BIOMECHANICAL ASPECTS OF SPINAL SAGITTAL BALANCE IN ACHONDROPLASIA PATIENTS DURING ILIZAROV LIMB LENGTHENING

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Objective. To review specific features of spinal sagittal balance in achondroplasia patients at stages of lower limb lengthening using the Ilizarov method.

Material and Methods. Cross-sectional clinical and radiological study was performed in 29 achondroplasia patients prior to lower limb lengthening and at lengthening stages using the Ilizarov method. Parameters of sagittal balance of the spine and pelvis were evaluated radiologically. Clinical evaluation included examination, and assessment of neurological status and pain level.

Results. Clinical manifestations of sagittal imbalance included hypokyphosis of the thoracic spine in 44.8 % of cases and increased lumbar lordosis in 55.2 %. No neurological disorders were diagnosed in patients. Pain scores 2 to 4 were observed in 17.2 % ofcases. After staged lower limb lengthening by 19.8 $\pm$ 3.3 cm, it was revealed that the values of the thoracic kyphosis, lumbar lordosis and the angle of the sacrum tilt improved and approached those of healthy peers. Vertical sagittal alignment measurements correlated with those of thoracic kyphosis. Thoracic kyphosis showed a correlation with lumbar lordosis. Pelvic indices had a moderate correlation with lumbar lordosis.

Conclusion. Biomechanically substantiated transosseous compression-distraction osteosynthesis by Ilizarov technique used for lower limb lengthening in achondroplasia patients improves spinal sagittal balance parameters.

Key Words: achondroplasia, lower limb lengthening, transosseous compression-distraction osteosynthesis by Ilizarov technique, spinal sagittal balance.

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Achondroplasia is a common form of skeletal dysplasia with a rate of 1 case per 15,000 population [1, 2]. This genetic autosomal dominant disease is characterized by a trunk to limb disproportion and spinal anomalies [3–5].

Often, achondroplasia-associated changes in the spine are characterized by spinal canal stenosis and sagittal imbalance [3, 5–8] as well as synostosis disorders, wedge-shaped vertebrae, sacral underdevelopment, and changes in pedicle size [9–11].

Over the last years, there have been conducted active studies of the spinal sagittal balance and its relationship with the pelvis, both in healthy people and in various pathological conditions, primarily for understanding the physiological and pathophysiological aspects of diseases [5, 12, 13]. There are single publications on sagittal balance in achondroplasia patients [1, 5].

Shortening of the limbs in achondroplasia is not just a cosmetic problem, but a gross functional impairment that significantly limits the everyday activities of patients. The Ilizarov method of transosseous compression-distraction osteosynthesis enables lengthening of the lower limbs by 28–30 cm. The question arises: how this affects spine condition?

The purpose of this study was to analyze the spinal sagittal balance features in achondroplasia patients at different stages of Ilizarov lower limb lengthening.

Design: cross-sectional study.

Due to the staged course of treatment, the period of lower limb lengthening takes 4–6 years, on average. At present, we do not have a radiological archive of patients at all stages of treatment. A retrospective analysis takes a prolonged period of time. Also, there is no comparison group due to the lack of another osteosynthesis method enabling comparable lengthening of the limbs.

The study inclusion criteria were as follows: achondroplasia patients at different stages of limb lengthening by the Ilizarov apparatus.

The study exclusion criteria were as follows: patients with other systemic diseases during limb lengthening by the Ilizarov apparatus.

Study setting: the study was performed at the Department of Traumatology and Orthopedics No. 17 of the Russian Ilizarov Scientific Center for Restorative Traumatology and Orthopaedics (Kurgan).

The study was performed in the period between October 2016 and November 2017.

The study was conducted in accordance with the 1964 Helsinki Declaration principles. Patients or their parents provided informed voluntary consent for participating in diagnostic studies and medical interventions and using the obtained data for scientific purposes.

Material and Methods

We performed a clinical X-ray study of 29 achondroplasia patients; of these, 2 patients were examined twice at different treatment stages. Ten patients were examined before treatment; 12 patients were examined at different stages of lower limb lengthening; 9 patients were assessed after completion of lower limb lengthening.

Lower limb lengthening technique. Limb lengthening was performed in stages by mono- or bilocal distraction osteosynthesis, sequentially or crosswise. Single-stage lengthening was performed as follows: the tibia was lengthened 7–8 cm, and the femur was lengthened 8–9 cm. In the case of crosswise lengthening of segments, the tibia was repeatedly lengthened.

X-ray examination. X-ray was used to evaluate the following parameters: the scoliosis Cobb angle, thoracic kyphosis angle (TK at the T4–T12 level), thoracolumbar kyphosis angle (TLK at the T10–L2 level), lumbar lordosis angle (LL1 at the L1–S1 level and LL2 at the L2–S1 level), sagittal vertical axis (SVA), pelvic incidence (PI), pelvic tilt (PT) angle, and sacral slope (SS) angle. X-ray measurements were performed using the Surgimap v2.2.12.1 software.

Clinical examination. Clinical evaluation was based on a physical examination and assessment of the neurological status and pain level (Wong-Baker Faces Pain Rating Scale).

The data were processed with the Microsoft Office Excel (2016) statistical software package. The arithmetic mean (M), deviation of the mean (± m), Pearson correlation coefficient r with evaluation on the Chaddock scale were calculated; the Student's t-test with calculation of the statistical confidence was used to assess the significance of mean differences.

Results

A general examination of the body revealed flattening of thoracic kyphosis in all patients. Increased lordosis of the lumbar spine was detected in 16 (55.2 %) patients, and kyphosis at the thoracolumbar junction level was found in 8 (27.5 %) patients (Table 1).

Chest deformity in the form of uni- or bilateral indrawing of the anterior rib portions was diagnosed in 16 (55.2 %) cases; scoliotic spinal deformity was present in 4 (13.8 %) cases.

An analysis of X-ray films demonstrated wedge-shaped deformity of the vertebral bodies at the thoracolumbar junction level (T12–L1–L2) with the formation of local kyphosis in 8 (27.5 %) patients and sacral hypoplasia in one case (Fig. 1).

The neurological status was without serious pathology. The pain score in five patients ranged from 2 to 4.

Radiological parameters of patients at different stages of treatment are presented in Table 2.

Radiological and clinical outcomes at different stages of treatment are presented in Fig. 2 and 3, respectively. Given the small number of patients, the Pearson correlation coefficient for sagittal balance indicators was calculated at different stages and at the end of lengthening. The sagittal vertical axis SVA had a moderate correlation ($r = 0.5$) with TK. The TK parameter moderately correlated with LL1 ($r = 0.4$). The pelvic parameters PI, PT, and SS had a moderate correlation with LL1 ($r = 0.6$). The value of lower limb lengthening moderately correlated with LL1 ($r = 0.4$), PI ($r = 0.4$), and PT ($r = 0.4$).

Discussion

Achondroplasia is a systemic skeletal disease that is characterized by impaired endochondral osteogenesis, dwarfism, shortening of the limbs (with the trunk length being normal), limb and spine deformities, and macrocephaly.

Thoracic kyphosis is a common (up to 94 % of cases) form of spinal deformity in achondroplasia children under the age of 1 year. As the child grows and begins walking, the kyphosis decreases during the first 10 years, but then gradually progresses and is diagnosed in approximately 11 % of patients [14–16]. Kyphotic spinal deformity and grade 1 scoliosis are detected in 76 % and 17 % of children with achondroplasia, respectively. Since the age of 7 years, 15 % of children have wedge-shaped vertebrae whose degree of deformity varies from 10° to 18° [17]. In our study, kyphotic spinal deformity in the setting of wedge-shaped deformity of the T12, L1, and L2 vertebral bodies was diagnosed in 8 (27.5 %) cases with local angular kyphosis of 13.1° ± 7.2° at the T10–L2 level. Scoliotic deformity before treatment was 1.4° ± 2.3°, on average, with an almost two-fold increase during lower limb lengthening (3.2° ± 3.4°), which was associated with the staged course of osteosynthesis (sequential or crosswise lengthening of lower limb segments). At the end of osteosynthesis, the deformity value almost did not differ from the initial one (1.9° ± 1.7°) and indicated symmetrical lengthening of the limbs. Clinical manifestations of sagittal imbalance in the study patients were thoracic hypokyphosis (44.8 % of cases) and an increase in lumbar lordosis (55.2 % of cases). No neurological disorders were diagnosed in the patients.

After staged lengthening of the lower limbs in the study patients, a change in SVA, an increase in TK and PI, and a decrease in LL1 and LL2 were observed. The other sagittal balance indicators differed at stages of lower limb lengthening and did not significantly differ after lengthening completion.

Due to a small archive of our own measurements of the sagittal balance in achondroplasia patients before treatment (10 people), we conducted an additional comparative analysis with the literature data. There are few publications on this topic. The sagittal spinal balance in achondroplasia patients at the age of 17–36 years compared to that in healthy patients was studied by Hong et al. [5]. Karikari and co-authors [1] investigated sagittal balance parameters in achondroplasia children but at the age of 2.6 years. For comparison, we used the data of healthy people (13.1 ± 2.1 years) from studies by Mac-Thiong et al. and Marty et al. [18, 19] (Table 3).

In achondroplasia patients after lower limb lengthening, TK, LL1, and SS values approach those of healthy peers as the age increases. At the same time, the sagittal balance parameters before treatment are comparable with the results of Hong et al. [5] for achondroplasia patients.

According to V.I. Shevtsov et al. [20], lower limb lengthening was accompanied by an increase in the lumbosacral angle up to normal values in all age groups. Our study confirmed this fact.

To understand the mechanisms of changes in the sagittal balance parameters during lower limb lengthening, let us consider biomechanical models

Table 1

General characterization of achondroplasia patients

Parameter	Value
Age, years	12.6 ± 4.8 (from 5 to 35)
Males : females, n	12 : 17
Height before lengthening, cm	107.0 ± 11.8
Height at examination (n = 21), cm	127.9 ± 13.7
Amount of lower limb lengthening at examination (n = 21), cm	15.9 ± 5.1
Increase in lumbar lordosis (clinically), n (%)	16 (55.2)
Decrease in thoracic kyphosis (clinically), n (%)	13 (44.8)
Kyphosis at the thoracolumbar junction level (clinically), n (%)	8 (27.5)
Chest deformity, n (%)	4 (13.8)
History of pain, n (%)	5 (17.2)

Table 2

X-ray parameters of sagittal balance in achondroplasia patients at different stages of treatment

Parameter	Before treatment (n = 10)	At stages of lengthening (n = 12)	After completion of lengthening (n = 9)
Lower limb lengthening, cm	–	13.0 ± 4.7 (from 5 to 25)	19.8 ± 3.3 (from 12 to 25)
Cobb scoliosis, degree	1.4 ± 2.3 (from 0 to 14.2)	3.2 ± 3.4 (from 0 to 10)	1.9 ± 1.7 (from 0 to 6) p = 0.07
Global sagittal balance, mm	-0.26 ± 21.6 (from -61.6 to 38.5)	35.9 ± 24.4 (from 0 to 116.5)	35.1 ± 29.8 (from -0.5 to 98.1) p = 0.04
Thoracic kyphosis T4–T12, degree	16.6 ± 3.2 (from 4.6 to 21.7)	12.8 ± 4.0 (from 3.4 to 26.0)	22.2 ± 7.7 (from 6.2 to 48.4) p = 0.09
Thoracolumbar kyphosis T10–L2, degree	10.6 ± 4.8 (from 0.8 to 24.1)	9.7 ± 5.1 (from 0.5 to 31.8)	11.7 ± 6.2 (from 0.8 to 27.1) p = 0.07
Lumbar lordosis L1–S1, degree	58.9 ± 9.3 (from 38.9 to 72.3)	51.1 ± 8.8 (from 24.0 to 64.4)	50.4 ± 8.9 (from 31.8 to 66.3) p = 0.10
Lumbar lordosis L2–S1, degree	60.2 ± 8.0 (from 39.1 to 71.5)	50.1 ± 5.7 (from 32.7 to 64.4)	52.2 ± 7.1 (from 39.2 to 69.3). p = 0.10
Pelvic incidence, degree	50.9 ± 6.8 (from 36.2 to 63.9)	50.9 ± 5.6 (from 26.0 to 65.6)	53.2 ± 3.6 (from 44.0 to 58.3) p = 0.04
Pelvic tilt, degree	10.9 ± 5.1 (from 1.6 to 21.4)	8.7 ± 4.3 (from -2.0 to 18.0)	11.0 ± 4.3 (from 1.5 to 20.3) p = 0.09
Lumbosacral angle, degree	42.1 ± 5.8 (from 28.9 to 53.6)	44.8 ± 8.2 (from 25.6 to 62.9)	41.2 ± 6.0 (from 29.0 to 56.8) p = 0.08

The significance of differences between groups before treatment and after completion of lengthening according to the difference (Student) t-test.

Fig. 1
Spondylogram of a 9-year-old female patient B. in a lateral projection at the second stage of lower limb lengthening (total lengthening is 14 cm): wedge-shaped deformity of the L1 and L2 vertebral bodies and thoracolumbar kyphosis of 5.7°

of achondroplasia patients before and after treatment. The sagittal balance of the human body is provided by mutual arrangement of the pelvis, spine, and lower extremities [21]. In this case, the pelvis position is determined by the state and mobility of the femoral heads for maintaining the vertical body posture [22, 23]. The femoral heads form a point through which the axial load is transferred from the upper spinal parts to the lower extremities [22, 24]. Thus, it is the condition of the hip joints that determines the position of the pelvis, spine, and whole body.

The anatomical features of achondroplasia patients include shortening of the limbs, O-shaped curvature of the lower limbs with lateral instability of the knee joints, and flexion contractures of the hips [25]. Limited mobility in the hip joints triggers compensatory mechanisms for correction of sagittal imbalance: the pelvic tilt angle, lumbar lordosis, and thoracic kyphosis change.

A feature of Ilizarov osteosynthesis upon lengthening of the femur at its proximal third level is directional formation of antecurvation deformity with an angle value corresponding to the value of excessive lumbar lordosis [26] (Table 4).

An increase in the limb length and a decrease in hip contractures with an increase in mobility of the femoral heads change pelvic parameters, which, in turn, leads to changes in other sagittal balance parameters due to correlations found in the study (Fig. 4).

Conclusion

Achondroplasia patients are characterized by a decrease in thoracic kyphosis and an increase in the lumbar lordosis, pelvic incidence, and angle of the pelvic tilt and vertical body axis. In the study, significant correlations were found between sagittal balance parameters: vertical sagittal axis and thoracic kyphosis, thoracic kyphosis and lumbar lordosis, as well as lumbar lordosis and pelvic parameters. The value of lower limb lengthening moderately correlated with lumbar lordosis, pelvic incidence, and pelvic tilt.

Ilizarov lower limb lengthening provides mobility of the femoral heads, which leads to changes in pelvic parameters, a decrease in lumbar lordosis, an increase in thoracic kyphosis, and correction of the sagittal body imbalance.

Comparative analysis of the results in study patients at all stages of observation requires further research.

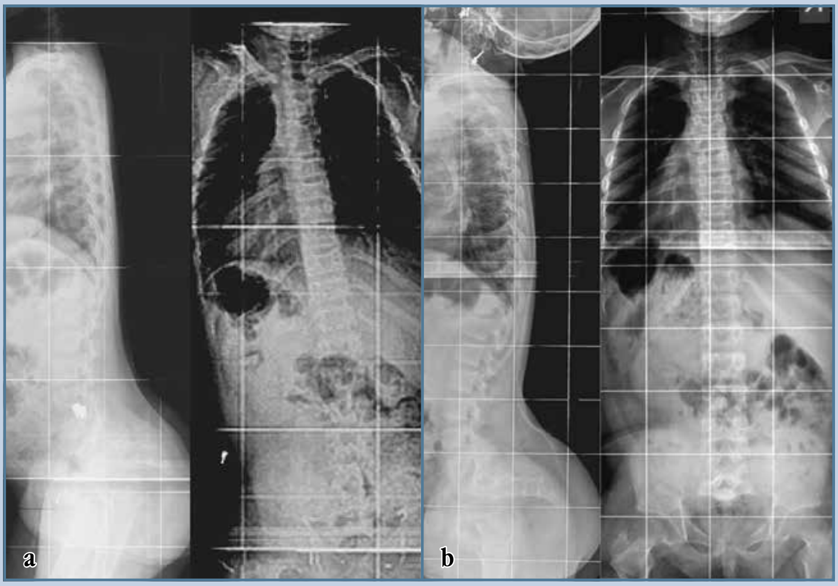

Fig. 2
Spondylograms of a 9-year-old female patient I. at the second stage of lower limb lengthening: **a** – after osteotomy and osteosynthesis with the Ilizarov apparatus; **b** – after completion of staged lengthening and disassembly of the Ilizarov apparatus (total lengthening is 15 cm)

Fig. 3
A 10-year-old male patient I.: **a** – before treatment; **b** – after staged lengthening and disassembly of the Ilizarov apparatus (total lengthening is 12 cm)

Table 3

Comparison of sagittal balance parameters in study patients with literature data

Evaluated parameter	Present study, achondroplasia, before treatment (n = 10)	Present study, achondroplasia, lower limb lengthening (n = 21)	Hong et al. [5], achondroplasia (n = 32)	Hong et al [5], healthy subjects (n = 24)	Mac-Thiong et al. [18], Marty et al. [19], healthy subjects (n = 145)
Age, years	9.5 ± 3.5	14.1 ± 4.8	17–36	17–32	13.1 ± 2.1
Global sagittal balance, mm	-0.26 ± 21.60	35.10 ± 29.80	22.20 ± 10.60	10.30 ± 8.76	–
Thoracic kyphosis T4–T12, degree	16.60 ± 3.20	22.20 ± 7.70	19.52 ± 10.30	33.40 ± 10.92	44.20 ± 10.30
Thoracolumbar kyphosis T10–L2 degree	10.60 ± 4.80	11.70 ± 6.20	10.30 ± 12.42	0.39 ± 5.47	–
Lumbar lordosis L1–S1, degree	58.90 ± 9.30	50.40 ± 8.90	56.12 ± 11.44	38.37 ± 19.67	49.20 ± 12.40
Lumbar lordosis L2–S1, degree	60.20 ± 8.00	52.20 ± 7.10	46.37 ± 14.03	50.96 ± 8.29	–
Pelvic incidence, degree	50.90 ± 6.80	53.20 ± 3.60	43.10 ± 17.47	52.47 ± 13.06	49.30 ± 11.20
Pelvic tilt, degree	10.90 ± 5.10	11.00 ± 4.30	0.42 ± 12.73	13.28 ± 8.59	7.90 ± 7.70
Lumbosacral angle, degree	42.10 ± 5.80	41.20 ± 6.00	44.03 ± 9.46	39.51 ± 7.6	41.40 ± 8.50

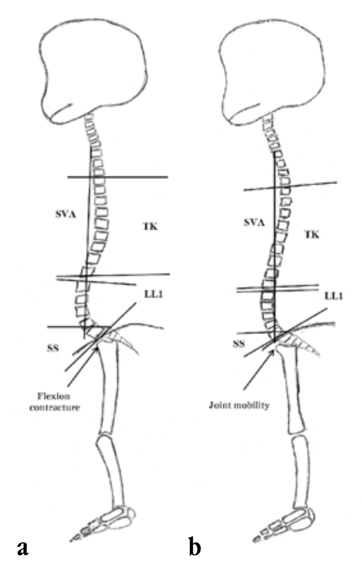

Fig. 4
Biomechanical scheme of the sagittal spinal balance in achondroplasia patients: **a** – before treatment; **b** – after lower limb lengtheningй

Table 4
Biomechanical features of achondroplasia patients before and after treatment

Parameter	Before treatment	After treatment
Length of hips and tibias	Shortening	Lengthening, increased levers
Hip joints	Flexion contractures	Decreased contractures, increased mobility of the femoral heads
Pelvic tilt	Increase	Increase
Lumbosacral angle	Increase	Close to normal value
Lumbar lordosis	Aggravation	Decrease
Thoracic kyphosis	Decrease	Close to normal value
Vertical body axis	Significant deviation	Close to normal value

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