SURGERY OF CERVICAL SPINE TUMORS IN THE CLOSE VICINITY TO THE VERTEBRAL ARTERY: LITERATURE REVIEW

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Objective. To analyze the literature on cervical spine tumors located in the region of the vertebral artery.

Material and Methods. Sixty five publications containing data on the surgical treatment of 101 patients with tumors of the cervical spine located in the area of V1—V3 vertebral artery segments were selected for the literature review.

Results. The analysis of publications was performed according to the following criteria: demographic data, complaints, histological type of tumor, involvement of the vertebral artery, the performed occlusion test, final embolization or ligation of the artery during surgery, and postoperative complications. The group of patients described in selected publications consisted of 66 men and 35 women, whose average age at the time of surgery was 38.7 years. Acute development of neurological symptoms at the prehospital stage caused by vertebral artery compression was observed in two cases. The C2—C4 vertebrae were most often affected, and the malignant process was confirmed in 70.3 % of cases. In 27 cases, patients underwent an occlusion test of the involved vertebral artery. At the preoperative stage, the final embolization of the artery was performed in 9 cases, the artery was tied during the main operation and removed together with the tumor — in 25, and damaged — in 7. Neurological disorders associated with stopping blood flow through the vertebral artery were noted in two cases. In a number of cases, patients underwent vascular anastomosis with preservation of blood flow at follow-up examination.

Conclusion. Taking into account the obtained data and careful preoperative planning will improve the treatment of this group of patients, increase survival, and reduce the risks of possible neurological disorders and tumor recurrence.

Key Words: tumor, cervical vertebrae, vertebral artery, surgery.

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Surgical interventions on the cervical spine are considered one of the most difficult procedures due to the anatomical peculiarities, of the great vessels providing blood supply to the brain: the carotid and vertebral arteries. Carotid arteries, which are located in soft tissues, have a large diameter and are sufficiently mobile and accessible for potential surgical manipulations. Vertebral arteries are, on the contrary, embedded in the bone matrix, which significantly limits the surgeon's range of motion.

The conventional anatomical variant of the blood supply with complete set of vertebral, carotid, and connective arteries is found only in 34.5 % of the population. Other 83 variants include from completely isolated carotid and vertebral systems to complex systems of vascular collaterals [1]. For this reason, clinical manifestations of blood flow disorders in the vertebral arteries are not entirely predictable and can vary from asymptomatic disease to severe cerebral ischemia [2–4].

When it comes to surgical treatment of cervical spine tumors, preservation of vertebral arteries is always opposed to radical resection of the neoplasm. There are different types of the relationship between the tumor and a vessel (from compression to the circular coverage of the vessel), which requires accurate planning of surgery, prevention of bleeding, and considering options for intraoperative hemostasis.

The aim of the study was to analyze the literature on cervical spine tumors located in the region of vertebral artery.

Material and Methods

A systematic literature search was performed in the databases E-library.ru, Pubmed, and Google Scholar (date of the last inquiry: June 1, 2020) without restrictions on the retrospective time and type of publication for the following key words: "cervical spine tumor" and "vertebral artery". The inclusion criteria were the following:

1) tumors of the cervical spine;

2) data on its relationship with the vertebral artery.

The exclusion criteria were as follows:

1) neurogenic neoplasms of the cervical spine;

2) unavailability of the full-text version of the article;

3) lack of a description of the relationship between the neoplasm and the artery, as well as the specificities of the procedures in the area around it during a diagnostic procedure or surgery;

4) the tumor is located at the level of the V4 segment of the vertebral artery (above the loop at C1–C2 level).

Results

Selection of publications. The algorithm for selecting publications using the inclusion and exclusion criteria is

presented in Fig. 1. The final sample included 65 articles, of which only 11 papers described a clinical series, including from two to 10 cases each [5–15], while the remaining 54 studies were single case reports [16–69].

In total, 65 publications include data on 101 patients (66 men and 35 women) with the average age at the time of surgery of 38.7 years (min, 5 years; max, 83 years). There were 21 (20.8 %) pediatric patients (up to 18 years old) presented in 14 studies in total [5–8, 16–25]. The largest series in the articles were comprised of three children [5–7].

Clinical and morphological features. By the time of surgical treatment, there were a wide range of the patients' complaints, with the leading ones being pain in the neck as well as sensory and motor disorders in the upper extremities (Table 1). Clinical manifestations of vascular pathology were noted only in two cases: it was manifested by dizziness and bow hunter syndrome in one case [26] and acute cerebral symptoms (headache, vertigo, nausea, and vomiting) and gait disturbance due to an ischemic lesion in the cerebellum in another case [27]. In both cases, the complaints were caused by occlusion of the vertebral artery due to osteochondroma.

Apparently, the rarity of the pathology and non-specificity of the complaints, including the absence of acute neurological symptoms in most cases, complicate the diagnosis: the average diagnostic pause in the group was 17.9 months (min, 2 days; max, 10 years).

All patients underwent CT and MRI for visualization and characterization of the lesion, as well as determination of its topographic features. In a total of 57 (56.4 %) patients, more than one (from 2 to 5) adjacent vertebrae were affected. According to the segmental structure of the vertebral artery, neoplasms were located at the level of the cervicothoracic junction (V1 segment of the *a. vertebralis*) in 12 cases, at the level of the cervical vertebral canal (V2 segment) in 40, and at C1–C2 (V3 segment) in 49 ones (Fig. 2).

Biopsy as the first stage of invasive interventions was performed in 61 out of 101 cases, while in the remaining 40 patients, it was combined with the main stage of the operation. The most frequent histological variants of tumors were chordomas (55 (54.5 %) patients) and osteoblastomas (11 (10.9 %) patients).

The malignant nature of the tumor was confirmed in 71 (70.3 %) cases (Fig. 3).

Features of preoperative examination. The extent of the planned surgical interventions depended on the type of tumor and its relationship with the surrounding anatomical structures. For this reason, in a significant number of publications, the authors point to the need for an additional contrast-enhanced study: MR angiography (20 articles), CT angiography (35), and X-ray angiography (41). A total of 18 publications described a combined use of angiographic methods.

In cases of a significant risk of injury to the vertebral artery or its loss during *en bloc* resection, temporary occlusion test is recommended. The test was performed in 27 patients of the considered group; in 20 cases, it was performed by an endovascular surgeon [13–18, 28–34]. The patient's condition was evaluated based on the data of neurophysiological monitoring or changes in the neurological status for 30 minutes if the manipulation was performed under local anesthesia. During the test, one patient developed hemiparesis, which required correction of the surgical treatment plan [28]. Final embolization of the involved artery after the occlusion test was performed in three patients (in one case, it was conducted on both sides) [18, 29, 30] and without the test in six cases [9, 35–39]. In seven cases, the test was carried out during the primary surgery with clamp fixation of the vertebral artery for 30 min with neurophysiological monitoring of the evoked motor and sensory potentials [10, 11, 19, 40–42].

Features of surgical interventions. In most patients, the extent of surgery included tumor resection through the combined approach with anterior and posterior stabilization. Types of the surgical interventions performed are presented in Table 2.

A correlation between the number of affected vertebral segments assessed according to the Weinstein–Boriani–Biagini (WBB) classification [70] and the extent of surgical intervention shows a very weak relationship ($r = 0.25$). Extravertebral spread of the tumor (layer A according to WBB) was observed in 66 cases, intracanal spread (layer D) and intradural penetration (layer E) were noted in 58 and three patients, respectively. Of special interest is the fact that layer F (vertebral artery canal) was involved in the tumor process only in seven cases [9, 11, 15, 21, 37]. Layer F was distinguished specifically to describe the tumor spread in the cervical spine in detail in the amended WBB classification [71]; this layer is not described in the original article by Boriani S. et al.

En bloc resection was performed in 37 out of 101 patients. Intraoperative neurophysiological monitoring was used in 32 cases.

The involved vertebral artery was ligated and removed together with the tumor mass during the main stage of surgical treatment in 25 patients who had not previously undergone embolization [10–16, 19, 31–33, 40, 41, 43–46]. In 15 cases, the planned cessation of blood flow through the involved artery was performed without preliminary occlusion test and neurophysiological monitoring [9, 12, 14, 35–39, 43–46]. After removal of the involved part of the vertebral artery, three patients underwent its plasty with a venous autograft without specifying the indications [6]. Intraoperative damage to the artery occurred in seven cases: the artery was ligated in two cases, plugged in two, coagulated in one, and one patient underwent application of a vascular suture [7, 20, 47–49]. In another case, damage to the vertebral artery by a metal structure occurred after surgery and was manifested by convulsions and tetraparesis due to the detected ischemic lesion in the brain: a shunt was placed between the distal part of the damaged vertebral artery and the external carotid artery with a venous autograft, which allowed relieving the neurological symptoms [6]. Convulsive syndrome alone was noted in one case in the postoperative period after ligation of the vertebral artery [12].

In addition to the vertebral artery, from one to four involved spinal roots were ligated in 31 cases, with five patients having the procedure on both sides. No significant neurological complications

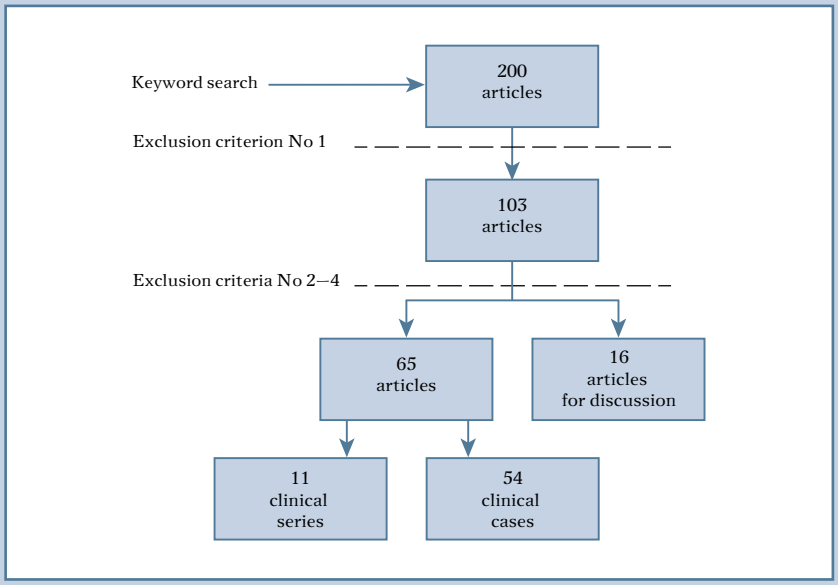

Fig. 1
The algorithm for selecting publications

Table 1
Clinical manifestations of spinal tumors in the region of vertebral artery

Clinical manifestations	Cases, n (%)
Cervicalgia	62 (61.4)
Radiculopathy	52 (51.5)
Myelopathy	19 (18.8)
Dysphagia	15 (14.9)
Stiffness	7 (6.9)
Torticollis	6 (5.9)
Dyspnoea	4 (4.0)
Headache	3 (3.0)
Dysphonia	2 (2.0)
Throat discomfort	1 (1.0)
Non-focal neurological symptoms, ataxia	1 (1.0)
Bow hunter syndrome	1 (1.0)

were noted upon ligation of the C1–C3 roots; three and two patients out of 14 had diaphragmatic palsy and pneumonia, respectively, upon C4 root ligation. As expected, ligation of the C5 and caudal roots was manifested by a corresponding neurological deficit. The observed postoperative complications are presented in Table 3.

The mean duration of postoperative follow-up was 30.3 months (min, 2; max, 169). In patients with angioplasty, blood flow was preserved in the area of reconstruction [6]. Four patients received radiation therapy for residual tumor (three chordomas, one giant cell tumor) [50–53], 10 patients had a relapse: among them, there were two cases of osteoblastoma and one case of epithelioid hemangioendothelioma (taken for follow-up examination) [20, 48, 54]; seven individuals were diagnosed with chordoma (three patients underwent radiation therapy [9, 13], two patients had repeated tumor resection [6, 40], and two patients died due to the disease progression [8]). Of the patients with metastatic lesions of the spine, three died in the absence of local tumor recurrence in the area of intervention [6], and one patient with residual tumor was taken for follow-up examination [38].

Discussion

The performed literature analysis demonstrates a small number of reports of cervical spine lesions due to a malignant process in the region of vertebral arteries: even fewer works consider this category of pediatric patients.

In contrast to older patients, in whom primary malignant and metastatic processes prevail, in childhood (from 0 to 17 years old) and juvenile age (from 18 to 29 years old) groups, there is a significant variation in histological types of tumors with a predominance of benign processes.

In case if the pathology is detected in vertebral arteries, it is advisable to perform angiography by any of the available methods. This will make it possible to plan the upcoming surgery [15, 30, 72]. However, the variability of the blood supply to the brain does not allow relying on angiography data only when deciding the fate of the vertebral artery: among the 101 cases considered in the literature, including 51 cases of temporary (occlusion test) or permanent blockage of the artery, ischemic brain disorders were noted only in three patients, which comprises 5.9 % [6, 12, 28]. As noted above, prehospital neurological symptoms were observed in only two patients [26, 27].

In our opinion, data from 16 publications may be of some interest. These articles summarize rather large clinical series, which we did not include in the analyzed sample due to their inconsistency with the parameters we estimated. However, these studies describe direct manipulations on the vertebral artery in patients with tumors. Some of these works are on successful preventive endovascular embolization [4, 73, 74],

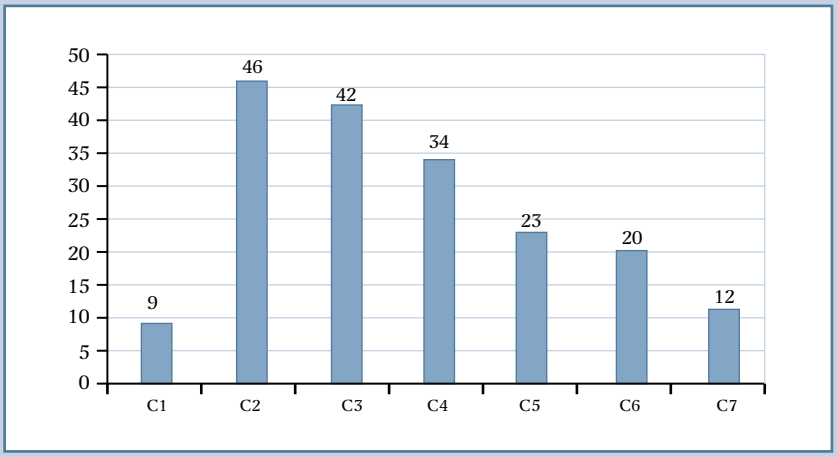

Fig. 2
Affected vertebrae (based on the results of 65 articles)

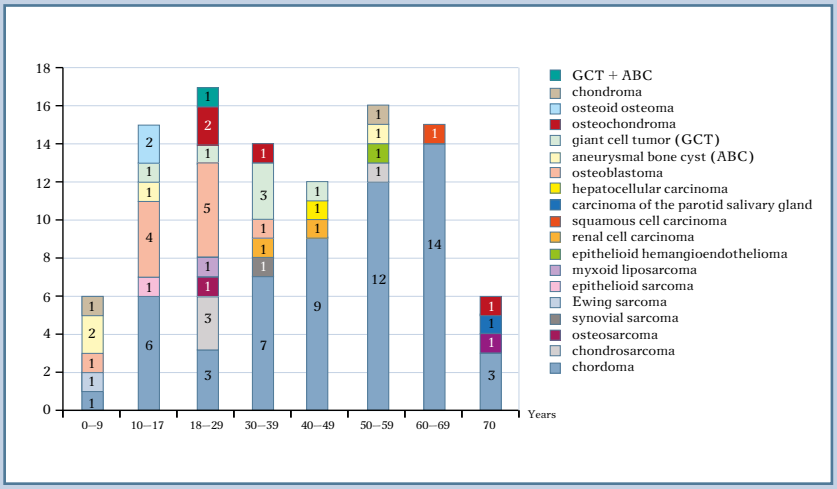

Fig. 3
Age distribution of histological types of tumors of the cervical spine

Table 2
Types of surgical interventions

Type of treatment	Patients, n (%)
Tumor resection using the combined approach with anterior and posterior stabilization	73 (72.3)
Tumor resection through the posterior approach with stabilization	3 (3.0)
Tumor resection through the posterior approach with stabilization and vertebroplasty	2 (2.0)
Tumor resection through the anterior approach with stabilization	7 (6.9)
Tumor resection	15 (14.8)
Vertebroplasty	1 (1.0)

while others mention late (up to 11 days) ischemic complications [75, 76]. Despite this, all authors recommend vertebral artery embolization as an effective and safe procedure.

Isolated cases of vertebral artery injury during tumor resection are also mentioned in the literature among general complications with the absence of neurological symptoms in the postoperative period [77–82]. Choi et al. [83] report the occurrence of an ischemic lesion in the brain in one case in a series of 97 patients. Hoshino et al. [84] carried out unilateral ligation of non-dominant vertebral arteries for treating recurrent chordoma during the occlusion test of the remaining vertebral artery, which was also involved in the tumor process. After the procedure, the authors observed loss of visual fields with restoration. For this reason, artery ligation and removal with bypass grafting between its distal fragment and external carotid artery was performed during repeated tumor resection. Some authors cite successful experience of using a surgical microscope to isolate and preserve the vertebral arteries of interest [8, 85, 86]. Mattei et al. [87] recommend conducting posterior instrumented fixation of the cervical spine at a separate stage, prior to any manipulations with the vertebral artery. The authors explain such choice by a possible risk of vertebral artery injury during placement of pedicle screws and when preserving blood flow in the basilar artery in case of complications. The above-mentioned data and complications correspond to those presented in the main publication sample.

Despite the low rate of complications in unilateral cessation of blood flow through the vertebral artery, their nature itself is disabling. Although a number of authors [4, 75] refer to a small sample and the occurrence of long-term complications and thus argue the effectiveness of the temporary occlusion test for predicting the ischemic consequences, its implementation using either endovascular or open approach with neurophysiological monitoring can be justified in order to avoid some acute neurological complications [2, 3, 6, 28, 83, 84].

Performing biopsy before the main stage of surgical treatment is optimal for understanding the nature of the process and choosing the right treatment strategy. The question of choosing the biopsy technique remains open and depends on the topography of the tumor, technical equipment, and the surgeon's skills.

Based on the experience of treating 67 patients with tumors in the region of vertebral artery, Westbroek et al. [88] proposed an algorithm that considers oncological (type of tumor, presence of metastases), mechanical (degree of vertebral artery involvement), and vascular (features of the circle of Willis and radiculomedullary arteries, bilateral involvement) criteria. The authors recommend using this algorithm when deciding on whether to sacrifice or isolate and preserve the involved vertebral artery. In our opinion, amendment of the algorithm with taking into account the possibility of carrying out vascular anastomoses presented by other researchers [6, 84] can make it possible to achieve a more radical resection of the tumor (if necessary) and improve the patient's prognosis.

Table 3
Types of postoperative complications

Complications	Patients, n
Neurological:	27 (26.7 %)
radiculopathy	16
paresis of the diaphragm	3
convulsions	2
cerebrospinal fluid leakage	2
tetraparesis	1
Horner syndrome	1
meningocele	1
dysphonia	1
Orthopedic:	6 (5.9 %)
migration of metal structures	3
neck stiffness	2
deformity	1
Wound:	12 (11.9 %)
wound defect	8
infection	3
hematoma	1
Others:	14 (13.9 %)
dysphagia	6
pneumonia	4
sepsis	1
gastrointestinal bleeding	1
pain syndrome	1
atrophy of *m. splenius capitis* (at the site of approach)	1

Conclusion

The presented literature analysis illustrates the complexity of choosing diagnostic and therapeutic strategies in patients with cervical spine tumors. The possibility of performing the diagnosis and surgery by a multidisciplinary team explains the advisability of concentrating such patients in specialized surgical centers. In addition, when planning such surgeries, technical actions for preventing possible bleeding and a rational approach to choosing the method of preventive embolization and managing intraoperative bleeding should be discussed, which will ultimately minimize the risk of rare but potentially severe complications, including the neurological ones.

Limitations on the reliability of the completeness of the literature review

1. A detailed analysis of the peculiarities of the disease course and treatment of spinal tumors located in the region of vertebral arteries in children is deliberately beyond the scope of this review. This age group is noted only in 14 publications containing 21 cases in total. Having our own experience comparable to the literature, we have decided to present an analysis of this age group in a separate publication.

2. Assessing the degree of the involvement of the vertebral artery based on the data presented in publications is very difficult, since only a few authors provide a description in the text, while the figures presented in the articles are not always informative.

3. The review does not provide the analysis of the peculiarities of surgical approaches (which depend on the level of cervical spine involvement and are described in detail in the literature) and the effectiveness of tumor treatment (according to the articles, it was carried out based on the protocols corresponding to the histological type).

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