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Spinal osteotomy for children with congenital scoliosis with unilateral unsegmented bar: Preliminary results

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BACKGROUND: Segmentation disorder of the vertebral body lateral surfaces develops progressive spinal deformity. Surgical interventions in different variants are the only effective way of treatment. Thus, this study reviews the use of corrective vertebrectomy in patients with congenital scoliosis with impaired segmentation of the vertebral body lateral surfaces.

AIM: To evaluate the surgical treatment outcomes in children with congenital scoliosis with impaired segmentation of the vertebral body lateral surfaces.

MATERIALS AND METHODS: A single-center retrospective study following the Department of spine surgery and neurosurgery from 2014 to 2020 included 33 patients with 17 girls and 16 boys. The age range was 36–211 months. All patients underwent a one-stage corrective wedge vertebrectomy. Statistical processing was performed by comparing the reliability of differences in distributions using the Wilcoxon *t*-criterion.

RESULTS: The median Cobb preoperative scoliotic deformity was 31° (interquartile interval [IQR] = 30.5). The median preoperative lordotic deformity was 29° Cobb (IQR = 29.5). The correction magnitude of the scoliotic deformity component was 64% (median value after intervention: 5° according to Cobb, IQR = 14.5). The correction magnitude of pathological thoracic spine lordosis was 42% (median value after intervention: 17° according to Cobb, IQR = 14.5). The obtained results were statistically significant ($p < 0.05$).

CONCLUSIONS: Corrective wedge vertebrectomy is an effective method for surgical treatment of children with congenital spinal deformity with impaired segmentation of the vertebral body lateral surfaces. This treatment method achieves an average of 64% correction of scoliotic deformity and 42% correction of pathological lordosis.

Keywords: congenital scoliosis; unsegmented bar; children; surgical treatment; vertebrectomy; spine.

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Остеотомия позвоночника в лечении детей с врожденным сколиозом при нарушении сегментации боковых поверхностей тел позвонков (предварительные результаты)

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Обоснование. Нарушение сегментации боковых поверхностей тел позвонков приводит к развитию прогрессирующей деформации позвоночника. Единственным эффективным способом лечения является хирургический в различных вариантах. В работе рассмотрен метод корригирующей вертебротомии, использованный при лечении пациентов с врожденным сколиозом и нарушениями сегментации боковых поверхностей тел позвонков.

Цель — оценить результаты хирургического лечения детей с врожденным сколиозом на фоне нарушения сегментации боковых поверхностей тел позвонков с применением корригирующей вертебротомии.

Материалы и методы. Моноцентровое ретроспективное исследование на базе клиники патологии позвоночника и нейрохирургии НМИЦ детской травматологии и ортопедии им. Г.И. Турнера за период с сентября 2014 по сентябрь 2020 г. В исследование включены 33 пациента: 17 девочек и 16 мальчиков. Возрастной диапазон — от 36 до 211 мес. Всем пациентам выполняли оперативное вмешательство в объеме одномоментной корригирующей клиновидной вертебротомии. Статистическую обработку материала проводили путем сравнения достоверности различий распределений с помощью *t*-критерия Уилкоксона.

Результаты. Медианное значение величины сколиотической деформации по Cobb до операции — 31°, межквартельный интервал (IQR) — 30,5. Медианное значение величины лордотической деформации до хирургического лечения — 29° по Cobb, IQR — 29,5. Величина коррекции сколиотического компонента деформации — 64 % (медианное значение после вмешательства — 5° по Cobb, IQR — 14,5). Величина коррекции патологического лордоза грудного отдела позвоночника — 42 % (медианное значение после вмешательства — 17° по Cobb, IQR — 14,5). Полученные результаты были статистически достоверными ($p < 0,05$).

Заключение. Корригирующая клиновидная вертебротомия является эффективным методом хирургического лечения детей с врожденной деформацией позвоночника при нарушении сегментации боковых поверхностей тел позвонков. Данный метод позволяет достигнуть в среднем 64 % коррекции сколиотической деформации и 42 % коррекции патологического лордоза.

Ключевые слова: врожденный сколиоз; нарушение сегментации; дети; хирургическое лечение; вертебротомия; позвоночник.

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ЭКО•ВЕКТОР

BACKGROUND

Unilateral blockage of the lateral surfaces of the vertebral bodies is a congenital abnormality of spinal development, which is distinguished by rapid deformity progression [1, 2]. Partial blockage of the anterior surfaces of the vertebral bodies resulted in pathological kyphosis, and impaired segmentation of the posterior and lateral parts of the vertebrae leads to pathological lordoscoliosis [3, 4]. An unsegmented bar is considered in case of impairment of the segmentation of the lateral surfaces of the vertebral bodies, which represents the undifferentiated lateral surfaces of two or more vertebral bodies (Fig. 1).

Anomaly development is possibly caused by impaired embryonic development in the first trimester of pregnancy [1]. The anomaly is caused by a unilateral disruption of the segmentation of the primary vertebra, represented by the mesenchyme, resulting in the formation of an unsegmented bar connecting two or more adjacent vertebrae, while the opposite side, on which the growth zones and part of the intervertebral disc are preserved, develops normally [5, 6]. In this case, the deformity is caused by the multidirectional growth of the vertebral bodies on the concave and convex sides. The synostosis of the ribs that exists in this case often leads to the asymmetric development of the chest and aggravates the deformity [4, 7].

The exact incidence of congenital spinal deformity, associated with the unilateral impairment of the segmentation of the lateral surfaces of the vertebral bodies in the Russian Federation, is still unknown [7–9].

Chest deformity in combination with the abnormal development of the ribs in the form of multiple synostoses on the side of the unsegmented bar causes progressive respiratory failure, which, according to some authors, is the most significant factor in reducing the quality and life expectancy of patients with congenital scoliosis [3, 6, 10]. Moreover, the objective assessment of respiratory function in younger children has not been fully investigated [7, 8].

Conservative methods of treating patients with congenital spine deformities and impaired segmentation of the lateral surfaces of the vertebral bodies are ineffective [1, 4, 7].

Naturally, the rapid progression of spinal deformity up to 8°–10° annually in this defect type leads to the formation of severe and rigid curvatures at an early age [1] and necessitates surgical treatment [4, 9].

One of the treatment options of patients with impaired segmentation of the lateral surfaces of the vertebral bodies is surgery to destroy the growth zones and intervertebral discs on the side opposite to the unsegmented bar at age 18 months [3]. This "preventive" surgery helps avoid the progression of spinal deformity and, ultimately, reduce the risk of neurological complications during the final corrective surgery in adolescence [3]. However, this surgical intervention disrupts the growth and development of the thoracic spine, which further leads to the disruption of lung development [7, 9].

Fig. 1. Schematic representation of impaired segmentation of the lateral surfaces of vertebral bodies

To address the impaired development of the chest, performing staged surgery using distractible costo-costal, costo-vertebral, and costo-pelvic structures was recommended; consequently, according to several authors, it is possible to slow the progression rate of spinal deformity and improve the quality of life by increasing the space for lung growth and development [8, 11, 12]. However, with these interventions, the incidence of destabilization of the hardware and revision surgeries is high [9].

Generally, single-staged corrective multilevel osteotomies of the spine at the apex of the deformity are the final stage of surgical treatment; they are performed after the bone growth has ended [12, 13]. However, in recent years, with the improvement of spinal implants and anesthesia, this treatment method can be also applied in young children.

The surgical treatment of pediatric patients with impaired segmentation of the lateral surfaces of the vertebral bodies is a complex and urgent issue that does not have a clear solution [9].

This study presents the outcomes of surgical treatment of 33 pediatric patients with congenital spine malformation associated with impaired segmentation of the lateral surfaces of the vertebral bodies and the methods and results of surgical treatment according to the literature.

This study aimed to evaluate the outcomes of surgical treatment of pediatric patients with congenital scoliosis associated with impaired segmentation of the lateral surfaces of the vertebral bodies using corrective vertebrectomy.

MATERIALS AND METHODS

This study was conducted as a retrospective single-center cohort study. This study analyzed the surgical outcomes of patients with congenital scoliosis associated with impaired segmentation of the lateral surfaces of the vertebral bodies, which underwent surgery to correct the curvature through one-stage corrective vertebrectomy in the Department of Spinal Pathology and Neurosurgery from September 2014 to January 2020. The average follow-up period was 3 (range, 1.5–6) years.

The impaired segmentation of the lateral surfaces of the vertebral bodies (unsegmented bar) is considered a congenital spinal deformity caused by a bone block of the lateral surfaces of the vertebral bodies, transverse processes, and abnormal development of the posterior bone structures in the same level.

The inclusion criteria were as follows: congenital deformity of the spine caused by impaired segmentation of the lateral surfaces of the vertebral bodies, an unsegmented bar located in the thoracic spine, absence of neurological disorders, surgical intervention through one-stage corrective wedge osteotomy (Fig. 2) followed by posterior instrumental fixation; and patients' age >2 years and <18 years at the time of intervention.

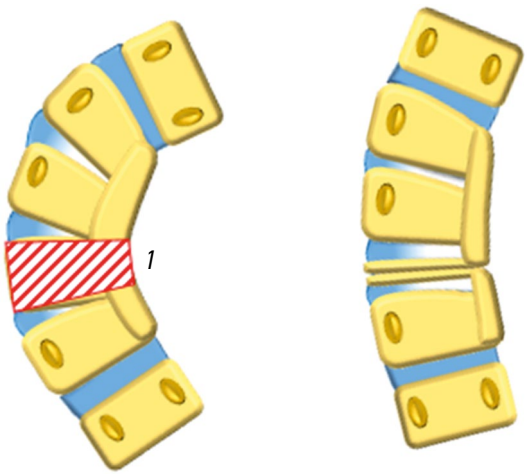

Fig. 2. Scheme of a one-stage corrective wedge osteotomy at the top of the unsegmented bar. *1,* area of vertebrectomy

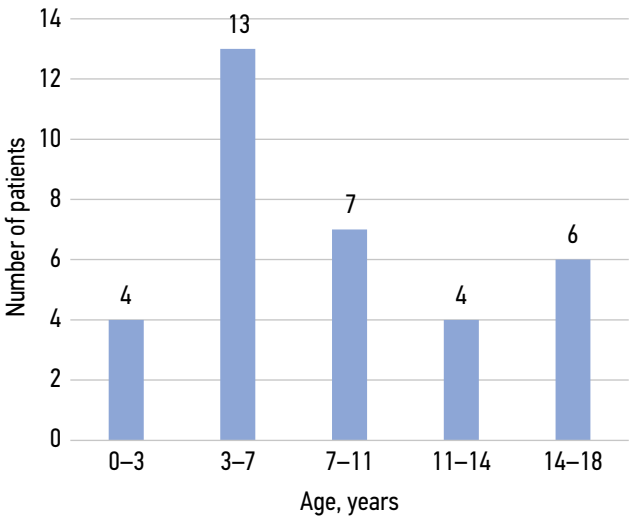

Fig. 3. Histogram of the distribution of patients by age groups

The exclusion criteria were as follows: spinal deformity caused by other abnormalities of development, abnormalities in the development of the spinal cord and spinal canal, neurological deficit, severe concomitant somatic pathology of the internal organs, and refusal of the patient or his/her representative from surgical treatment and participation in the study.

The following data were collected: case histories, radiological examination (X-ray images, multispiral computed tomography data), and magnetic resonance imaging of 33 patients with congenital deformity of the spine associated with segmentation disorders of the lateral surfaces of the vertebrae. Regarding gender, there were 17 girls and 16 boys. The average age at the time of surgical treatment was 107.5 (minimum, 36; maximum, 211 months; $M = 101$) months. The patients were distributed by age based on the classification proposed by Gundobin (Fig. 3).

Table 1 and Fig. 3 show that younger children dominated the study population, which indicated the formation and progression of gross spinal deformity at an early age.

All patients underwent a comprehensive clinical and radiological examination before and after surgery. Case follow-up after surgery was performed twice a year. Based on the X-ray imaging of the spine, performed in two mutually perpendicular projections, the variant of the developmental anomaly and location and length of the unsegmented bar were specified. The size of the scoliotic and lordotic components of the spinal deformity was estimated using the Cobb method before and after surgery. Multispiral computed tomography was performed to rule out intracranial pathology, clarify the length of the unsegmented bar, plan the level of vertebrectomy and spinal implant placement, and confirm the correct position of the hardware. Magnetic resonance imaging can rule out malformations of the spinal cord and spinal canal.

Statistical data processing was performed in the Wolfram Mathematica 11.0 program. The study parameters were tested for distribution normality using the Shapiro–Wilk test. For all parameters, the level of two-sided significance was $p > 0.05$; thus, it was impossible to apply the normal distribution criteria, and the results were presented as histograms of distributions (Fig. 3). The median values and interquartile intervals were also used. The significance of the differences was assessed using the Wilcoxon signed-rank test.

All patients underwent one-stage corrective wedge osteotomy at the apex of the spinal deformity, followed by correction and stabilization of the deformity using a double-rod multi-support hardware.

Surgical technique. A skin incision was made from the dorsal approach along the midline in the projection of the spinous processes of the vertebrae; thereafter, the dorsal structures of the vertebrae were skeletonized. Following visual control and radiography using X-ray contrast marks, the vertebrectomy site was determined, i.e., at the level of the apical vertebra. Channels were formed in the bodies of adjacent vertebrae for the installation of pedicle screws. If transpedicular fixation was not possible, laminar hooks were used. Vertebrectomy was started after X-ray control of the correctness of the position of supporting elements. The transverse process of the apical vertebra, a part of the arch on the convex side of the deformity, and a fragment of a rib were removed over a distance of 2.0 cm from the costovertebral joint. Then, wedge-shaped resection of the vertebral body was performed transpedicularly using small curets and conchotomes with the apex facing the concave side of the curvature, while the cranial and caudal bone plates were preserved. Then, the bars bent in accordance with the physiological profile of the spine were fixed in the support elements installed on both sides of the deformity, and segmental correction was performed (Fig. 4). The amount and position of correction of the supporting elements were assessed using intraoperative radiography (Fig. 5). The procedure was completed with spinal fusion using autologous bone, and the wound was drained and sutured.

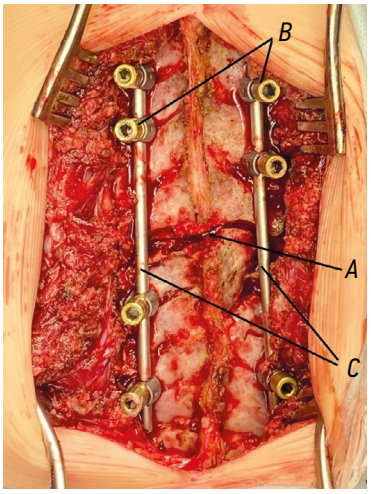

Fig. 4. Surgical intervention site after wedge osteotomy: *A*, vertebrectomy site; *B*, supporting elements installed on both sides of the deformity, with fixed bars; *C*, bars bent in accordance with the physiological profile of the spine after segmental correction

RESULTS

In all patients, congenital spinal deformity was caused by impaired segmentation of the vertebrae in the thoracic spine. The median age (*M*) of patients at the time of surgery was 101 (range, 36–211) months.

The deformity apex in patients was localized in the Th_4–Th_9 vertebrae and always corresponded to the middle spinal motion segment included in the unsegmented bar.

On average, there were six vertebrae in an unsegmented bar (4–8 thoracic vertebrae).

The indications for surgical intervention to correct the curvature of the spinal column were progressive congenital scoliotic deformity and pathological lordosis. Pathological lordosis in the thoracic spine is caused by the nature of the congenital curvature and impaired development of the bone structures of the vertebrae that occurs at this level and the spine as a whole, during the growth of the child.

The mean value of scoliotic deformity according to Cobb was 31.4° (minimum 20°, maximum 80°), with a median

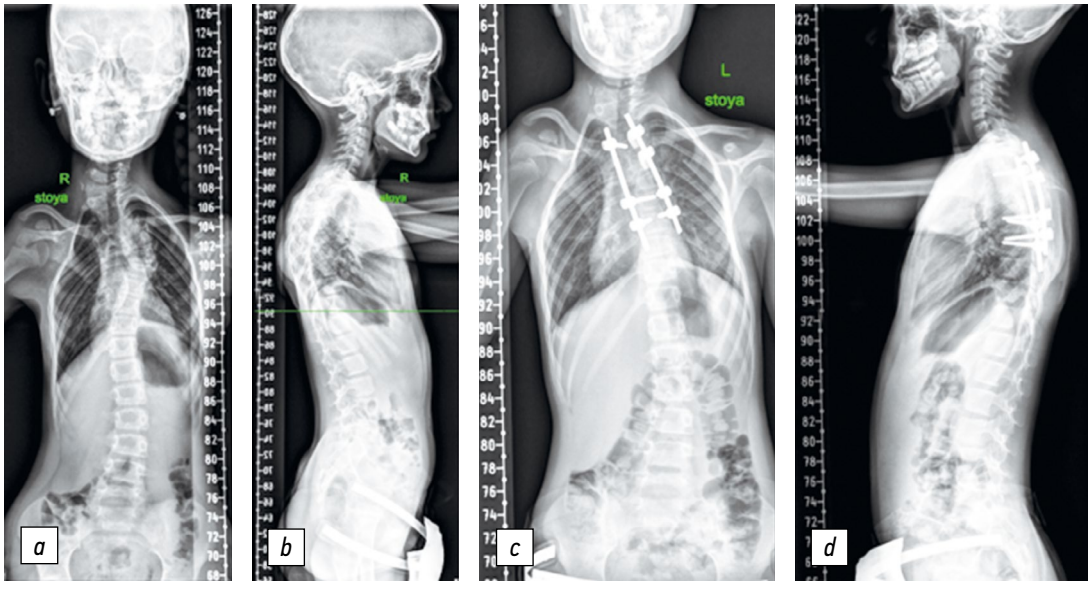

Fig. 5. Panoramic radiograph of the spine in frontal and lateral projections before (*a*, *b*) and after (*c*, *d*) surgery. A 9-year-old patient; after corrective wedge osteotomy with correction and stabilization of spinal deformity using a multi-support corrective system

Table 1. Spinal deformity degree in the studied patients before surgery

Deformity	Age of patients, years				
	0–3 years	3–7 years	7–11 years	11–14 years	14–18 years
Local scoliotic	23.8	28.6	31.7	45.3	33.2
Local lordotic	32.8	30.8	37.9	41.8	40.3

Table 2. Spinal deformity degree in the studied patients after surgery

Age, years	Deformity, degrees		Deformity correction, %	
	Local scoliosis	Local lordosis	Local scoliosis	Local lordosis
0–3	13.3	21.8	44.2	33.6
3–7	8.5	15.1	70.2	51.0
7–11	12.4	17.0	60.8	58.4
11–14	14.0	24.5	69.1	41.3
14–18	13.7	29.8	58.8	26.0

of 31° and interquartile interval (IQR) of 30.5. The mean value of pathological lordosis in the thoracic spine before surgery was 34.7° (minimum 15°, maximum 56°), with median of 29° according to Cobb and IQR of 29.5. No structural compensatory curves in patients with this type of congenital deformity were recorded before surgery. Table 1 presents the distribution of the average value of local spinal deformity depending on the age group.

All patients underwent wedge osteotomy at the apex of the curvature, followed by the correction of the deformity and stabilization of the results achieved. The main aim of surgery was the correction of congenital spinal deformity and creation of conditions for the restoration or improvement of the parameters of the frontal and sagittal profiles of the thoracic spine.

The degree of correction of the scoliotic component of the deformity was 61% (median correction, 84%) (median value after surgery, 5° according to Cobb, IQR 14.5). The magnitude of lordosis correction was 42% (median correction, 41%) (median value after intervention, 17° according to Cobb, IQR 14.5). The distribution of these values according to age groups is presented in Table 2. The significance of

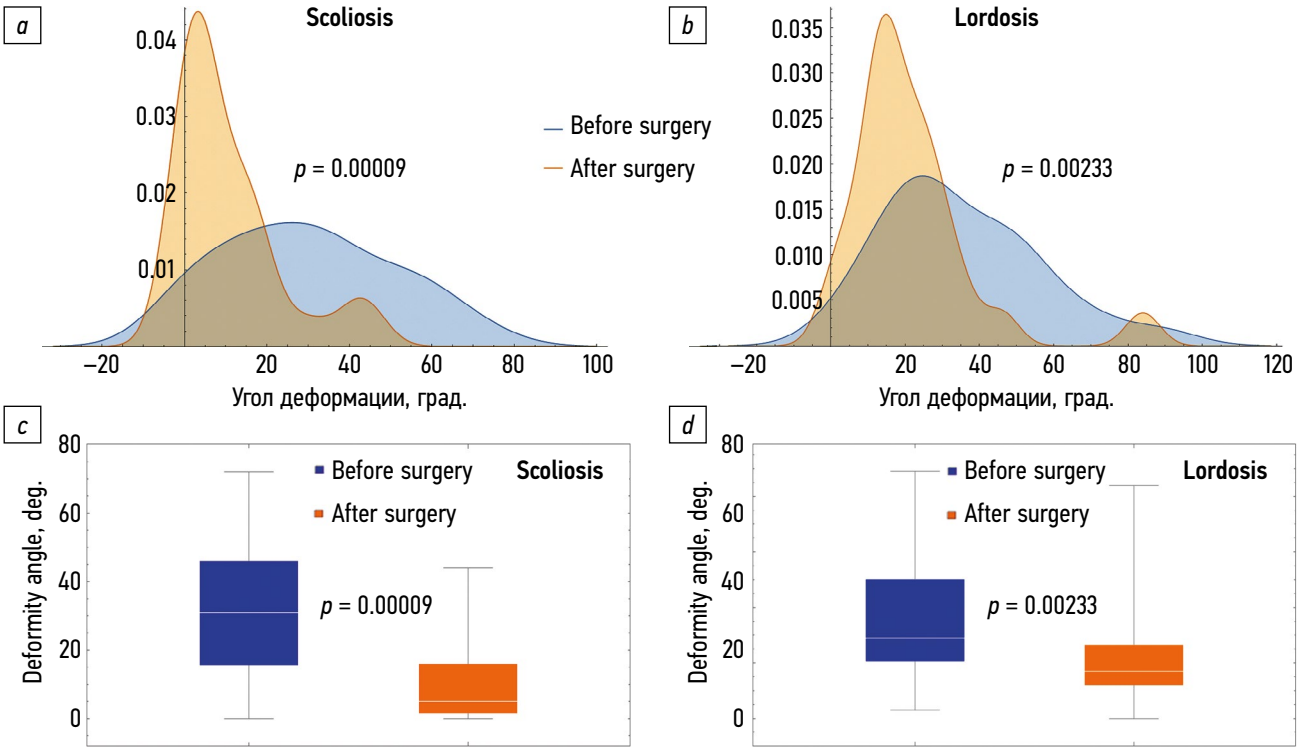

Fig. 6. Distribution of deformity values of the thoracic spine (scoliosis and lordosis) before and after the intervention: *a, c,* distribution of deformity angle values in histogram and box plot for patients with scoliosis; *b, d,* distribution of deformity angle values in histogram and box plot for patients with pathological lordosis

differences in distributions using the Wilcoxon t-test was set at $p < 0.05$.

In the postoperative period, patients in the younger and middle age groups wore a corset to facilitate adaptation; the effect of the orthosis on deformity correction was not evaluated in this study.

The follow-up period for patients ranged from 2 to 6 years, and the average follow-up period was 3 years after surgery. Control examination and radiography of the spine were performed every 6 months. Over the follow-up period, no progression of the deformity was noted; one patient ha destabilization of the hardware due to the initial deformity of the spine of >100° and non-compliance with the orthopedic regimen.

For all parameters, the level of two-sided significance was $p > 0.05$, which indicates that applying the normal distribution criteria was not possible; therefore, the results are presented as histograms of distributions, and median values and interquartile intervals were calculated.

When comparing the significance of distribution differences using the Wilcoxon *t*-test, the values of the spinal deformity before and after surgery were significantly different ($p < 0.05$). Graphs of the distribution of these values are presented in Fig. 6.

In the early postoperative period, two patients with an unsegmented bar located at the Th_2–Th_5 level developed a transient neurological deficit, which was manifested by a unilateral decrease in the strength of the upper limb on the side of the transpedicular wedge osteotomy and development of Horner's syndrome. During the conservative treatment, neurological symptoms regressed by day 14 after surgery in both patients. In one patient, destabilization of the hardware was recorded 3 months after surgery in the form of a rod fracture, which was due to a violation of the orthopedic regimen. In all other patients, the hardware position was stable and correct throughout the follow-up period according to multispiral computed tomography.

The average length of the instrumental block in patients included in the study ranged from 5 to 9 vertebrae, with an average of six vertebrae.

During the follow-up period, the achieved correction of spinal deformity was not lost in patients included in the study.

DISCUSSION

Impaired segmentation of the lateral surfaces of the vertebral bodies is one of the variants of the vertebral development anomaly, leading to the formation of severe and rapidly progressive deformity of the spinal column. The fastest progression rates are noted during periods of active growth [9, 14].

A few studies and the variety of clinical and radiological manifestations of the disease cannot draw clear conclusions regarding the optimal methods of surgical correction of congenital spinal deformity in this defect type [6, 12, 15].

Nearly all studies known present a small series of cases with groups of 4–20 pediatric patients with congenital spinal deformity associated with impaired segmentation of the lateral surfaces of the vertebral bodies and synostosis of the ribs.

The classical methods of surgical treatment of severe spinal deformities, including congenital deformities, use the 360° approach [13], namely, removal of intervertebral discs, followed by dorsal instrumentation. This technique is currently of historical interest and has not been used in recent years [16]. Given the limited growth of the thoracic spine in such patients, lung development is inevitably impaired, which can subsequently decrease the quality and expectancy of life because of secondary pulmonary hypertension [8].

In the late 1980s, devices have been proposed to provide simultaneously to allow the development of the organs of the chest and correct indirectly the congenital deformity. However, they need to perform staged distractions under general anesthesia, has high frequency of hardware destabilization, and causes development of infectious complications because of trophic disorders of the skin in the implantation area [17, 18].

Corrective vertebrectomy in young patients is limited by the need for extended instrumental fixation, which in itself also disrupted the normal development of the thoracic spine [17, 19]. However, with the improvement of spinal implants, surgical technique, provision of anesthesia, and emergence of blood-saving technologies, this variant of surgical intervention is increasingly used in young patients [9].

For the first time, an osteotomy of the spine through a hemivertebra removal was described in 1922 by MacLennan. Subsequently, this surgical method has been changed, modified several times, and acquired various forms. The authors reported deformity correction, balance restoration, and extremely low complication rate [1, 6]. The advent of modern low-profile segmental instrumentation enables the use of modern highly effective methods of surgical treatment in pediatric patients [8].

Thus, Li and Shen [19] presented the treatment results of 31 and 12 patients, respectively, with severe scoliosis (average, 98°). During the studies, patients underwent anterior release at the height of the spinal deformity, followed by posterior fixation of the operated segment. The surgical result was the correction of the initial curvature by an average of 50%. Moreover, the authors indicate a high injury rate and several complications associated with damage to the chest organs.

One of the possible methods of surgical treatment proposed by Chao Li et al. is a two-level osteotomy of the spine beyond the zone of an unsegmented bar from the dorsal approach in combination with the release of the spinal costal joints from

the concave side of the curvature [1]. The authors described the treatment results of 10 patients with an average age of 17.1 years. The average degree of the frontal plane correction was 66%, and the average correction of kyphosis was 61%.

Suk et al. used a different approach in the treatment of congenital deformity of the thoracic spine associated with impaired vertebral segmentation [20]. The correction of congenital spinal deformity by posterior VCR, followed by posterior fixation, was performed in 16 patients. Given the surgical aid, nearly 60% of the deformity correction was achieved; the average value of the scoliotic component of the spinal deformity before surgery was 109° according to Cobb. After surgery, it was 46° according to Cobb. However, the authors indicate a high incidence (up to 25%) of severe complications (including persistent neurological ones) because of inevitable translation and compression of the spinal cord during osteotomy.

Our results demonstrate the efficiency of one-stage corrective vertebrectomy in pediatric patients with spinal deformity associated with impaired vertebral segmentation. With the use of transpedicular fixation, the length of the hardware was minimized, and a significant correction was obtained, which ensured conditions for continued growth and development of the thoracic spine. The minimum number of complications confirmed that this technique is relatively safe. However, our study was limited by the small number of patients, which can be due to the low incidence of this type of defect.

CONCLUSION

The rapid progression of spinal deformity and the lack of a universal surgical treatment of patients with unilateral impairment of the segmentation of the lateral surfaces of the vertebral bodies require searching for the most optimal surgical technique. Simultaneous corrective vertebrectomy in pediatric patients with spinal deformity associated with impaired segmentation of the lateral surfaces of the vertebral bodies is an effective surgical treatment and avoids severe neurological complications. With this method, on average, 61% correction of the scoliotic component of the deformity and 41% correction of the lordotic deformity, which creates optimal conditions for the spinal column development in the near future after surgery, can be achieved. None of the patients required staged surgery during the follow-up period.

The preliminary results of simultaneous corrective vertebrectomy in pediatric patients with this type of defect showed the possibility of effective correction of congenital deformity and provided conditions for further development of the spine in a physiologically favorable position and retention of treatment outcomes.

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 the H.Turner National Medical Research Center for Children's Orthopedics and Trauma Surgery of the Ministry of Health of Russia (Protocol No. 20-3 dated 11/20/2020). Written consent was obtained from patient representatives for the processing and publication of personal data.

Author contributions. *S.V. Vissarionov* formulated the aim and developed the study design and performed surgery on the patients. *M.S. Asadulaev* wrote all sections of the article, collected and analyzed the data, and analyzed the literature. *M.A. Khardikov* collected and analyzed the data, analyzed the literature, performed staged editing of the text of the article, and arranged the reference list. *A.S. Shabunin* performed statistical processing of the data, staged editing of the article, and translated the abstracts and information about the authors into English. *N.O. Khusainov* performed staged and final editing of the article text and performed surgery on the patients. *K.A. Kartavenko* performed staged editing of the article, collected the data, and performed surgery on the patients.

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

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