Literature Review
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# Multi-stage correction of shortening and deformities of the lower leg in children and adolescents with an external fixator according to Ilizarov: a systematic literature review

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## Introduction

The main principles of transosseous osteosynthesis, developed by G.A. Ilizarov (originally for the treatment of fractures), are: 1- reposition of fragments and good contact of the spliced areas of the bones, 2- stable fixation, which is maintained in any typeloads, 3 - early loading with active function of adjacent joints, 4 - absence of trophic disorders, 5 - minimal trauma of the fixation method.[1] In the classification of transosseous osteosynthesis, 3 large groups are distinguished - compression, distraction, compression-distraction, in which 2 subgroups are distinguished - mono- and polylocal, combined or alternating. Bilocal alternating distraction-compression and bilocal combined compression-distraction osteosynthesis is used to replace defects in long tubular bones. Multi-stage transosseous osteosynthesis should be considered any of the above options, which initially or during treatment cannot lead to the final result (satisfying the needs of the patient and the doctor's requests) and requires repeated surgical intervention.

## Materials and methods

Every year, thousands of surgical interventions are performed around the world for congenital/acquired shortening and/or deformity of the lower leg bones. It is not possible to find absolute figures for the number of operations performed on the global Internet network. Indirectly, the scale of this section of surgery is indicated by the frequency of congenital diseases that cause low growth or deformities of the lower extremities (achondroplasia 1/15000-40000 newborns, hypochondroplasia 1/30000, pseudoachondroplasia 1/100000,[2] Lobstein-Vrolik disease 4-7/100000, Gaucher disease 1/50,000, bone tumors, inflammatory diseases, etc.). According to some authors,[3] it ranges from 1.33% to 2.47%, of which 57.9–61.1% are accounted for by disorders in the development of the lower extremities.

In addition, already at the stage of pregnancy, it is possible with a certain degree of probability to predict the birth of a child with a proportional (both height and weight) developmental delay. Fertility analysis showed a direct relationship between the quality of life and the number of underdeveloped children. An interesting correlation with the level of systemic arterial pressure was also revealed: at a SBP level of 108 mm Hg. Art. developmental delay is up to 13%, while at a SBP level of 115 it is only 6%.

Additionally, it is necessary to take into account the initial length of the segments, since there is a direct positive strong relationship between the latter and the speed, and in the rudiments of the limbs (for example, at the age of 5 the leg length was 12 cm), there is no growth of the limb in length at all. In general, this dependence is described by the following formula: V =-0.48 + 0.08xL cm / year for the shoulder, forearm and thigh, while for the lower leg it is -0.51+ 0.13xL cm / year. At the birth of a child with congenital malformations (for example, amniotic constriction, which resulted in the amputation of the entire segment or part of it), a deficit of 4 cm is partially compensated for by the growth retardation of the intact limb, while shortening of more than 12 cm does not affect the length of the healthy segment in any way.[4]

It is especially worth highlighting cases of short stature and massive bone defects, since with them there is a high probability of repeated surgical interventions, due to the fact that it is often not possible to obtain the final result at once for various reasons (severe pain syndrome, contractures of adjacent joints, the mental state of the patient , changes in the soft tissues of the segment (scars, burns, contractures, inflammatory changes in nearby tissues), "poor" bone regeneration, etc.). In these cases, the patient's treatment is divided into several stages to achieve the desired result.

## Features of multi-stage surgical interventions

### Determination of indications for correction

In fact, all reconstructive interventions on the musculoskeletal system aim to bring certain characteristics of the patient closer to the standards of the environment and bring the biomechanical parameters of the musculoskeletal system closer to such values that there is no

excessive overload of the articular surfaces, ligamentous apparatus and muscles, which causes them long-term satisfactory performance. To determine the minimum size of the limbs, it is necessary to use the concept of percentile - this is a hundredth of the measurements of a population of people, which corresponds to a certain value of an anthropometric trait. This criterion is paramount for the patient and secondary for the attending physician. When correcting the length of the limbs, it will be correct to focus on the 5th percentile for women. In this category, the length of the lower limb is 786 mm with a minimum height of 150 cm and, accordingly, is the target indicator for interventions on the lower limbs.[5] The second equivalent indicator (significant for a traumatologist-orthopedist) is the creation of optimal relationships in the joints (especially the lower extremities). There are average indicators and ways to determine them to create an optimal load on the articular surfaces, ligaments of the joints and muscles.[6] Relatively controversial is the question of the absolute shortening of one of the lower extremities. Most orthopedic surgeons share the position of Marx V.O. that shortening of more than 2 cm causes gait disturbance, compensatory changes in posture and overload of the joints of the shortened limb due to the so-called. "Falling lameness". In the latter case, it is not always possible to resort to equalizing shortening, since the compensatory reactions of the muscles may be insufficient. Acute shortening of the femur by 5– 6 cm and the tibia by 3 cm is considered relatively safe.[7]. The classical setting is the need for surgical intervention with a difference in limb length of more than 4 cm, from 2 to 4 cm, surgery is possible, but not necessary, and less than 2 cm does not require surgical treatment.[8]

## Determining the optimal timing

In the works of many authors, it has been shown that it is necessary to start lengthening the limbs as early as possible, due to the high elasticity of soft tissues and the high reparative ability of bones in children aged 6-9 years. Limb lengthening up to 10 years of age avoids interference in the educational process in adolescence.[9] Carrying out the first stage of treatment at the age of 5-7 years is important for achieving a positive result of treatment.[10] It should be noted that in patients with comorbidities, the picture changes. According to studies of patients with rickets-like diseases,[11] the most favorable period in terms of correction of deformity and length is the period of the first growth spurt from 6 to 14 years; in this interval, at the age of 7 to 10 years, relapses of deformities are most often observed.

## Making a decision on the multi-stage correction

The initial limitation for one-stage treatment is the fact that, unlike natural growth, surgical lengthening reduces muscle strength, joint mobility, and neuropathies occur.[12–16] Also, by itself, the muscle traction that increases with elongation limits the activity of growth zones (Volkamn-Huter principle), which "takes" additional centimeters of natural growth from the patient.[17] This principle was previously noted by the staff of the Ilizarov Center (Kurgan) and Bari-Ilizarov Orthopaedic Centre (Bangladesh). In the work of O.V. Kolchev, D.Yu. return to baseline within 12 months, type III - an increase in growth rate followed by a slowdown to rates below the initial rate, type IV - a sharp slowdown or stoppage of growth with a gradual slight recovery after 9-12 months from the moment of surgery, type V - a gradual decrease growth rate with a possible slight recovery after 9-12 months.[18] Separately, it is worth mentioning the changes in the distraction regenerate itself. The well-known expression in the Ilizarov Center "1 month per 1 cm" is justified by the fact that the time of consolidation of the distraction regenerate increases disproportionately depending on its length. The main indicators used in the assessment of distraction osteosynthesis are: 1-DI-distraction index (calculated as the ratio of the length of the regenerate in mm to the number of days of distraction, in other words, the speed of distraction), 2-DCT-distraction-consolidation time (number of months from the onset of distraction before the formation of three of the four cortical plates of the regenerate), 3-CI - consolidation index (the ratio of the number of days from the operation to the dismantling of the external fixation device to the length of the regenerate in centimeters), 4-LI - elongation index (the number of days of fixation to the length of the regenerate in centimeters). It should be noted that only 2 indicators help to determine the tactics of treatment at various stages, while the rest are purely descriptive. In their work, Lukas Zak et al.,[19] when evaluating the results of lengthening in 19 patients, showed that with an average lengthening of 4 cm, regardless of age and gender, the distraction index is 0.71 mm/day, the lengthening index is 3.4 months/cm, the distraction-consolidation index is 8.4 months, the consolidation index is 86 days/cm, which differs from the indicators of the Ilizarov Center. There are many studies of distraction regenerate fractures. The Simpson-Kenwright classification is known, in which 4 types are distinguished: Ia - compression of the regenerate zone, Ib - fracture in the growth zone of the regenerate with displacement, II - fracture in the "base" of the regenerate, III - fracture of the maternal bone proximal / distal to the regenerate zone, IV – fracture of the adjacent segment.[20] Also, much attention is paid to the X-ray morphometric parameters of the distraction regenerate, in particular, the Ru Li classification,[21] which identifies 5 main callus morphotypes: 1. "Fusiform" (the area of the regenerate exceeds the area of interfragmentary diastasis). 2. "Cylindrical" (the area of the regenerate corresponds to the area of interfragmentary diastasis). 3. "Concave" (the regenerate is formed according to the "hourglass" type.) 4. "Lateral" (the regenerate has a marginal defect), 5. "Central" (the regenerate is represented by a thin column in the central part. The second, third and fifth types belong to unstable, due to which they require additional timely surgical intervention, for example, intramedullary TEN reinforcement.[22,23] On average, the frequency of regenerate fractures is about 11% in all age groups. The best option for preventing this complication is a multi-stage lengthening. With repeated lengthening of the lower leg, the radiographic picture of reparative osteogenesis at the proximal level of the tibia does not fundamentally differ from that during its first lengthening. A densitometric study of the regenerate showed a greater intensity in its proximal sections, which is associated with a large volume of soft tissues and better blood supply.[24] YES. Popkov, Prof. J.Prévot in his work,[25] guided by the fact that complete restructuring and adaptation of the musculoskeletal system after surgical lengthening occurs after 5-6 years, examined 18 patients who underwent multiple lengthening of the upper and lower extremities. The total average increase was 24.4 cm. Of these, more than half are satisfied with the results, a third are very satisfied. All patients received their driver's license without any problems, which made their life much easier. At the same time, the restructuring of the regenerates themselves, according to Dyachkov K.A. et al.,[26] occurs after 1.5-2 years, which creates a good basis for reoperation. In general, the use of the Ilizarov method to replace bone defects and correct deformities is highly appreciated.[27]. When assessing the replacement of post-traumatic defects, good and satisfactory results were noted in 89.3% of patients. Some authors achieved 100% good and satisfactory results, but it is worth noting that most of the patients for the entire period of distraction and correction were hospitalized under the supervision of the attending traumatologist-orthopedist.[28–30] A more realistic picture in terms of the availability of this method of treatment is reflected in another study that compared the results of treating patients with temporary hemiepiphysiodesis with transosseous osteosynthesis and an isolated

method of transosseous osteosynthesis.[31] At the same time, the percentage of complications in the first group was 53% in the period from 6 to 24 months. In the second group, the number of complications was significantly higher and amounted to 81%. The same authors noted one significant disadvantage of eight-shaped plates - growth correction occurs only in one plane, and correction of the torsion component and length is impossible. In addition, the use of a plate is absolutely ineffective if the vertex of the deformity is located in the projection of the joint gap. Similar results were demonstrated in their study by A.A. Shchukin et al.[32] After studying the results of multi-stage segment lengthening in 41 patients and evaluating them according to the Lascombs complication criteria, the following observations and conclusions were made: 1-the frequency of unsuccessful results increases with repeated lengthening in patients older than 14 years; 2 - the frequency of adverse treatment results with repeated lengthening increases with lengthening more than 50% of the initial length of the segment; 3-The frequency of adverse outcomes increases with a high rate of distraction (Osteosynthesis Index less than 20 days/cm). The authors themselves noted in their series the successful results of elongation according to Labscomes, provided that the relative elongation of the segment did not exceed 50%. Based on the above data that the use of the Ilizarov technique improves the quality of life of patients, contractures, neurotrophic complications and failure of the distraction regenerate remain the only obstacle to its use. All of these complications can be easily avoided by separating the process of lengthening and correction in time (creation of stages). At the same time, establishing the number of stages is extremely difficult to predict and communicate to patients. In the work of A.V. Popkov,[33] when evaluating the treatment of almost 300 patients, it was noted that with approximately the same percentage lengthening of the femur and lower leg at two stages, the restoration of movements in adjacent joints a year after the dismantling of the apparatus is lower with repeated lengthenings. Thus, with femur lengthening by 13.7% at the first stage and 17.3% at the second stage, the restoration of range of motion in the knee joint was 80.3% and 37.8%, respectively. For the lower leg, with lengthening at the first stage by 19.2%, and at the second by 17.6%, the recovery of movements in the knee joint was 95.8% and 74.3%, in the ankle joint 77.3% and 44.6%, respectively. In general, a huge number of studies of various complications accompanying distraction osteosynthesis.[34–46] In most cases, complications of distraction osteosynthesis in the Caton classification belong to types I and II and require minor additional interventions by the attending physician (removal/repositioning of transosseous elements, administration of systemic and local antibiotic therapy, increased volume of exercise therapy and physiotherapy procedures after removal of the device, re-osteosynthesis). Category III complications range from 0 to 13% in various studies and, as a rule, arise due to inconsistency between the actions of the doctor and the patient. In extremely rare cases, the physiological characteristics of the patient are present (increased susceptibility to thrombosis may in worst cases lead to amputation of the segment or massive PE, increased susceptibility to a systemic inflammatory response may lead to DIC, compartment syndrome). A separate group should include cases of incorrect arrangement of the apparatus and the insertion of transosseous elements, since, in fact, this is a violation of the Ilizarov segment lengthening technology. If we talk about the choice of the level of conducting basic transosseous elements, then there are 8 levels on the tibia according to the MUOCHO,[47] the division into which is conditional and is used to detail the description of the technology. The bottom line is to use the maximum possible length of bone fragments for the stability of the apparatus supports. In the work of A.V. Popkova et al.[48,49] when testing the rigidity of fixation with various variants of the layout 1 of the support, it was shown that when only 2 crossed spokes were inserted in the plane of the ring, the displacement of the latter was maximum (with an applied load of 5 kg, the displacement was mm). When an additional spoke is inserted 1 cm from the ring, the stiffness increases (with a load of 5 kg, the displacement was 5 mm). Removing the spokes additionally to an even greater distance proportionally increases structural rigidity. The diameter of the ring must also be adequately selected. In cases with a large body weight and a large volume of soft tissues of the segment, it is necessary to resort to duplication of rings, an increase in their number, and additional transosseous elements (according to the authors, in such cases it is necessary to duplicate the basic transosseous element, because, for example, removing it in the case of the development of an infectious process, it will lead to structural instability and displacement of bone fragments). To reduce traumatization and transfixation of all layers of soft tissues (especially near the joints), the duplicating basic transosseous element must be carried out in the same position according to the MUFSO, in which the main one is located. In cases of treatment of a pathology accompanied by a decrease in local bone mineral density (for example, Olier's disease), there is a problem of eruption of the wires through the "soft" bone and subsequent instability of the frame of the device. This problem is solved either by inserting additional transosseous elements (including threaded rods) to increase the contact area, or by intramedullary reinforcement, which removes part of the shear forces from the spokes of the apparatus, or by introducing an autograft into the area of reduced density (preferably with a cortical layer) and conducting already through its transosseous elements.[50] The latter method has an undeniable advantage due to the fact that no additional transosseous element is performed and there is no additional traumatization and transfixation of the soft tissues of the operated segment. The same researcher assessed the distraction regenerate of 67 segments of the lower extremities and noted that even the presence of chondromatous foci at the level of the deformity apex is not a contraindication for osteotomy, since in the process of distraction osteoneogenesis, pathological foci are replaced by bone tissue. From a financial point of view, 2 main directions for Western orthopedists - lengthening with an external fixator and insertion of an intramedullary nail and lengthening over a magnetic nail are almost the same in terms of costs. [51–53]

## Conclusion

The urgency of the problem of correction and lengthening of the limbs (including the lower leg) is growing equivalent to an increase in the population of people, a decrease in the quality of life and an increasing average age of parents. In the Euro-Asian region in the coming decades, the Ilizarov method will remain the predominant method of treating deformities and shortening of the lower leg, which further stimulates the study of the features of its application. Often, to meet the needs of the patient and the orthopedic traumatologist, one has to resort to repeated surgical interventions on the same segment, which is called a multi-stage correction. To reduce the number of stages, correct preoperative planning is necessary, as well as patient management in the postoperative period to reduce the number of complications that can interrupt or delay the completion of any of the stages of correction. The last conclusion once again emphasizes the need for constant contact between the doctor and the patient.

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## Conflicts of interest

The authors declare no conflicts of interest.

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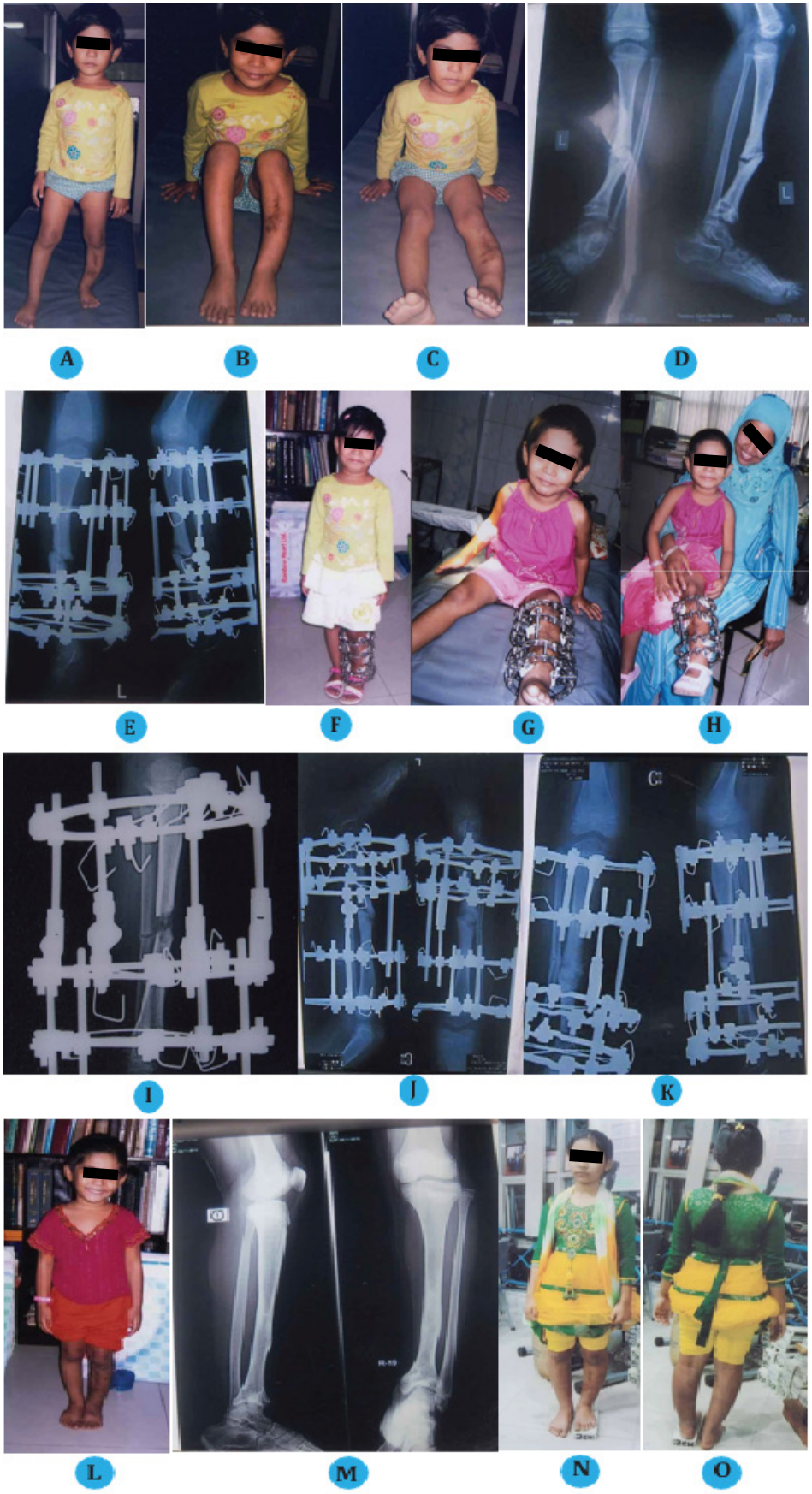


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**Case 1:**

A) 4 years old girl with stiff non -union of left tibia with é procurvatum and varus deformity with LLD

B, C) Visible varus deformity with LLD

D) X-ray of Lt. leg showing stiff nonunion è varium & Procurvatum varus deformity with LLD

E) X-ray after application of Ilizarov apparatus F, G, H) Patient living with the Ilizarov frame

I, J, K) Using hinger and clinical compression-distraction technique to correct the deformity.

L, M) Removal of frame after 4months

N, O, P) At age 11yrs. 3cm LLD has developed on the left side. Q) After application of Ilizarov frame

R, S, T, U) Corticotomy of upper tibia and gradual lengthening done as seen in the X-rays.

V,V1) Full correction of LLD at after 8 months

W, X, Y, Z, Z1) Final follow-up after 13 years (patient age- 17 yrs.) showing full correction

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