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Journal Article



Assessment of the respiratory system in children with congenital scoliosis by impulse oscillometry and computed tomography (preliminary results)

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BACKGROUND: Segmentation disorder of the vertebral body lateral surfaces and rib synostosis are severe variants of congenital pathology of the spine and thorax. They lead to the development of thoracic insufficiency syndrome and are manifested by the inability of the thorax to provide normal respiratory mechanics.

AIM: This study presents the preliminary results of functional and radiological (CT-morphometric) methods of lung examinations in patients with congenital thoracic spine scoliosis with impaired segmentation of the lateral surfaces of the vertebral bodies and unilateral rib synostosis.

MATERIALS AND METHODS: This design is represented by a small clinical series. This study is a prospective study of 10 patients aged 3 to 7 years with congenital spinal deformity, with impaired segmentation of the lateral surfaces of vertebral bodies and unilateral rib synostosis. This paper presents the preliminary results of the pulmonary function assessment by pulse oscillometry and CT morphometry in a 3D reconstruction of multispiral computer tomography (MSCT) of the thorax.

RESULTS: The study of respiratory function using pulse oscillometry revealed no respiratory impairment in seven observations, also reflected in the CT morphometry results. According to the Institute of Medicine (IOM), three children with detected ventilatory abnormalities showed the following parameters with the most significant changes: total respiratory impedance, resonance frequency, and frequency dependence of the resistive component. In all patients, the morphometric indexes of the lung scoring revealed during 3D modeling of the lung were completely consistent with the results of the lung function study by the IOM method.

CONCLUSIONS: Further study of the problem of respiratory function assessment in children with congenital scoliosis seems promising in diagnostic terms and for evaluating effective surgical treatment.

Keywords: segmentation disorder; congenital scoliosis; thoracic insufficiency syndrome; pulse oscillometry.

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Научная статья

Оценка состояния респираторной системы у детей с врожденным сколиозом методом импульсной осциллометрии и компьютерной томографии (предварительные результаты)

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Обоснование. Нарушение сегментации боковых поверхностей тел позвонков и реберный синостоз — один из самых тяжелых вариантов врожденной патологии позвоночника и грудной клетки, которые приводят к развитию синдрома торакальной недостаточности, проявляющегося в неспособности грудной клетки обеспечивать нормальную дыхательную механику.

Цель — представить предварительные результаты функциональных и лучевых (КТ-морфометрических) исследований легких у пациентов с врожденным сколиозом грудного отдела позвоночника при нарушении сегментации боковых отделов тел позвонков и одностороннем синостозе ребер.

Материалы и методы. Дизайн исследования — малая клиническая серия. В проспективное исследование включены данные импульсной осциллометрии и КТ-морфометрии при 3D-реконструкции данных мультиспиральной компьютерной томографии органов грудной клетки 10 пациентов в возрасте от 3 до 7 лет с врожденным сколиозом грудного отдела позвоночника при одностороннем нарушении сегментации боковых поверхностей тел позвонков и одностороннем синостозе ребер.

Результаты. При исследовании дыхательной функции с применением импульсной осциллометрии в 7 клинических случаях не выявлено дыхательных нарушений. У 3 детей с вентиляционными нарушениями по данным импульсной осциллометрии наиболее значимые изменения касались параметров общего дыхательного импеданса, а также резонансной частоты и частотной зависимости резистивного компонента. У всех пациентов морфометрические показатели оценки легких, выявленные по 3D-модели легкого, соответствовали результатам исследования легочной функции методом импульсной осциллометрии.

Заключение. Дальнейшее изучение проблемы оценки функции дыхания у детей с врожденным сколиозом представляется перспективным как в диагностическом плане, так и для определения эффективности хирургического лечения.

Ключевые слова: нарушение сегментации; врожденный сколиоз; синдром торакальной недостаточности; импульсная осциллометрия.

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BACKGROUND

One of the most severe types of congenital malformation of the spine associated with an abnormality of the chest is a unilateral segmentation disorder of the lateral surfaces of the vertebral body (non-segmented rod) in combination with unilateral synostosis of the ribs, which leads to the formation and rapid progression of the deformity [1, 2].

The rapid progression of congenital spinal curvature is a mainstay in the development of thoracic insufficiency syndrome. This concept was developed by Campbell and is characterized by the chest's inability to ensure the physiological biomechanics of respiration and growth of lung tissue [3, 4].

During the growth and development of children with this defect, severe and rigid scoliotic deformity has already formed at an early age, and the quality and life expectancy are significantly reduced, mainly due to respiratory failure [5].

According to Tong et al., at present, approximately 4,000 children in the USA have thoracic insufficiency syndrome [6], whereas accurate data on its prevalence in the Russian Federation are not currently available [7].

The treatment of patients with this variant of congenital deformity is a complex and unresolved problem associated with the need to correct severe and rapidly progressive curvature of the spinal column and the need to consider the potential growth of the spine, chest, and cardiopulmonary system [8]. The rapid progression of spinal deformity, reaching over 8°–10° per year with this defect in a natural course, induces the formation of extremely severe curvatures already in period 2 of childhood, which confirms the ineffectiveness of conservative treatment and the expediency of using surgical methods for correcting congenital curvature in the first years of life [9, 10].

An important aspect that does not have definite solution is the assessment of the respiratory system condition at the start of surgical treatment and at its stages during growth [11–13]. In several clinical cases, the result of the analysis of the external respiration function can be used as a determining factor in the selection of approach for the treatment of children with congenital scoliosis with a unilateral non-segmented rod and costal synostosis [14, 15]. To evaluate external respiration in younger patients, resource-intensive technical support is required [16–18]; therefore, measurements are usually not performed, which complicates the interpretation of the efficacy of treatment results [19–21].

This study aimed to present preliminary results of functional and radiometric (computed tomography [CT] morphometric) methods of examining the lungs in patients with congenital scoliosis of the thoracic spine with unilateral impairment of the segmentation of the lateral surfaces of the vertebral bodies and costal synostosis.

MATERIALS AND METHODS

This study presents a small clinical series. The prospective study included the results of a preoperative examination of 10 patients with congenital scoliosis of the thoracic spine with unilateral impairment of segmentation of the lateral surfaces of the vertebral bodies and costal synostosis, who were examined and treated at the Department of Spinal Pathology and Neurosurgery of H. Turner National Medical Research Center for Children's Orthopedics and Trauma Surgery.

The inclusion criteria, in addition to the anatomical variant of the defect, were the absence of neurological disorders and age 3–7 years at the time of the study.

The exclusion criteria were spinal deformity caused by other types of congenital abnormalities, severe concomitant somatic pathology of internal organs, including malformations of the bronchopulmonary system, and refusal to participate in the study.

The preoperative examination, in addition to clinical and laboratory methods, included the use of radiation diagnostic methods, namely, standard radiography and multispiral computed tomography (MSCT) of the spine and chest organs (Fig. 1) and assessment of the external respiration function by impulse oscillometry (IOM) based on K.A. Rauhfus Children's City Multidisciplinary Clinical Center of High Medical Technology (St. Petersburg).

MSCT was performed using a Brilliance 64 (PHILIPS) device. The study protocol consisted of performing a topogram, a native study of the spine and chest organs to assess the condition of the bone tissue and lung parenchyma. In all patients, special pediatric protocols with positioning tools, filters, and programs with a decrease in the scanning field and tube voltage (70 kV or less) were used. Technical parameters, pediatric protocols, and dose-reduction technologies were selected based on the child's weight. MSCT was performed with the patient lying on his/her back in the craniocaudal direction from the level of the upper border of the clavicle through both costophrenic angles. Scanning was performed at the spiral mode parameters of quality reference 110 mAs, KV/effective mAs; rotation time 120/110/0.5 s; detector collimation, 1.5 mm; slice thickness, 1.0 mm; pitch factor, 0.3–1; increment, 2 mm.

To assess the type of the spinal and rib malformations and to calculate the hemithorax volume and densitometric characteristics of the lung parenchyma, post-processing programs were used, namely, three-dimensional multiplanar reconstruction, projections of maximum intensity, and surface-shaded image (Fig. 1, *a–d*). During the study, tomograph software was used, including the method of tissue segmentation with the differentiation of the lung parenchyma from the ribs, soft tissues of the mediastinum, and blood vessels, followed by mapping of the zones obtained and identifying

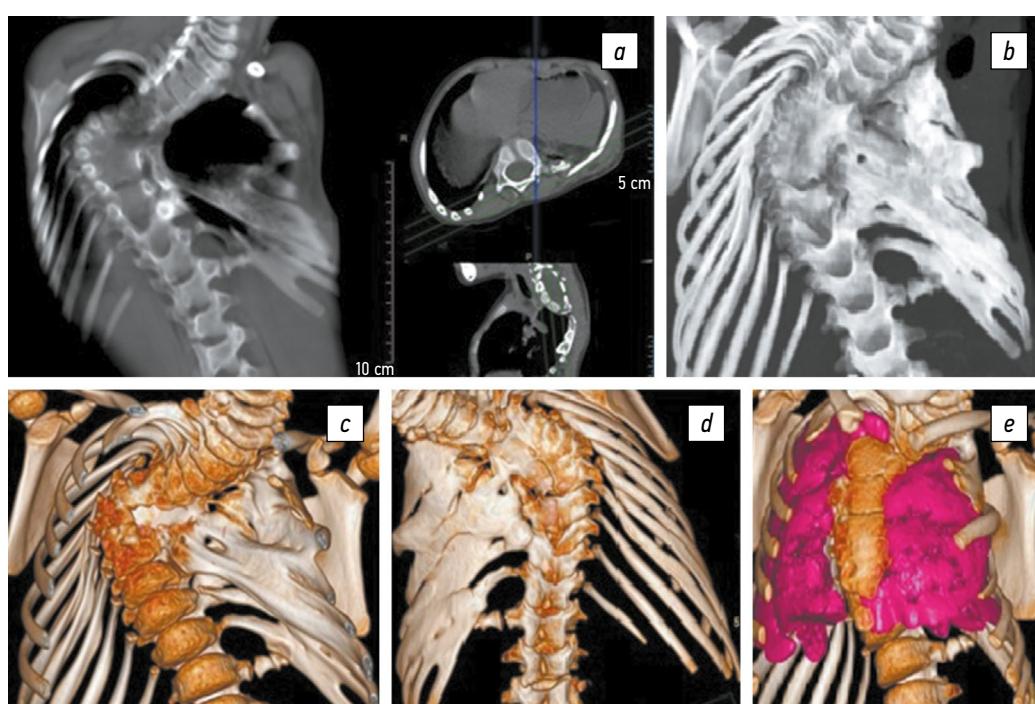


Fig. 1. Patient S., 5 years old. Multispiral computed tomography of the spine and chest organs in target structure modes. Multiplanar skew coronal reconstruction (*a*) and volumetric images show projections of maximum intensity (*b*). Surface-shaded image, front view (*c*), posterior view (*d*) of segmentation disorders of the lateral surfaces of the vertebral bodies with subtotal left-sided costal synostosis (posterolateral sections); reconstruction of the chest organs with isolated visualization of the bone tissue and lung parenchyma using tissue segmentation techniques (*e*)

the lung tissue volume (Fig. 1, *e*). The localization and extent of areas with reduced pneumatization were assessed, and changes in the densitometric characteristics of density less than -500 units HU were chosen as criteria. In areas with visually uniform density, the average lung area indicators were determined on the Hounsfield scale. In areas with uneven density, measurements were performed at several points, and average values were calculated. Lung volume (CT volumetry) was calculated using CT scanner software. "Thick" slices were used; for this, images were reconstructed with a slice thickness of 5 mm, with the reconstruction increment of 5 mm. The calculation principle was based on a modified Simpson formula.

A virtual model of the bronchopulmonary system was obtained using DICOM digital data processing (Fig. 2).

The identified changes in the lung tissue with hypoventilation zones were localized along the side of the lesion, in accordance with the prevalence (segment and lobe) and extent relative to the scoliosis apex in the craniocaudal direction and along the periphery (anteroposterior).

To assess the functional state of the lungs, all patients underwent IOM, which represents a variant of the forced oscillation technique and allows for a passive measurement of lung mechanics. The IOM study was performed on the Master Screen IOS unit (Viasys Healthcare, Germany). The principle of IOM is based on the superposition of sound waves on normal respiratory sounds, which leads to changes in the airway flow and pressure [22–24]. IOM was assessed based on respiratory impedance (total) at an oscillation frequency of 5 Hz (Zrs5) and resistive (frictional) component

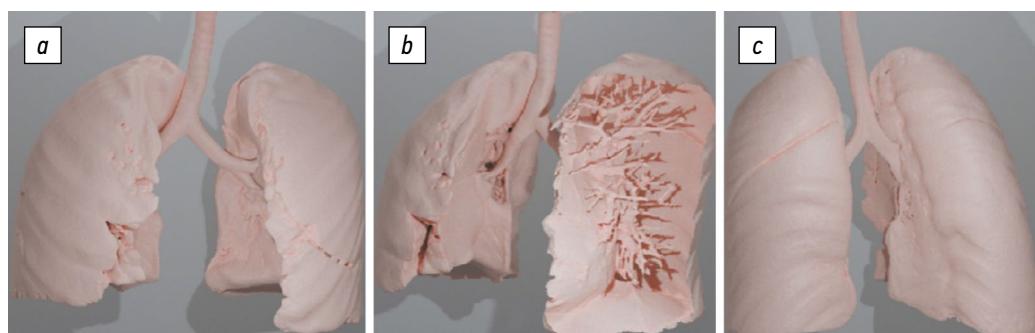


Fig. 2. Calculation of the lung tissue volume in Patient A (5 years old) by constructing a virtual 3D model of the bronchopulmonary system based on inspiratory multispiral computed tomography findings. Virtual model of the lungs: *a*, front view; *b*, lateral view; *c*, posterior view

of the respiratory impedance at an oscillation frequency of 5 and 20 Hz (Rrs5 and RRrs20). Deviations from the norm of IOM parameters were determined by changes in the basic indicators Rrs5 and Xrs5 [22, 24].

RESULTS

The scoliotic curve in the study patients ranged from 30° to 90°. In the sagittal plane, most patients had hypokyphosis, and some had pathological thoracic lordosis up to 15°.

In the study of the pulmonary function through impulse oscillography, ventilation disorders were not detected in seven cases. Despite the normal indicators of impulse resistance, the children were found to have impaired airway patency according to the speed parameters of the air flow. This is the so-called lower shunt, in which a part of the impulse oscillations (resistance) is lost due to lung tissue hyperinflation.

In three pediatric patients with ventilation disorders, according to IOM findings, the parameters of the general respiratory impedance changed the most, namely, the reactive and resistive components (Xrs5 and Rrs5, respectively) and frequency dependence of the resistive component (FR Rrs5-20) (Table 1). These impulse resistance parameters indicate not only the patency of the airways but also the elastic properties of the respiratory structures involved in the respiratory cycle. An increase in resistive component indicates a patency disorder at different levels of the tracheobronchial tree, a decrease in the parameter of the elastic part of the reactive component of the Xrs5 impedance, and indicates a change in the elastic properties of the respiratory structures.

In three patients with a decrease in morphometric parameters identified during the construction of a 3D model of the lungs and CT volumetry, the most significant changes in functional parameters were detected by IOM (Tables 1 and 2).

According to the literature, normally, a physiological asymmetry in the lung development is registered in children,

Table 1. Results of impulse oscillography

Patient	Age	Resistive component Zrs (Rrs5). Normal* from 0.78–1.36	Reactive component Zrs (Xrs5). Normal* –0.61 to –0.32	Frequency dependence of the resistive component (FD Rs5-20). Normal* from 0.25–0.57
1	3 years	1.32	–0.42	0.26
2	7 years	1.05	–0.35	0.28
3	3 years	0.72	–0.34	0.27
4**	5 years	1.48	–0.28	0.21
5**	4 years	1.51	–0.25	0.19
6	3 years	0.9	–0.34	0.26
7	4 years	0.81	–0.36	0.31
8	3 years	0.89	–0.31	0.28
9**	5 years	1.45	–0.27	0.18
10	7 years	0.9	–0.39	0.27

* Range of normal parameter values according to Elida Duenas-Meza et al. [44]. ** Patients with the most altered parameters.

Table 2. Results of digital radiographs and multispiral computed tomography

Patient	Age	Location of the non-segmented rod	Scoliotic deformity according to Cobb, deg.	Side of the main curve, D/S	Right lung volume, cm ³	Left lung volume, cm ³	Total lung volume, cm ³
1	3 years	Th ₅ –Th ₈	48	D	392.0	317.8	709.8
2	7 years	Th ₄ –Th ₇	61	S	324.3	365.6	689.9
3	3 years	Th ₇ –Th ₁₀	38	D	295.7	247.8	543.5
4*	5 years	Th ₁ –Th ₅	34	D	485.3	351.7	837.0
5*	4 years	Th ₆ –Th ₁₂	52	D	717.5	561.3	1278.8
6	3 years	Th ₃ –Th ₉	69	D	230.7	183.7	414.4
7	4 years	Th ₂ –Th ₉	44	S	334.4	392	726.4
8	3 years	Th ₄ –Th ₉	42	S	240.1	286.7	526.8
9*	5 years	Th ₄ –Th ₉	90	D	434.5	190.7	595.2
10	7 years	Th ₉ –Th ₁₁	30	D	583.4	486.4	1069.8

* Patients with the most pronounced changes identified during the study.

Table 3. Comparison of lung volumetric parameters according to multispiral computed tomography in the studied patients with indicators of the physiological norm [27]

Total lung volume, cm ³ (M; min–max)		Right lung volume, cm ³ (M; min–max)		Left lung volume, cm ³ (M; min–max)	
norm	studied patients	norm	studied patients	norm	studied patients
689.0; 313.0–2180.0	739.8; 414.4–1278.0	379.0; 192.0–1218.0	403.8; 230.7–717.5	310.0; 106.0–962.0	338.4; 183.7–561.3

which reaches 5% [25–27]. The difference in the lung volume in pediatric patients with segmentation disorder of the lateral surfaces of the vertebral body and costal synostosis ranged from 12% to 56.2%. A tendency for a decrease in the lung on the segmentation disorder side was also noted in these patients (Table 2).

When comparing the data obtained and the normal values based on literary sources, a tendency to a significant decrease in the possible maximum lung volume was noted, whereas the average value of lung volume exceeded the normal value, which may indicate the early activation of compensatory mechanisms. Data obtained are presented in Table 3.

DISCUSSION

Respiratory failure is the most serious complication of congenital spinal deformity because of impaired segmentation of the lateral surfaces of the vertebral bodies associated with unilateral synostosis of the ribs [28, 29].

Thoracic insufficiency may be associated with impaired development of lung function from early childhood, and this condition worsens as the child grows [5, 30, 31]. If left untreated, children are at increased risk of early death because of cardiopulmonary failure [32–34]. There are currently no clear quantitative or qualitative criteria for the diagnosis of thoracic insufficiency syndrome [5, 35].

Until recently, no noninvasive and reliable methods are available for assessing the respiratory function in younger children with congenital malformations [36–38]. The low compliance of children has long been an obstacle to the widespread use of functional tests [24]. One of the possible ways to bypass the restrictions was the use of drug sedation; however, due to the possible effect on the respiratory center and changes in the tone of the smooth muscle tissue of the respiratory tract, the study results were inaccurate; therefore, this method was not widely used [6, 22]. Our IOM method is devoid of the disadvantages described above.

Given the small number of cases, we cannot conclusively state the association of the respiratory disorders with the child's age and magnitude of the main curve of deformity. Clinically, patients generally did not actively complain about the respiratory dysfunction, which was due to the compensatory capabilities of the body; however, in early childhood,

a tendency to a progressive deterioration in the external respiratory function can be detected.

In the literature, patients with severe deformities of the spine and chest at an early age live with near-normal motor activity [39–41]; however, with growth spurt, characterized by an increase in body weight, respiratory failure inevitably develops [42, 43].

In seven clinical cases, patients with congenital spinal deformity associated with impaired segmentation of the lateral surfaces of the vertebral bodies and unilateral synostosis of the ribs did not have ventilation disorders. Three pediatric patients had abnormal changes in the general respiratory impedance, resonant frequency, and frequency dependence of the resistive component.

The decrease in the morphometric parameters of the lungs in three patients, which was revealed during the lung 3D model construction, corresponded to the results in the lung function study using the IOM method. A difference in lung volume from 12% to 56.2% was detected in our patients. Moreover, a tendency to an increase in the average value of the lung volume exceeding the normal values can be noted, which may indicate the early activation of compensatory mechanisms.

To date, the choice of treatment is based on clinical and radiological data; however, the need for a functional assessment of the respiratory system condition is clear.

Limitations of significance of the study results. The study is preliminary in nature. Statistical and factor analyses were not performed because of the small number of clinical cases included in the study. An additional restriction criterion is the selected age range.

CONCLUSION

IOM is a fast, simple, and noninvasive method for assessing the function of external respiration during spontaneous breathing in younger patients.

IOM and morphometric assessment of the lungs using a 3D-CT model provides new objective data on the respiratory system condition, which were previously inaccessible for pediatric patients with congenital scoliosis.

The presented methods are promising for both diagnosing and analyzing the efficiency of surgical treatment of congenital scoliosis in preschool children when assessing changes in lung functions.

ADDITIONAL INFORMATION

Funding. The study had no external funding.

Conflict of interest. The authors declare no conflict of interest.

Ethical considerations. The study was approved by the ethics committee of H. Turner National Medical Research Center for Children's Orthopedics and Trauma Surgery of the Ministry of Health of Russia (Protocol No. 20-3, November 20, 2020).

Written consent was obtained from the patient representatives for the processing and publication of personal data.

Author contributions. S.V. Vissarionov formulated the aims and developed the study design and performed surgical treatment of the patients. M.S. Asadulaev wrote all sections of the article, collected and analyzed the data, performed analysis of the literature, and conducted staged and final editing of the article

text. E.A. Orlova performed pulmonological examination of patients and staged editing of the article text. V.G. Toriya collected and analyzed X-ray examination data, performed staged editing of the text of the article, created 3D models of the lungs, and calculated the lung tissue volume. K.A. Kartavenko performed staged editing of the text of the article and collected the data. T.S. Rybinskikh performed staged editing of the article text and translated the abstract and information about the authors into the English language. T.V. Murashko described the MSCT results and performed staged editing of the article text. M.A. Khardikov prepared the list of references and performed staged editing of the article text. D.N. Kokushin performed staged editing of the article text.

All authors made a significant contribution to the study and preparation of the article and have read and approved the final version before its publication.

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