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Original Study Article

Assessment of the efficacy of treatment for children with congenital scoliosis with unsegmented bar and rib synostosis

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BACKGROUND: Treatment of children with congenital deformity with unsegmented rod and rib synostosis is an important and topical problem to date. Topical publications present the results of surgical correction efficacy and analysis of treatment complications. The extremely important aspect of treatment efficacy assessment regarding changes in the function of external respiration is still topical.

AIM: This study aimed to analyze the treatment results of children with congenital scoliosis and unilateral segmentation disorder of the lateral surfaces of the vertebral bodies and rib synostosis.

MATERIALS AND METHODS: This is a retrospective monocenter cohort study of the treatment outcomes of 30 patients aged 1–14 years. In the preoperative period, external respiration was evaluated by pulse oscillometry, multi-slice computed tomography, digital X-ray imaging of the craniopelvis in two projections. All patients underwent expanding thoracoplasty with osteotomy of the rib synostosis and fixation with a rib-costal or rib-vertebral distractor. Control studies were performed every 6 months after the surgical intervention. The average follow-up period was 2 years. Nonparametric analysis was applied to estimate the obtained data.

RESULTS: The median (Me) age at the start of treatment was 6 years (interquartile range, 4.25; IQR hereafter). The Me scoliosis before treatment was 74° (IQR, 22.75). The Me scoliosis correction after the first stage of treatment was 16° (IQR, 11) and the second correction achieved 6° (IQR, 13). The Me kyphosis was 15° (IQR, 32), the first intervention improved kyphosis by 4° (IQR, 16), and the second by 6° (IQR, 11).

Complications were represented by the destabilization of the metal construct in six cases, and trophic disorders of soft tissues were noted in four. The assessment of external respiratory function using IOM demonstrates reliable improvement of resistive component, reactive component, and frequency dependence of the resistive component ($p < 0,01$).

CONCLUSIONS: The assessment of the external respiratory function in young children and analysis of reference values may allow their use as absolute indications for surgical treatment in children with congenital scoliosis with unsegmented bar and rib synostosis.

Keywords: children; congenital malformation; surgical treatment; congenital scoliosis; segmentation disorder; rib synostosis; unsegmented bar; treatment results; pulse oscillometry.

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Оригинальное исследование

Оценка эффективности лечения детей с врожденным сколиозом при несегментированном стержне и синостозе ребер

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Обоснование. Лечение детей с врожденной деформацией при несегментированном стержне и синостозе ребер до настоящего времени остается важной и актуальной задачей. В тематических публикациях приведены результаты хирургической коррекции, проанализированы осложнения лечения. Оценка эффективности лечения с позиции изменения функции внешнего дыхания все еще не потеряла своего значения.

Цель — анализ результатов лечения детей с врожденным сколиозом при одностороннем нарушении сегментации боковых поверхностей тел позвонков и реберном синостозе.

Материалы и методы. Проведено ретроспективное моноцентровое когортное исследование, в которое вошли 30 пациентов в возрасте от года до 14 лет. В предоперационном периоде оценивали функцию внешнего дыхания методом импульсной осциллометрии, выполняли мультиспиральную компьютерную томографию, цифровую рентгенографию череп – таз в двух проекциях. Всем пациентам проведена расширяющая торакопластика с остеотомией реберного синостоза и фиксацией реберно-реберным или реберно-позвоночным дистрактором. Контрольные исследования осуществляли через каждые 6 мес. после оперативного вмешательства. Средний срок наблюдения составил 2 года. Для оценки полученных данных применяли метод непараметрического анализа.

Результаты. Медиана возраста на момент начала лечения составила 6 лет, межквартильный интервал равнялся 4,25 (IQR). Медиана сколиоза до лечения — 74°, IQR — 22,75. Величина коррекции сколиоза после первого этапа лечения — 16° (IQR — 11), после второй коррекции — 6° (IQR — 13). Медиана кифоза — 15° (IQR — 32), первое вмешательство позволило корригировать кифоз на 4° (IQR — 16), второе — на 6° (IQR — 11).

Осложнения представлены дестабилизацией металлоконструкции в 6 случаях, трофические нарушения мягких тканей отмечены в 4 наблюдениях. По результатам оценки функции внешнего дыхания с применением импульсной осциллометрии установлено достоверное улучшение показателей резистивного компонента, реактивного компонента и частотной зависимости резистивного компонента ($p < 0,01$).

Заключение. Результаты оценки функции внешнего дыхания у детей младшей возрастной группы и анализ референтных значений можно использовать в качестве абсолютных показаний к хирургическому лечению у детей с врожденным сколиозом при несегментированном стержне и синостозе ребер.

Ключевые слова: дети; врожденный порок; хирургическое лечение; врожденный сколиоз; нарушение сегментации; реберный синостоз; несегментированный стержень; результаты лечения; импульсная осциллометрия.

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BACKGROUND

Currently, no accurate information is presented on the prevalence of spinal malformations in the general population [1]. In principle, this indicator should be determined based on the proportion of spinal malformations, such as scoliosis or general orthopedic pathology, that is, 2–3‰ and 0.5–1‰, respectively [2–4].

The surgical treatment of pediatric patients with congenital scoliosis has a rich history based on the experience of both Russian [5–7] and international [2, 8, 9] researchers. The choice of surgical intervention depends on the type of vertebral anomaly, magnitude of the deformity arc, and age of the child [3, 10]. However, the issue of the surgical treatment of children with congenital scoliosis, unilateral impairment of the segmentation of the lateral surfaces of the vertebral bodies, and costal synostosis remains relevant [11–13]. The patients analyzed constitute a relatively small group [14, 15]. However, congenital scoliosis represents one of the most severe types of congenital deformities [10, 12, 16]. Such patients have a rigid and rapidly progressive spinal curvature [12, 17], reaching 10° annually [12], which results in a deformity exceeding 100°–130° by the termination of bone growth [1, 9, 10]. A distinctive characteristic of these patients is a maldevelopment of lung tissue and breathing biomechanics, which was described by Campbell as thoracic insufficiency syndrome (TIS) [12].

Several researchers believe that the management approach in such patients should be primarily determined by the increasing influence of the pathological process on lung development [12, 18, 19]. At present, the failure of conservative treatment of this pathology is indisputable [10, 20, 21].

The treatment of patients with congenital deformity is aimed at preventing its rapid progression [16, 22, 23], preserving the possibility of potential growth of the spine and lung tissue [24, 25], and improving the quality of life [13]. To date, the treatment approach is based on the provisions of the consensus statement in 2015 in relation to early-onset scoliosis [26].

International [24, 27] and Russian [5, 11, 28] studies have described the experience of treating such patients (expanding thoracoplasty) using hardware in various combinations (costocostal capture, costovertebral distractor, and system resting on the pelvic bones) [10, 29, 30]. Like any technique, initially, this surgical method was actively used in clinical practice for congenital scoliosis [11, 15] and other spinal deformities [23, 31, 32]. However, later, the only indication for the use of this type of hardware was clearly formulated, i.e., TIS [12, 17, 26]. This treatment method showed maximum efficacy in early childhood, when there is active growth and development of the lung tissue [10, 22]. Nevertheless, the efficiency of the surgical treatment results with respect to external respiration function in young children and the development of absolute criteria for choosing a treatment method remain unclear [33–35].

This study aimed to analyze the treatment results of children with congenital scoliosis, unilateral impairment of the segmentation of the lateral surfaces of the vertebral bodies, and costal synostosis.

MATERIALS AND METHODS

A single-center cohort study was conducted, which included a retrospective analysis of the results of surgical treatment of 30 children with congenital scoliosis of the thoracic spine and unilateral impairment of the segmentation of the lateral surfaces of the vertebral bodies and costal synostosis and a prospective study of external respiration function using the impulse oscillometry. All patients received staged surgical treatment at the Department of Spinal Pathology and Neurosurgery of the H.I. Turner National Medical Research Center for Children's Orthopedics and Trauma Surgery in the period from 2015 to 2021. The average follow-up period for 2 (1.5–6) years

The inclusion criteria were as follows: congenital deformity of the spine caused by impaired segmentation of the lateral surfaces of the vertebral bodies, transverse processes in combination with unilateral synostosis of the ribs or marked abnormalities, a congenital defect in the thoracic and thoracolumbar spine; absence of neurological disorders, and age of 1–14 years at the time of surgical treatment.

The exclusion criteria were as follows: spinal deformity caused by other variants of impaired development, severe concomitant somatic pathology of internal organs (including bronchopulmonary malformations), and refusal of the patient or his/her representative from surgical treatment and participation in the study.

The study analyzed case histories, radiation imaging methods (multispiral computed tomography and digital radiography), and magnetic resonance imaging of 30 patients with congenital scoliosis of the thoracic spine, impaired segmentation of the lateral surfaces of the vertebral bodies, and unilateral synostosis of the ribs. There were 19 girls and 11 boys. The median age at the start of surgical treatment was 6 years, and the interquartile interval (IQR) was 4.25 (minimum, 12 months; maximum, 13 years). The patients were distributed by age based on N.P. Gundobin's classification (Fig. 1).

All patients underwent a comprehensive clinical and radiological examination before the surgery, after the surgery, and at follow-up. After the surgery, examinations were performed every 6 months. The variant of the developmental anomalies, localization and length of the blocked vertebrae, and number of ribs involved in synostosis were specified using digital radiography of the spine in two mutually

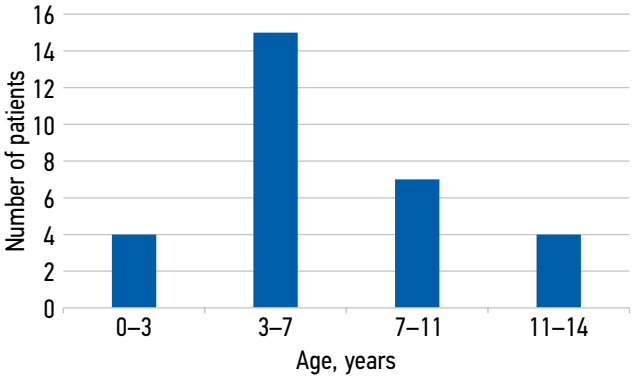

Fig. 1. Histogram of the distribution of patients by age groups at the start of surgical treatment

perpendicular projections. The Cobb method was used to measure the magnitude of the scoliotic, kyphotic, or lordotic components of the deformity before and after the surgery. Multislice computed tomography could exclude bone intracanal pathology, assess the volume of lung tissue before and after the surgery, and plan the level of installation, length of implants, and the degree of correctness of their placement. Magnetic resonance imaging was used to rule out malformations of the spinal cord and spinal canal.

Statistical analysis was performed using the Wolfram Mathematica 11.0 program. The normality of distribution was tested using the Shapiro–Wilk test. For all parameters, the level of two-sided significance was $p > 0.05$, which indicates the impossibility of applying the normal distribution criteria. Median values and quartile intervals were calculated. The statistical significance of differences was assessed using the Wilcoxon signed-rank test.

All patients underwent expanding thoracoplasty to correct the shape of the chest and increase its volume using bifocal costocostal grips and costovertebral distractors. A total of 69 surgical interventions were performed. Staged surgical treatment was performed within 6 months to 1.5 years from the initial corrective surgery. The number and timing of surgical interventions depended on the magnitude of the deformity and rate of progression along with the child's growth. The layout of the hardware was chosen depending on the location of the rib anomalies. In the case of costal synostosis located in the upper and middle thoracic regions and the presence of fully formed supporting ribs in the lower thoracic region, the rib–rib arrangement was used, whereas in costal synostosis located in the lower thoracic region, a variant of the rib–spine distractor was used.

Technique of surgical intervention. With the patient in the lateral position, a hockey stick-shaped incision was made around the angle of the scapula (Fig. 2). Then, the synostotic areas of the ribs were isolated in layers with the formation of a full-thickness flap of soft tissues for the subsequent closure of the hardware. Using a curved raspatory and wet cotton swabs, the periosteum was detached in the area with subsequent installation of the rib grips. Careful skeletization was required to preserve the integrity of the parietal pleura. In this study, a grip on two ribs was used. Costal synostosis osteotomy was performed, and the number of osteotomies depended on the costal block length. In the variant of the costovertebral hardware, skeletization of the dorsal structures of the vertebrae on the side of the nonsegmented nail was additionally performed along the line of the spinous processes below the zone of the nonsegmented nail. After visual and X-ray control, the site for placing pedicle screws was determined. Then, channels for pedicle screws were formed in the vertebrae. In the X-ray control, the correctness of the position of the supporting elements was assessed, and the distracting device installation was started. After the rib grip installation (during the placement of the costovertebral hardware), a channel was bluntly formed

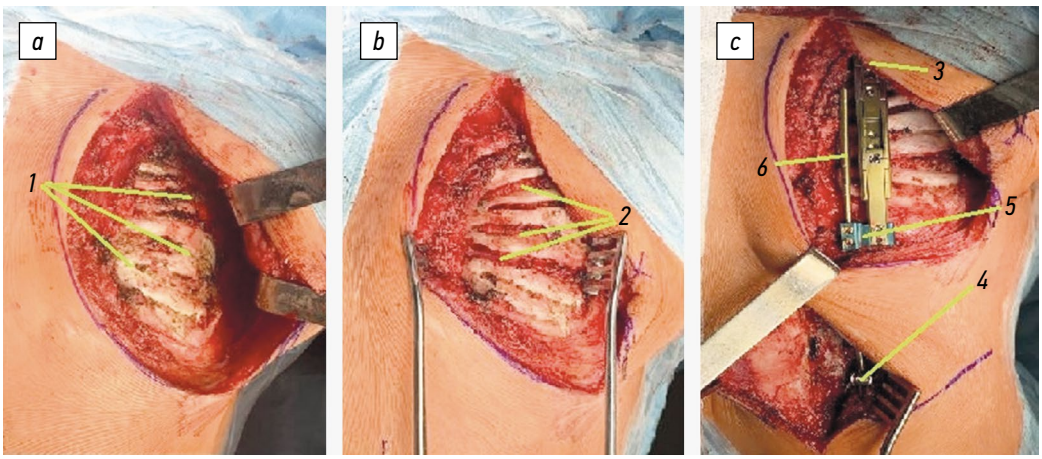

Fig. 2. Synostosis of the ribs (*a*). Appearance of the ribs after osteotomy of the synostosis of the ribs (*b*). Surgical intervention site after endoprosthesis replacement of the hardware and correction of the deformity (*c*): *1*, zones of synostosis of the ribs; *2*, osteotomy site; *3*, area of the rib capture installation; *4*, area of pedicle screw installation; *5*, rib system connector; *6*, length of the rod required to perform staged surgical interventions

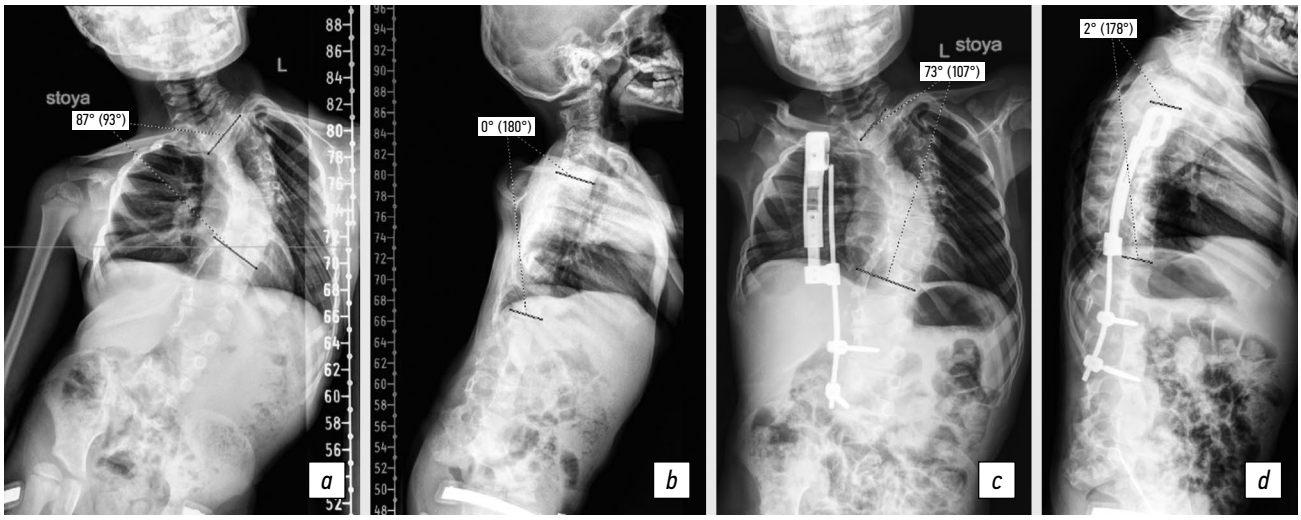

Fig. 3. Panoramic radiograph of the spine in frontal and lateral projections before and after the surgical treatment. Patient, 6 years old: *a, b,* scoliosis before the surgery (87°), lordosis of the thoracic spine (0°); *c, d,* scoliosis after the surgery (73°), kyphosis (2°)

to pass the nail to the site of pedicle screw installation. Distraction was performed along the hardware to correct indirectly the spinal deformity and increase the hemithoracic volume. The amount of correction and correct position of the supporting elements were determined by intraoperative radiography. The surgical intervention was completed by Redon drainage and layered closure of the wound.

The fixation length depended on the severity of the developmental anomaly, length of the nonsegmented nail, and number of ribs in the synostosis. The patients were verticalized on day 3 after the surgery. In younger and middle-age children, a brace was used in the postoperative period to facilitate adaptation. In this study, the effect of the orthosis on deformity correction was not evaluated.

In 25 cases, the deformity was localized in the thoracic region, whereas in the remaining five patients, the deformity was located in the thoracolumbar region.

As in primary surgical correction, indications for staged surgical treatment were severe congenital deformity of the spine and/or rapid progression of curvature (>10° per year), TIS progression, pathological kyphosis in the thoracic spine, and pathological lordosis (Fig. 3). In the case of correction loss and hardware destabilization, staged surgical treatment aimed at restoring the hardware stability was also performed.

The spinal deformity apex was localized at the level of the vertebrae from Th_{III} to Th_{X} and corresponded to the middle spinal motion segment involved in the nonsegmented nail. The median number of vertebrae in a nonsegmented nail was 4 (IQR 3). Costal synostosis included fusion of four ribs on average (minimum, 3; maximum, 12). An equal number of patients had left-sided and right-sided localizations of curvature arcs.

The average value of scoliotic deformity in the study patients was 74° (IQR 22.75). The patients with scoliotic deformity had hypokyphosis reaching an average of 15° (IQR 32°), whereas some patients had pathological lordosis of the thoracic spine up to 20°.

A total of 69 surgical interventions were performed, with an average of 2.3 staged surgeries. In 24 cases, a costocostal distractor was used, and in 45 cases, a costovertebral distractor was applied.

RESULTS

The amount of scoliotic deformity correction after stage 1 of treatment was 16° (IQR 11°). The median magnitude of scoliosis after the initial surgical intervention was 60° (IQR 27.25°). Stage 2 corrected the scoliosis by 6° (IQR 13°). For scoliotic deformity, the *p*-value was 0.0145528, which indicated statistically significant differences between scoliotic deformity before and after the surgery. During the treatment, the deformity did not progress.

The median kyphosis was 15° (IQR 32°). After stage 1, the kyphosis was corrected by 4° (IQR 16°). The median kyphosis after stage 1 of surgery was 20° (IQR 20.5°). After stage 2 of the surgical intervention, the kyphosis correction reached 6° (IQR 11°). The *p*-value for kyphosis was 0.679892, which indicates that the surgical treatment did not affect significantly the magnitude of kyphosis, but a positive trend was found in the sagittal profile improvement.

According to the analysis of CT volumetry (Fig. 4), the median total lung tissue volume before surgery was 860.6 cm^3 (IQR 415.2) cm^3. After the surgery, the median total lung volume reached 951.3 (IQR 441.3) cm^3. After 6 months, the lung volume on the side of costal synostosis increased by 13% and that on the contralateral side by 4%. Thus, the lung volume tended to increase to a greater extent on the side of the costal synostosis, but it did not reach a statistically significant level. The *p*-values for the lung volume values

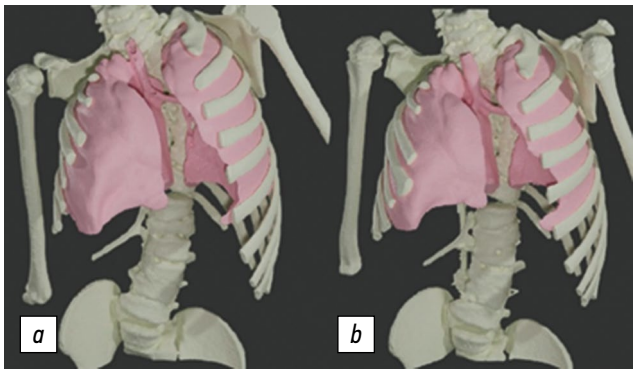

Fig. 4. CT volumetry. Patient, 6 years old: *a,* the lungs before the surgery; *b,* appearance 6 months after the surgery: a change in the spatial position of the shoulder girdle, an increase in the hemithoracic volume on the concave side, and an increase in the right lung by 72.8 cm^3 (22.6%) and the left lung by 62.3 cm^3 (20.7%). The total lung volume 6 months after the surgery increased by 21.7% (135.1 cm^3)

were 0.204973 on the synostotic side and 0.35883 on the contralateral side.

According to the correlation analysis of lung volumes on the side of the costal synostosis and the contralateral side before the surgery, the *p*-value was 0.880099, whereas according to the analysis of volumes after the surgery, it was 0.925588, which indicates a high dependence of volumes. The lung volume on the concave side is always less than that on the convex side. The increase in the lung size after the surgery occurred approximately equally (Fig. 5).

By analyzing the functional results of impulse oscillometry, a significant difference was revealed in the parameters before and after the surgery ($p < 0.01$) (Fig. 6). The resistive component in patients before the surgery was 1.28 (IQR 0.503), whereas after the surgery, the resistive component decreased to 1.0 (IQR 0.4). The reactive component before the surgery was –0.27 (IQR 0.14), whereas after

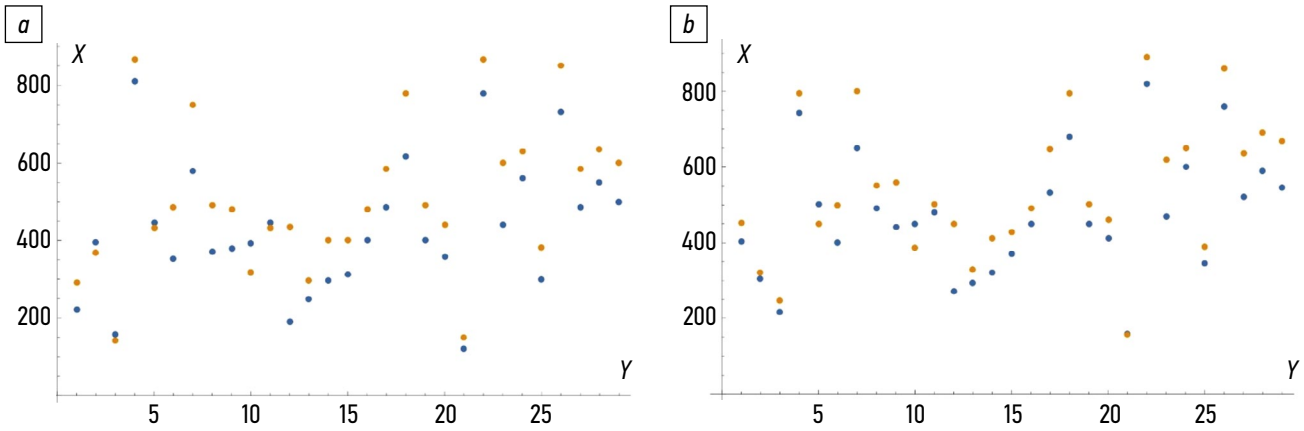

Fig. 5. Correlation of the volumes of the right and left lungs before and after the surgery: *a,* volumes before the surgery; *b,* volumes after the surgery. *X* is the volume cm^3, *Y* is the serial number of the patient, yellow indicates the healthy side, and blue indicates the volume on the costal synostosis side. Before the surgery, the lung volume on the concave side was less than that on the convex side

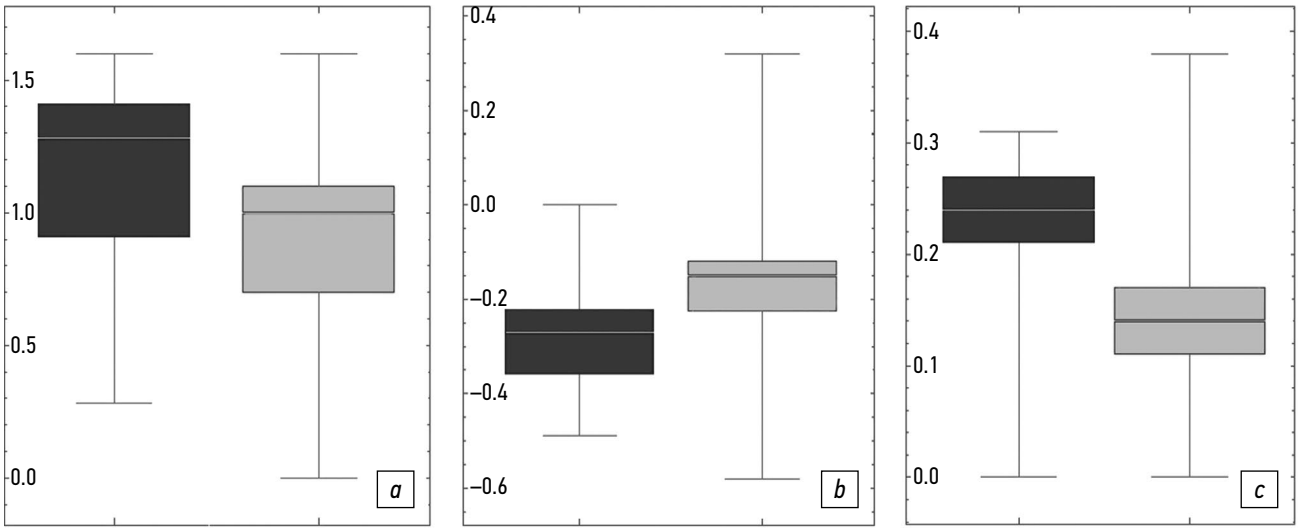

Fig. 6. Distribution of impulse oscillometry parameters in box-plot format before and after the surgery is indicated in gray and light gray, respectively: *a,* resistive component; *b,* reactive component; *c,* frequency dependence of the resistive component

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the surgery, the indicators improved to –0.15 (IQR 0.108). The frequency dependence of the resistive component before the surgery was 0.24 (IQR 0.06); after the surgery, the median of the indicator was 0.14 (IQR 0.06). Thus, in the future, the assessment of the external respiration function, including younger children, will provide reference data on the functional state of the lungs both before the surgery and at its stages.

The results revealed that changes in lung volume during the follow-up does not provide reliable data in contrast to impulse oscillometry, whose indicators improve conclusively and significantly and can be used as criteria for evaluating surgical treatment.

Complications of surgical treatment included surgical hardware destabilization in six cases 1–6 months after the surgery, accompanied by the loss of the achieved correction of the deformity, which necessitated repeated surgical intervention to restore system integrity. Generally, destabilization was preceded by a gross disruption of the orthopedic regimen. Trophic disorders of soft tissues were noted in four patients after the second correction and subsequent correction and were controlled by conservative treatment.

DISCUSSION

The amount of scoliotic deformity correction after stage 1 of treatment was 16° (IQR 11°). Stage 2 enabled the correction of scoliosis by 6° (IQR 13°). For scoliotic deformity, the *p*-value was 0.0145528, indicating a statistically significant difference between scoliotic deformity before and after the surgery. These data are comparable with the results of both Russian [6] and international [35] authors. The decrease in the efficiency of subsequent interventions is also consistent with the indicators presented in the literature.

In addition, the absence of statistically significant results of the correction of the kyphotic component of the deformity does not contradict scientific data. The correction of the sagittal component is not typical for the systems analyzed.

The complications that occurred in the present study are comparable with those of other authors and are due to the large magnitude of the curvature at an early age and noncompliance with the orthopedic regimen due to the pronounced motor activity of the patients.

In several studies, the severity of TIS is determined by the number of ribs included in the synostosis [12, 24, 35], which can be due to the progressive deterioration caused by the three-plane deformity of the chest.

Volumetry using multislice computed tomography can supplement the diagnostic map in the preoperative period and at stages of surgery. However, the results of the analysis of preoperative treatment data and at its stages revealed no significant difference in the indicators. Moreover, a relationship was established between lung volumes on the side of costal synostosis and healthy side both before and after the surgery. Before the surgery, the lung volume on the concave side was less than that on the convex one, taking into account the physiological difference in size between the right and left lungs. The lung size after the surgery on the convex and concave sides increased in approximately equal extent.

The analysis of respiratory function indicators provides previously inaccessible data in patients in the first decade of life. The decrease in the parameters analyzed indicates the normalization of the external respiration function after stage 1 of the surgical intervention. To develop clear criteria for staged surgical treatment, further study is necessary to determine the reference values of the parameters of impulse oscillometry.

Study limitations. Given the small number of cases and the nonnormal distribution of data, nonparametric analysis methods were used in the study. Another study limitation is the wide range in patient age.

CONCLUSION

The preliminary results of the staged surgical interventions demonstrate the possibility of preventing the progression of chest deformity and creating conditions for lung development. In the assessment of the functional state of the lungs, impulse oscillometry revealed a significant difference in indicators, which suggests improvement in the respiratory system.

Staged surgical interventions in young patients should be performed only if impaired external respiration function is confirmed by impulse oscillometry. The rapid progression and severity of the deformity without changes in external respiration function could indicate the need for the early correction of spinal deformity. Further analysis of the reference values reveals absolute indications of pediatric patients with congenital scoliosis, nonsegmented nail, and costal synostosis for surgical treatment.

CT volumetry is advisable for a dynamic assessment of the condition and accumulation of data to determine reference boundaries of the norm and pathology.

Further research will help formulate absolute criteria for choosing a staged treatment, which will reduce the frequency of surgical interventions and the risks of complications.

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 local ethics committee of the H.I. Turner National Medical Research Center for Children's Orthopedics and Trauma Surgery of the Ministry of Health of Russia (Protocol No. 20-3 dated 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, developed the design of the study, and performed surgical treatment of the patients. *M.S. Asadulaev* wrote all sections of the article, collected and analyzed the data, and analyzed the literature. *E.A. Orlova* performed pulmonological examination of all the patients included in the study and conducted staged editing of the article text. *P.A. Ivanova* collected and analyzed the retrospective data and translated the abstract into English. *A.S. Shabunin* performed statistical processing of data. *T.V. Murashko* analyzed X-ray data and performed staged editing of the article text. *M.A. Khardikov* performed staged editing of the article text and formatted the references. *V.G. Toriya* collected and analyzed the radiation study data. *T.S. Rybinskikh* performed staged editing of the article text and translated the text into English. *K.N. Rodionova* performed statistical processing of the findings. *D.N. Kokushin* performed staged editing of the article text and collected the data.

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