Journal of Bone Research

Batur et al., J Bone Res 2017, 5:2
DOI: 10.4172/2572-4916.1000182

Research Paper Open Access

Our Results of Modified Sofield Procedure in Lower Extremity Deformities with Osteogenesis Imperfecta

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Received Date: July 19, 2017; **Accepted Date:** September 20, 2017; **Published Date:** September 27, 2017

Abstract

Introduction: Osteogenesis Imperfecta (OI) is a connective tissue disorder, which is the most common cause of genetic osteoporosis characterized by long bone deformities and fractures. The key defect is the qualitative and quantitative lack of type I collagen. The primary target in these patients is to fix deformities and to prevent fractures. The surgical application is often a basic surgical technique known as “Shish Kebab” osteotomy and mainly based on the principle of multiple osteotomies, fixation, and intramedullary nailing. In the present study, we present our results of the modified Sofield procedure for osteogenesis imperfecta-related lower extremity deformities.

Materials and methods: The study retrospectively examined 12 OI patients who were surgically treated between March 2006 and November 2011. The modified Sofield procedure was applied to a total of 29 lower extremity bones including 21 femurs and eight tibias. Double intramedullary K-wire and single K-wire were used in surgeries for the femur and the tibia, respectively.

Results: The median age of the patients was 7 (range: 6 to 16) years. The mean follow-up was 3.18 (range: 1 to 5) years. The revision rate was 42.8% for femoral surgery, 62.5% for tibial surgery, and 48.2% for all surgery. The mean time of union was 47.3 (range: 30 to 60) days including 46.3 (range: 30 to 60) days for femoral, and 49.3 (range: 30 to 60) days for tibial surgeries. The mean time from one surgery to another was 58 (range: 4 to 96) months for femoral revision, and the mean time of tibial revision was 27.7 (range: 9 to 60) months. The mean time required for total revision was 28 (range: 4 to 96) months. The complication rate was 39% and 46% for femur and tibia, respectively. The overall complication rate was 41%.

Conclusion: Our study results suggest that this method is an effective and reliable method in the surgical treatment of osteogenesis imperfecta, as it is easy to practice and access as well as being cost-effective. Although surgical treatment does not change the patient ambulation, it maintains mobilization, reducing the incidence of fractures and deformities. The double-rod application used at the femur decreases the number of revisions and prolongs the time of revision.

Key words:

Osteogenesis imperfect; Deformity; Shish kebab osteotomy; Limited incision; Intramedullary fixation

Introduction

Osteogenesis Imperfecta (OI) is a genetic connective tissue disease with an incidence of 1/20,000 births, which causes long bone fractures and deformities as the main clinical symptom accompanied by other clinical symptoms such as blue sclera, hearing loss, dental problems and ligamentous laxity [1-4]. As the most common genetic cause of osteoporosis, it involves frequent long bone fractures and vertebral compression fractures following a simple trauma. Clinically, it includes a wide range of forms from a perinatal fatal one to a mild one that can be detected in adulthood [2,5,6]. The key defect is often the qualitative and quantitative defect of type I collagen. Most of the OI forms are associated with mutations in the type I collagen genes [1,7-9]. Although several surgical techniques have been identified to treat deformities due to OI, the basic surgical principle is multiple osteotomies, fixation, and intramedullary nailing [10-12]. In the present study, we aimed to present our results of the modified Sofield procedure for OI-related lower extremity deformities.

Materials and Methods

This single-center study was conducted in accordance with the principles of the Declaration of Helsinki. A written informed consent was obtained from each patient. A total of 12 patients diagnosed with OI and presented at our clinic due to lower extremity deformities between March 2006 and November 2011 were retrospectively analyzed. All patients who have referred to our clinic were included in the study.All patients were classified based on the Sillence and Shapiro classification (Tables 1 and 2) [5,13]. Except one, all patients were under treatment by Pediatric Endocrinology and receiving Pamidronate. The walking pattern of the patients was assessed using the Ambulation Scale described by Hoffer and Bullock [14].

In total, 48 surgical treatments were applied to 29 lower extremity long bones. Three of these revisions were performed due to nonunion surgery (bilateral femur in one patient and right femur in one patient). The indication for primary surgery was angulation, new fracture, and nonunion fractures despite conservative therapy in patients with OI, compared to new fractures, increased angulation, pseudarthrosis and wire migration for revision surgery.

As the basic surgical technique, the technique known as Sofield procedure which was first described by Sofield and Miller [15] was preferred. However, the entire bone was not subperiostally peeled off and osteotomy was performed in a number that would correct the deformity. Double rots were used in femoral surgeries, while single rot was used in tibial surgeries. Intramedullary fixation was performed using the Kirschner wire (K-wire) with an appropriate thickness in all patients, except one. Double intramedullary K-wire and single K-wire were used in the surgeries performed for the femur and the tibia, respectively (Figures 1 and 2). In one patient for whom a K-wire was used for intramedullary fixation, plate osteosynthesis was performed upon fracture formation. In one patient who was previously administered femoral intramedullary nail, revision was performed again using an intramedullary nail. Surgical interventions were performed under general anesthesia and over a radiolucent table allowing fluoroscopic imaging. The number of interventions was limited in cases of excessive bleeding during surgery. The patients with a hemoglobin <8 g/dL or hematocrit <25% due to bleeding were administered 8 to 10 mL/kg of whole blood. Blood transfusion was made in 22 patients from of an overall 48 surgical operations. The patients had external fixation using a plaster-splint following surgery. After becoming hemodynamically stable, the patients were discharged after a mean time of 3.68 (2-8) days. All patients were called for follow-up on postoperative days 15, 30, 45, 60, and 90. In an average of four weeks, the plaster-splint was removed and articular motions were started, and the patients were encouraged to controlled mobilization.

Results

Of the patients, five (42%) were males and seven (58%) were females. The mean age of the patients was 7 years (range: 6 months to 16 years). Four of these patients previously underwent several surgeries at external clinics. Other patients underwent their first surgical intervention at our clinic (Table 3). In total, 48 surgical interventions were applied to 29 lower extremity bones of 12 patients. Of these lower extremity bones, 21 (72.4%) were femurs and eight (27.6%) were tibias. Revision surgery was applied to nine (42.8%) of the femurs and five (62.5%) of the tibias. In total, revision surgery was performed on 14 (48.2%) of 29 bones. Surgery was primary in 25 (52%) and revision in 23 (48%) of the overall 48 surgeries. In total, 23 revision surgeries were applied to 14 lower extremity bones undergoing revision. Among revision surgery patients, there was nonunion following bilateral femoral surgery in one patient and right femoral surgery in another patient. Union was observed following all other surgeries. The mean follow-up was 3.18 (range: 1 to 5) years.

The mean time of union was 47.3 (30-60) days including 46.3 (range: 30 to 60) days for femoral and 49.3 (range: 30 to 60) days for tibial surgeries. The mean time from one surgery to another was 58 (range: 4 to 96) months for femoral revision, and the mean time of tibial revision was 27.7 (range: 9 to 60) months. The mean time required for total revision was 28 (range: 4 to 96) months. The complication rate was 39% and 46% for femur and tibia, respectively. The overall surgical complication rate was 41% (Table 4).The walking patterns and mobilization degrees of the patients are summarized in (Tables 5 and 6) respectively.

Discussion

A multidisciplinary approach is essential to treat OI, and even the patient family should be included in the management team [1,4,7]. As osteoporosis is the main problem in this disease, medical treatment is important, and such treatment is based on bisphosphonates (particularly pamidronate) and daily vitamin D and calcium requirement [1,3,7]. In our series, all patients, except one, were on pamidronate. The goal of surgical treatment of OI patients is to fix fracture-related deformities, to prevent new fracture formation, and to improve the functional status. The most common condition requiring treatment is long bone deformations in these patients. Intramedullary rod implantation is the most attractive option among available treatments to restore pre-existing deformities and to fix the fractures [1,9,16,17]. Although plate osteosynthesis seems to be an option for treating the deformities and fractures of children with OI, it is disadvantageous, compared to intramedullary nailing, due to high rates of complication, short revision duration, and the unknown effect on the bone growth [18]. Implant failure and new fracture formation are common complications in the plate method [2,7,9]. We performed plate osteosynthesis only in one patient who developed new fracture, despite the intramedullary fixation and had appropriate bone quality. Under normal circumstances, however, we believe that plate osteosynthesis does not have a place in OI cases scheduled for primary surgery. The surgical method most commonly performed and recognized for long bones is intramedullary fixation along with osteotomy. This method was first defined by Sofield and Millar [15]. In this technique, the deformed bone is divided into flat pieces and aligned around a rod. Since this resembles “Shish Kebab”, it is also called "Shish Kebab” osteotomy. Thereby, both the formation of recurring fracture is prevented and a proper extremity is achieved by means of the inside rod. In this procedure, although not often, complications such as rod migration, nonunion or delayed union as the bone is revascularized too much may occur [1,15]. Since the flat nails used in a classical Sofield technique falls short over time due to the increased bone length, new implant designs have been investigated. Bailey and Dubow [19] designed a new telescopic nail that can be fixed to the bone with T-shaped ends and get longer as the bone becomes longer. The incidence of re-operation reduced, since the bone was not without fixation due to the nail getting longer with increased bone length. However, these nails are reported to cause high rates of complications such as migration, disintegration of the T-parts and rod protrusion to the soft tissue. To resolve, particularly the T-part breakage issue, the Baily-Dubow nails were modified and Sheffield rods were developed. Wilkinson et al. [20] reported that these new Sheffield rods result in less epiphyseal damage and has lower rates of complication. Later, it was reported that less joint damage, less invasive procedure, and lower mechanical complications are achieved with the Fassier-Duval nail developed by Fassier and Duval [21]. Besides, these nails have an advantage that removes knee arthrotomy in femoral surgery and ankle arthrotomy in tibial surgery. The nail is placed with small incisions under fluoroscopy along with osteotomies. However, this is a challenging technique with a steep learning curve [1,8,9,22,23].

In a classical Sofield procedure, the diaphysis is opened lengthwise to fix the bone deformity. However, Li et al. [24] suggested performing osteotomy with limited incision upon establishing the deformity location using a monitor. Thereby, they aimed to achieve less bleeding and less periosteal damage, and to minimize the devascularized bone rate. The technique used in the present study has similarities with the modified Sofield technique used by Li et al. [24]. The deformity apex was established under scopy and the deformity was fixed with limited incision and osteotomy. Thus, less periosteal damage, less blood loss and infection were observed. Therefore, we also recommend limited incision, limited periosteal damage, and limited osteotomy to minimize the devascularized bone rate.

The mostly discussed complication of classical Sofield technique is rod migration. The study by Tiley and Albright [25] reported 20 rod migrations in 112 surgeries. They reported that the main reason of this was the use of a short rod. They indicated that the short rod cannot prevent deformity formation and causes deformity formation by creating a stress point. Williams [26] reported that rod migration may be longitudinal or lateral, and distal migration could be prevented using a hook at the proximal; however, it was unable to be done in the tibia. In our series, distal migration of the rod was observed in two femur cases. In the first case, the wire in the distal joint was removed and the proximal wire was maintained in its place. In the second case, the femur was revised again and the proximal and distal wires were replaced. In our cases, the ends of the wires used in the femur were bended (a hook-like shaped) and embedded into trochanter at the proximal and femoral notch at the distal. No proximal loosening or migration was observed; however, there might be distal migration, if the distal wire is not placed properly in the cartilage. Likewise, the wire was embedded into the cartilage of tibial process by bending its end in our technique. Wire migration was not observed in any of our tibia cases. Furthermore, the age of surgery of patients is still controversial in the literature. The earliest surgery age is not reported; however, the youngest age was six months in the series of Sofield et al. [15], and 10 months in the series of Williams [26]. It was reported that, although the deformity would progress and the length loss would increase in case of a delayed surgery, it should be noted that very early surgery would increase the number of revision [26]. Ryöppy et al. [27] suggested that early intervention to children without a lower limit of age by using non-extending nails improved motor development of the lower extremity. Tiley and Albright [25], in turn, reported that the time to stand up was the age to start fixation for the lower extremity. The mean age of eight patients who had their first surgery at our clinic was three years. The youngest and oldest ages undergoing surgery was six months and five years, respectively. Since there is not any consensus on the age of starting surgery in the literature, we believe that the major factor in deciding surgery is the clinical condition of an individual patient. We considered nonunion of fraction by conservative method and/or prevention of walking ability and resulting problems such as significant shortness and pain due to deformity as the criteria for starting initial surgery. Due to these reasons, we believe that it is difficult to determine the age of starting surgery beforehand, and it may vary by patient.

Conservative therapy was recommended to the patients whose age and bone sizes were not eligible for open osteoclasis and intramedullary fixation. However, fixation with closed osteotomy or osteoclasis can be performed for young children requiring fixation [1]. Morel et al. [28] suggested closed intramedullary nailing following closed osteoclasis rather than external support with plaster-splint; however, it is a highly challenging technique to advance a nail in a closed manner through a medullary canal that is already narrow. In the present study, no patient had deformity fixing surgery with closed osteoclasis. Additionally, we followed with closed reduction and plaster splints before performing revision if there was not any severe deformation of the bone in fractures developed after nailing. This technique seems effective, particularly in young children who need deformity or fracture fixation, but cannot tolerate surgery and blood loss. Nonunion is not rare in OI patients. The first and broadest study on nonunion was published by Gamble et al. [29]. The authors observed 12 (19%) nonunion cases in 10 of 52 patients. They reported that nonunion occurred mostly in the bones where there were several fractures in the same site and a progressive deformity was developed. In our series, nonunion was identified only in two patients after surgery. There was bilateral femoral nonunion in one patient and right femoral nonunion in another patient. According to the postoperative nonunion, the rate of nonunion was 6% in the present study. We performed revision surgery in two patients with nonunion. In revision surgery, the existing wires were removed, sclerotic areas in the nonunion site were cleared, and the wires were replaced with large-size wires. Union was achieved without grafting.

Due to the bone growth following deformity fixation with non-extending rods, it is seen hat problems such as rod migration based on the implant, move of the rod beyond the cortex and formation of deformity and/or fracture upon non-rod bone growth, and rod bending or breakage are common. These complications and associated rates vary by surgical technique and implants used [30]. Porat et al. [31] reported that the complication rate was 50% after fixation with non-extending nails and 72% with Bailey-Dubow nails using classical Sofield procedure. Gamble et al. [32], in turn, reported a complication rate of 55% with non-extending nails and 69% with Baily-Dubow nails. Wilkinson et al. [20] reported a complication rate of 20% for fixation with Sheffield nails. In the present study, the main problem in tibial deformities fixed with an intramedullary K-wire was the fracture itself and increased angulation in the distal bone which was not supported by an implant. In femoral deformities, in turn, although no site was left without an implant by using double-wire, the wire diameter and length were observed to fail to prevent the deformation of the bone in some cases due to bone growth. In certain cases, there was deformity and/or fracture formation related to a stress point at the bone due to the insufficiency of the wire diameter after the wire got out of the lateral cortex or the child grew. There was no need of revision in all cases due to the stability provided by the use of double-wire, except one case with fracture observed upon angulation.

In a study, Joseph et al. [30] reported that there was similarity in fracture incidence between double-rod and extendable nails and fixation. The authors showed that the double-rod filling the medullary canal provided support as much as the single wire; however, bending was observed at the femur as a result of the wires getting out of the cortex in double-wire technique. This can be attributed to the bending of the bone due to overload or muscle contractures. They also reported that not performing an ankle arthrotomy based on the extendable nail was an advantage in the single rod technique used for the tibia. In another study, Luhmann et al. [32] reported that the complication rate was 39% with extendable nails, whereas there was no complication with double-rod. In the present study, complication was established in 20 of 48 surgeries. We believe that the implant-dependent high complication rate in our cases results from the exposure of bone to an increased load stress due to early mobilization and the sufficient muscle strength that can deform the bone. Irrespective of the complication rates, whether this causes revision is the most critical concern [30]. Femoral angulation upon the K-wire migration from the cortex was also common among our patients; however, revision was not performed on all cases. In the literature, several articles on surgeries with non-extending nails report that the mean duration of revision was 2 to 2.5 years [11], and the mean time was 28 months for total revision surgeries in our series.

In addition, walking capacity is an important issue in OI patients. However, there are several factors with effect on patient's walking ability, which precludes predicting it beforehand. Daly et al. [17] concluded that walking potential was high in a child who was able to independently sit at the age of 10 months. Khoshhal and Ellis [14] indicated that factors such as motivation, increased body mass index, and reduced muscle strength might be important factors in mobilization. They highlighted that prolonged hospital stays and multiple operations had adverse effects on child development. Jones et al. [7] showed that the stabilization of long bones might help a child with a walking potential to walk. In the present study, the mobilization states of the patients were determined according to the ambulation scoring of Hoffer and Bullock. The mobilization state did not significantly change after surgery in any of our patients. However, the fracture risk reduced, and the immobilized patients had an improved articular motion and quality of life due to reduced pain after the surgeries. In mobilized patients, in turn, the fracture and deformity incidence reduced, and the maintenance of mobilization was ensured. We believe that mobilization cannot be significantly improved with surgeries; however, the existing status can be maintained by reducing the fracture and deformity risk.

Congenita A	Fractures at birth with deformation of bones,
Congenita B	Fractures at birth without deformation of bones
Tarda A	Fractures before walking
Tarda B	Fractures after walking

Table 1: Shapiro classification.

Type IA	Autosomal dominant, mild involvement or normal stature, most frequently, blue sclerae
Type IA	As above but dental involvement
Type II	Autosomal recessive, a fatal form seen in the perinatal period
Type IIIA	Autosomal recessive, heaviest form that can survive the newborn period, vertebral deformities, very short stature and multiple fractures, triangular face, fatal lung problems, normal sclerae
Type IIIB	As above but dental involvement
Type IVA	Autosomal dominant, mild and moderate involvement, white sclerae
Type IVB	As above but dental involvement

Table 2: Sillence classification

	Birth time	Right Femur	Left Femur	Right Tibia	Left Tibia
Case 1	1999	9-10-13 years	7 years	7-9-14 years	8 years
Case 2	1998	Ø	6-12 years	Ø	7-9-12 years
Case 3	1995	9 years	Ø	14-16 years	Ø
Case 4	2008	3 years	3 years	Ø	Ø
Case 5	2006	4 years	4-6 years	4-6 years	4 years
Case 6	2004	5 years	7 years	7 years	4-5-7 years
Case 7	2010	1-2 years	1 years	Ø	Ø
Case 8	2005	4-4,5 years	4-4,5 years	Ø	Ø
Case 9	2002	5 years	5 years	Ø	Ø
Case 10	2006	0.5-4-4,5 years	0.5-4-5 years	Ø	Ø
Case 11	2007	3 years	3 years	Ø	Ø
Case 12	2001	8-10 years	Ø	Ø	Ø

Table 3: The age distribution of the patients and the age at which the surgical operation was performed on which bone.

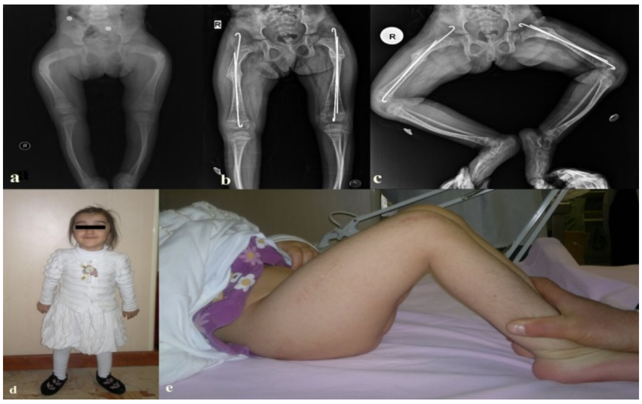

Figure 1: a) Appearance of patient with bilateral femur pseudoarthrosis and 90° anterolateral angulation that prevents to walking, b) anteroposterior radiological appearance after surgery, c) lateral radiological appearance after surgery, d) limited incision and approach, e) Patient's standing appearance.

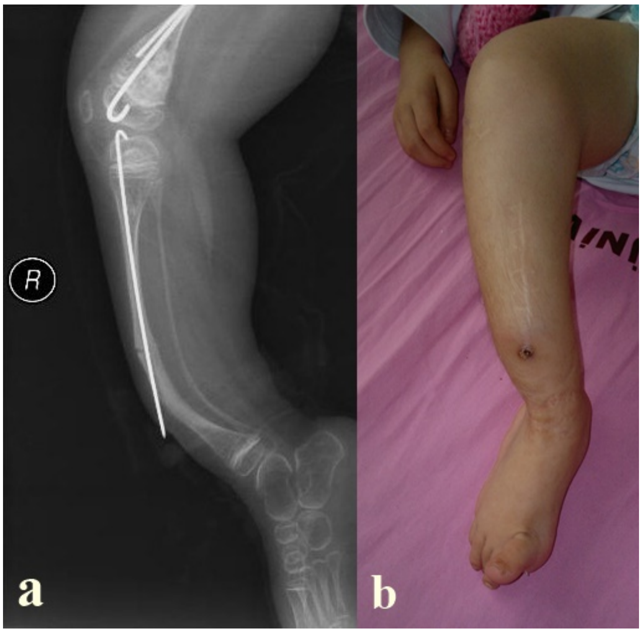

Figure 2: Appearence of patient with complication after tibial surgery. a) Increased angulation at distal part of the k-wire, b) out of the skin and cortica.

	Femur (n=33)	**Tibia (n=15)**	**Total (n=48)**
Nonunion	3	0	3
Kirschner wire migration	2	0	2
Fracture	3	1	4
Angulation and fracture	1	3	4
Angulation and cortical migration	4	3	7
Total	13	7	20

Table 4: Complications.

Grade I	Community walking
Grade II	Home walking
Grade III	Functional walking
Grade IV	Independent on wheelchair
Grade V	Dependent on wheelchair
Grade VI	Bed-dependent, non-ambulatory

Table 5: Classification of walking pattern according to Hoffer and Bullock.

Case no.	**Sillence**	**Shapiro**	**Ambulation Level**
Case 1	Type IA	Tarda B	Grade 1B
Case 2	Type IB	Tarda B	Grade 1B
Case 3	Type III	Tarda A	Grade 4
Case 4	Type IA	Congenita B	Grade 1A
Case 5	Type IVA	Congenita B	Grade 3B
Case 6	Type III	Congenita B	Grade 5
Case 7	Type IVB	Congenita B	Grade 3B
Case 8	Type IA	Tarda B	Grade 2A
Case 9	Type IVA	Congenita B	Grade 1A
Case 10	Type IA	Congenita B	Grade 1A
Case 11	Type IA	Congenita B	Grade 1A
Case 12	Type IA	Tarda A	Grade 1A

Table 6: Ambulation levels of patients.

Conclusion

In conclusion, although the technique and implants first described by Sofield have undergone several modifications to date, osteotomy, revision, and intramedullary nailing are basically the gold standard in preventing fractures and deformations of long bones. The major postoperative problem in OI patients is the high complication rates and inevitable need of revision. Most of the complications are due to the implant. Although the nonunion incidence is low, we believe that the most important reason is excessive periosteal stripping and implant failure. The double-rod application used at the femur decreases the number of revisions and prolongs the time of revision. Surgical treatment does not change patient ambulation; however, it maintains mobilization, reducing the incidence of fractures and deformities. In immobilized patients, in turn, it reduces pain and provides improved articular motion and quality of life. For patients with frequent fractures, ortheses that help walking can be limitedly used to help mobilization.Based on our study results, we suggest that this type of surgical technique using K-wires is a simple, cost-effective, and easy to access implant and is a technically applicable, is an effective and reliable method in treating OI, particularly in the conditions of Turkey, compared to other implants and surgeries.

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