THREE COLUMN VERTEBRECTOMY OUTSIDE THE APICAL ZONE AS A METHOD FOR CORRECTION OF CERVICOTHORACIC JUNCTION DEFORMITIES: ANALYSIS OF CLINICAL SERIES AND LITERATURE DATA

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Objective. To perform a retrospective analysis of early treatment results in patients with malformation and malsegmentation of vertebrae in the cervical and upper thoracic spine. Level of evidence – IV.
Material and Methods. The study included retrospective monocentre series of 8 patients aged 2–15 years. Inclusion criteria were: age at the time of surgery less than 15 years, deformity in the frontal plane, performed three-column osteotomyies, and presence of full X-ray history.
Results. Patients with multiple developmental abnormalities including vertebral malsegmentation and malformation as leading components prevailed in a series. Violations of the sagittal balance were not noted. Preoperative magnitude of the scoliotic curve ranged from 30° to 66 ° (mean value – 46.1°) according to Cobb, with a frontal imbalance in 6 (75 %) patients. After surgery, residual scoliosis magnitudes were from 3° to 34° (mean value – 15.3°), the frontal balance was restored in all cases. The amount of correction ranged from 49 to 90 % (mean 69.4 %). Neurological status of patients was clinically normal, deviations from the norm were insignificant and recorded only based on ENMG data.
Conclusion. The use of vertebrectomy outside the apical zone in children with multiple vertebral malformations in the cervical and upper thoracic spine allows an adequate deformity correction with restoration of the spine balance, and minimizing the risk of neurological complications due to the leading compression maneuver of correction. This reduces the area of instrumental fixation, which is important for maintaining axial growth.
Key Words: cervicothoracic junction, spine deformity, congenital scoliosis, spine osteotomy, balance, spine malformations.

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The unique anatomy and biomechanics of the highly mobile cervicothoracic spine explain a particular interest to surgical procedures in this area [10, 23, 33, 41, 46]. The literature highlights the experience in the treatment of adult patients with Larsen syndrome [31, 38], Goldenhar syndrome [49], neurofibromatosis [56], ankylosing spondylitis [13, 44, 51], vertebral fractures [5, 9, 14, 16, 20, 46], malignant processes [10], and proximal junctional kyphosis (PJK) [1, 35, 36]. Rare publications concern congenital disorders at the cervicothoracic spine [2, 3, 37], which are mostly devoted to torticollis associated with Klippel-Feil syndrome [27, 28] or unfavorable prognosis of mixed vertebral anomalies of nonsegmented bar with contralateral hemivertebrae [15, 22, 27, 36, 40, 41, 48].

The surgical treatment of this pathology includes various methods:

1) vertebra excision without fixation [21];

2) anterior and/or posterior instrumental fixation [7, 9, 12, 14, 16, 19, 20, 24–26];

3) posterior instrumental fixation combined with osteoplastic fusion [11, 12, 22, 29, 35, 50, 53, 54, 55];

4) anterior vertebrectomy, corporectomy, and release with defect substitution using different implants and posterior instrumental fixation [8, 10, 39, 44];

5) posterior vertebrectomy with instrumental fixation [6, 36].

A diversity of surgical techniques can be related to dissatisfaction with the outcomes when using each of these procedures. These variations motivated the authors to analyze own series of clinical cases and present their option of treating the patients with congenital scoliosis associated with vertebral malformation and malsegmentation at the cervicothoracic junction.

The aim of the study is to analyze retrospectively the early treatment outcomes of patients with vertebral malformations and malsegmentation of the cervical and upper thoracic spine.

The reference sources were searched using PubMed (NCBI), e-Library, and Google scholar databases using keywords: cervical thoracic deformity, cervical thoracic area, osteotomy in abnormality zone spine deformity, cervicothoracic deformation, cervicothoracic spine,

cervicothoracic abnormalities etc. The Russian references were searched in a similar way.

Material and Methods

A retrospective monocentre cohort included 8 children aged 2–15 years (the mean age was 8 years) operated on in 2014–2016 at the Clinic of Spinal Pathology and Rare Diseases, Russian Ilizarov Scientific Center for Restorative Traumatology and Orthopaedics for congenital scoliosis of the cervicothoracic spine. The long-term outcomes were followed-up at a period 1 year to 2 years 1 month. The level of evidence – IV.

The inclusion criteria were:

– age at the time of surgery less than 15 years;

– deformity in the frontal plane;

– monocentre cohort;

– the use of the same type surgical technique – three column wedge-shaped vertebrectomy through a posterior approach;

– presence of full X-ray history.

The patients underwent clinical examination. The neurological status during stages of treatment was assessed using the Frankel grading system. Radiography study included CT and MRI. Functional tests included diagnostic screening (ENMG) and intraoperative neuromonitoring.

The type of abnormality, the leading component in multiple anomalies, and the preoperative and postoperative angle of scoliotic curve were assessed. Neurological status over time was graded using the Frankel scale. The Cobb angles of curves were estimated using the Surgimap software.

An analysis of surgical outcomes did not include data of one female patient. In our opinion, the reconstruction of the apical main curve would not provide proper correction in this patient and we thus used dynamic constructs that allow influencing the secondary dysplastic curves of the thoracic and lumbar spine (Fig. 1).

Preoperative planning. The findings of radiography examination were used to identify the leading component of abnormality according to the proposed method [3]. Functional methods, clinical and paraclinical evaluation of the patient complemented an integrated assessment of the condition and the risk of a surgical intervention. The instrumentation area was planned and the level and type of spinal osteotomy were simulated using the Surgimap software. An important factor in planning was the need for bilateral transpedicular fixation of at least two cranial vertebrae above the osteotomy area for the formation of cranial support base for correction, solid fixation and creating the conditions for fusion. The level and number of fixation points of the distal base were planned according to the level of the distal junction vertebra, frontal imbalance with offset of the C7 and vertebrae of the area of curvature from the central sacral vertical line (CSVL), and sagittal balance. The latter was assessed by the C7 vertebra offset from the posterior sacral vertical line – a plumb line drawn from the C7 vertebra through the posterior-superior corner of the sacrum on the lateral spondylogram (PSVL), and the T1 slope angle.

Surgical procedure. Surgery was performed through a posterior bilateral approach. After exposure of the posterior structures, fixation points were determined and electron-optical image converter-control to sense screw placement was performed. The area of vertebrectomy was located according to preoperative planning. Next, three-column transpedicular vertebrectomy was performed [4] using asymmetric PSO (type III on the Schwab classification), COWO (Schwab IV type), or VCR (Schwab V type) outside the apical zone in children with multiple vertebral developmental abnormalities of the cervical and upper thoracic spine [18, 47]. Metal constructs were then inserted with deformity correction using compression maneuver on the convex side of the scoliotic curve. The operation was completed with 360° fusion using autologous bone (Fig. 2). All manipulations were performed under neurophysiological control.

The volume of blood loss and operative time were varied depending on the amount of instrumental fixation and corresponded to the data obtained during thoracic and lumbar hemivertebrae excision using transpedicular approach [6].

Results and Discussion

Patients with multiple developmental abnormalities including vertebral malsegmentation (n = 5) and malformation (n = 3; Table 1) as leading components prevailed in a series.

Violations of local or global balance, deviations of neither C7 from PSVL nor T1 slope were noted in the patients. The sagittal profile was within either the normal range or local hypokyphosis in the area of abnormality in 4 (50 %) patients. This was the reason for the assessment of sagittal balance in general in the cervicothoracic junction of the spine, which was preserved in all patients before and after surgical treatment. Hypokyphosis (12° according to Cobb) improved to normokyphosis (28°) postoperatively in 1 patient and it remained at the initial levels in 7 patients.

The preoperative Cobb angle of scoliotic curve varied from 30 to 66° (the mean magnitude was 46.1°) and there was frontal imbalance with C7 plumb line offset of 3 to 5 cm in relation to the CSVL in 6 (75 %) patients. The postoperative residual magnitudes of scoliosis were 3 to 34° (the mean magnitude was 15.3°); however, in all cases after surgery, C7 vertebra aligned with the CSVL line and the primary curve vertebrae were in the Harrington stable zone. Curve correction was within 49–90 % (mean, 69.4 %; Table 1).

The pre- and postoperative neurological status of the patients corresponded to Frankel grade E, all deviations from the norm were insignificant and recorded only based on ENMG data. There were no clinical changes.

Intraoperative neuromonitoring was performed in 5 patients. At baseline, all patients elicited stable M-responses. The intraoperative responses remained stable in 2 patients; there were unstable responses and a moderate decline in response amplitude – in 1 and short-term sub-critical changes – in 1. Steady

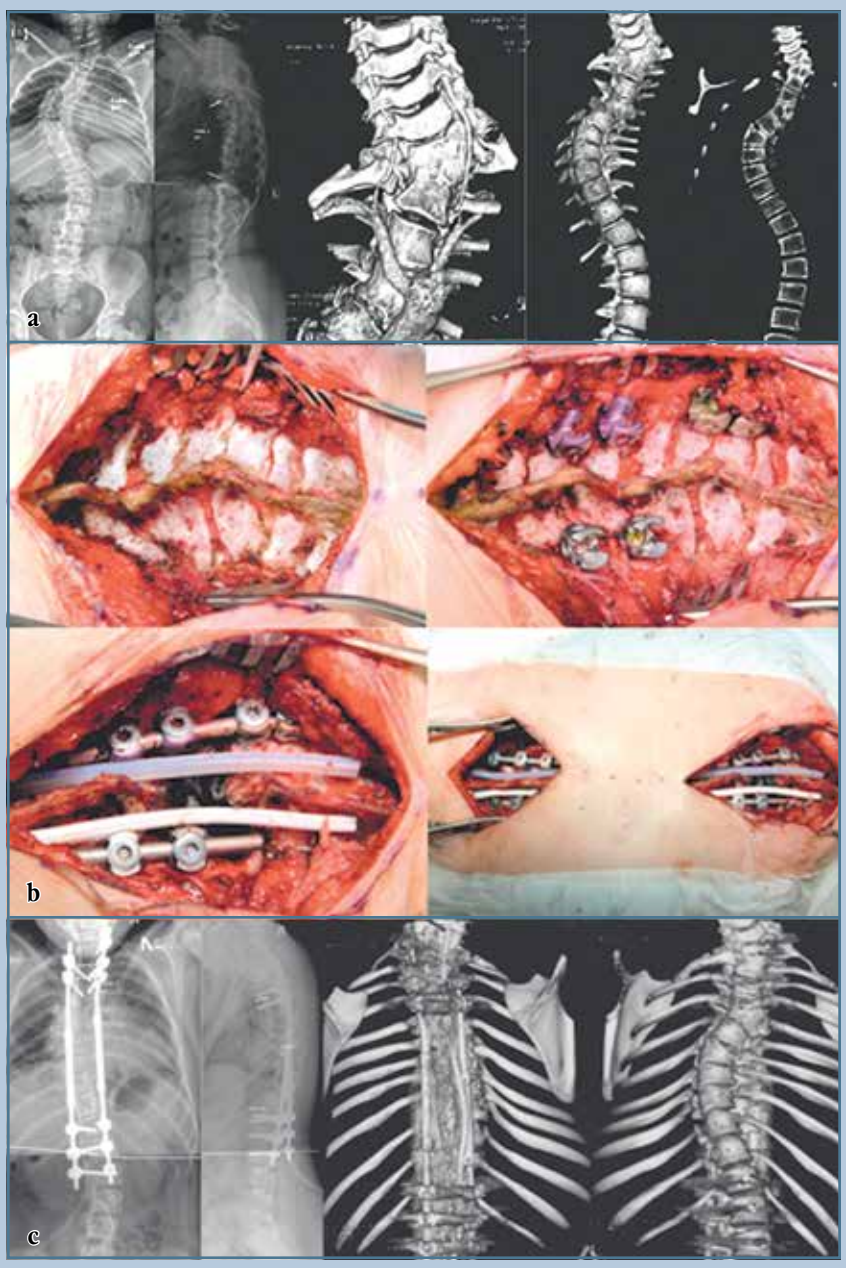

Fig. 1
A female patient aged 11 years, with congenital kyphoscoliosis, the leading abnormality – malformation and malsegmentation of the C7–T5 vertebrae: **a** – preoperative radiography and CT: S-shaped combined kyphoscoliotic C6–T2 curve with the apex at the C7 hemivertebrae (arbitrary), scoliotic component – 47° according to Cobb, local kyphosis at the T3–T10 level – 43°; T3–T11 thoracic scoliotic curve with the apex at the T5 hemivertebrae – 45°; **b** – the main stages of a surgical intervention: asymmetric PSO at the T1 level, posterior instrumental fixation using a TSRH dynamic system, curve correction; **c** – postoperative radiography and CT: scoliotic deformity angle – 5°/20°, correction – 90/69 %, kyphotic curve angle – 12°, correction – 78 %

reduction of M-responses at the end of surgery to 30 % of the original values remained in 1 patient and M-responses recovered to the initial level in the rest children.

The sample of patients was characterized by the location of the curvature apex at the C7–T5 level with the involvement of cervical vertebrae in the primary curve. We preferred bilateral transpedicular fixation due to the formation of stiffness triangle on the each level allowing postsegmental correction. A key factor in planning was the need to insert transpedicular screws into the bodies of two or more cranial vertebrae above the osteotomy area for the formation of cranial support base for correction, solid fixation, and prevention of pseudarthrosis.

The cervicothoracic spinal balance has been analyzed very poorly in the literature. Mainly two parameters were assessed: the sagittal indicator – T1 slope angle [32, 42] and the frontal indicator – shoulder balance [17, 30, 34, 43, 45, 52]. The indicators of the cervicothoracic spinal balance secondary to developmental abnormalities in children are not analyzed in the literature; therefore, we chose curve vertebrae offset from the CSVL line as the main indicator to assess frontal balance and curve vertebrae deviation relative to the PSVL line and T1 slope – for sagittal balance. This allowed us to plan the type of three-column osteotomy and the fixation length. Complete curve correction is not always possible. The focus should be hence on balance restoration at the cervicothoracic junction and the normalization of the above parameters rather than complete correction of local deformity. In this regard, with the location of the leading monosegmental component of abnormality in the cervicothoracic junction or in presence of vertebral arch hypoplasia or aplasia, vertebrectomy outside the apical zone allowed us in all cases to perform curve correction with the restoration of frontal balance, minimize the instrumentation area, and reduce the risk of neurological complications due to the leading compression maneuver of correction. This fact is confirmed by intraoperative neuromonitoring, which shows

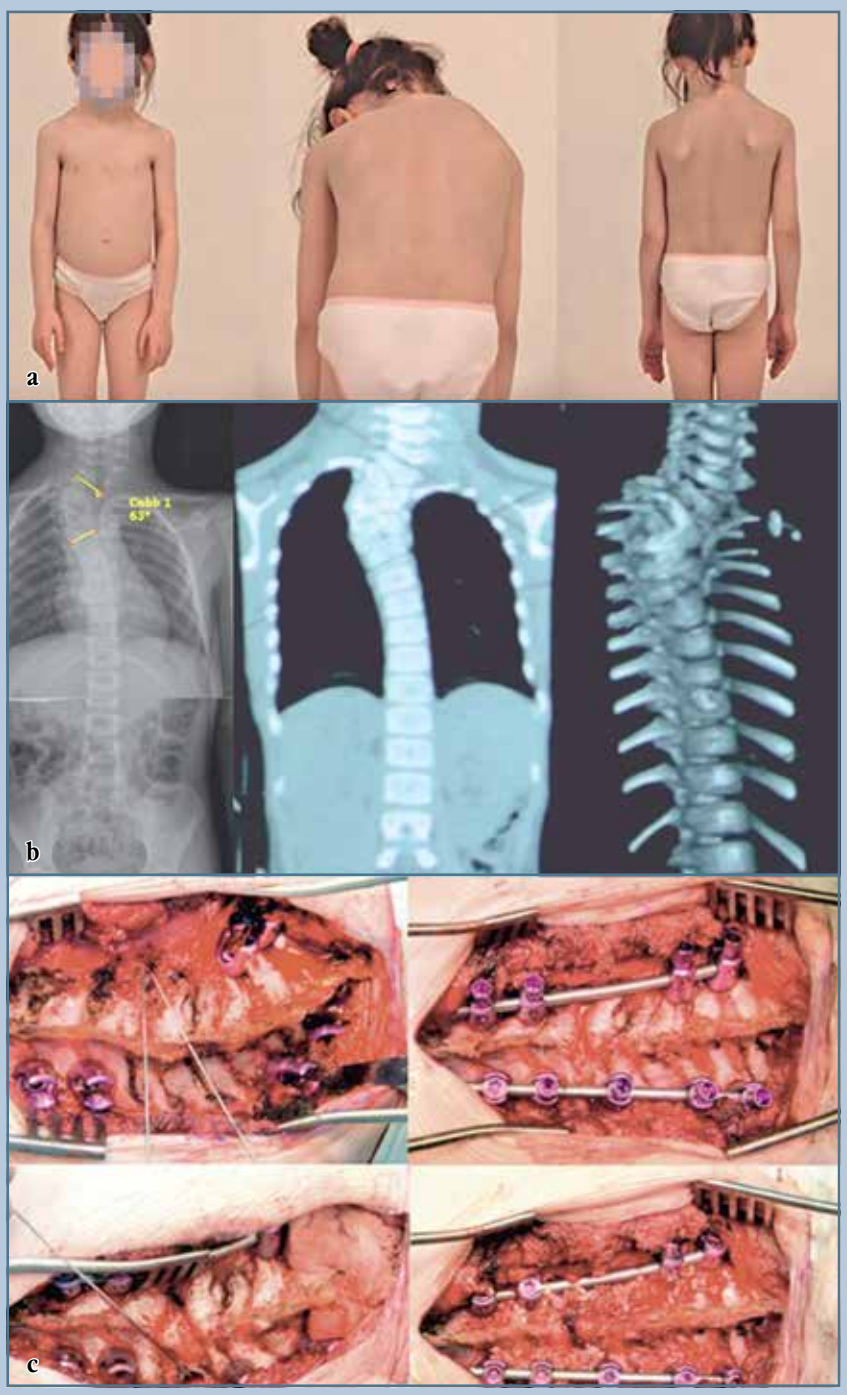

Fig. 2
A female patient aged 6 years 9 months with congenital scoliosis, the leading abnormality – malformation and malsegmentation of the C7–T5 vertebrae: **a** – before treatment; **b** – preoperative radiography and CT: congenital spinal anomalies, T1–T3 lateral hemivertebrae and the T4 butterfly hemivertebra, concrescence of posterior part of the III–IV, V–VI ribs, concrescence of T2–T5 vertebrae, C-shaped scoliosis of 63°; **c** – the main stages of a surgical intervention: asymmetric PSO with the resection of the right portion of T4 butterfly vertebra, posterior instrumental fixation using a pediatric system, correction of deformity, 360° spinal fusion

the absence of any significant intraoperative deviations from the norm.

Conclusion

An essential parameter in surgical correction of abnormalities at the cervicothoracic junction is the local balance reconstruction in the frontal and sagittal planes rather than a complete correction of the local deformity.

Vertebrectomy outside the apical zone in children with multiple developmental abnormalities of the cervical and upper thoracic spine allows one to perform a satisfactory correction of the deformity (approximately by 69.4 %) with spinal balance recovery and to minimize the risk of neurological complications. This reduces the area of instrumental fixation compared to classic methodologies of scoliosis fixation, which is important for maintaining axial growth.

The study is not a sponsored project. The authors declare that they have no conflict of interest.

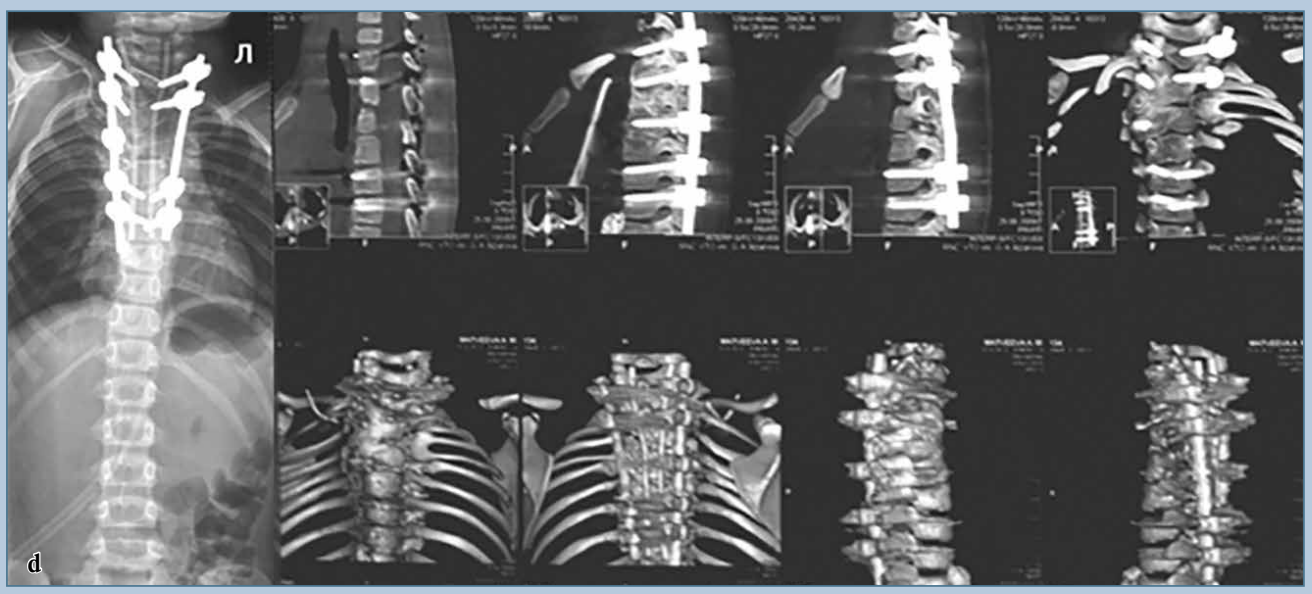

Fig. 2 (end)
A female patient aged 6 years 9 months with congenital scoliosis, the leading abnormality – malformation and malsegmentation of C7–T5 vertebrae: **d** – postoperative radiography and CT: scoliotic curve angle – 25°, correction – 72 %

Table
The outcomes of surgical treatment

Patient	Age	Type of the leading scoliogenic abnormality	Scoliotic deformity, degrees		Correction, %	Curve apex/ vertebrectomy level
			preoperative	postoperative		
1st	7 years	A lateral hemivertebra	39	13	67	T3/T4
2nd	2 years	A lateral hemivertebra	66	34	49	T2/T3
3rd	15 years	A lateral hemivertebra	53	19	64	T5/T6
4th	5 years 4 months	Multiple developmental abnormalities: the leading abnormality — asymmetric malsegmentation	30	3	90	T1/T2
5th	6 years 5 months	Alternating hemivertebrae	51	26	49	T2/T3 and T5
6th	11 years	A lateral hemivertebra	44	5	89	C7/T1
7th	6 years 9 months	Multiple developmental abnormalities: the leading abnormality — asymmetric malsegmentation	47	10	79	T3/T4
8th	11 years	Multiple developmental abnormalities: the leading abnormality — asymmetric malsegmentation	39	12	69	T1/T2 and T3

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