their fracture rate in patients with achondroplasia was significantly higher in those aged under nine years. In our study, patients who underwent femoral lengthening for short stature (including achondroplasia) had a regenerate fracture rate of 10.1% (2 fractures out of 19 lengthening procedures).¹⁹ The median age at time of surgery in this group in our study was 13.0 years and median lengthening was 23 mm.

In limb-lengthening procedures, educating the patient and their parents regarding the surgery and the post-operative mobilisation regime is important and a limb reconstruction clinical nurse specialist is fundamental for follow-up and management of these patients. Reinforcement of the need for protected full weight-bearing during

the period that the patient has the external fixator is vital. Development of a good relationship between the patient and orthopaedic team allows for any queries or potential complications to be highlighted and addressed at an early stage. Awareness of the increased risk of fracture after the external fixator is removed is emphasised to the patient especially in the initial four-week period.

In our study, the percentage of femoral lengthening in the fracture group was 18.7%. A previous study showed that femoral lengthening more than 6 cm, or exceeding 20% of the original length in congenital cases, increases the risk of fracture.²⁰ However, others have recommended that congenital lengthening should not be more than 15% of the original length of the femur.²¹ Devmurari et al showed that regenerate fractures occurred in their study of congenital patients when lengthening was more than 30%.²² In short stature patients, studies have recommended 4 cm to 6 cm of lengthening or 8% to 22% of the initial bone length.²³⁻²⁶ In our study, lengthening of more than 50 mm was associated with an increased risk of regenerate fracture regardless of disease aetiology.

It is our opinion that the post-operative rehabilitation regime is a major factor in the formation of good quality callus formation. In our institution, we advise a fully protected weight-bearing mobilisation programme. Loading the regenerate bone, especially in the consolidation phase, has also been advised by other authors, including Ilizarov.^{1,4} Inadequate bone loading may lead to the development of osteopenic bone in the femur, including the regenerate bone, and leaving it more susceptible to fracture. The maintenance of knee and hip movement may also reduce the possibility of adhesions between the regenerate bone and the quadriceps muscle.²⁷ If this develops, it may cause additional stress to the regenerate bone and so increasing the possibility of fracture. This stress would be greatest while doing physiotherapy exercises following circular or mono-lateral frame removal. When inserting screws or wires into the femur, it is recommended that the knee is flexed to aid with comfort while doing hip and

knee mobilisation exercises, and thus reducing the development of these adhesions.²⁸ It has also been suggested that a proximal femoral osteotomy above the bulk of the quadriceps muscle would reduce this muscle scarring and adherence.²⁰

Additional support of the regenerate bone by lengthening over a rush nail has shown to significantly reduce the rate of regenerate fracture.²⁰ Placement of a rigid or flexible intramedullary nails has also been proposed.¹⁹ These techniques have shown to reduce the duration of time in external fixator, but are associated with complications such as failure or incarceration of the nail and osteomyelitis.^{29,30} Our fracture rate of 4.5% remains acceptable without this combined technique, although it is an option to be considered in select cases. Also the bone healing index in our study was 47.2 days/cm which is similar to femoral lengthening over an intramedullary nail with an external fixator (43 days/cm).²⁹

This study is limited by the fact that it is a single surgeon, single institution, retrospective observational study. Furthermore other variables that can contribute to a regenerate fracture were not assessed such as shape, type, length and width ratio of the regenerate bone, nutritional status and body mass index at time of regenerate fracture.³¹ In this study, we advocate the use of the Fishgrund principles when deciding when the external fixator is removed. In our hands, femoral lengthening of more than 50 mm is associated with a significant risk of regenerate fracture independent of the disease aetiology. Protected full weight-bearing during and after removal of the external fixator is advocated to load the regenerate bone and improve its overall quality. We recommend gentle physiotherapy during the initial four weeks following external fixator removal to minimise the risk of regenerate fractures.

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COMPLIANCE WITH ETHICAL STANDARDS

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ETHICAL STATEMENT

This study has been approved by the Institutional Review Board.

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee

and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

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