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Ilizarov technique in the management of bilateral tibia nonunion in an adult with osteogenesis imperfecta

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Abstract
The purpose of this study is to present our surgical strategy and describe the advantages and outcomes of the Ilizarov method in the treatment of bilateral tibia nonunion in an adult with OI. The major of bone fracture fixation drawback is the difficult placement of nails due to abnormal bony anatomy, non-linear and imperforate canals, marked bowing and unusually short limbs. A female adult aged 18yo with bilateral tibia nonunion affected with OI type V was treated with the Ilizarov method. The tibia was in varus (15° at left, 14° at right) and recurvatum (18° on the left, 27° on the right). Surgical technic was same for both tibia in one-time procedure and it's compounded: primary fibula osteotomy, tibia and osteotomy taking out interfragmentary fibrosis, reduction and fixation by Ilizarov frame. Full weight-bearing was allowed at 3 months. The complications that occurred were minors. Bone's consolidation was obtained at 5 months and the frames were removed at 6 months in the office without anesthesia. At the follow-up visit 5 months after removal, the ASAMI functional and radiological score was excellent on the left side and good on the right side. The use of the Ilizarov method for treating a tibia bilateral nonunion in an adult patient with moderate OI provides good stability. This technique ensures early weight full bearing, above joint motion, and recovery functional necessary to normal bone tissue regeneration. This is an alternative to avoid difficulties and complications due to internal fixation in deformities correction in adults with acquired bone-weakening conditions.

Keywords: Osteogenesis imperfecta type V, Ilizarov technique, tibia nonunion

Introduction
The management of fractures in osteogenesis imperfecta (OI) included a variety of methods allowing the correction and stabilization of long bone deformities: plaster, open reduction and intramedullary nailing, or plating [1]. Treatment by intramedullary nailing remains the most common technique in children and adults [1]. Load-sharing intramedullary devices are strongly preferred over rigid plate constructs. Plate constructs promote stress shielding and can lead to complete bony resorption, failure via screw pullout, fracture at the end of the plate. Although the use of intramedullary devices is preferable, their implementation can be challenging because of abnormal bony anatomy, non-linear or imperforate canals, marked super physiologic bowing, and unusually short limbs they may require the use of pediatric or custom implants [2]. The Ilizarov method in adults is a well-known technique for correcting deformities, however poorly reported in the management of fractures and nonunion in an adult with OI. The purpose of this study is to present our surgical strategy and describe the advantages and outcomes of the Ilizarov method in the treatment of bilateral tibia nonunion in an adult with moderate OI.

Case presentation
Miss M. Y. is an 18-year-old patient studying in secondary school who wishes to become a lawyer. She had consulted for pain in both legs with absolute functional impairment after a fall at home 3 months previously. Unsuccessful indigenous treatment was performed because of a lack of means.
In this history, walking was independent before the fall. She had two episodes of fracture to her right tibia at 7 and 10 years old, which were treated indigenously.

She is the second of five siblings. Two of them were deceased because of OI: the eldest died at 20 years old; the younger sister at 15 months years old. Two younger sisters are currently alive and well health aged of 7-year-old with an OI and a 4-year-old with a normal morphotype. The father died of an unknown cause. There is no notion of inbreeding. She was never be monitored for the OI.

On physical examination, the height was 132 cm. She presented: hyper-pigmented spots on the skin, a broad, slightly rounded forehead, no blue sclera, good oral hygiene with normal dentition, and good hearing. In both pelvic limbs, there was localized pain in the middle 1/3 of the legs with a skin opposite healthy and slight swelling. The bilateral tibial were in varus deformity. There were no ligament hyperlaxity and no other deformities on the upper limbs or the spine. The AP and lateral X-ray views of the tibia showed (Fig. 1): bilateral hypertrophic nonunion with a varus and recurvatum deformities. On the left tibia, the varus was 15 ° and the recurvatum 18 °. On the right tibia, the varus was 14 ° and the recurvatum 27 °.

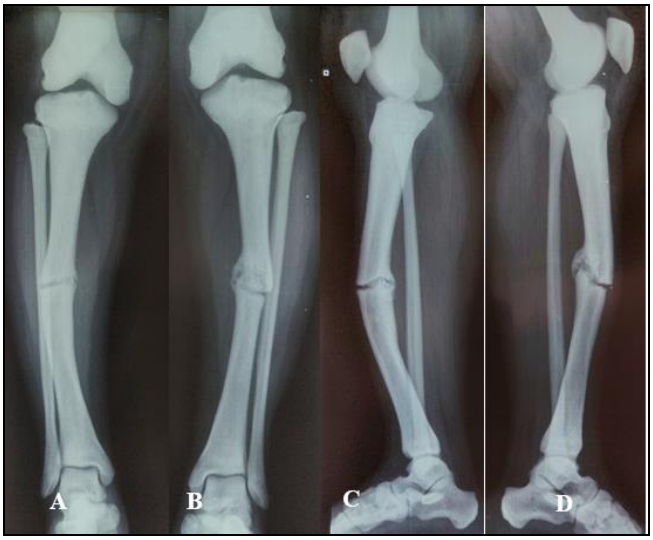

Fig 1: radiographic aspect showing bilateral nonunion. A: AP view right tibia (14° varus). B: AP view left tibia (15° varus). C: Lateral view right tibia (28° recurvatum). D: Lateral view of left tibia (18° recurvatum)

The pre-therapeutic biological blood test showed: a normal blood count, no inflammatory syndrome (sedimentation rate: 10 mm the first hour, C-reactive protein <3 mg / l); calcemia: 106 mg / l; phosphoremia: 49mg / l; magnesemia: 21 mg / l; T3 = 2.4 Pg / ml; T4 = 8.8 Pg / ml; TSH = 0.48 µIU / ml (euthyroidism 0.39 - 6.16 µIU / ml).

In total, she presented four fractures during 11 years, ie an annual incidence of 0.36 fractures. The diagnosis retained was bilateral tibia nonunion in an adult affected with osteogenesis imperfecta classified as type V [3]. The procedure planning was: open procedure, correction of deformities, reduction of the fracture, and bone fixation by an Ilizarov external fixator.

Surgical technique

The same surgical procedure was performed on each tibia in one operating time. The C-arm was not used. The limb was under a tourniquet. We performed a primary osteotomy of the fibula of approximately 1.5cm. Then for the tibia, we proceeded by an anterolateral approach to expose non-union. We performed under periosteum bone resection approximately 5 mm triangular in a wedge with an external apex taking out the interfragmentary fibrosis. The tibial varus and recurvatum were corrected by reducing valgus and flexum. Fixation was done by a circular external fixator. The frame was tibiotibial including one circular 14 cm diameter ring on either side of the fracture. Each ring was fixed by two 6mm pins placed on either side of the ring. There was no additional graft or decortication (Fig. 2). The immediate postoperative outcome was normal. The hospital staying was 7 days. The pins track care was done by a nurse twice a week. Clinical and radiological control showed a correction of both tibia deformities. AP and lateral X-ray view showed on right the leg a valgus and flexum hypercorrection (Fig. 3). It was corrected by external manipulation of the frame.

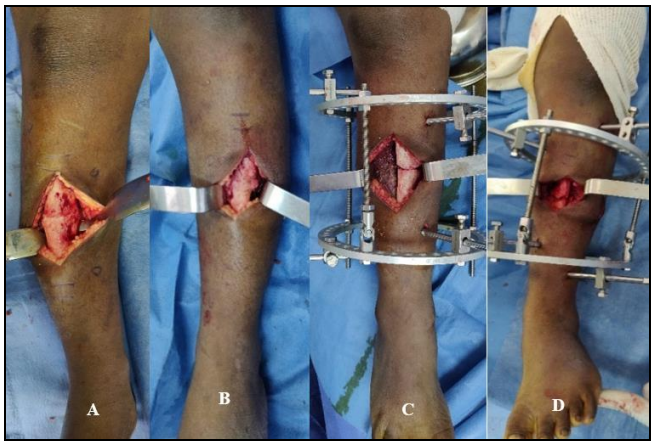

Fig 2: A + B: intra-operative aspect showing left and right tibia non-union. C + D: intra-operative aspect showing fixation by Ilizarov frame after deformities correction

Fig 3: A+ B: post-operative AP X-ray view showing normal correction in the left tibia and valgus hypercorrection in the right tibia.

Follow-up

The final follow-up was at 20 months. The follow-up was done during the successive outpatient controls. Partial weight-

bearing was allowed at days 50 postoperatively under canes protection. Full weight-bearing was allowed at three months (Fig. 4). According to Paley, the complications occurring were: pain in her right leg on walking and pins grade 2 track infection [4]. Satisfaction was moderate. The patient said the cumbersome device but was not hampered in everyday life. Consolidation was obtained in 5 months on both sides. Walking was independent without a cane before the fixator was removed at 6 months without anesthesia in outpatient control. The digital-to-analog scale per removing was 8 and after removing was 5.

Fig 4: clinical outpatient aspect showing patient standing without a cane in total weight bearing

The outcomes were evaluated according to the Association for the Study and Application of the Method of Ilizarov (ASAMI) score [4]. At the 18-months follow-up after removing the frame, walking was independent. The ASAMI functional score was excellent in the left leg and good in the right leg (Figure 4). On the radiological control, this score was excellent for the left leg and good for the right leg. The leg X-ray showed a normal tibia on the left, centered in the AP and lateral view. On the right tibia, there was an 8 ° valgus and a 14 ° flexum (Figure 5).

Fig 5: outpatient control. A+B: front and lateral aspects in standing patient

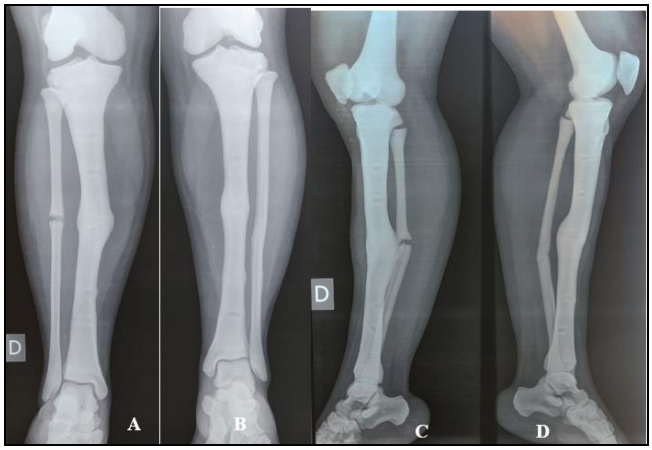

Fig 6: A + B: AP X-ray view showing a 5° valgus on the right tibia and normal axis on the left tibia. D+C: lateral X-ray view showing 14° hypercorrection flexum on the right tibia and normal axis on the left tibia.

Discussion

The literature review shows, in our setting the OI although rare and not exceptional, is poorly reported in publications that are mainly clinical cases. The management in these countries is characterized by: the lack of a referral center or institution, diagnostic delay inpatient whose most of the time living out in hard socio-economic conditions, the importance of antenatal ultrasound, the lack of multidisciplinary care; the unavailability of bisphosphonate, and finally, the rural environment which does not allow access to care [5-7].

The fractures incidence generally in OI declines with age due to skeletal maturity. From the age of 18, the prevalence is higher compared to the healthy population [8]. The Danish national register reports a prevalence of 154.4 fractures / 1000 patients / year, i.e an annual incidence of 0.15 fractures / patient [8]. The high prevalence of fractures in our patients [0.36 / year) was due to a lack of monitoring and prevention since the first episode of fracture. The long bones deformities lead to biomechanical properties impairment and cause a high bending moment making them more susceptible to fractures [9]. In our case, the patient presented with a bilateral recurvatum and varus, which explains the occurrence of low energy fractures by falling from at home.

In the management of fractures, nonsurgical treatment is preferable in equivocal situations. We can consider that in our case the patient received the equivalent of plaster immobilization for the first two fractures in childhood. Prolonged immobilization is not beneficial as it results in osteopenia and reduced mobility,y which can lead to refracture creating a vicious circle [1]. Surgical treatment of fracture in adults with OI should take into account bone deformity, increased risk of fracture, risk of shortening. Fixation on each tibia was done in our casa by the Ilizarov method after osteotomy to correct deformities and reduction. This method offered several advantages in our case: it was a less invasive implant compared to the plate or the intramedullary nail, its implantation was neither hampered by the varus and recurvatum deformations which modified the axis, nor by the imperforated canal, which would make intramedullary nailing hard. In addition, the fixator allowed: fracture compression, secondary correction of frame postoperatively, early mobilization of the above joints, and early weight-bearing, removal without another anesthesia after fracture healing [10].

Most often in OI, bone fixation is performed by nail or plate. But they lead to frequent non-union after open reduction [1, 11]. This is particularly true as patients age, the weight and length

of the bones increase while the medullary canal remains small, requiring a relatively small diameter nail to be chosen [12]. This achieves insufficient stability and therefore leads to non-union. To overcome this insufficiency, authors have proposed combining a sandwich allograft [12], or reinforcement of the intramedullary nail with a unilateral locked plate [13]. These methods are suitable for severe forms of OI, but are invasive, expensive, not available in our setting, and required another procedure for implant removal. The Ilizarov technique is well documented in the correction of long bong deformities and management of defects without grafts, fixation of bone fragments in fractures, and nonunion [10]. We adopted the Ilizarov method, which is less expensive, minimally invasive for a moderate form of OI. In children, the external fixator is also reported in associating with the intramedullary nail (telescopic nail or not) [14]. Indeed, intramedullary elastic nail bone fixation alone in a child with OI does not ensure rotational and longitudinal stability, does not allow immediate weight-bearing [14]. Popkov *et al*. [15] reported good results from this combination in a series of 12 patients by combining elastic titanium nailing and an external fixator. As we have pointed out, they noticed that this combination allowed longitudinal, rotational, and angular stability, thus allowing early weight-bearing. Both conditions promoted the good regeneration of bone tissue [15].

There were no intraoperative complications like excessive bleeding because we operated under tourniquets and the technique was external fixation without intramedullary procedure. The only complications observed in our case were minor and included pain, minor pin track infection, discomfort crowding of the frame. In children, gait abnormalities were noted due to frame crowding, especially in the leg, but which did not limit mobility [15]. The rate of non-union after nailing is high, reaching 24% in the event of a fracture and 52% after osteotomy [16]. Both tibias healed in a reasonable time. The radiological outcome was good at the long term follow up.

Conclusion

In our setting, the management of OI is most of time-limited to the specialty where the patient had been seen with the main clinical manifestations. Treatment considerations in adults living with OI vary from case to case. Poor bone quality and architectural modifications in the bone due to OI limit the use of nail or screw plate fixation techniques for bone fixation in an adult with OI. In this case report, Ilizarov's method gave good clinical and radiological outcomes in a moderate form of OI. It allowed correction of deformities and obtained stable fixation until union without extending the time to union.

Clinical significance

The circular external fixator is an alternative to avoid difficulties and complications due to internal fixation in deformities correction in adults with acquired bone-weakening conditions. Further studies involving a larger sample are needed to confirm the positive role of the circular external fixator in nonunion and osteotomies in adults with osteogenesis imperfecta.

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The authors declare that they have no competing interest concerning this manuscript.

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