

First use experience with titanium telescopic rod in pediatric limb deformity correction in osteogenesis imperfecta**E.R. Mingazov, F.F. Gofman, A.V. Popkov, A.M. Aranovich, A.V. Gubin, D.A. Popkov**

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Objective Present and evaluate results from the first use of a Russian titanium telescopic rod in children with osteogenesis imperfecta. **Material and methods** A study group included 7 patients with osteogenesis imperfecta types III and IV who underwent femoral and tibial deformity correction with a combined use of titanium telescopic rod and reduced Ilizarov frame. The follow-up period was 6–12 months. **Results** The desired correction was achieved and maintained in all the cases at 6 to 12 months. The mean length of external fixation was 30.2 ± 7.5 days. No complications related to external fixation were observed. Telescoping was present in all cases. No secondary bone displacement, telescopic rod migration were noted. **Conclusion** Titanium telescopic rod applied in combined technology of long bone deformity correction in children with severe osteogenesis imperfecta showed high reproducibility of the technique, the possibility of desired correction and no problems related to rod migration. Telescoping of the rod occurred in all cases with no episodes of blocking. The correction achieved persisted throughout the whole period of observation.

Keywords: osteogenesis imperfecta, titanium telescopic rod, Ilizarov method

INTRODUCTION

Transphyseal telescopic rodding was offered for long-lasting intramedullary (IM) osteosynthesis of long bones in children with osteogenesis imperfecta (OI) [1, 2, 3, 4]. Most successful systems include telescopic rods (Fassier-Duval, Bayley-Dubow) [5, 6, 7, 8], and outcomes with telescopic systems using transphyseal elastic nails have also been reported [7, 9, 10]. The major limitation of any telescopic rod is its inherent rotational instability [11, 12, 13] which, being combined with longitudinal instability of the construct and delayed consolidation and/or considerably severe orthopaedic pathology, can aggravate clinical scenario and result in new iatrogenic orthopaedic problems [14]. In addition to that, axial weight-bearing on the operated limb cannot be allowed at a short term postsurgery with standalone telescopic IM system [8, 11, 15]. This can lead to a secondary decrease in bone mass, aggravated osteoporosis and predispose to secondary fractures in patients with OI [2, 12, 13, 16]. The presence of metal

telescopic constructs raises concerns in producing magnetic resonance imaging (MRI) [17, 18, 19, 20]. Finally, the cost of foreign telescopic rods remains extremely high.

Combination of intramedullary telescopic systems and reduced external fixation applied at the Kurgan Ilizarov Center was shown to be effective overcoming disadvantages of longitudinal and rotational instability of standalone telescopic systems [21, 22]. This approach facilitated appropriate axial loading at early postoperative period [23]. Russian telescopic rod made of titanium alloy has been made commercially available in Russia since July 10, 2017 and has been used at the Kurgan Ilizarov Center since February 2018 as part of combined osteosynthesis.

The purpose of the report is to present and evaluate results from the first use of a Russian titanium telescopic rod in children with osteogenesis imperfecta at a 6-to-12-month follow-up.

MATERIALS AND METHODS

The study included 7 children with Sillence types III and IV OI [24] who underwent femoral deformity correction (6 segments) or tibial deformity correction (2 segments) with combined technique

using Russian titanium rod (intramedullary telescopic rod, reg. certificate № RZN 2017/6876 dtd 10.07.17. Designer: ООО “Metis”, Tomsk; manufacturer: “Osteosynthes”, Rybinsk). The rod was made from

✉ Mingazov E.R., Gofman F.F., Popkov A.V., Aranovich A.M., Gubin A.V., Popkov D.A. First use experience with titanium telescopic rod in pediatric limb deformity correction in osteogenesis imperfecta. *Genij Ortopedii*, 2019, T. 25, No 3, pp. 297-303. DOI 10.18019/1028-4427-2019-25-3-297-303. (In Russian)

VT-6 titanium alloy, GOST 19807-91. The binding inclusion criterion was length of observation of at least 6 months postsurgery.

Sillence type III OI was diagnosed in two children and five had type IV OI. Mean age of the patients at the time of surgery was 9.4 ± 1.5 years. Emergency operative treatment as a primary procedure was produced in two cases for femoral fracture of malaligned femur. Revision rodding was performed in the rest of the cases after elastic nailing (5 segments: 3 femurs, 2 tibiae) or telescopic stainless steel nailing (1 femur). Indications to revision rodding included recurrent clinically important deformity with low ambulation capabilities, low physical function, absence of telescopic steel rod and the development of a metadiaphyseal deformity. Surgical steps included removal of a construct in revision rodding procedure, introduction of a guidewire with correcting osteotomies consecutively performed, reaming of intramedullary canal, placement of telescopic rod and fixation of threaded ends in the epiphysis (apophysis of greater trochanter), wound suturing, osteosynthesis with reduced Ilizarov external fixation device using distal and proximal rings and 2–3 fixation components.

Introduction of telescopic rod started with the threaded end of the inner part primarily located at the level of distal metaphysis followed by placement of the external component to be screwed into the greater trochanter at femoral rodding or proximal tibial epiphysis. Then the inner component was placed by screwing the outstanding portion into the distal epiphysis and the outstanding part was cut off. The sequence of the placement excluded protrusion of the inner component into the distal joint with regard

to the rodding segment as described in the manual on classic Fassier-Duval telescopic intramedullary system [11].

Outcomes of surgical intervention were evaluated by:

1) length of surgery, blood loss, decrease in RBC and hemoglobin counts in the first 48 hours postsurgery (the least values), blood transfusion;

2) length of external fixation;

3) reference angles [25, 26] measured preoperatively, postoperatively, after removal of the Ilizarov frame, at 6–12 months postsurgery (mLPFA – mechanical lateral proximal femoral angle, aMPFA – anatomical medial proximal femoral angle, mLDFA – mechanical lateral distal femoral angle, aLDFA – anatomical lateral distal femoral angle, mMPTA – mechanical medial proximal tibial angle, aMPTA – anatomical medial proximal tibial angle, mLDTA – mechanical lateral distal tibial angle, aLDTA – anatomical lateral distal tibial angle, PDFA – posterior distal femoral angle, PPTA – posterior proximal tibial angle, ADTA – anterior distal tibial angle). Anatomical angles between the articular line and telescopic rod placed along the anatomical axis were measured postoperatively. The approach reflected a possible change in orientation of articular ends with regard to the segment axis during residual growth and/or owing to remodeling of pathological bone around the nail or nail migration;

4) length of rod telescoping;

5) adverse effects, errors and complications.

Statistical data analysis was performed using *AtteStat* 13.1 computer program (Russia). Statistical analysis included descriptive statistics: the arithmetic mean (M) and standard deviation (SD).

RESULTS

The average operating time was 136.1 ± 38.7 minutes on the whole, 130 ± 35.4 minutes for primary rodding and 137.9 ± 42.1 minutes for revision rodding combined with reduced Ilizarov external fixation. Estimated blood loss was 143.9 ± 55.7 mL including 163.3 ± 58.9 mL in femoral procedures and 41.7 ± 10.4 mL in tibial

surgeries. Table 1 shows preoperative RBC and hemoglobin counts and the minimal values recorded in the first 48 hours postsurgery. Blood transfusion was required in revision femoral procedures ($n = 2$) and in primary femoral rodding ($n = 1$) with difficulties in use of tourniquet.

Table 1

RBC and hemoglobin count

Parameter	Preoperative count	First 48 hours postsurgery
RBC count in femoral deformity correction; $\times 10^{12}/\text{mL}$	4.2 ± 0.5	3.1 ± 0.5
Hemoglobin in femoral deformity correction; g/mL	122.3 ± 13.1	90.7 ± 15.9
RBC count in tibial deformity correction; $\times 10^{12}/\text{mL}$	4.39 ± 0.29	4.12 ± 0.16
Hemoglobin count in tibial deformity correction; g/mL	118.0 ± 7.1	113.7 ± 5.5

Length of external fixation was 30.2 ± 7.55 days (28.7 ± 4.03 days for the femur and 33.4 ± 12.86 days for the tibia). All patients were verticalized using crutches or walkers with full axial loading on the operated limb during the first seven postoperative days. Plaster cast with free hip and ankle joints was applied for 3–4 weeks to all patients after frame removal. They continued walking with full weight-bearing with the cast on. Radiological and anatomical measurements of the

operated femur and tibia are presented in Tables 2 and 3.

The mean reference angles achieved were shown to be normal or close to normal values throughout the whole period of observation (Fig. 1 and 2). A slight decrease in mLPFA caused by moderate varus deformity of the femoral neck was accompanied by abduction of the femur of at least 40° . Increased measurements of ADTA were associated with dorsal flexion of the foot of at least 20° that excluded functional limitations.

Table 2

Radiological and anatomical measurements of the femur, reference angles ($^\circ$)

Description	mLPFA	aMPFA	mLDFA	aLDFA	PDFA
Preoperative	100.5 ± 13.4	80.7 ± 8.4	101.8 ± 7.3	87.5 ± 11.3	70.3 ± 11.1
Postsurgery	85.7 ± 14.8	89.3 ± 12.97	89.3 ± 4.4	85.8 ± 6.2	81.5 ± 6.5
Frame removed	86.2 ± 15.4	88.3 ± 16.2	91.3 ± 4.6	85.2 ± 4.5	81.8 ± 7.5
6–12-month follow-up	85 ± 15.2	89.3 ± 15.9	92.8 ± 6.2	86.2 ± 5.2	82.3 ± 6.3

Table 3

Radiological and anatomical measurements of the tibia, reference angles ($^\circ$)

Description	mMPTA	aMPTA	mLDTA	aLDTA	PPTA	ADTA
Preoperative	92 ± 4.2	87 ± 3.7	82 ± 7.1	84.7 ± 2.5	76 ± 8.5	121 ± 12.7
Postsurgery	88.5 ± 2.1	88.5 ± 2.1	82 ± 8.5	82 ± 8.5	85 ± 0.7	104.5 ± 6.4
Frame removed	89.5 ± 0.7	89 ± 0.5	86.5 ± 0.71	85 ± 1.4	80.5 ± 0.7	104 ± 5.7
6–12-month follow-up	89 ± 1.4	90.5 ± 0.7	86.5 ± 0.71	84.5 ± 0.7	81.5 ± 0.7	103.5 ± 4.9

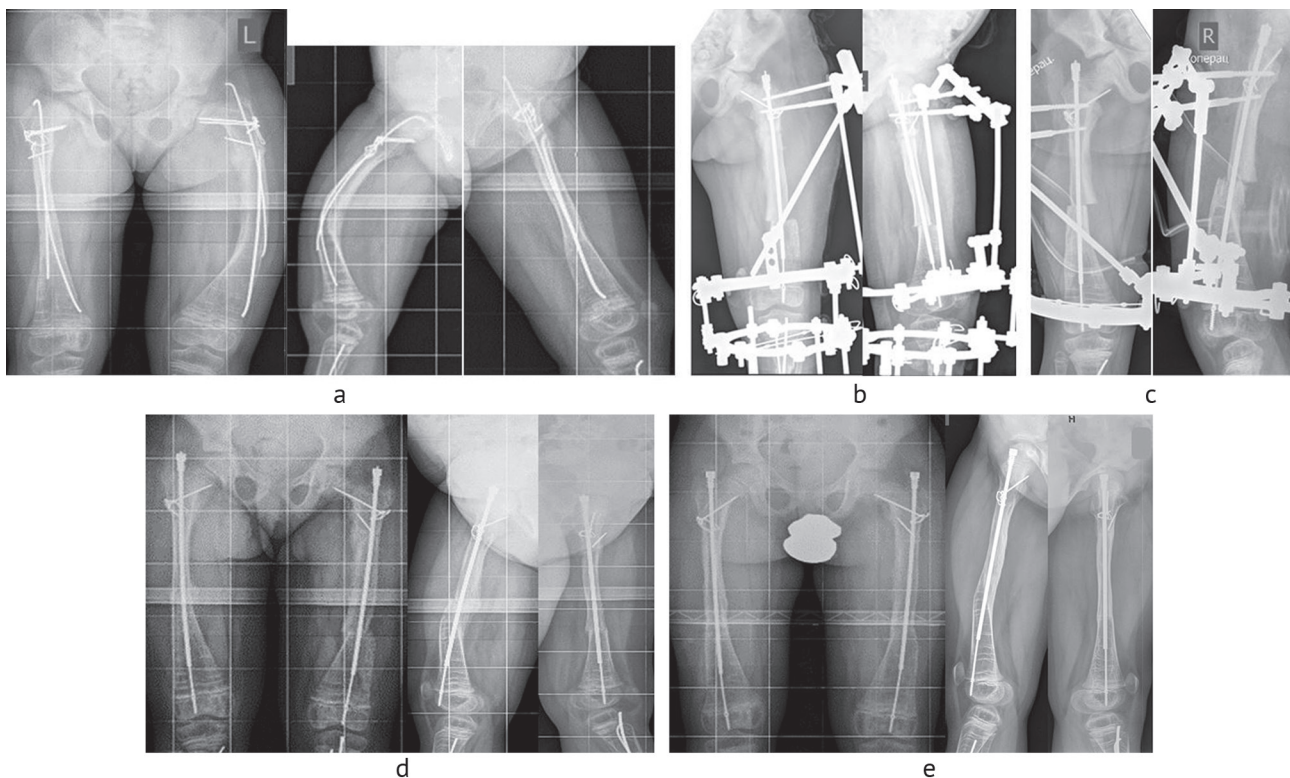


Fig. 1 An instance of revision correction of femoral deformities showing (a) preoperative radiographs; (b) deformity of the left femur being corrected with combined technique; (c) deformity of the right femur being corrected with combined technique; (d) external fixation devices removed; (e) rod telescoping and no signs of migration at 12 months after removal of the frame from the right femur and at 13 months after removal of the frame from the left femur

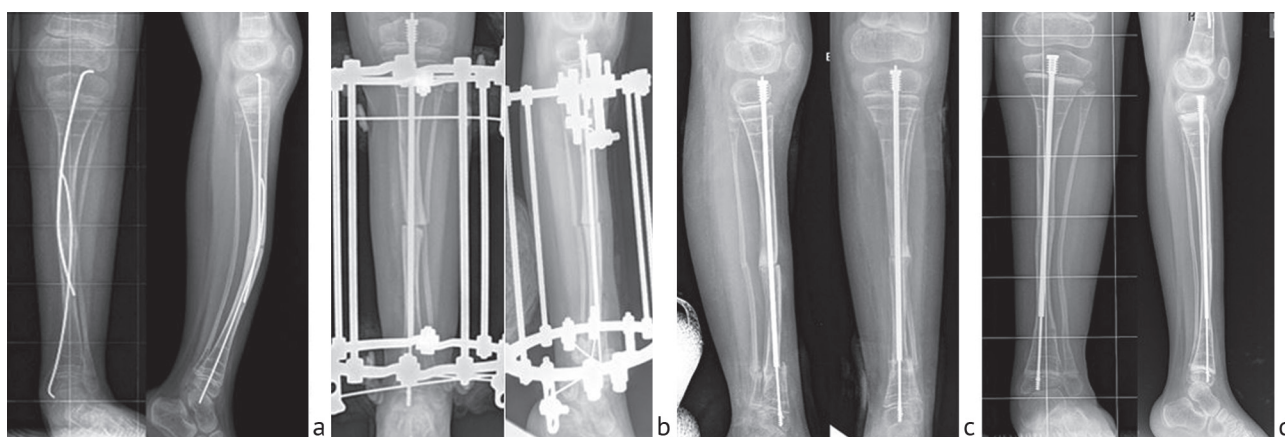


Fig. 2 An instance of revision correction of tibial deformity showing (a) preoperative radiographs; (b) combined osteosynthesis with the Ilizarov apparatus and titanium nail; (c) external fixation devices removed; (d) rod telescoping and no signs of migration at 12-month follow-up

Telescoping length measured in patients with a follow-up of at least 12 months was 13.5 ± 2.8 mm in the tibia and 15 ± 1.4 mm in the femur. Adverse events and complications included undisplaced fracture at the distal femoral metaphysis ($n = 1$) that occurred at positioning of the patient on the operating table. Neither additional intervention nor changes in the preoperative planning were required in the case, and the complication did not affect the final outcome. Another patient developed partial recurrence of varus deformity ($n = 1$) at the correction level due to moderate compression forces created between rings of the frame at early postoperative period (Fig. 3). The deformity was deemed to occur due to insufficient diameter of

the rod placed into a wider intramedullary canal. The complication did not affect the final result of the treatment because of bifocal correction that allowed us to achieve normal mechanical reference angles of the femur and proper alignment of the lower limb.

No inflammatory events were recorded around the Ilizarov components throughout the observation period. Rod telescoping occurred in all cases. Neither loss of threaded fixation in the femoral and tibial epiphyses and apophysis of the greater trochanter nor migration of the rod to the knee and ankle joints were observed in the patients. No secondary rotational or longitudinal bone displacement was noted in the cases.

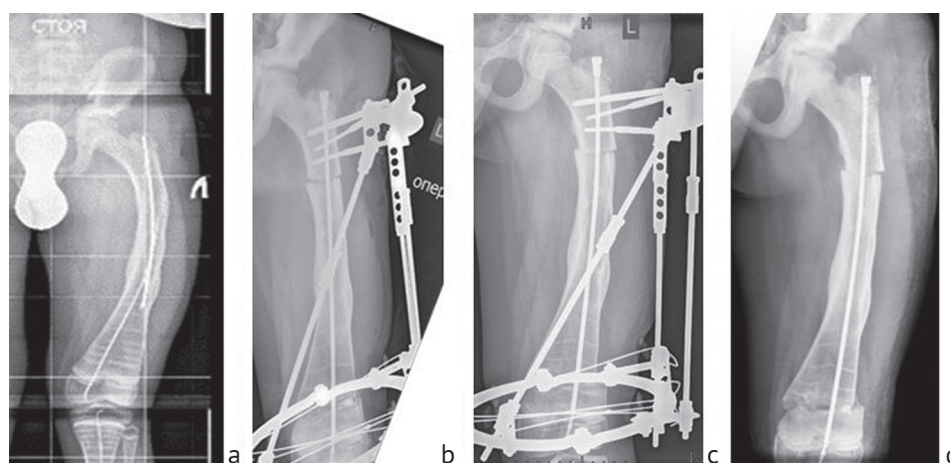


Fig. 3 An instance of partial loss of deformity correction at the level of the proximal osteotomy showing (a) preoperative radiograph; (b) intraoperative radiograph; (c) an angular deformity of 7° developed at the level of the proximal osteotomy following compression forces created with the rings of the external fixator; (d) radiograph of the femur after frame removal

DISCUSSION

Telescoping rodding is considered to be the most successful solution in creation of intramedullary transphyseal telescopic system applied for correction

of long bone deformities in children with OI. However, the complication rate is reported to range from 35 % to 55 % in standalone application of

telescopic systems [8]. The reported complications [9, 27, 28] include migration of the rod or construct parts in 10.5–23.7 % of the cases, inability to telescope in 2.1 %, fractures of nails in 6.9 %, delayed or failed consolidation of osteotomized bone fixed with telescopic IM nail in 20–25 %. Standalone application of intramedullary constructs suggests a 4-to-6-week period when the patient should avoid substantial weight-bearing on the limb to prevent secondary deformity. This aspect is unfavorable for the bone since it results in additional decrease in BMD due to immobilization with orthosis or plaster cast and absence of axial loading [6, 7, 9, 29, 30]. In addition to that, telescopic rods and elastic nailing cannot prevent the development of secondary torsion deformity (bone twisting at the level of osteotomy) or postoperative loss of torsional correction [5, 30, 31]. A recent article [32] on OI with F. Fassier being one of co-authors indicates to the use of telescopic rods versus regular and elastic rods as being controversial with technically demanding technique and high costs of telescopic systems. The reoperation rate with telescoping rods is reported to be close to 50 % compared with 58–87 % with regular rods.

Combined technique developed at the Kurgan Ilizarov Center for pediatric correction of limb deformities using transphyseal rodding and reduced external fixation allowed us to avoid disadvantages of standalone application of telescoping constructs. The system completely excluded risks of secondary rotation instability providing the possibility of

appropriate axial loading on the operated limb at early postoperative period that is an important aspect of the system according to the Ilizarov principles [23, 33]. However, transphyseal elastic nailing has disadvantages of a higher rate of reoperation compared to that of telescopic Fassier-Duval rodding to achieve desired outcomes being 87.5 % with elastic nailing [23] and 50 % with telescopic rods [32]. That is why the Russian titanium telescopic rod is comparable to the best foreign designs and is a more prospective implant than elastic nails.

First use experience with titanium telescopic rod in combined practice of deformity correction in children with severe OI showed high reproducibility of the technique, the possibility of achieving required outcomes and lack of problems associated with migration of rod components. Telescoping occurred in all the cases without construct blocking. The correction achieved persisted throughout the observation period. Dimension ratio between diameters of the rod and intramedullary canal must be ensured with the construct to prevent secondary angular deformities. 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 [17, 18, 19, 20]. Titanium alloy that is used to manufacture the construct is safe for MRI examination with minimum artifacts. Another significant benefit with the Russian telescopic rod is the low cost, now it is 6.25 times less expensive than Fassier-Duval rod.

CONCLUSION

Titanium telescopic rod applied in combined technology of long bone deformity correction in children with severe osteogenesis imperfecta showed high reproducibility of the technique, the possibility of desired correction and no problems related to rod migration. Telescoping of the rod occurred in all cases with no episodes of blocking. The correction achieved persisted throughout the

whole period of observation. No complications associated with specific application of titanium telescopic rod were observed at placement and throughout the observation period of 6-12 months postsurgery. Further observation and evaluation of outcomes are required for more accurate assessment of the possibilities, advantages and disadvantages of the system.

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Received: 20.06.2019

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