

# SURGICAL TREATMENT OF TUMORS AND SPONDYLITIS OF THE CERVICAL SPINE IN CHILDREN: WHAT DOES 20-YEAR MONOCENTRIC EXPERIENCE WITH THE SYNDROMIC APPROACH REVEAL?

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**Objective.** To analyze short- and long-term results of surgical treatment of children with tumor and infectious destruction of the cervical vertebrae.

Material and Methods. The study included data from 94 children. The following parameters were evaluated: the effectiveness of perioperative use of halo-fixation, surgical approaches, methods of reconstruction and stabilization of the spine, dynamics of pain syndrome and neurological disorders, and the structure of postoperative complications. The material was statistically processed using nonparametric analysis methods. Statistical analysis and visualization of the obtained results were carried out using the R programming language. Results. A halo-fixation was used in 12.8 % of cases due to significant deformations and stenosis of the spinal canal. Of the 34 resections that required combined approach, 30 were performed in a single surgical session. The median duration of the surgical stage was 162.5 minutes, the median blood loss volume -100 ml. Reconstruction of the anterior column of the spine was required in 62.8 % of cases, and posterior instrumental fixation - in 42.6 %. Local pain syndrome was eliminated in all cases. Four patients with neurological disorders showed an improvement in neurological status by one or more levels according to the Frankel scale, two patients - worsening by one level, and no significant change in the neurological picture was observed in nine cases. Complications were revealed in 26.6 % of cases, while only five patients required repeated surgery. The outcome of surgical treatment is affected by the destruction of three or more vertebrae (p = 0.0298). Conclusion. In the treatment of children with cervical vertebrae destruction, a unified syndromic approach is justified and associated with a minimal frequency of clinically significant perioperative complications.

**Key Words:** cervical spine; surgical treatment; tumor; spondylitis; osteomyelitis; children.

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Most hospitals focusing on chronic destructive skeletal lesions in children and adults specialize in the treatment of specific diseases, such as tumors and non-specific or tuberculous osteomyelitis. For many years, the specific feature of the Clinic for Pediatric Surgery and Orthopedics of the Saint-Petersburg Research Institute of Phthisiopulmonology has been a predominantly syndromic approach to destructive skeletal pathology, where diagnosis and treatment strategy depend on the prevalence of destruction, involvement of surrounding structures, and the presence of complications rather than on the etiology of the disease (with the exception of malignant disorders). This is most appropriate for rare localizations of lesions, in which a sufficient number of cases for analysis can only be obtained by long-term or multicenter data collection. This localization includes lesions of the cervical spine in children.

The objective is to analyze shortand long-term results of surgical treatment of children with destructive pathology of the cervical spine.

Study design: a retrospective and prospective monocenter cohort.

Class of Evidence: III (Oxford CEBM, 2011).

Object of study: children with tumors as well as infection and inflammatory lesions of the cervical vertebrae.

Inclusion criteria:

- localization of the lesions: cervical vertebrae, including Oc/C1–C7;
- patients younger than 18 years old at the time of surgical treatment;
- a single center for surgery: the Clinic for Pediatrics Surgery and Orthopedics of the Saint-Petersburg Research Institute of Phthisiopulmonology;
  - period of surgeries: 2001–2021;
- duration of postoperative follow-up: at least one year after surgery;
- morphologically or bacteriologically verified origin of vertebral lesions:

tumors and infectious inflammatory diseases.

Exclusion criteria: congenital malformations or traumatic injuries of the cervical vertebrae; primary neurosurgical pathology (tumors and congenital malformations of the spinal cord or skull base, including any types of myelodysplasia).

# **Material and Methods**

The above inclusion criteria were fulfilled by the data of 101 children whose etiology and clinical features were given in detail in a previously published article [1]. Seven children (two cases each of generalized tuberculosis, nonspecific multifocal osteomyelitis, and multifocal Langerhans cell histiocytosis, and one case of generalized aspergillosis) showed multiple skeletal lesions; however, destruction of the cervical vertebrae did not result in instability and local complaints, and the diagnosis was verified by biopsy of more accessible foci with further systemic etiopathogenetic therapy of the disease. These patients were excluded from further analysis. Therefore, the study group consisted of 94 children who underwent surgery for a destructive process of the cervical spine after the diagnosis was clarified (Fig. 1): in a 20-year retrospective and prospective sample, 35 patients had cervical vertebral tumors, 28 patients had nonspecific osteomyelitis, 25 patients had spinal tuberculosis, four patients had nonbacterial osteomyelitis, and two patients had mycotic spondylitis.

Parameters under evaluation: indications and effectiveness of perioperative use of halo-fixation, surgical approaches, techniques of spine reconstruction and stabilization, progress of pain syndrome and neurological disorders, and the structure of postoperative complications.

The data were subjected to statistical processing using nonparametric analysis; the values of the sample parameters are given as Me (Q1–Q2), since the Shapiro-Wilk test and Lilliefors test (Kolmogorov-Smirnov with Lilliefors correction) revealed statistically significant deviations of the studied groups of parameters

from the normal distribution. Accumulation, correction, and systematization of initial data were performed in spreadsheets in LibreOffice Calc (version 3.7.2, MPL v.2). Statistical analysis and visualization of the results were performed using the R programming language (version 3.5.1, GNU GPL v.2) in the RStudio Desktop software development environment (version 2022.02, GNU AGPL v.3).

# Results

Peculiarities of halo fixation application

The junctional areas of the spine were involved in 10 out of 12 cases of halo-fixation: four cases of craniovertebral and six cases of cervicothoracic ones.

The halo fixator was used in five cases for preoperative stabilization and attempted stage correction of deformity at the level of the upper cervical spine (Fig. 2) because of severe deformity and spinal canal stenosis. The halo-ring was applied under anesthesia with simultaneous performance of transoral trephine biopsy. Following the awakening of the patient, stage deformity correction was performed with evaluation of the neurological status. In seven cases, the halo device was used only for intraoperative temporary stabilization of the spine during resection of affected vertebral bodies, the median number of which was 4.5 (min 1; max 9).

Peculiarities of the surgical treatment
The surgical treatment was performed
with consideration of the syndromic
approach regardless of the medical conditions. Because of the complexity of the
anatomical area and the multiplicity of
closely located vital structures, a confirmed malignant tumor was removed in
compliance with the principle of ablastics, if possible; in other cases, intralesional or marginal resection was performed.

The main surgical stage included removal of pathological tissues and spinal reconstruction. The used surgical approaches are summarized in Table 1 and Fig. 3.

Among 34 resections in which the surgery was performed from the combined approach, 30 were performed in one surgical session and 4 were performed in

a staged manner. The median duration of the surgery was 162.5 (123.8–215.0) minutes with a blood loss volume in the cohort of 100 (50–200) ml and no statistically significant difference between medical conditions (Kruskal–Wallis test:  $\chi^2 = 5.1836$ , df = 4, p = 0.2690).

Reconstruction and stabilization of the spine

Reconstruction of the anterior spinal column associated with initial or developed instability after resection of pathological tissues was required in 59 (62.8 %) cases. The most commonly used graft was a titanium implant filled with autogenous bone (Table 2).

Indications for posterior instrumented fixation were spinal deformity, junctional area lesions (C0-C2 and C7-T1), resection of more than two spinal motion segments, and initial neurological deficit. Posterior instrumented fixation was subsequently performed in 40 (42.6 %) patients, with inclusion of the occipital bone in 5 (5.3 %) cases. The choice of posterior instrumentation technique depended on the time of surgery: the first fixations in children were performed 20 years ago using laminar (hook) systems. Thereafter, there was a trend towards both a reduction in the instrumentation extent and a shift to the use of screws, which were more often placed in lateral masses or into C2 in a translaminar manner because of the small anatomical dimensions of the vertebrae (Fig. 4).

Changes in the main clinical manifestations of pathology after surgery

The postoperative changes in the main clinical manifestations of the disease (pain syndrome and neurological disorders) are given in Table 3.

Being the most frequent complaint, local pain syndrome was relieved in all cases (Wilcoxon signed-rank test: V = 171, p = 0.0002).

Among 15 patients with neurological disorders, improvement of the neurological status by one level or more according to the Frankel scale [2] was observed in four patients; neurological status of patients worsened by one level in two cases. There were no changes in the neurological picture in nine cases with

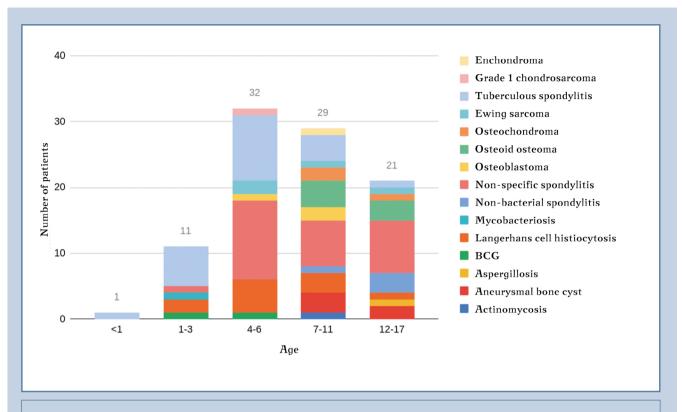


Fig. 1
Etiological structure of destructive lesions of the cervical vertebrae in children included in the study

initially minimal neurological disorders (Frankel type D). Moreover, no statistically significant correlation was found between postoperative neurological outcomes and the duration of the diagnostic pause ranked according to three time periods: up to 3 months, 3 to 6 months, and more than 6 months (Fisher's exact test, p = 0.4675).

Complications and long-term outcomes of surgical treatment

In accordance with the most common Satava [3] and Clavien-Dindo [4] classifications, 32 (31.6%) adverse events were reported in 25 (26.6%) cases during and after surgery; 27 cases required local or systemic drug treatment. Repeated surgery was required only in five cases corresponding to Clavien–Dindo class IIIB: two cases in the early postoperative period and three case in the late one.

The fatal outcome was recorded in three cases in patients with malignant tumors: two cases associated with Ewing sarcoma (one case with local progression on the background of adjuvant chemotherapy within a month after surgery; and one case with distant multiple metastases without signs of local recurrence after radiotherapy on the background of anti-relapsing chemotherapy 11 months after surgery); and one case associated with acute respiratory complications 2.5 years after treatment for Grade 1 chondrosarcoma recurrence.

The accumulation of adverse events and the period of their onset after surgery (besides mortality and complications that required surgical treatment) are shown in the Kaplan–Meier curve (Fig. 5).

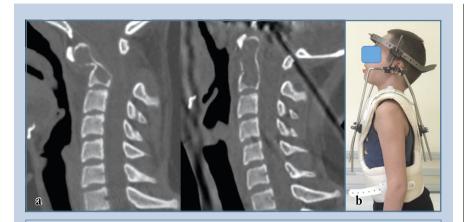
All three patients with late postoperative complications underwent primary extended (three or more vertebrae) resections for cervicothoracic tuberculous spondylitis and anterior allograft fusion at the age of 5–7 years. During the period from 18 to 25 months postoperatively, on the background of children's

grew up without recurrence of destruction or development of pseudarthrosis, kyphotic deformity formation with impaired gaze angle occurred requiring staged corrective posterior vertebrotomy to improve the head position.

A multiple logistic regression model was built to evaluate factors influencing the surgical outcome, considering the patient's age at the time of surgery, the etiology, level, and extent of the lesion, as well as the type of anterior reconstruction and the presence of posterior instrumentation. The results are shown in Table 4.

The built model revealed that the only factor with a statistically significant effect on surgical outcome was the extent of destruction (p = 0.0298), with a threshold value of three or more vertebrae damaged. According to our cohort, all other factors have not significant effect on surgical outcomes (p > 0.05).

The obtained results provided the basis for formulating an algorithm of



**Fig. 2**Preoperative correction using a halo device of upper cervical kyphosis associated with an aneurysmal bone cyst of the C2 odontoid process, complicated by a pathological fracture: **a** – sagittal CT section before and after deformity correction, **b** – the child's appearance in a halo device

Table 1
Cervical spine approaches used for surgeries in children

Approach	Absolute quantity, n	Relative quantity, %	
Anterolateral	29	30.8	
Transoral	9	9.6	
Posterior	22	23.4	
Combined,	34	36.2	
including transoral	3	3.2	

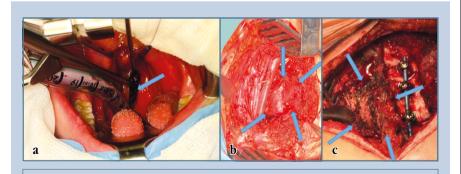


Fig. 3
Surgical approaches to the spine: **a** – transoral approach, the arrow indicates a titanium implant installed after resection of the C2 aneurysmal bone cyst; **b** – anterolateral approach, arrows indicate the boundaries of the C3 Ewing sarcoma; **c** – posterior approach, arrows indicate the boundaries of the C5–C6 aneurysmal bone cyst with contralateral preventive instrumental fixation

surgical care for children with destructive lesions of the cervical spine (Fig. 6).

# Discussion

Only seven articles on surgical treatment of destructive pathology of the cervical spine in children were available, representing the largest samples (from 5 to 29 people) and reflecting the treatment outcomes of children with only one etiology of the process, such as tumors or spondylitis [5–11]. These small numbers prove the rarity of cervical vertebral lesions in children. Therefore, the analyzed surgical outcomes of 94 patients aged 11 months to 17 years represent the largest selective clinical sample.

The use of the halo device should be considered as an additional or stepwise treatment of such pathology. The main indications for its use are engagement of junctional areas, extended destruction, and pronounced deformity of the cervical spine with neurological disorders. For patients with a suspected malignant process with mechanical or neurological spinal instability, the use of halo-fixation as a preliminary stage of treatment provides a safe waiting period for biopsy findings.

According to our data, anterior column reconstruction associated with spinal instability was required in 62.8 % of cases, and posterior instrumentation was required in 42.6 % of cases with extended resection, deformity, involvement of the junctional area, and neurological disorders.

It should be noted that surgical treatment of this category of patients provided complete pain relief in all children, while neurological signs were improved in only 4 out of 15 children. There was no significant effect of the duration of the diagnostic pause on the neurological picture and its postoperative changes.

Within this analysis, cases of vertebral artery involvement in the pathological process are deliberately not considered. The features of such patients and the treatment procedure for the artery have been presented earlier [12].

Although the number of reported adverse perioperative events was quite

Table 2
Reconstruction of the anterior column of the cervical spine in children after resection of the destruction zone

Reconstruction technique	Absolute quantity, n	Relative quantity, %	
Titanium mesh with an autograft	30	50.8	
Allograft	16	27.1	
Autograft	9	15.3	
Autograft + allograft	4	6.8	

Table 3

Assessment of clinical outcomes of treatment of cervical destructive lesions in children

Parameter	Before	After	Assessment
	surgery	surgery	
Severity of pain syndrome	5 (3.5-7.0)	0 (0.0-0.0)	V = 171;
according to VAS			p = 0.0002
Neurological impairment	B-1	$B \rightarrow D - 1$	${\rm Improvement}-4;$
(type according to Frankel, number	C-2	$C \rightarrow D - 1$	no change — 9;
of patients)	D - 12	$C \rightarrow B - 1$	${\rm deterioration}-2$
		$D \rightarrow E - 2$	
		$D \rightarrow C - 1$	

VAS-a 10-point visual analogue pain scale; B,C,D,E-assessment of neurological status according to the Frankel scale; V-Wilcoxon test.

high, the vast majority of them did not affect the treatment outcome. Additional surgeries because of postoperative complications/consequences were required in only five (5.3 %) cases, including two in the early postoperative period and three between 18 and 25 months after treatment. Three or more vertebral lesions were found to be a factor influencing the risk of their development, which is an indication for 360-degree stabilization.

# Conclusion

The syndromic approach to surgical treatment of tumors and infectious and inflammatory lesions of the cervical spine in children provides relief of pain syndrome that is the main clinical complaint of patients.

Neurological complications of cervical vertebral destruction are found in 16 % of cases, rarely associated with severe paresis/paralysis, but still remain in most cases after decompression and stabilization surgery.

A significant factor for severe peri-/ postoperative adverse events was involvement of three or more vertebrae

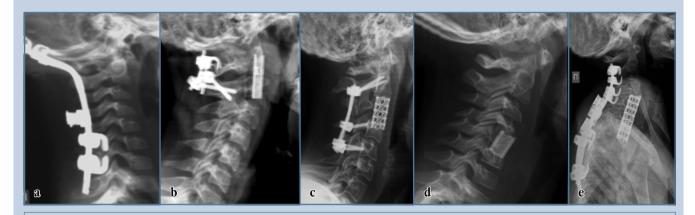


Fig. 4

Variants of reconstruction and posterior instrumentation of the cervical spine:  $\bf a$  – anterior occipital cervical fusion of the C0–C2 vertebrae using an allograft, posterior instrumental fixation of the C0–C7 vertebrae (the first craniovertebral fixation in a child with tuberculous spondylitis of Oc–C1–C2 performed in 2004);  $\bf b$  – anterior spinal fusion of the C1–C2 vertebrae using a porous titanium mesh with an autograft, posterior instrumental fixation of the C1–C2 vertebrae using translaminar screw placement;  $\bf c$  – anterior spinal fusion of the C2–C4 vertebrae using a mesh with autogenous bone, posterior instrumental fixation of the C2–C5 vertebrae;  $\bf d$  – anterior spinal fusion of the C4–C6 vertebrae using a porous titanium mesh with an autograft;  $\bf e$  – anterior spinal fusion of the C4–T3 vertebrae using a mesh with autogenous bone, posterior instrumental fixation of the C2–T6 vertebrae using a laminar structure

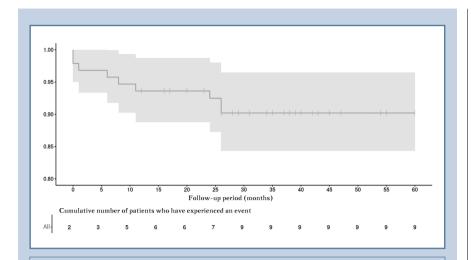


Fig. 5

Overall event-free postoperative course in the cohort

 $\label{thm:continuous} \begin{tabular}{ll} Table 4 \\ The influence of various factors on the outcome of surgical treatment of cervical destructive lesions in children \\ \end{tabular}$ 

Factor	Value	z score	p
Age (under 7 years, 7 years and over)	-0.1703	-0.096	0.9237
Non-specific spondylitis	-14.9517	0.000	0.9996
Non-bacterial spondylitis	-32.9192	-0.001	0.9993
Tuberculous spondylitis	-23.2909	-0.001	0.9994
Mycotic spondylitis	-30.9552	-0.001	0.9992
Tumors	-18.7339	-0.001	0.9995
Level (junction area)	-8.5394	-0.868	0.3853
Extension of destruction	3.0032	2.173	0.0298
Reconstruction using a mesh	40.5755	0.007	0.9942
Reconstruction using a graft	6.6644	0.001	0.9993
Presence of posterior fixation	2.5010	1.027	0.3044

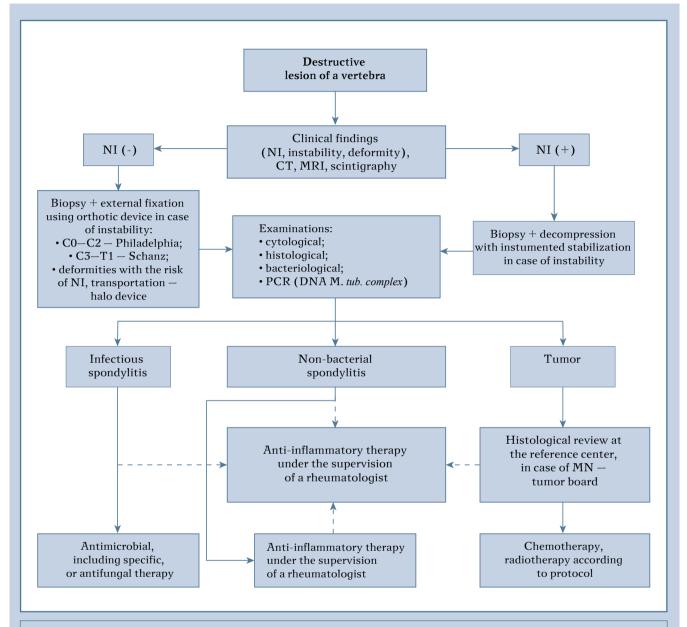
(p = 0.0298), and risk factors for longterm (more than 1.5 years after procedures) orthopedic outcomes were cervicothoracic lesions and preschool age of children at the time of surgery.

The obtained data can be used in the planning of surgical treatment, follow-up of patients, and informing their parents about the possible outcomes of treatment of destructive lesions of the cervical vertebrae.

The study had no sponsors. The authors declare that they have no conflict of interest.

The study was approved by the local ethics committee of the institution.

All authors contributed significantly to the research and preparation of the article, read and approved the final version before publication.



 $\label{eq:Fig.6} \textbf{Algorithm for providing surgical care to patients with destructive lesions of the cervical spine: NI - neurological impairment, MN - malignant neoplasm$ 

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