Combined technique of titanium telescopic rods and external fixation in osteogenesis imperfecta patients: First 12 consecutive cases

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ABSTRACT

Introduction: The major limitation of any intramedullary telescopic system is rotational and longitudinal instability. The combination of telescopic system with an external fixator in patients with osteogenesis imperfecta (OI) demonstrated advantages of stability, early weight-bearing and rehabilitation. This study aimed to examine the outcomes of deformity correction by combined technique uniting titanium telescopic rod and reduced Ilizarov frame in children with types III or IV of OI with a minimum 1-year follow-up.

Material and methods: The study included 12 children with OI who underwent femoral deformity correction (20 segments) or tibial deformity correction (4 segments) by combined technique. The children ranged in age between 2 years and 3 months and 12 years and 4 months (mean: 8.9 ± 2.02 years) at the time of the rodding. Parameters of surgery, clinical examination data, data of 3D gait analysis were assessed in the study.

Results: External fixation lasted 35.8 ± 13.2 days in average. Neither loss of threaded fixation in the distal femoral and tibial epiphyses and apophysis of the greater trochanter nor migration of the rod into the knee and ankle joints were observed in follow-up. No secondary rotational or longitudinal bone displacement was noted. Telescoping gain related to spontaneous growth assessed at one-year follow-up control was 13.7 mm in the tibia and 15.9 mm in the femur. There were no deep infection or neurologic complications. The alignment measured by radio anatomical reference angles was maintained throughout the follow-up period.

Gait abnormalities in postoperative period were caused by bulk and weight of EF: external hip rotation, slight external angle of foot progression, increased stride width and increased hip abduction angle. The second feature was reduced ROM in sagittal plane at all levels associated with significantly reduced ankle plantarflexion, hip and knee joint moments in comparison to kinetics of limb without EF. These abnormalities resolved by the one-year assessment.

Conclusion: The combination of titanium telescopic rod with reduced external fixation is reliable advantage in reconstructive orthopaedic surgery for OI children. Reduced external fixation allows to overcome inconveniences of longitudinal and rotational instability of telescopic systems. Children were able to walk with weight-bearing since early postoperative period because of external fixation. Gait temporary changes were influenced by external device size and by strategy to reduce pin site pain.

1. Introduction

Osteogenesis imperfecta (OI) is a heterogenous group of genetic generalized connective tissue disorder with an incidence of 1 in 10.000 to 1 in 20.000 births.^{1–4} The bone fragility and refracture, osteopenia, progressive bone deformity and varying degree of short stature are primary clinical manifestations representing involvement the skeleton.^{1,2}

The telescopic rodding and transphyseal nailing provide a long-lasting osteosynthesis in a growing long bone.^{5–8} The most used system of Fassier-Duval telescopic rod allows to decrease the complication and

reoperation rate, and to reduce the blood loss in comparison to other telescoping systems.^{9–11} The Fassier-Duval rod has advantage of avoiding knee and ankle arthrotomy in femur and tibia surgery.¹²

However, the major limitation of any telescopic system is related to its inherent rotational and longitudinal instability.^{13–16} Furthermore, all telescopic systems do not allow an immediate weight-bearing in early postoperative period.^{12,17,18} This inconvenience of the system does not prevent secondary bone mass reduction, disuse osteoporosis and further fractures.^{6,14,15,19} Furthermore, the presence of stainless-steel telescopic constructs (e.g., Fassier-Duval rod) in children with osteogenesis imperfecta raising concerns for magnetic resonance

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imaging (MRI) study is one more disadvantage.^{20–23}

Since February 2018, we have used a telescopic rod made of titanium alloy in combination with minimal external fixation. In literature, the combination of telescopic flexible intramedullary nailing with an external fixator in patients with osteogenesis imperfecta or other severe underlying bone pathology demonstrated advantages of stability, early weight-bearing and rehabilitation.^{9,13,16,24}

The objective of the current study was to examine the outcomes of deformity correction by combined technique in children with severe and moderate-to-severe types of OI classification with a minimum 1-year follow-up. The accuracy of alignment, walking ability in early postoperative period by computed gait analysis, maintain of deformity correction in one-year follow-up, telescoping of rods, complications were assessed in the study.

2. Material and methods

The study included 12 children with Silience types III and IV OI²⁵ who underwent femoral deformity correction (20 segments) or tibial deformity correction (4 segments) by combined technique.

The Silience type III OI was diagnosed in two children, other 10 patients had type IV of OI. The children ranged in age between 2 years and 3 months and 12 years and 4 months (mean: 8.9 ± 2.02 years) at the time of the rodding.

Urgent telescopic rodding was produced for five fractures of previously malaligned femurs (three fractures happened over FIN). For six segments (5 femurs and 1 tibia), the rodding was the primary surgical procedure. Rod insertion was associated with removal of previously inserted intramedullary material (flexible nails in 10 segments and stainless-steel rods in four segments) for 14 segments. Nine patients in this study were treated with bisphosphonates (pamidronate or zoledronic acid) as in standard practice before study. Bisphosphonates were continued or initiated in middle postoperative period in all patients.

3. Surgical technique

Surgical steps included removal of previous material that was followed by osteotomy (-ies) performed percutaneously or in an open technique, and insertion of a guidewire for reaming. Guide wire was inserted through greater trochanter or proximal tibial epiphysis. Alternatively, the guide wire was placed through osteotomy site or fracture. Once the bone is aligned and reamed, the guide wire was replaced by male rod. The male rod was never screwed first into distal epiphysis in order to avoid intraarticular protrusion. The female rod was cut to size always intraoperatively and then inserted over the male rod. Female rod was always screwed first in proximal tibial epiphysis/greater trochanter with the female driver. At the last step of rodding, the male rod was screwed into distal epiphysis under x-ray control (C-arm) using the male driver. In all patients, the titanium alloy telescopic rod (Intramedullary Telescopic Rod, reg. certificate № RZN 2017/6876, dated 10.07.17., Designer: OOO “Metis”, Tomsk, Russia) was used. The diameter of female rod varied from 4.2 to 5.5 in our series. An external fixation using reduced Ilizarov frame was done as final step of surgery. A proximal short arc with 2–3 half-pins or half-wires and distal 2/3 ring with 3 wires were applied for femur. In tibia, two rings, each with 3 wires, were used. Rings and arc were connected in definitive position by the end of surgery ensuring rotational and longitudinal stability for bone fragments and required torsional deformity correction. In early postoperative period patients were encouraged for standing up and walking with weight bearing since 3rd to 4th day using a walker or crutches. No manipulations or external frame adjustment were performed after surgery. Frame removal was indicated and done while radiological signs of uninterrupted periosteal and endosteal callus were observed.

Outcomes of surgical intervention were evaluated by:

- 1) duration of surgery,
- 2) blood loss, decrease in RBC and hemoglobin in the first 48 post-operative hours, blood transfusion,
- 3) external fixation period,
- 4) reference radiological angles^{26,27} measured on the standard anteroposterior and lateral radiographs preoperatively postoperatively, and in 12-month follow-up post surgery (mLPFA, aMPFA, mLDFa, aLDFa, mMPTA, aMPTA, mLDTA, aLDTA, aPDFa, mPPTA, mADTA. Anatomical angles between the articular line and telescopic rod placed along the anatomical axis were measured postoperatively. That approach reflected orientation of articular line with regard to the segment axis during residual growth or due to remodeling of pathological bone around the nail or nail lateral migration,
- 5) length of rod telescoping,
- 6) adverse effect and complications.

At each assessment, the children underwent a standardized clinical examination that included joint ROM measurements by goniometry. The range of internal and external rotation of the hips in the prone position was used to measure rotational malunion. The Gillette Functional Assessment Questionnaire²⁸ was used to measure ambulation. Scoring was done before surgery, by the end of the first post-operative week and just prior to frame removal and in a year after treatment.

Three-dimensional gait analysis was also carried out on 9 subjects walking barefoot who underwent femoral surgery. Kinematic and kinetic variables were assessed evaluated on 6th to 10th day of post-operative period and approximately in a year after frame removal. We used 6-camerax Qualisys Oqus system and one AMTI force plate (Advanced Mechanical Technology Inc., Watertown, MA) to collect motion analysis data. In the Ilizarov Gait Analysis Laboratory the IOR model was used for markers.²⁹

A reduced Ilizarov frame configuration allowed to attach retro reflective markers in standard points according to anatomical landmarks. Marker FTC was placed right below proximal arc. The distal femoral and both tibial rings were placed out of anatomical landmarks leaving available space for markers FME, FLE, FAX, TTC, FAL and TAM (Fig. 1). So, that external frame configuration did not interfere with kinematic data collection.

Temporospatial parameters, joint kinematics and kinetics were calculated for each patient. The gait parameters of operated limb were compared with ones of limb without external fixator. AtteStat 12.0.5 software was used for the statistical analyses. The statistical values described the mean and standard deviation. Data were compared by using Wilcoxon signed-ranks test for matched pairs. The test was two-tailed with a 0.05 level of significance.

This research was approved by the Ilizarov Center Review Board. The study complies with the Declaration of Helsinki statement on medical protocol and ethics. Representatives of all patients enrolled in the study provided oral and written informed consent.

4. Results

Telescopic rodding was done for one segment per surgery. All patients who underwent two or more surgeries were operated on consecutively. There was no reoperation due to complications for the follow-up period. Two patients underwent three operations, rodding of two femurs was done consecutively in 6 children. In 6 patients only 1 femur was operated on. In five cases, telescopic rod insertion was done simultaneously with Ilizarov frame removal from the segment previously operated. In other cases of consecutive surgeries, an interval between operations varied from 2 to 6 months.

The average time for surgery was 135.9 ± 40.8 min: 115.2 ± 25.8 min for primary rodding combined with external fixation, and 149.2 ± 37.4 min for revision rodding combined with reduced Ilizarov external fixation. Intraoperative blood loss was assessed

Fig. 1. Retro reflective markers attached on patients with EF.

Table 1

Indices of hemogram.

Parameter	Preoperative mean value	In 24 h	In 48 h
Number of erythrocytes ($\times 10^6/\text{mL}$)	4.46 ± 0.49	3.49 ± 0.58	3.57 ± 0.51
Hemoglobin (g/l)	122.6 ± 12.35	101.6 ± 15.12	106.1 ± 10.46

as 119.3 ± 53.8 mL including 152.1 ± 27.5 mL in femoral procedures and 53.4 ± 11.6 mL in tibial surgeries. Table 1 demonstrates number of erythrocytes and hemoglobin in pre- and postoperative period (during first 48 h). Blood transfusion became indicated only in two cases of revision femoral procedure and in one primary femoral rodding.

In total, external fixation lasted 35.8 ± 13.2 days in average (range, 21–73 days). The mean external fixation time was 35.8 ± 13.2 days (range, 21–73 days) for the femur and 36.7 ± 8.6 days for the tibia (range, 29–46 days).

All patients were verticalized and encouraged for walking using crutches or walkers with full axial loading on the operated limb since 4th–7th postoperative day. Molded orthosis or plaster cast with free hip and ankle joints was applied for 3–4 weeks to all patients after frame removal. Patient continued walking with full weight-bearing with the orthosis on.

Radiological measurements of the operated femur and tibia are presented in Tables 2 and 3. The mean radio-anatomical angle values reflecting achieved alignment did not significantly changed throughout follow-up period (Figs. 2 and 3). A slight decrease in mLPFA was not accompanied by hip abduction less than 40° . Increased measurements of mADTA were associated with normal ankle dorsiflexion, more than 20° that excluded functional impairment due to reduced ROM. The mean rotational hip ROM were 40.4° for internal rotation and 46.2° for external rotation at frame removal. It remained well-balanced in one

Table 2

Radiological measurements of femoral reference angles (M $\pm$ SD).

Angle	Preoperative	Postoperative (after frame removal)	In 12 months
mLPFA; $^\circ$	103.4 ± 13.9	83.5 ± 13.2	86.3 ± 16.2
aMPFA; $^\circ$	79.5 ± 19.9	91.1 ± 14.02	87.8 ± 17.5
mLDFA; $^\circ$	94.5 ± 6.5	91.8 ± 5.4	90.8 ± 4.9
aLDFA; $^\circ$	79.3 ± 5.9	85.5 ± 4.4	85.3 ± 3.98
PDFA; $^\circ$	89.0 ± 6.3	84.7 ± 5.4	84.0 ± 4.7

Table 3

Radiological measurements of tibial reference angles (M $\pm$ SD).

Angle	Preoperative	Postoperative (after frame removal)	In 12 months
mMPTA; $^\circ$	93.3 ± 4.04	91.0 ± 2.0	90.9 ± 2.1
aMPTA; $^\circ$	90.3 ± 1.5	89.0 ± 3.6	89.4 ± 2.7
mLDTA; $^\circ$	79.3 ± 9.3	83.7 ± 7.6	83.1 ± 5.4
aLDTA; $^\circ$	83.3 ± 3.5	82.3 ± 7.5	83.3 ± 7.2
mPPTA; $^\circ$	71.0 ± 7.9	81.0 ± 3.6	82.4 ± 2.8
mADTA; $^\circ$	113.0 ± 8.9	100.3 ± 2.9	101.1 ± 4.6

year of follow-up (42.7° for internal rotation and 53.7° for external rotation) reflecting avoidance of rotational malunion.

In two femurs we noticed a partial varus deformity recurrence at osteotomy site due to small diameter of a rod that was insufficient for wide intramedullary canal.

Neither loss of threaded fixation in the distal femoral and tibial epiphyses and apophysis of the greater trochanter nor migration of the rod into the knee and ankle joints were observed in the patients. No secondary rotational or longitudinal bone displacement was noted in our series. During follow-up, in 1 patient MRI was required for diagnostic reason of pituitary adenoma. The presence of titanium alloy telescopic rod did not interfered with magnetic resonance study.

Telescoping gain related to spontaneous growth assessed at one-year follow-up control was 13.7 ± 3.1 mm in the tibia and 15.9 ± 2.3 mm in the femur (Figs. 2B and 3C).

There were no deep infection or neurologic complications either. There was discharge from the proximal and distal pin sites in two cases required a half-pin and a wire removal at three weeks after surgery. One patient fractured tibia over telescopic rod without displacement of bone fragments. The fracture was spiral and healed in plaster cast. Other adverse events included non-displaced fracture at the distal femoral metaphysis ($n = 1$) that occurred at installation of patient on the operating table. In one case preoperative planning did not accounted procurvatum deformity at distal third of femoral shaft. It caused modification of surgery while performing operation.

A

B

Fig. 2. Telescopic rodding of femur, combined technique used for alignment procedure after flexible intramedullary nailing: A - Consecutive radiographs with dates demonstrate result of deformity correction and outcome in 1 year 3 months after surgery; B – amount of telescoping since frame removal is of 16 mm.

The pre-operative mean FAQ score in group was 4.3 (range, 1 to 8). During the period of external fixation (Fig. 4) mean FAQ raised to 5.2 (range, 4 to 7). We should emphasize that after six surgeries patients reduced their level of FAQ from 8 to 6 due to applied external frame. Nevertheless, the mean FAQ score increased up to 6.8 by 1-year-point of follow-up. Only two patients did not advance their walking capacity remaining scored of 7 and 8 at one year.

Mean walking speed at the first study, with the external fixator (EF) in place, was 0.45 m/s; this significantly increased to 0.86 m/s at 1 year after treatment (Table 4). Cadence and stride length were reduced when wearing external fixator. They improved in long-term follow-up. The gait profile score revealed a progressive improvement of overall walking ability in children by 12 months after rodding. While EF wearing, the stride width was significantly increased on the EF side due

to the distal ring. Regarding stance and swing time, there was significant decrease of stance time and increase of swing time for operated side. But this had improved by the time of final follow-up.

Gait analysis (Fig. 4) revealed that sagittal kinematics were characterized by asymmetry between external fixator side and contralateral side (Tables 5 and 6). There was significantly reduced ROM at all levels on the EF side. Knee, hip patterns were abnormal in all subjects. At EF side, knee kinematics demonstrated full extension during stance phase and significantly reduced peak flexion angle in swing phase in comparison to contralateral limb despite clinical examination demonstrating greater passive knee ROM (more than 50° of flexion). Ankle position at initial contact was usually planiflexion on both sides and was followed by reduced dorsiflexion through mid to terminal stance. Decreased ROM on the EF side in sagittal plane correlated with grossly

Fig. 3. Telescopic rodding of tibia, combined technique used for alignment procedure after flexible intramedullary nailing: A,B - Consecutive radiographs with dates demonstrate result of deformity correction and outcome in 1 year 6 months after surgery; C – amount of telescoping since frame removal is of 19 mm. The level of upper extremity of female rod is indicated by dashed arrow.

Fig. 4. Gait with EF: A – patient G., walking with EF at the right femur, consecutive photos, EF side; B - patient G., walking with EF, consecutive photos, contralateral side; C – Gait cycle of patient G., mean data of kinematics and kinetics. EF site indicated by arrow. Decreased ROM on the EF side in sagittal plane is associated with reduced ankle plantarflexion moment, hip extension and flexion moment. Power generation on the EF side was significantly reduced at the ankle and knee joints; D – patient K., consecutive photos demonstrating gait using walker.

Fig. 4. (continued)

reduced ankle plantarflexion moment, hip extension and flexion moment. Power generation on the EF side was significantly reduced at the ankle, knee and hip in comparison to kinetics of limb without EF. These abnormalities resolved by the one-year assessment.

On the EF side, transverse plane kinematics were variable. In general, hip rotation was consistently external. It was associated with foot progression angle slightly externally rotated. These changes in transverse plane reflected gait adaptation to bulk and weight of Ilizarov frame as well as to pain and joint stiffness in sagittal plane.

5. Discussion

Improvement of mobility, self-care, functional skills and functional independence are the main goals ensuring better quality of life for children with OI.^{1,30} Only long-term multidisciplinary approach including bisphosphonate therapy, orthopaedic treatment, and rehabilitation (muscular strengthening and amelioration of range of motion) provide their achievement.^{12,31–33}

Telescoping rodding is recognized the most successful system for long-lasting intramedullary osteosynthesis dedicated to correction of long bone deformities in children with OI. However, the reoperation rate varies from 13% to 53% related to patient growth and subsequent fracture, migration of the rod or construct parts and joint intrusion (in

10.5–23.7% of the cases), non-telescoping (in 2.1%).^{13,17,34–37} A recent review article³⁸ on OI indicates to the use of telescopic rods versus regular rods or elastic nails as being controversial with technically demanding technique and high costs of telescopic systems.³⁹ The reoperation rate with telescoping rods is reported to be close to 50% compared with 58–87% with regular rods.⁴⁰

Avoidance of the operated limb during walking may adversely impact joint and muscle conditioning, as well as delay bone union due to a lack of axial loading.⁴¹ This aspect is unfavorable for the osteoporotic bone as it results in secondary bone mass reduction and disuse osteoporosis that can more handicapping than the disease itself.^{42,43} Use of telescopic rods suggests a 3-to-8-week period without weight-bearing. Weight-bearing is authorized by the end of this period, only with the locked KAFO (it means without active motions in standing position) and on a tilt table.^{12,18} In alignment reconstructive surgery, Munns et al.,¹⁸ Anam et al.¹⁷ suggested to start weight-bearing with the aid of orthoses 3 weeks after rodding only on condition of radiological evidence of bone callus formation.

Nevertheless, the rate of delayed union or non-union is still high in patients with osteogenesis imperfecta. It varies from 11% to 42% depending on bisphosphonate therapy, method of osteotomy, age of patient etc.^{17,18,36} We may speculate that delayed bone healing could be also related to primary longitudinal and rotational instability of any

Fig. 4. (continued)

telescopic system.^{8,44} Furthermore, telescopic rods are prone to secondary torsion displacement at osteotomy site or postoperative loss of torsional correction.^{13,37,43}

The combination of transphyseal telescopic rodding with reduced external fixation in our series allowed to overcome above mentioned disadvantages of telescoping rod: primary rotational and longitudinal instability and related secondary displacement, delayed loading of operated limb by weight-bearing, delayed bone healing.

First use experience with titanium telescopic rod in deformity correction in children with severe and moderate-to-severe OI demonstrated high reproducibility of the technique, the possibility of achieving required outcomes and lack of problems associated with migration of rod components. Telescoping of rod parts occurred in all the cases without construct blocking. The correction achieved persisted throughout the follow-up period. We insist on respecting the

appropriate medullary canal/telescopic rod diameter ratio.

Two more advantages of the titanium rod are worth mentioning. Neuroimaging studies with MRI are reported to be substantial for several neurological implications that children with OI can present.^{20–23} Titanium alloy that is used to manufacture the rod is safe for MRI examination with minimum artifacts. Another significant benefit with the titanium alloy telescopic rod is of the low cost.

Regarding assessment of gait in postoperative period, there are few published studies of gait analysis in patients wearing external fixator and after frame removal.^{45–47} In biomechanical analysis simulating the use of Ilizarov frame at tibia Layton et al.⁴⁶ reported the quantitative influence of bulk and weight of external fixator on gait abnormalities. Study revealed that increased inertia due to Ilizarov fixator causes alterations to step length. Increased stride width and abduction angle are related to the size of frame. Author found increased peak knee extensor

Fig. 4. (continued)

Table 4

Temporospatial data; me (25% ÷ 75%).

Variable	Gait wearing external fixator	At one year after surgery
Gait speed; m/s	0.45 (0.30 ÷ 0.52)	0.86 (0.77 ÷ 0.92) ^b
Stride width; m	0.19 (0.18 ÷ 0.22)	0.11 (0.09 ÷ 0.12) ^b
Stride length; m	0.60 (0.52 ÷ 0.69)	0.85 (0.66 ÷ 1.05)
Cadence; steps/s	0.85 (0.77 ÷ 0.95)	0.90 (0.83 ÷ 0.93)
Gait Profile Score	14.9 (12.4 ÷ 16.7)	12.3 (10.2 ÷ 14.4)

	Limb with External fixator	Contralateral limb	Both limbs
Cycle time; s	1.34 (1.28 ÷ 1.51)	1.33 (1.27 ÷ 1.53)	1.14 (1.03 ÷ 1.16)
Stance time; %	61.2 (57.5 ÷ 61.8)	71.2 (68.4 ÷ 75.6) ^a	66.5 (62.9 ÷ 71.4)
Swing time; %	38.9 (38.0 ÷ 41.1)	28.7 (24.7 ÷ 31.9) ^a	35.1 (34.0 ÷ 36.7)
Double support time; %	31.9 (26.0 ÷ 39.3)	33.4 (26.3 ÷ 39.5)	31.3 (25.8 ÷ 37.3)

^a - significant difference in comparison to value of contralateral leg by Wilcoxon signed-ranks test ($p < 0.05$).^b - significant difference in comparison to value of first study (while Ilizarov frame) by Wilcoxon signed-ranks test ($p < 0.05$).

Table 5

Kinematics and kinetics of ankle; Me (25% ÷ 75%).

Variable	Gait with external fixator	At one year after surgery	
	Limb with external fixator	Contralateral limb	Both limbs
Ankle Initial Contact; °	− 4.0 (− 4.6 ÷ 3.8)	− 1.9 (− 8.7 ÷ 4.0)	4.8 (1.5 ÷ 8.4)
Ankle ROM in Stance; °	22.1 (18.3 ÷ 27.4)	18.0 (16.4 ÷ 21.7)	22.2 (17.3 ÷ 27.3)
Ankle ROM in Swing; °	9.0 (4.8 ÷ 14.0)	16.6 (9.6 ÷ 20.2) ^a	16.1 (13.3 ÷ 22.5)
Timing of Max in Stance; %	52.0 (47.0 ÷ 72.0)	52.0 (49.0 ÷ 55.0)	50.0 (47.7 ÷ 55.3)
Foot progression angle; °	− 28.3 (− 30.3 ÷ − 16.3)	− 11.6 (− 15.5 ÷ − 4.4)	− 12.7 (− 16.2 ÷ 3.3)
Ankle plantarflexion moment; N*m/kg	0.29 (0.09 ÷ 0.31)	0.45 (0.25 ÷ 1.15) ^a	0.69 (0.52 ÷ 0.93)
Ankle Power; W/kg	0.38 (0.11 ÷ 0.73)	1.05 (0.29 ÷ 1.38) ^a	1.6 (1.32 ÷ 2.33) ^b

^a - significant difference in comparison to value of contralateral leg by Wilcoxon signed-ranks test ($p < 0.05$).^b - significant difference in comparison to value of first study (while Ilizarov frame) by Wilcoxon signed-ranks test ($p < 0.05$).

and plantarflexion moments that may be due to a requirement for an increased magnitude of propulsion in order to swing the EF leg. Authors recognize the limitation of the study that does not take into account the effect of injury, pain and wires attaching the frame. This study justifies redesigning the Ilizarov frame in a way to reduce diameter of rings.

Gait abnormalities magnify when the frame is attached on injured leg. Wong et al. reported a series of children with femoral fracture managed by Ilizarov frame.⁴⁵ Gait analysis was done just prior to Ilizarov frame removal and in middle- and long-term follow-up comparing results to patients treated with early hip spica cast. In EF group patients demonstrated a gait called “quadriceps avoidance” aimed to reduce distal pin site pain: knee maintained close to full extension throughout stance and swing, absence of push-off, reduced ankle moment and power. In coronal plane, gait abnormalities included

significant hip abduction and marked lateral trunk shift over injured leg reflecting antalgic gait strategy as well. Author emphasize rapid resolve of gait abnormalities after frame removal explained by short time in the EF. In contrast to EF treatment, patients managed by hip spica cast demonstrated like “crouch gait” due to quadriceps weakness caused by the period of immobilization in cast and disuse muscle atrophy. On the other hand, functional recovery was rapid in healthy children. We suppose that early functional activity in OI children must be crucial in contrast to above mentioned groups of patients.

Our study of gait demonstrated the fact that OI patients are able to walk in early postoperative period with weight-bearing. Combination of telescopic rodding with reduced external fixation ensured mechanical loading of operated limb. So, the gait may be quantitatively assessed.

In our series, OI patient demonstrated gait abnormalities caused by

Table 6
Knee and Hip kinematics and kinetics; Me (25% ÷ 75%).

Variable	Gait with external fixator		At one year after surgery
	Limb with external fixator	Contralateral limb	Both limbs
Knee Initial Contact angle; °	2,5 (–2,0 ÷ 10,0)	12,2 (5,9 ÷ 18,0)	14,1 (13,0 ÷ 20,0)
Knee ROM; °	18,0 (13,8 ÷ 27,8)	48,0 (39,7 ÷ 53,2) ^a	40,6 (38,6 ÷ 46,6)
Knee extension moment; N*m/kg	0,05 (0,04 ÷ 0,06)	0,10 (0,05 ÷ 0,11)	0,54 (0,46 ÷ 0,52)
Knee flexion moment; N*m/kg	0,07 (0,06 ÷ 0,08)	0,19 (0,06 ÷ 0,28)	0,09 (0,015 ÷ 0,17)
Knee Power; W/kg	0,22 (0,14 ÷ 0,42)	0,53 (0,48 ÷ 0,78) ^a	2,09 (1,24 ÷ 2,81)
Hip initial contact angle; °	36,3 (30,9 ÷ 46,7)	44,3 (31,7 ÷ 49,3)	36,2 (34,4 ÷ 36,9)
Hip ROM in sagittal plane; °	18,2 (14,0 ÷ 25,8)	40,7 (32,9 ÷ 43,0) ^a	42,4 (35,0 ÷ 46,1)
Hip Rotation; °	–25,0 (–29,8 ÷ –21,2)	–28,1 (–35,0 ÷ –18,5)	–19,6 (–24,8 ÷ –14,8)
Hip abduction; °	11,7 (9,4 ÷ 14,2)	4,3 (0 ÷ 14,8) ^a	8,1 (4,5 ÷ 11,0)
Hip ROM in coronal plane; °	11,7 (9,4 ÷ 14,2)	13,8 (12,9 ÷ 19,0)	13,1 (9,4 ÷ 19,3)
Hip extensor moment; N*m/kg	0,18 (0,08 ÷ 0,24)	0,43 (0,33 ÷ 0,61) ^a	0,81 (0,66 ÷ 0,95)
Hip flexion moment; N*m/kg	0,13 (0,12 ÷ 0,15)	0,18 (0,17 ÷ 0,28) ^a	0,31 (0,27 ÷ 0,41)
Hip Power; W/kg	0,3 (0,2 ÷ 0,37)	0,7 (0,59 ÷ 0,95) ^a	1,63 (1,44 ÷ 1,89) ^b
Pelvic Tilt; °	24,5 (18,4 ÷ 27,0)		11,1 (8,3 ÷ 14,3)

^{ca} - significant difference in comparison to value of contralateral leg by Wilcoxon signed-ranks test ($p < 0,05$).

^b - significant difference in comparison to value of first study (while Ilizarov frame) by Wilcoxon signed-ranks test ($p < 0,05$).

bulk and weight of EF: external hip rotation, slight external angle of foot progression, increased stride width and increased hip abduction angle. The second feature was that reduced ROM (especially almost permanent knee extension throughout full gait cycle on the EF side) at all levels was associated with significantly reduced ankle plantarflexion moment, hip extension and flexion moments in comparison to kinetics of limb without EF. In our study plantarflexed ankle position at initial contact was never followed by excessive dorsiflexion in stance as it was reported by Wong et al.⁴⁵ Abnormalities in OI children improved by the one-year assessment. We can state that gait abnormalities in OI children in postoperative time are close to those of children with femoral fracture managed with EF.

The limitation of the study is related to relatively small sample size of the cohort, nevertheless it included consecutive cases. The assessment of results was done with follow-up of one year. Further observation may reveal obstacles and problems related to long-term patient growth. On the other site, study with longitudinal follow-up would strengthen our findings too. This study did not include a control group as withholding surgery for appropriate candidates would be deemed unethical.

6. Conclusion

We conclude that despite the abnormal bone, the combination of titanium telescopic rod with reduced external fixation is reliable advantage in reconstructive orthopaedic surgery for OI children. Limited in time and reduced in bulk external fixation allows to overcome inconveniences of telescopic system related to longitudinal and rotational instability as it was predicted by Birke et al.¹³ Titanium alloy telescopic rod is not prone to limited telescoping, deformity relapse and rod migration. Children demonstrated abilities for walking with weight-bearing since early postoperative period because of external fixation. Gait temporary changes were influenced by external device size and by strategy to reduce pin site pain.

Ethical approval

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. Informed consent was obtained from all individual participants included in the study.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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