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CLINICAL AND RADIOLOGICAL ASPECTS OF THE SAGITTAL BALANCE OF THE SPINE IN CHILDREN WITH ACHONDROPLASIA

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Background. Changes in the spine with achondroplasia are represented by disorders of synostosis, the presence of wedge-shaped vertebrae, underdevelopment of the sacrum, changes in the size of the roots of the arches, stenosis of the spinal canal, and changes in the sagittal balance.
Aim. To investigate the clinical and radiological features of the sagittal balance of the spine in children with achondroplasia.
Materials and methods. We performed a cross-sectional clinical and radiological study of 16 patients with achondroplasia aged 6–17 years (mean, 9.2 ± 3.3 years). Radiographically, the parameters of the sagittal balance of the spine and pelvis and scoliosis were evaluated. Clinical evaluation included orthopedic and neurological status and back pain syndrome.
Results. The anatomic features of patients with achondroplasia are limb shortening, O-shaped curvature of the lower extremities with lateral instability of the knee joints, and flexural contractures of the hip joints. With restriction of mobility in the hip joints, compensatory mechanisms for correcting sagittal imbalance are triggered: pelvic incline, lumbar lordosis, and thoracic kyphosis change. The clinical manifestations of sagittal imbalance in enrolled children were hypokyphosis of the thoracic spine in 100% and an increase in lumbar lordosis in 56.25% of patients. In 50% of patients, wedge-shaped deformation of vertebral bodies was diagnosed at the level of the thoracolumbar transition with the formation of local kyphosis. Neurological disorders have not been diagnosed in children.
Conclusions. The anatomical features of the lower limbs and hip joints in achondroplasia reflect the biomechanical features of the relationship between the spine, pelvis, and lower limbs, which should be considered when planning for orthopedic and spinal surgery after prediction.

Keywords: achondroplasia; sagittal balance of the spine; parameters of the spine and pelvis.

КЛИНИКО-РЕНТГЕНОЛОГИЧЕСКИЕ АСПЕКТЫ САГИТТАЛЬНОГО БАЛАНСА ПОЗВОНОЧНИКА У ДЕТЕЙ С АХОНДРОПЛАЗИЕЙ

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Обоснование. Изменения позвоночника при ахондроплазии представлены нарушениями синостозирования, наличием клиновидных позвонков, недоразвитием крестца, изменением размеров ножек корней дуг, стенозом позвоночного канала и изменениями сагиттального баланса.
Цель — изучить клинико-рентгенологические особенности сагиттального баланса позвоночника у детей с ахондроплазией.
Материалы и методы. Проведено кросс-секционное клинико-рентгенологическое исследование 16 пациентов с ахондроплазией в возрасте от 6 до 17 лет (средний возраст — 9,2 ± 3,3 года), поступивших для первичного удлинения нижних конечностей. Длительность исследования: с октября 2016 по апрель 2018 г. Рентгенологически оценивали параметры сагиттального баланса позвоночника и таза, сколиоз. Клиническая оценка включала ортопедический и неврологический статус, болевой синдром в спине. Для обработки данных применяли пакет

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статистических программ Microsoft Office Excel (2016): вычисление средней арифметической (*M*) и отклонение среднего (±*m*), коэффициент корреляции Пирсона (*r*) с оценкой по шкале Чеддока.
Результаты. Анатомическими особенностями больных с ахондроплазией являются укорочение конечностей, О-образное искривление нижних конечностей с боковой нестабильностью коленных суставов и сгибательные контрактуры тазобедренных суставов. При ограничении подвижности в тазобедренных суставах запускаются компенсаторные механизмы коррекции сагиттального дисбаланса: изменяются угол наклона таза, поясничный лордоз, грудной кифоз. Клиническими проявлениями сагиттального дисбаланса у исследуемых детей были гипокифоз грудного отдела позвоночника в 100 % случаев и увеличение поясничного лордоза у 56,25 % больных. У 50 % пациентов диагностирована клиновидная деформация тел позвонков на уровне грудопоясничного перехода с формированием локального кифоза. У детей не обнаружено неврологических нарушений. Выраженность болевого синдрома в спине у 5 человек колебалась от 2 до 4 баллов.
Заключение. Для детей с ахондроплазией характерно уменьшение грудного кифоза, увеличение поясничного лордоза, тазового индекса, угла наклона таза и вертикальной оси туловища. Анатомические особенности нижних конечностей и тазобедренных суставов при ахондроплазии отражают биомеханические особенности взаимоотношений позвоночника, таза, нижних конечностей, что необходимо учитывать при планировании ортопедических вмешательств и прогнозировании состояния позвоночника после операций.

Ключевые слова: ахондроплазия; сагиттальный баланс позвоночника; позвоночно-тазовые параметры.

Background

Achondroplasia, a genetic skeletal dysplasia, is characterized by disproportionate short stature along with spinal pathology [1–3]. Spinal changes in achondroplasia are represented by synostosis disorders, wedge-shaped vertebrae, sacral underdevelopment, changes in the sizes of the pedicle radices of arches, spinal stenosis, and abnormal sagittal balance [4–9].

Many publications discuss surgical correction of kyphotic deformities with a wedge-shaped deformity of the vertebral bodies and laminoplasty for spinal stenosis [9–13]. The study of the sagittal balance of the spine and its relationship with the pelvis and lower extremities is essential for understanding of the physiological and pathophysiological aspects of achondroplasia, planning of orthopedic interventions, and predicting the state of the spine after surgery [14–17].

On that account, the present **study aimed** to analyze the clinical and radiological aspects of the sagittal balance of the spine in pediatric patients with achondroplasia.

Methods

Study design: cross-sectional study.

Acceptance criteria

Inclusion criteria: patients under 18 years of age with achondroplasia.

Exclusion criteria: patients over 18 years of age or patients with diseases other than achonodroplasia systemic diseases.

Study period

The study was performed from October 2016 to April 2018.

Research methods

Radiological examination

Radiological assessment parameters included angle of scoliosis (Cobb), angle of thoracic kyphosis (*TK*, at the level of vertebrae Th_4–Th_{12}), angle of thoracolumbar kyphosis (*TLK*, at the level of vertebrae Th_{10}–L_2), angle of lumbar lordosis (LL_1, at the level of vertebrae L_1–S_1, and LL_2, at the level of vertebrae L_2–S_1), sagittal vertical axis (*SVA*), pelvic index (*PI*), pelvic tilt (*PT*), and lumbosacral angle (*SS*). Radiological measurements were performed using the Surgimap v2.2.12.2 program.

Clinical study

Orthopedic and neurological statuses were assessed by using the previously published methods [18], whereas the pain syndrome at the back was measured with Wong-Baker Numerical Rating Scale of Pain [19].

Statistical analysis

Statistical software package Microsoft Office Excel (2016) was used for data processing. The arithmetic mean (*M*), the deviation of the mean (±*m*), and the Pearson correlation coefficient (*r*) with assessment by the Cheddock scale were calculated.

Results

The mean age of patients was 9.2 ± 3.3 years (range 6–17 years). The male to female ratio in patients was 6 : 10.

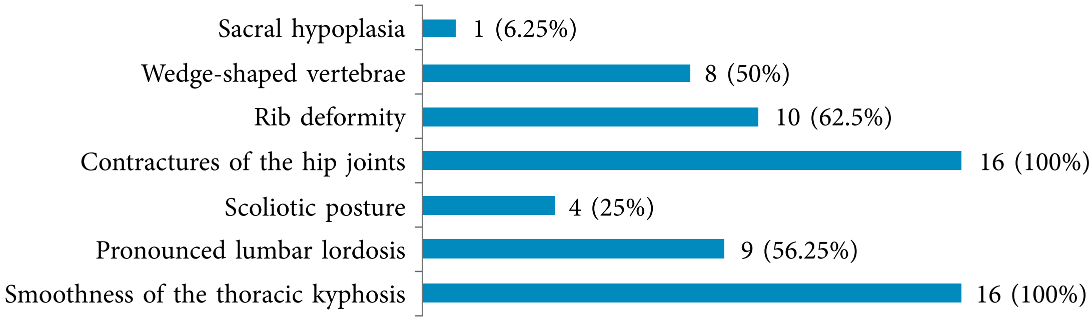


Fig. 1. Diagram of the orthopedic status of patients

During clinical examination of the trunk, smoothness of the thoracic kyphosis was determined in all patients. As shown in Fig.1, increased lordosis of the lumbar spine was detected in 9 patients (56.25%), scoliotic spinal deformity in 4 patients (25%), and deformity of the chest in the form of retraction of the anterior segments of ribs (from 2 to 6) was noted in 10 cases (62.5%).

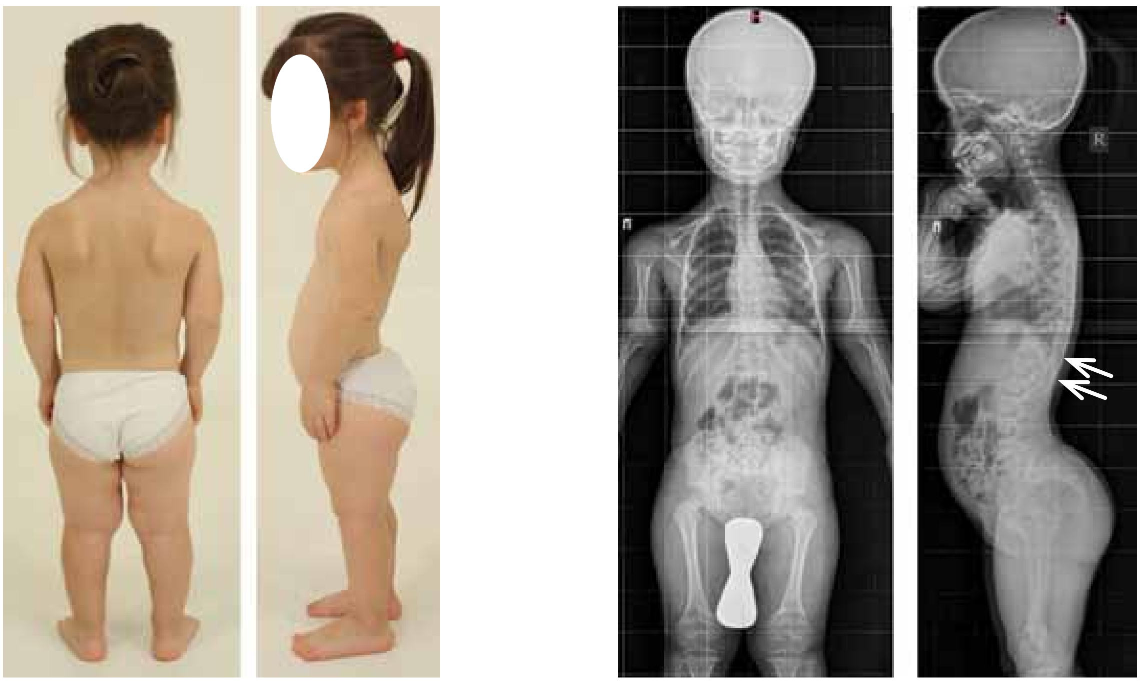
Radiographically, wedge-shaped deformity of the vertebral bodies at the level of the thoracolumbar transition (Th_{12}–L_1–L_2) with formation of local kyphosis was diagnosed in 8 patients (50%) (Fig. 2), and sacral hypoplasia was diagnosed in one patient.

During clinical assessment of the condition of the lower extremities, O-shaped curvature of the lower extremities with lateral instability of the knee joints and flexion contractures of the hip joints were revealed in all children. The range of motion in the right and left hip joints is tabulated in Table 1 below.

Patients did not show any gross violations in their neurological status. Moreover, the severity of the pain syndrome at the back of 5 patients (aged 9–14 years) ranged from 2 to 4 points [19].

Radiological parameters of the sagittal balance are presented in Table 2.

When determining the relationship between the sagittal balance indices, it was revealed that the *SVA* showed a moderate correlation ($r = 0.5$) with *TK*. The index of *TK* was moderately correlated with lumbar lordosis LL_1 ($r = 0.4$). In addition, the pelvic coefficients *PI*, *PT*, and *SS* showed an average correlation force with the values of lumbar lordosis LL_1 ($r = 0.6$).



a *b*

Fig. 2. Photo (*a*) and spondylograms (*b*) of a 7-year-old patient with achondroplasia. The thoracic kyphosis was smoothed, the lumbar lordosis was intensified, and the wedge-shaped deformity of the vertebral bodies Th_{12}, L_1 was evident (*arrows*)

Table 1

The range of motion in the hip joints

Types of movement	Right	Left	Normal indices for children 9/11 years [18]
Flexion (with the leg straightened in the knee joint in the prone position) (°)	130.9 ± 2.6 (125–140)	132.1 ± 3.0 (130–140)	146/38
Extension (°)	22.5 ± 2.6 (15–25)	22.5 ± 2.6 (15–25)	27/25
Abduction (°)	44.6 ± 3.3 (36–50)	44.9 ± 4.1 (35–55)	49/45
Adduction (°)	27.1 ± 5.8 (20–40)	26.8 ± 6.1 (20–40)	28/29
External rotation (°)	42.8 ± 3.8 (35–55)	42.5 ± 3.5 (35–55)	43/42
Internal rotation (°)	51.5 ± 4.0 (45–60)	51.5 ± 3.6 (45–60)	54/48

Table 2

Radiological parameters of the sagittal balance

Evaluation parameters	Results obtained	Healthy children, aged 7.3 ± 1.8 (Mac-Thiong J.M., 2004) [15]
Scoliosis (°), Cobb	1.5 ± 2.3 (0–14.2)	–
SVA (mm)	1.3 ± 20.9 (–61.6–38.5)	–
TK Th_4–Th_{12} (°)	16.2 ± 4.3 (4.1–23.8)	38.3 ± 9.8
TLK Th_{10}–L_2 (°)	16.4 ± 3.2 (4.6–21.7)	–
LL_1, L_1–S_1 (°)	57.5 ± 10.1 (38.9–72.3)	45.6 ± 12.1
LL_2, L_2–S_1 (°)	58.8 ± 8.9 (39.1–71.5)	–
PI (pelvic index) (°)	50.5 ± 6.4 (36.2–63.9)	44.6 ± 10.6
PT (pelvic tilt) (°)	11.2 ± 4.9 (1.6–21.4)	4.3 ± 8.1
SS (lumbosacral angle) (°)	40.7 ± 5.7 (22.9–53.6)	40.3 ± 8.7

Discussion

Achondroplasia is characterized by impaired endochondral osteogenesis, dwarfism, shortening of the extremities with normal body height, deformities of the extremities and spine, and macrocephaly [2–4, 20].

There are limited publications about the aspects of the state of spine in pediatric patients with achondroplasia. Studies on surgical correction of thoracolumbar kyphosis [10, 12, 21–23], decompression in stenosis of the spinal canal [9, 11, 13], and neurological complications [21] are predominant.

In a study conducted by I.O. Karikari et al. (2012), a retrospective analysis of the indices of the sagittal balance of pediatric patients with achondroplasia aged from 1 month to 10 years was conducted. The average age was 2.6 ± 2.1 years [24].

J.-Y. Hong (2011) conducted a study comparing the sagittal balance of the spine in adults with achondroplasia (aged 17–36 years) and healthy patients [8].

In the present study, the spinal–pelvic relationship in pediatric patients with achondroplasia within the age range of 6–17 years (average age was 9.2 ± 3.3 years) was analyzed. No spinal changes were detected in patients who were within this age group.

The data of J.M. Mac-Thiong for healthy children aged 7.3 ± 1.8 years were taken as a reference (2004) (Table 3) [15, 16].

As shown in Table 3, in pediatric patients with achondroplasia, the thoracic kyphosis *TK* increases with age; however, this indicator remains half of that found in healthy peers, regardless of age.

Table 3

Comparative analysis of the indices of sagittal balance

Evaluation parameters	I.O. Karikari et al. (2012) [24], achondroplasia (n = 40)	Present study, achondroplasia (n = 16)	J.M. Mac-Thiong (2004) [15], healthy (n = 35)	J.-Y. Hong (2011) [8], achondroplasia (n = 32)
Age of patients (years)	2.6 ± 2.1	9.2 ± 3.3	7.3 ± 1.8	17–36
SVA (sagittal vertical axis) (mm)	–	1.3 ± 20.9	–	–22.2 ± 10.6
TK, Th_4–Th_{12} (°)	13.26 ± 18	16.2 ± 4.3	38.3 ± 9.8	19.52 ± 10.3
TLK, Th_{10}–L_2 (°)	37.4 ± 15.8	16.4 ± 3.2	–	10.3 ± 12.42
LL_1, L_1–S_1 (°)	58.8 ± 15.9	57.5 ± 10.1	45.6 ± 12.1	56.12 ± 11.44
LL_2, L_2–S_1 (°)	–	57.5 ± 10.1	–	46.37 ± 14.03
PI (pelvic index) (°)	36.4 ± 16.6	50.5 ± 6.4	44.6 ± 10.6	43.1 ± 17.47
PT (pelvic tilt) (°)	6.68 ± 25.5	11.2 ± 4.9	4.3 ± 8.1	0.42 ± 12.73
SS (lumbosacral angle) (°)	36.1 ± 15.2	40.7 ± 5.7	40.3 ± 8.7	44.03 ± 9.46

In agreement to J.-Y. Hong, the index of kyphosis of the thoracolumbar transition *TLK* is 3.5 times lower than in children whose average age is 2.6 ± 2.1 years [8, 24]. Kyphosis of the thoracolumbar spine is a common form of spinal deformity in achondroplasia, with an incidence rate of 94% in children under 1 year of age. Nonetheless, growth and walking can cause a regression in kyphosis during the first 10 years of childhood [10, 17, 23]. When the child begins to walk confidently, the final formation of hypokyphosis of the thoracic region occurs, and the compensatory mechanisms for the correction of the sagittal balance will be triggered.

Starting at the age of 7, 11%–15% of children showed the formation of wedge-shaped deformities of the vertebrae, the degree of deformity ranges from 10° to 18° [12, 25]. In the present study, the wedge-shaped deformity of the vertebral bodies Th_{12}, L_1, and L_2 was diagnosed in eight cases (50%).

In reference to other studies, the lumbar lordosis index *LL* (LL_1 and LL_2), in the patients of our study have exceeded normal values and practically did not change with age. The increase in lumbar lordosis is the main clinical and radiological characteristic of the sagittal balance of patients with achondroplasia.

In the age group studied, an increase in the *PI* in comparison with young children was revealed, as well as its approximation to the values of adult patients with achondroplasia and healthy people of all ages. The *PI* indicates an anatomical relationship between the sacrum and hip joints. The *PI* increases during childhood, becomes unchanged after reaching its final growth, and henceforth determines the remaining vertebral pelvic parameters (*SS*, *PT*, *LL*, and *TK*) [24].

Based on the data obtained, pelvic balance indices *PT* and *SS* in pediatric patients with achondroplasia increase with age, but they remain relatively lower in healthy peers. According to I.O. Karikari et al. (2012), children aged 2.6 years showed ambiguous results where patients with negative and positive *PT* were identified. The researchers attribute these ambiguous results to the patient age and the lack of prospectivity of the study [24].

In the present study, the sagittal vertical axis in adult patients with achondroplasia had large magnitude of values. The numerical parameters of the indicator in pediatric patients with achondroplasia are not presented in the present study.

Scoliotic spinal deformity has been detected in minority pediatric patients with achondroplasia (17%) [25]. In our group of patients, scoliosis was diagnosed in 3 of them with a magnitude of 1.5 ± 2.3°.

Clinical manifestations of sagittal imbalance in the studied patients were hypokifosis of the thoracic spine in 100% of the patients and an increase in lumbar lordosis in 50% of the patients. No neurological disorders were diagnosed in the patients.

The sagittal balance of the human body ensures an appropriate interposition of the pelvis, spine, and lower extremities [26]. The position of the pelvis depends on the condition and mobility of the femoral heads and determines the vertical position of the body [14–16].

Anatomical aspects of the patients with achondroplasia include limb shortening, O-shaped curvature of the lower limbs with lateral instability of the knee joints, and flexion contractures of the hip joints [12, 13, 24]. When the mobility of the hip joints is limited, compensatory mechanisms for the correction of sagittal imbalance will be triggered, including the pelvic tilt, lumbar lordosis, and thoracic kyphosis change. In the group of children studied, there was a decrease in thoracic kyphosis, an increase in lumbar lordosis, and changes in pelvic parameters, which naturally reflect the biomechanical relationship of the spine, pelvis, and lower extremities in pediatric patients with achondroplasia.

Conclusion

Pediatric patients with achondroplasia are characterized by a decrease in thoracic kyphosis, an increase in lumbar lordosis, a pelvic index, a pelvic tilt, and a vertical axis of the body. It is clinically manifested by smoothed thoracic kyphosis of the thoracic spine and pronounced lumbar lordosis. In the present study, significant correlations between the indicators of sagittal balance were established, including the vertical sagittal axis and thoracic kyphosis, thoracic kyphosis and lumbar lordosis, lumbar lordosis, and pelvic parameters.

The anatomical aspects of the lower limbs and hip joints in achondroplasia indicate a biomechanical relationship between the spine, pelvis, and lower limbs, which must be considered when planning orthopedic surgeries and predicting the state of the spine after interventions.

Additional information

Source of funding. The study was performed without financial support from sponsors.

Conflict of interest. The authors declare no obvious and potential conflicts of interest related to the publication of this article.

Ethical review. The examinations were conducted in accordance with the requirements of the 1964 Helsinki Declaration. Patients or their parents gave informed voluntary consent to diagnostic manipulations and medical interventions and use the data obtained for scientific purposes.

Contribution of the authors

O.G. Prudnikova was engaged in research design, collection, and processing of materials, analysis of the data obtained, and writing the text.

A.M. Aranovich created the concept of the study and performed the analysis of the data obtained.

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