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*ORIGINAL STUDY*

# Complications associated with bone lengthening of the lower limb by callotasis

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**The present study aims to assess the incidence of complications related to bone lengthening procedures and to identify factors that may predict these complications. We retrospectively studied 51 lengthening procedures in 39 patients (mean age 13 years) from 2001 to 2015. A circular external fixator was used in 37 procedures and a monolateral fixator in 14 procedures. Duration of distraction, fixator's time, days of treatment, lengthening percentage, bone healing index, distraction regenerate length, distraction index, risk factors and complications were evaluated. The mean follow-up was 5 years. Complications occurred in 84 % of the procedures. Duration of distraction, fixator's time, days of treatment and distraction regenerate length were predictors of complications. Close follow-up is necessary during distraction and healing period and after fixator removal.**

**Keywords** : callotasis ; complications.

## INTRODUCTION

Callotasis is a method of limb lengthening by progressive callus distraction used in the context of limb length discrepancy (LLD), limb deformities or bone defects. Limb lengthening is generally performed when the LLD is expected to exceed 4-5 cm at maturity. LLD is a relative common orthopaedic problem with multifactorial etiologies. Some etiologies cause a decreased while others cause an increased limb length. Etiologies are commonly classified as congenital, developmental and acquired. Congenital LLD is present at birth and usually remains stable in percentage during growth (but the absolute value increases). Developmental LLD may or may not be present at birth but will increase during growth (in percentage). Acquired LLD is not present at birth but follows a triggering event (trauma, infection, tumor, etc.). LLD can have significant psychological and functional consequences for the affected patient.

In 1905, Alessandro Codivilla was the first to introduce surgical practices for lengthening of the lower limbs; however, these techniques were hindered by a high rate of complications and unsatisfactory results *(26)*. Subsequently Ilizarov developed his method based on the biology of the bone and the ability of the surrounding soft tissues to regenerate under tension (*16,17*). Ilizarov's technique improved

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the results by reducing the need for secondary bone grafts and minimizing the rate of late fracture. Although complications (including joint contractures, traction injuries to nerves and vessels, infection) and fixation failures remained possible with this technique, their rate and severity significantly improved (*22,24*). Indices to predict the complications were described. Modifications in preoperative planning, operative techniques and postoperative care were proposed to decrease complications incidence (*15,17*). Although improvements in the callotasis method and in fixation method have substantially reduced the complication rate, there remains a very high to nearly certain risk of complications that surgeons would not tolerate in other reconstructive procedures. Any surgeon undertaking leg lengthening must be thoroughly versed in the myriad complications that can arise (*1,2,3,4,5,6,7,9,10,11,12,13,14*). Therefore, we performed this study to evaluate the incidence and type of complications related to this procedure. We searched for factors that could help in predicting these complications.

## MATERIALS AND METHODS

We retrospectively studied 51 lengthening procedures in 39 patients at the authors' institution from 2001 to 2015. There were 25 male and 14 female patients with a mean age of 13 years (range 1-39 years). The etiology of limb shortening was congenital in 18 procedures, developmental in 17 procedures and acquired in 16 procedures. There were 27 femoral and 24 tibial lengthening procedures. A circular external fixation distraction apparatus (Ilizarov®, TSF®, TLHex®) was used in 37 procedures and a monolateral fixator in 14 procedures (LRS Orthofix®). In 1 procedure a combination of lengthening with and external fixator and FIN (Flexible Intramedullary Nailing) was used. In order to render our sample size more homogenous, we decided to exclude humeral, ulnar, metacarpal and metatarsal lengthening procedures and procedures with an intramedullary lengthening rod (ISKD®). Bone transports to treat non-unions were also excluded in the same optical. The mean follow-up was 5 years (range 1-15 years). The hospital charts and radiographs were reviewed for all patients.

### Limb lengthening procedure

Following the application of the external fixator to the bone segments, the corticotomy was carefully performed by drilling a series of holes in the bone at the desired level and connecting the holes with an osteotome, as described by De Bastiani and co-workers *(8)*. We took care to preserve the periosteum and soft tissue envelope. The location of the osteotomy was diaphyseal in 37 procedures (72 %), metaphyso-diaphyseal in 11 procedures (22 %) and metaphyseal in 3 procedures (6 %). In 11 tibial lengthening procedures (8 with a circular fixator and 3 with a monolateral fixator), the lateral malleolus was fixed to the tibia to prevent ascent of the lateral malleolus. In 4 lengthening procedures, a controlateral shortening was performed by epiphysiodesis or acute shortening.

Lengthening was initiated at a mean of 5 days (range 1-10 days). The distraction rate was adjusted according to the efficiency of new bone formation or patients' tolerance *(15,17)*. The mean rate of lengthening was 1 mm/day (range 0.2-2.1 mm/day). All patients were encouraged for full weight bearing as soon as tolerated after surgery. Removal of the fixator was done when satisfactory consolidation of the new bone had occurred (dense bone formation with corticalization in 3 of 4 cortices). In 9 of the 51 procedures (18 %), the fixator was removed prematurely (before consolidation) and an osteosynthesis plate was applied. In 32 of 51 procedures (63 %), a cast was applied just after removal of the external fixator to protect the regenerate.

### Distraction indices

Duration of distraction, fixator's time, days of treatment, lengthening percentage, bone healing index, distraction regenerate length and distraction index (*15,17*) were evaluated.

Duration of distraction was defined as the time from the beginning until the end of the distraction. The fixator's time was defined as the day from insertion until removal of the distraction apparatus. The days of treatment were defined as the time from surgery to formation of dense regenerated bone and complete union to allow the patient to walk without

crutches. The lengthening percentage was calculated as the ratio of the distraction regenerate length to the initial bone segment length. The healing index was defined as the ratio of the total number of days of treatment to the total lengthening (days/cm). The distraction index was defined as the ratio of the duration of distraction to the total lengthening (days/cm) (*19*).

## Risk factors for complications

Risk factors for complications were classified as minor and major (*6,9*). Minor risk factors were considered those, which do not compromise the end result (*6*). Major risk factors were considered those, which can significantly alter treatment plans and can seriously compromise end results (Table I) (*6*).

## Complications

Complications were recorded at each follow-up evaluation and at the last follow-up. Any unwanted event happening during lengthening or after apparatus removal was considered a complication to the patient (*19,24*), despite complications can vary in nature and severity. The severity of a complication was assigned a grade of minor or major (*9*). Minor complications did not affect outcome or require ex-

Table I — Minor and major risk factors for complications related to distraction osteogenesis.

| **Minor risk factors** | **Major risk factors** |
|---|---|
| Angulation, translation, rotation | Complex or congenital deformity |
| History of prior infection | Active infection |
| Contracture in an adjacent articulation prior to surgery | Preoperative instability |
| Neurological deficits | History of previous lengthening procedure on the same limb |
| Poor nutrition (BMI < 18,5 kg/m2 for adults and percentile < 25 for children) | History of previous multiple surgeries |
| Obesity (BMI > 30 kg/m2 for adults and percentile > 95 for children) | Presence of nonunion or bone loss |
| Age > 18 years | Smoking |

Table II. Common minor and major complications related to distraction osteogenesis.

| **Complications** | **Minor** | **Major** |
|---|---|---|
| Pin tract problems | Loosening, pin-tract infection | Break, sequestration |
| Infection | Superficial | Deep, osteomyelitis |
| Vascular | - | Vascular laceration or occlusion requiring repair |
| Neurological | Hypoesthesia | Neurapraxia, complete palsy |
| Medical | Pneumonia | Deep vein thrombosis, cardiac arrest |
| Psychological | Stress | Depression |
| Premature union | - | Requiring repeat corticotomy |
| Delayed union / non union | - | Requiring bone grafting or internal fixation |
| Fracture | Requiring immobilization in a cast | Requiring internal fixation or osteotomy |
| Axis deviation | 6-10° | > 10° |
| Joint subluxation | Temporary | Permanent |
| Joint contractures | < 10° | > 10° |
| Premature interruption of the lengthening procedure | - | For mechanical raison, unbearable pain, etc |
| Ascent of the lateral malleolus | - | |

tensive intervention, whereas major complications required major unplanned surgery or resulted in major permanent sequelae (Table II) (*19,22*).

### Statistical analysis

A Kolmogorov-Smirnov test was performed to test normality of groups. Comparison of the mean between groups was performed by using unpaired Student t-test. Chi-square test was performed to test the occurrence of complication between groups. A P-value less than 0.05 was considered to be statistically significant.

## RESULTS

The mean duration of distraction was 50 days (range 18-104 days). The mean fixator's time was 146 days (range 46-326 days). The mean days of treatment were 201 (range 73-433 days). The mean distraction regenerate length was 4.8 cm (range 1-8.5 cm). The mean lengthening percentage was 19 % (range 3-57 %). The mean bone healing index was 35 days/cm (range 11-101 days/cm). The mean distraction index was 12 days/cm (range 5-44 days/cm). Complications occurred in 43 of the 51 procedures (84 %). Minor risk factors were observed in 38 of the 51 procedures (74 %) and major risk factors in 37 of the 51 procedures (72 %). The most common major complications were axis deviations (14 %), delayed unions or nonunions (12 %), joint contractures (12 %) and fractures (12 %). The most common minor complications were pin tract problems (47 %), psychological (18 %) and medical (14 %). (Table III)

### Influence of distractor type on complications

We found more overall complications and more major complications ($p = 0.06$) in the lengthening procedures with monolateral fixators (compared with circular frames).

In the three cases of ascent of the lateral malleolus, one was fixed and the two others were not. (Table IV)

### Influence of anatomical segment on complications

The level of osteotomy (diaphysis, metaphysodiaphysis or metaphysis) or specific anatomical segment (femur or tibia) did not statistically influence the rate and type of complications, except for major axial deviations. We observed more major axial deviations with tibial lengthening procedures ($N = 1$ of 27 femur versus $N = 6$ of 24 tibias, $p = 0.027$).

Table III. — Encountered complications.

| Complications | Minor | Major |
|---|---|---|
| Pin tract problem | 24 | 5 |
| Infection | - (included in pin tract problems) | 2 |
| Vascular | 0 | 0 |
| Neurological | 6 | 0 |
| Medical | 7 | 0 |
| Psychological | 9 | 1 |
| Premature union | 0 | 3 |
| Delayed union / nonunion | 0 | 6 |
| Fracture | 4 | 6 |
| Axis deviation | 1 | 7 |
| Joint subluxation | 0 | 1 |
| Joint contractures | 4 | 6 |
| Premature interruption of the lengthening procedure | 0 | 5 |
| Ascent of the lateral malleolus | 0 | 3 |

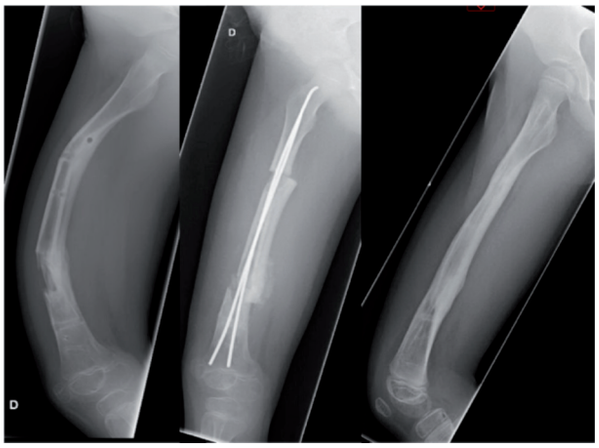

*Fig. 1.* — Fracture and flexum deviation after a 4 cm-lengthening procedure in a 6-year-old girl. Osteotomies and intramedullary nailing were performed with a good end result. Left: lateral view of femur after fixator removal. Middle: double osteotomies and nailing. Right: end result.

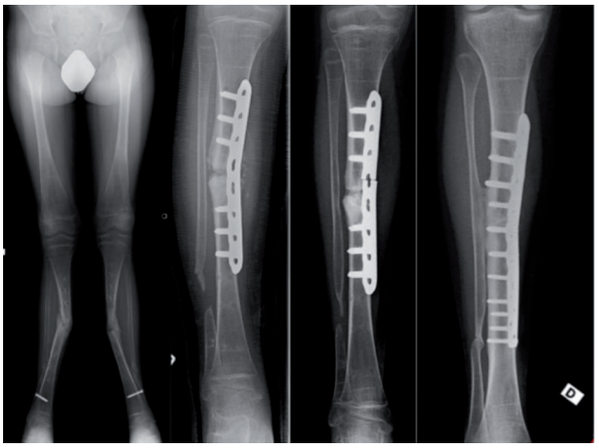

*Fig. 2.* — 12-year-old boy with Blackfan-Diamond syndrome (erythroblastopenia). After a bilateral 5 cm-lengthening procedure and removal of the monolateral fixator, the patient sustained a valgus deviation of both tibias. B: Despite plate osteosynthesis, the patient developed a pseudarthrosis (C). D: Finally, healing was obtained after intertibiofibular grafting and new osteosynthesis.

### Influence of age on the complications

When we compared the procedures done on the group of patients younger than 18 years (N = 44) with the procedures done on the group of patients older than 18 years (N = 7), no significant difference was found according to the global rate of complications. But we observed more major fracture complications in the group older than 18 years (Table IV).

### Influence of distraction indices on complications

When we compared the group of patients who experienced a complication to those who did not, we found a statistical significant difference concerning duration of distraction ($p < 0.0001$), fixator's time ($p < 0.0001$), days of treatment ($p = 0.001$) and distraction regenerate length ($p = 0.019$). They were all higher in the group that experienced one or more complications.

When we compared the group of patients with major complications and the group without major complications, we found the same significant difference concerning duration of distraction ($p < 0.0001$), fixator's time ($p < 0.0001$), days of treatment ($p = 0.001$) and distraction regenerate length ($p = 0.019$).

Lengthening procedures of 5 cm or more expose to the occurrence of complications.

There was no significant difference for the distraction index, the healing index and percentage of lengthening.

## DISCUSSION

Distraction osteogenesis has been widely used to increase length of a segment but has been associated with significant complications (*20*). In the literature the rate of complications varies from 14 % to 100 % (*8,21*). These widely divergent reports are a reflection of the different definitions of complication in relation to limb lengthening and the diligence with which these events are sought. Ilizarov defined a complication as any adverse event or unexpected condition or effect that alters the care plan or reduces the quality of results (*16,17*). Paley classified these untoward events as problems (not requiring operative intervention to resolve), obstacles (requiring operative intervention but without sequelae) or complications (intraoperative injury or anything resulting in permanent sequelae) (*22*). More recently, complications were considered any adverse effect that a treatment can have on the patient (*6*). In our study, every unwanted event occurring during

Table IV. — Influence of distractor type on complications.

| Complications | P value |
|---|---|
| Major nonunion<br>– circular frames: N = 2 of 37<br>– monolateral fixators: N = 4 of 14 | 0.041 |
| Ascent of the lateral malleolus<br>– circular frames : N = 0 of 37<br>– monolateral fixators : N = 3 of 14 | 0.017 |
| Major fracture<br>– circular frames : N = 2 of 37<br>– monolateral fixators : N = 4 of 14 | 0.041 |
| Major axis deviation<br>– circular frames : N = 3 of 37<br>– monolateral fixators : N = 4 of 14 | 0.08 |

lengthening or after removal of the distractor was considered a complication (*6,25*). As previously described, the severity of a complication was assigned a grade of minor or major (*9*). Minor complications did not affect outcome or require extensive intervention, whereas major complications required major unplanned surgery or resulted in major permanent sequelae (*19,22*).

In the present study, complications occurred in 43 of the 51 procedures (84 %). As expected, the most common minor complications were pin tract problems (47 %). The most common major complications were axis deviations (14 %), delayed unions or nonunions (12 %), joint contractures (12 %) and fractures (12%). Major fracture complications were more prevalent in adults (older than 18 years).

The overall complication rate of 84 % is rather high. We explain this high rate of complications because we included all minor and major complications that our patients experienced, and included patients of variable age and medical comorbidities. In order to render our sample size more homogenous, we decided to exclude humeral, ulnar, metacarpal and metatarsal lengthening procedures and procedures with intramedullary lengthening rods. Bone transports to treat congenital pseudarthrosis of the tibia or to treat acquired pseudarthrosis were also excluded in the same optical.

We evaluated the callotasis technique and related complications and risk factors and drew useful conclusions. Although distraction osteogenesis may provide excellent results, it is associated with a high rate of complications. The surgeons should be aware of these complications and carefully select and follow their patients at long term after removal of the distraction osteogenesis device. Careful follow-up, good pin care, adjusting distraction rhythms according to callus formation, encouraging patients to bear weight can contribute to better healing and less problems.

Furthermore, we found that the duration of distraction, fixator's time, days of treatment and distraction regenerate length were significantly increased in patients who experienced complications. This finding suggests that the fixator apparatus should be removed once radiological evidence of bone formation is observed and should not be prolonged further. Conversion to internal osteosynthesis or cast application after removal of the external fixator can help in reducing the fixator's time. Plate osteosynthesis represents an additional risk of deep infection because of the possible skin colonization by pathogens due to the fixator pins but in the 9 cases of our series, we didn't observe any deep infection. Addition of FIN or bifocal lengthening procedures are also possible means to decrease fixator's time (*23*). Conversion from a circular to a monolateral fixator is an option to minimize the discomfort of a circular frame and it also stimulates bone healing as previously described by Laumen et al (*18*).

Lengthening procedures of 5 cm or more expose to an increased risk of complications. The surgeon should opt for two smaller successive lengthening procedures at some years of interval instead of one important one (for example 2 x 4 cm instead of 1 x 8 cm).

Distraction index, healing index and percentage of lengthening were not associated with increased incidence of complications. The level of osteotomy (diaphysis, metaphyso-diaphysis or metaphysis) or specific anatomical segment (femur or tibia) did not statistically influence de rate of complications either.

On the other hand, major fracture complications, nonunions and axis deviations were more prevalent in procedures with monolateral fixtators than with circular frames. We explain this by the fact that a monolateral fixator is less stable than a circular frame. We also observed more problems of lateral malleolus ascent with monolateral fixators.

Table V. — Influence of certain risk factors on certain complications.

| Risk factors | Complications | P value |
|---|---|---|
| Poor nutrition | Major infection (osteomyelitis)<br>– N = 2 of 7 with poor nutrition<br>– N = 0 of 44 with normal nutrition | 0.016 |
| History of previous lengthening procedure on the same limb | Delayed union / nonunion<br>– N = 3 of 5 with previous lengthening<br>– N = 3 of 46 without previous lengthening | <0.0001 |
| | Minor neurological<br>– N = 2 of 5 with previous lengthening<br>– N = 4 of 46 without previous lengthening | 0.039 |
| Age > 18 years | Major fracture<br>– N = 3 of 7 > 18y<br>– N = 3 of 44 < 18y | 0.006 |

## CONCLUSION

A close follow-up is mandatory during the distraction period, the bone healing process and after implant removal. Shorter periods of fixator could minimize the rate of complications related to distraction osteogenesis. Lengthening procedures should not exceed 5 cm. Surgeons should be aware of the high incidence of complications related to bone lengthening procedures by callotasis and should be alert to recognize and treat these complications promptly and properly when they occur.

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