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NONSPECIFIC CRANIOVERTEBRAL SPONDYLITIS: FEATURES OF SURGICAL TACTICS

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Objective. To evaluate the effectiveness of surgical treatment of nonspecific spondylitis in the area of craniovertebral junction taking into account the clinical and pathomorphological features of the disease.

Material and Methods. The study included 11 patients with nonspecific craniovertebral spondylitis: 4 women and 7 men aged 31 to 75 years. In 8 patients, the clinical picture was represented by neurological symptoms, and in 3 — by pain syndrome. Combined interventions were performed in 7 patients who had compression of the spinal cord and medulla oblongata. Of these, 5 patients first underwent transoral decompression, and then occipitospondylodesis. In the rest patients, the sequence of surgical treatment had the reverse order. In 6 cases, combined operations were performed simultaneously, and in one case — with an interval of 7 days. In one patient, the first stage was external ventricular drainage of hydrocephalus due to occlusion of the cerebrospinal fluid pathways by a displaced odontoid process, and the second stage was transoral decompression. Patients with pain syndrome underwent occipitospondylodesis. In one case, after combined surgery, ventriculoperitoneal shunting was performed for aresorptive hydrocephalus that developed after meningitis.

Results. In 10 patients, the pain intensity according to VAS decreased by 5–7 points (6.3 on average) in the postoperative period. Out of 8 patients with conductor symptoms, regression of neurological disorders after surgery was achieved in 3, complete recovery (Frankel E) in 4, and one patient died of purulent ventriculitis. Control examination of 10 patients confirmed the relief of the inflammatory process and the absence of compression of the spinal cord and medulla oblongata. Among the early postoperative complications, one case of postoperative liquorrhea and the divergence of the edges of the wound of the posterior pharyngeal wall were recorded. Fracture of metal structure elements was observed in one patient 3 years after surgery.

Conclusion. In nonspecific craniovertebral spondylitis, an active surgical tactic is justified. Occipitospondylodesis at an early stage of the disease allows to eliminate atlantoaxial instability and prevent the associated risk of neurological complications. In conduction disorders, if the patient's condition allows, simultaneous transoral decompression and craniocervical fixation is the best option for surgical treatment of nonspecific spondylitis of the craniovertebral region. Preservation of mobility in the cervical spine facilitates the transoral stage, therefore occipitospondylodesis is advisable to be performed at the second stage.

Key Words: nonspecific craniovertebral spondylitis; transoral decompression; occipitospondylodesis.

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Nonspecific spondylitis of the craniovertebral junction (CVJ) is an uncommon infectious spinal pathology that can lead to serious neurological complications, including death. Makins and Abbott first described the disease in 1896 using the findings of a pathological and anatomical study [1]. In the structure of purulent inflammatory pathology of the spine associated with nonspecific microflora, nonspecific CVJ spondylitis accounts for only 0.7 % [2]. The number of articles on this issue is extremely low in both domestic and foreign literature. All existing publications contain single cases or small series of observations of this disease [3–6].

According to some authors [5–10], the disease process is associated with hematogenous dissemination of infection; others believe that the cause for the development of nonspecific CVJ spondylitis is direct bacterial invasion from nearby inflammatory foci through vascular basins connecting the venous plexus of the pharynx with the upper cervical vertebrae.

Nonspecific purulent processes of the CVJ are diagnosed in the advanced stage of the disease in 50 % of patients, which is influenced by a number of both objective and subjective causes [5, 6, 10]. The late diagnosis results in a high incidence of neurological complications, ranging from 34 to 89 % of cases, and high mortality [3, 5, 6, 10, 11].

The location of the pathological process and its surgical accessibility, as well as the adjacency of vital parts of the central nervous system, define the complexity of surgical treatment of nonspecific CVJ spondylitis.

The objective is to evaluate the effectiveness of surgical treatment of nonspecific spondylitis in the area of the craniovertebral junction, taking into account the clinical and pathomorphological features of the disease.

Material and Methods

The surgical outcomes of 11 patients (4 women and 7 men) aged 31 to 75 years (mean 56.6 years) with nonspecific CVJ spondylitis were included in the retrospective (period from 2008 to 2024) analysis. The duration of the disease from the manifestation of the first symptoms to the developed clinical picture ranged from 4 to 16 weeks (mean 9.09 weeks).

The clinical examination included assessment of the degree of neurological disorders according to the Frankel scale and pain syndrome according to VAS. The diagnosis was confirmed by radiography of the spine, MRI, and CT data. Transoral needle biopsy to identify the causative agent was performed in two patients with pain syndrome; in one case, it was positive. In case of long-tract signs in patients, this diagnostic procedure was not performed because of time limitations. The basis of laboratory diagnostics and monitoring of the activity of the infectious process was bacteriological examination of blood, determination of white blood cell count, ESR level, C-reactive protein, and procalcitonin. Laboratory and bacteriological examination of spinal fluid was performed in patients with meningitis. Basic information about clinical symptoms, duration of the disease, risk factors, and sources of infection is given in Table 1.

Patients with pain syndrome underwent occipitospondylodesis. The combined surgeries were performed in seven patients with neurological deficit; five of them underwent transoral decompression first, followed by occipitospondylodesis; in two cases, the surgical treatment was performed in the reverse order. The combined surgeries were performed simultaneously in five patients and with an interval of 7 days in one patient because of the severity of the condition. A patient with depression of consciousness to sopor associated with obstructive hydrocephalus due to inclination of the odontoid process into the great occipital foramen underwent an external ventricular drainage as a first treatment stage and transoral removal of the odontoid process 5 days later. Ventriculoperitoneal shunting was performed in one patient concerning postoperative aresorptive hydrocephalus associated with passed purulent meningitis.

During the decompression stage, bacteriologic, microscopic, and histopathological studies of the biopsy specimen were mandatory. The specific origin of the disease was excluded in all patients according to the results of laboratory tests. The duration of antibiotic therapy in patients with identified pyogenic microflora was 4 weeks. Empiric therapy with broad-spectrum antibiotics was performed for 8 weeks in two patients with an undiagnosed pathogen. The course of clinical symptoms, laboratory markers of inflammation, and data of instrumental methods of examination were monitored to evaluate the effectiveness of treatment in the postoperative period.

Results

A detailed study of preoperative radiography of the spine, CT, and MRI data in 11 patients revealed the cause-effect relations of the ongoing inflammatory changes and the main features of the pathomorphology of nonspecific CVJ spondylitis, which are shown in Table 2.

Despite the variety of pathomorphologic manifestations, CT and MRI findings revealed destruction of one of the paired atlantoaxial joints in all patients, the mechanism of occurrence of which is identical to the pathogenesis of nonspecific spondylodiscitis. The hematogenous route of dissemination was the most likely source of infection since the patients had no history of inflammatory processes of orofacial location. According to this circumstance, it is the purulent arthritis of the lateral atlantoaxial joint that triggered the chain of pathologic transformations typical of nonspecific spondylitis of the CVJ. Subsequent changes were already a consequence of the spread of infection from the joint cavity to the osteoligamentous structures of the CVJ, to the epidural and retropharyngeal space. In four cases, there was an ipsilateral lesion of the atlantooccipital joints, which can be explained by the contact mechanism of infection spread from the atlantoaxial joint cavity because of destruction of the lateral mass of the atlas.

According to analysis of the radiography of the spine, MRI, and CT data, all patients showed atlantoaxial dislocation, and basilar impression resulted from lysis of the capsular ligamentous apparatus of the CVJ. Moreover, only three patients had significant dislocations of the spinal cord and medulla oblongata as the direct cause of gross compression.

As a consequence of purulent fusion of the atlantoaxial joint, rotational subluxation of the atlas was also detected in five patients. In these cases, atlantoaxial dislocation and basilar impression had maximum values.

Osteomyelitis of the odontoid process was found in eight patients, the pathomorphologic pattern of which was represented by marginal destruction and lytic lesions in the trabecular bone.

Retropharyngeal abscesses were detected in six cases. Epidural abscesses were the cause of compression in the majority of patients with neurological complications.

Postoperative follow-up periods ranged from 4 to 36 months. The main data on patients and surgeries performed are given in Table 3. The assessment of surgical treatment outcomes is given in Table 4. During the follow-up period, the pain intensity decreased in 10 patients on mean by 6.5 points according to VAS. Seven patients with spinal cord and medulla oblongata compression achieved regression of conduction disorders in the postoperative period; four of them showed complete recovery.

One patient died eleven days after transoral resection of the odontoid process because of purulent ventriculitis that developed after external drainage of the lateral ventricle.

Control examination of patients with conductor symptoms in the early postoperative period confirmed complete decompression of the spinal cord. According to CT and MRI data in the distant postoperative period, inflammatory changes have been suppressed (Fig. 1, 2). The remodeling of the bone structures of the CVJ was noted in two patients 24 and

Table 1
Basic information about clinical symptoms, duration of the disease, risk factors, and sources of infection in patients in the analyzed sample

Feature	Values
Mean age, years ± standard deviation	56.6 ± 14.02
Men, n (%)	7 (63.63)
Women, n (%)	4 (36.36)
Clinical symptoms at admission, n (%)	
Pain and stiffness in the neck	11 (100.00)
Torticollis	5 (45.45)
Hyperthermia	5 (45.45)
Conduction disorders	8 (72.72)
Neurogenic pelvic dysfunction	8 (72.72)
Respiratory disorders	2 (18.18)
Obstructive hydrocephalus	1 (9.09)
Purulent meningitis	1 (9.09)
Purulent ventriculitis	1(9.09)
Dysphagia	4 (36.36)
Mean duration of disease, weeks ± standard deviation	9.09 ± 4.52
Risk factors, n (%)	
Diabetes mellitus	5 (45.45)
Chronic renal failure	1 (9.09)
HIV	1 (9.09)
Source of infection, n (%)	
Intravenous drug use	3 (27.27)
Soft tissue infection	3 (27.27)
Urinary tract infection	1 (9.09)
Source not identified	4 (36.36)

36 months after occipitospondylodesis (Fig. 3).

The course of the early postoperative period in one patient was complicated by cerebrospinal fluid leakage from the dura mater defect, which was sealed with a free muscle graft and biologic adhesive, layered closure of the posterior pharyngeal wall, and lumbar drainage for seven days. After meningitis, the patient subsequently developed aresorptive hydrocephalus for which a ventriculoperitoneal shunting was performed. In one case, there was a divergence of the wound edges of the posterior pharyngeal wall, which healed by secondary intention. 36 months after craniocervical fixation, one patient had a fracture of the rods of the implanted instrumentation, which was removed.

Discussion

Nonspecific purulent pathology of the CVJ has been indicated by various terms in the literature to this day. Such diagnoses as osteomyelitis of the odontoid process, epidural abscess of the upper cervical spine, osteomyelitis of the odontoid process with epidural abscess, and atlantoaxial instability associated with purulent lesions of the upper cervical spine are limited to the definition of

Table 2
The main features of the pathomorphology of nonspecific spondylitis of the craniovertebral junction

Patient	Purulent arthritis of the AAJ	Purulent arthritis of the AOJ	Osteomyelitis of the odontoid process	AAD	BI	Rotational subluxation of the atlas	Epidural abscess	Retropharyngeal abscess
1	+	–	+	+	–	–	–	–
2	+	+	+	+	+	–	–	+
3	+	–	–	+	+	–	–	–
4	+	–	+	+	+	+	–	–
5	+	–	+	+	+	–	+	+
6	+	+	+	+	–	–	+	–
7	+	–	–	+	+	–	+	+
8	+	+	+	+	+	+	+	+
9	+	+	+	+	–	+	+	+
10	+	–	–	+	+	+	–	–
11	+	–	+	+	+	+	–	+

AAJ – atlantoaxial joint (paired); AOJ – atlantooccipital joint; AAD – atlantoaxial dislocation; BI – basilar impression.

Table 3
Basic information about the patients included in the study and the interventions performed

Patient	Gender	Age, years	Duration of disease, weeks	Clinical picture of disease	Compression trigger	Surgery	Microflora	Duration of antibiotic therapy, weeks	Follow-up period, months	Outcome
1	M	47	16	Pain	–	Occipitospondylodesis	–	8	12	Complete recovery
2	M	67	12	Pain, dysphagia	–	Occipitospondylodesis	–	8	36	Complete recovery
3	M	31	10	Pain	–	Occipitospondylodesis	*St. haemoliticus*	4	6	Complete recovery
4	F	68	8	Tetraplegia, respiratory distress, mechanical ventilation	AAD, BI	1. Transoral decompression 2. Occipitospondylodesis	*E. faecium*	4	24	Improvement
5	F	75	10	Pain, dysphagia, tetraparesis	Epidural abscess	Occipitospondylodesis + transoral decompression	*St. aureus, Klebsiella pneumoniae*	4	6	Improvement
6	F	37	4	Pain, tetraparesis	Epidural abscess	Occipitospondylodesis + transoral decompression	*St. haemoliticus*	4	6	Complete recovery
7	F	60	5	Pain, dysphagia, tetraparesis	Epidural abscess	Transoral decompression + occipitospondylodesis	*St. haemoliticus, Str. pyogenes D*	4	12	Complete recovery
8	M	72	18	Pain, dysphagia, tetraparesis	Epidural abscess	Transoral decompression + occipitospondylodesis	*MRSA*	4	6	Complete recovery
9	M	54	5	Pain, tetraparesis, meningitis	Epidural abscess	1. Transoral decompression + occipitospondylodesis 2. Ventriculoperitoneal shunting	*Str. constellatus*	4	4	Improvement
10	M	46	8	Pain, tetraparesis, obstructive hydrocephalus	AAD, BI	1. External drainage of the lateral ventricle 2. Transoral decompression	*MRSA*	2	–	Death
11	M	66	4	Pain, tetraparesis	AAD, BI	Transoral decompression + occipitospondylodesis	*MRSA*	4	4	Complete recovery

AAD – atlantoaxial dislocation; BI – basilar impression.

Table 4
Results of surgical treatment of patients in the study sample

Patient	VAS		Frankel	
	Before surgery	After surgery	Before surgery	After surgery
1	7	0	E	E
2	8	1	E	E
3	8	2	E	E
4	6	1	B	D
5	7	0	C	D
6	6	0	C	E
7	6	1	D	E
8	7	0	C	E
9	8	1	C	D
10	8	–	B	–
11	8	0	D	E

only individual manifestations of the disease without reflecting the etiology, pathogenesis, and morphofunctional details of purulent pathology of the CVJ [3, 4, 8–14]. The data obtained by us from radiological diagnostic techniques that explain the cause-effect relations between the inflammatory changes can be used to assert that the disease essence is most fully conveyed by the term nonspecific spondylitis of the CVJ. This reflects the involvement of both upper cervical spine and skull base structures in the pathologic process and also unites the mechanism of onset and development of the disease and its individual manifestations [5, 6].

World experience in the treatment of nonspecific spondylitis of the CVJ is based on single cases or small series of observations of this rare disease. According to the literature [3, 6, 8], the conservative treatment of purulent inflammatory pathology of the CVJ was the main option until the end of the 1980s. The high incidence of neurological complications and high mortality that occurred in this case became the reason for the introduction of surgical treatment, which was greatly contributed to by the rapid development and improvement of health technology.

Nowadays, conservative treatment of nonspecific spondylitis of the CVJ is indicated at the initial stage of the disease, when the symptoms and signs are either pain syndrome or mild pyramidal insufficiency. According to the literature [3, 15–18], antibiotic treatment, chosen empirically or according to the results of bacteriologic examination of blood or punctate, is performed from 6 to 12 weeks. The evaluation of the successful conservative treatment is based on the changes in pain syndrome, laboratory markers of inflammation, and data of instrumental methods of examination.

Transoral puncture biopsy, introduced into practice to verify the causative agent, has become an irreplaceable diagnostic study and significantly strengthened the possibilities of conservative treatment. A number of authors used puncture aspiration of pus for debridement and decompression of epidural abscesses of the CVJ. However, this technique has not been widely used as a therapeutic procedure because of its questionable efficacy, along with the unconditional hazard of the procedure [5, 6, 8, 13, 15].

Atlantoaxial instability is a typical pathomorphologic manifestation of nonspecific purulent pathology of the CVJ in consequence of destruction of osteoligamentous structures. The size of the dislocation is usually not reach significant values, so atlantoaxial dislocation is rarely a direct cause of compression of the spinal cord and medulla oblongata. In most cases, the onset of neurological deficit is associated with the formation of epidural abscesses due to atlantoaxial instability [8, 18, 19].

Since compression of the spinal cord and medulla oblongata in nonspecific spondylitis of the CVJ poses a threat of severe neurological complications, up to mortality, manifestation and progression of conduction disorders are an absolute indication for surgical treatment, the results of which directly depend on the timing of surgery. For example, according to Kobayashi et al. [18], in the group of patients operated on for nonspecific purulent pathology of the CVJ before the development of conduction disorders, persistent neurologic deficit and mortality were observed in 3.8 % of cases, and in patients operated on after, in 50 % of observations, which is strong evidence of the advantage of active surgical strategy [3, 8].

Moreover, failure of conservative treatment evaluated by clinical, laboratory, and instrumental data is considered the indication for surgical treatment. During the chronic stage of the disease, the indication for surgical treatment is also formed atlantoaxial instability associated with lysis of the capsular ligamentous apparatus of the CVJ [3–6, 8, 20, 21].

The surgical treatment strategy for nonspecific spondylitis of the CVJ is still a controversial topic because of the rarity of the disease and the complexity of surgical interventions, which require specific surgical skills and relevant experience

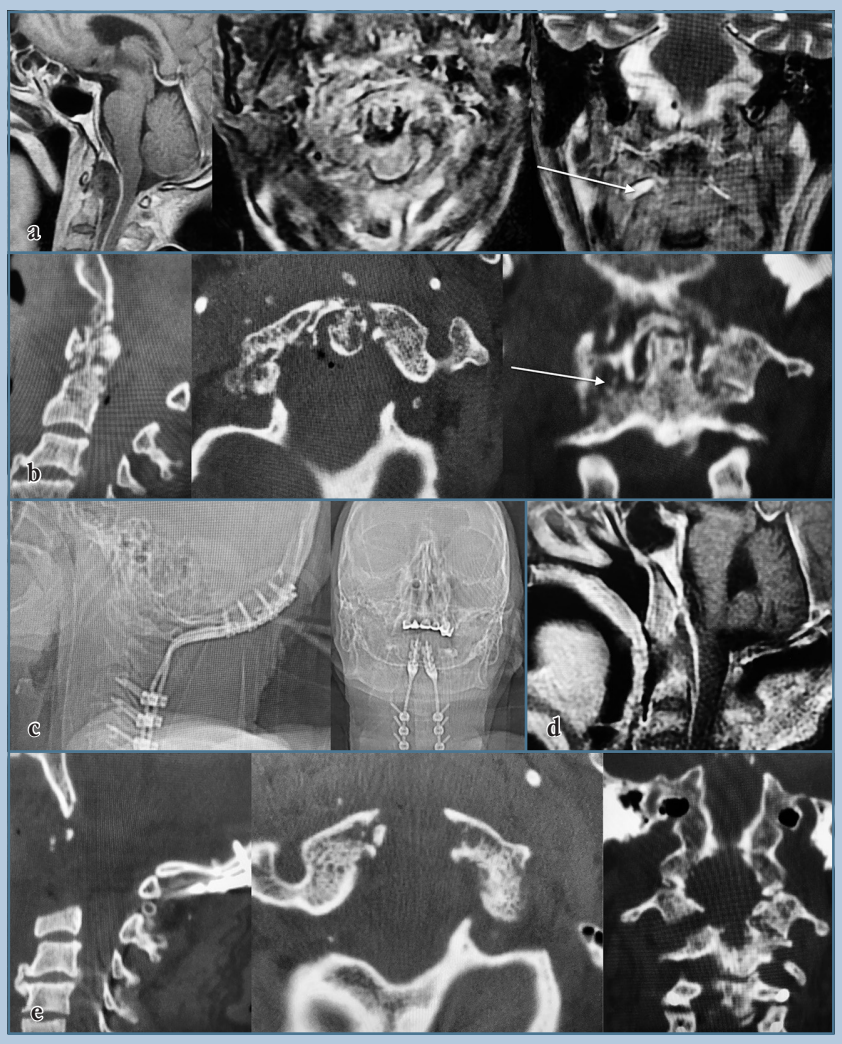

Fig. 1
Examination results of a patient before and after surgery: **a** – MRI scans in sagittal, axial and frontal planes show nonspecific spondylitis of the craniovertebral junction with severe compression of the spinal cord and medulla oblongata by epidural abscess, rotational subluxation of the atlas, purulent arthritis of the atlantoaxial and atlantooccipital joints (arrow); **b** – CT scans in sagittal, axial and frontal planes show osteomyelitis of the odontoid process, purulent arthritis of the right atlantoaxial and atlantooccipital joints (arrow) with the presence of atlantoaxial dislocation and basilar impression; **c** – radiography of the spine after transoral decompression and occipitospondylodesis in lateral and frontal planes; **d** – control MRI scans in sagittal plane 4 months after combined surgical treatment; **e** – control CT scans in sagittal, axial and frontal planes

[3, 5, 6, 8]. Until now, surgical treatment of purulent pathology of the CVJ has been associated with a high incidence of postoperative complications, reaching 18–26 % [5].

Surgical treatment of nonspecific purulent pathology of the CVJ has its history since the late 1980s, when some researchers made the first attempts to treat the purulent focus and decompress the spinal cord and medulla oblongata using anterior cervical and transoral approaches [4]. As experience has shown, the anterior approach to the cervical spine is associated with a number of technical challenges due to the angle of attack when performing this surgery and the deficiency of adequate visual control during resection of osteoligamentous structures. In this connection, the anterior cervical approach in its conventional version is currently practically not used in the surgical treatment of nonspecific spondylitis of the CVJ. Nevertheless, single cases of purulent focus debridement and decompression of the spinal cord and medulla oblongata through a minimally invasive anterior cervical approach using a tubular technique and microscope have been described in the literature [22].

The transoral technique is widely regarded as the gold standard in upper cervical spine surgery. Even though it has been known for a long time, it has not been easy to put it into practice. The first report of the use of the transoral approach was published back in 1917. In 1957, much later, Southwick and Robinson removed a C2 vertebral tumor from this approach [23]. In 1962, Fang et al. gave a detailed description of the transoral approach, which they used in the surgical treatment of rigid atlantoaxial dislocations, including tuberculous lesions of the CVJ [24]. The transoral approach to the upper cervical spine has obvious advantages over the anterior cervical approach and its minimally invasive modifications, as it provides a direct angle of attack, freedom of manipulation, and good visual control at all stages of surgery. Despite the undeniable advantages of the approach, the high incidence of complications at the first stages, primarily due to the absence of technical conditions for performing such surgeries, limited the use of this technique for a long time. The improvement of the surgical technique and the mastering of modern medical technologies, including the use of the surgical

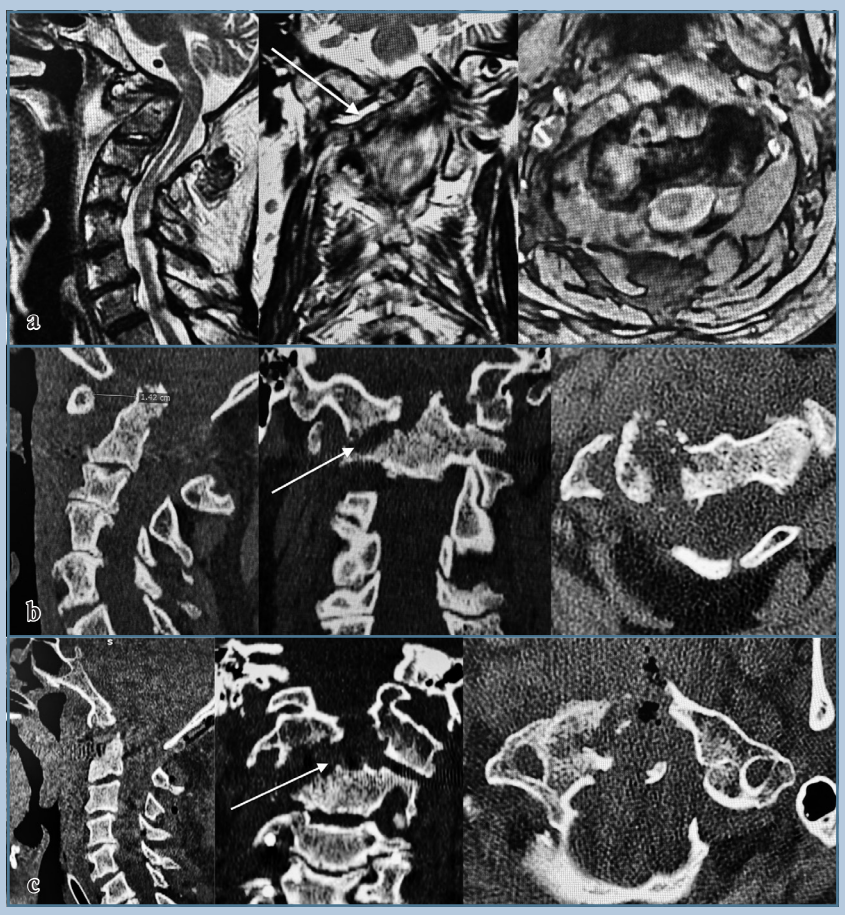

Fig. 2
Examination results of a patient before and after surgical treatment: **a** – MRI scans in sagittal, axial and frontal planes show nonspecific spondylitis of the craniovertebral junction with compression of the spinal cord and medulla oblongata, displaced odontoid process, purulent arthritis of the atlantoaxial joint (arrow) and rotational subluxation of the atlas; **b** – CT scans in sagittal, axial and frontal planes show significant atlantoaxial dislocation and basilar impression, osteomyelitis of the odontoid process, purulent arthritis of the lateral atlantoaxial joint (arrow) and rotational subluxation of the atlas; **c** – CT scans in sagittal, frontal and axial planes after combined intervention show transoral resection of the odontoid process and atlantoaxial joint (arrow), restoration of anatomical relationships in the craniovertebral junction

microscope, have reduced the incidence of complications and introduced the transoral approach into widespread practice, including surgical treatment of nonspecific purulent pathology of the CVJ [5, 6, 23].

During the last two decades, there has been a rapid development of endoscopic surgery of the skull base and the CVJ. Given this background, the endoscopic transnasal approach is successfully used for decompression of the spinal cord and medulla oblongata during surgical treatment of nonspecific spondylitis of the CVJ [25, 26].

Atlantoaxial instability is one of the topical issues of nonspecific purulent pathology of the CVJ; to eliminate it, both external immobilization and various modes of posterior fusion of the C1–C2 vertebrae as well as occipitospondylodesis with cerclage wire and autogenous bone graft were used. A significant progress in the surgical treatment of nonspecific spondylitis of the CVJ was achieved at the beginning of this century owing to the proposed instrumentation techniques that have made it possible to perform primary stable atlantoaxial or craniocervical fixation. Nowadays, occipitospondylodesis has become an imperative procedure in the surgical treatment of nonspecific spondylitis of the CVJ. It is used as an independent technique to prevent compression complications. In cases of severe spinal cord and medulla oblongata compression, the combination of occipitospondylodesis with ventral decompression surgeries has become a routine practice [4–6, 8, 14, 15, 20, 21].

Various opinions are given in the literature about the staging of combined surgical treatment [4–6, 14, 20, 21, 25]. Nowadays, combined surgical treatment is performed simultaneously or with an interval between stages ranging from 10 days to 6 months. Simultaneous performance of anterior decompression and craniocervical fixation is distinguished by its long duration and injury rate, while two-stage surgical treatment is associated with a high risk of neurological complications. The views on the order of surgeries were also divided. One strategy includes first performing a ventral decompression followed by occipitospondylodesis, the other one involves the reverse sequence.

Our choice of an active surgical approach for the treatment of nonspecific spondylitis of the CVJ is conditioned by the high rate of development of severe neurological complications. Occipitospondylodesis at the early stage of the disease in three patients allowed preventing atlantoaxial instability and implementing antibiotic treatment until clinical improvement and normalization of inflammatory markers in the blood.

The combination of epidural abscesses and atlantoaxial instability was the

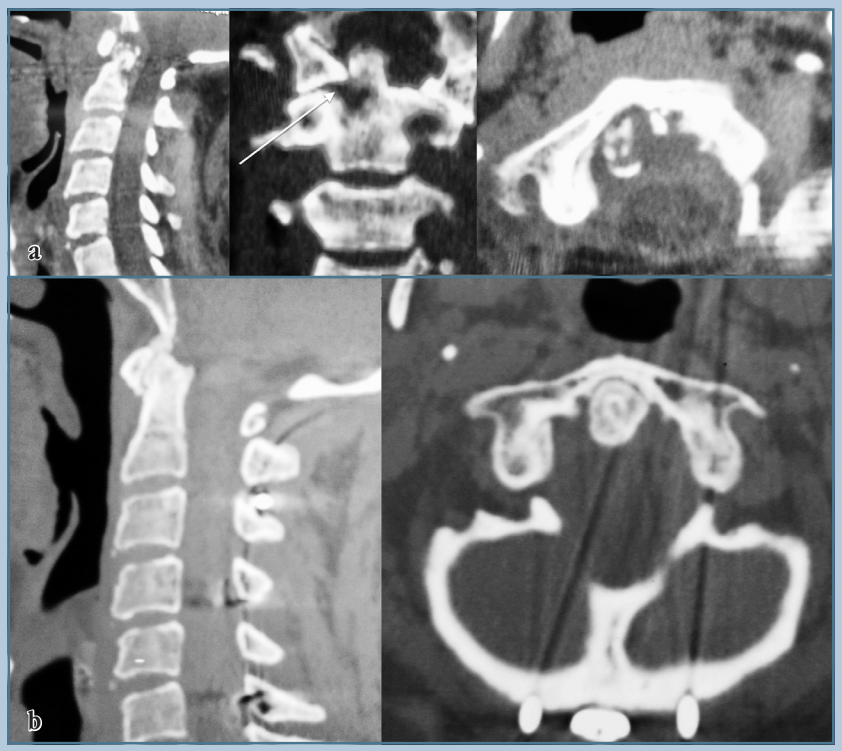

Fig. 3
Examination results of a patient before and after surgical treatment: **a** – CT scans in sagittal, frontal and axial planes show osteomyelitis of the odontoid process, arthritis of the atlantoaxial joint (arrow); **b** – control CT scans 3 years after occipitospondylodesis in sagittal and axial planes show remodeling of bone structures of the craniovertebral junction

main cause of conduction disorders in the majority of patients in our series; only three patients suffered from spinal cord and medulla oblongata compression associated with dislocated odontoid process. Both in one and in the other case, debridement of the site of infection and complete ventral decompression represented the main target of surgical treatment, the implementation of which was possible only via a transoral approach. Despite the duration and injury rate, in our opinion, the simultaneous performance of transoral decompression and occipitospondylodesis is the most appropriate surgical strategy for nonspecific spondylitis of the CVJ in the development of neurological complications; its application is limited only by the patient's condition. According to our experience with simultaneous surgeries, preserving mobility in the cervical spine is of great importance when performing the transoral stage. This is associated with the fact that overextension of the head during surgery eliminates atlantoaxial dislocation and simplifies the removal of the C2 odontoid process, and therefore occipitospondylodesis should be performed at the second stage.

Conclusion

An early detection of nonspecific purulent pathology of the CVJ is the guarantee of successful treatment. If nonspecific spondylitis of the CVJ is detected, active surgical strategy is justified. Occipitospondylodesis at an early stage of the disease allows to eliminate atlantoaxial instability and prevent the associated risk of neurological complications. If conduction disorders develop, simultaneous transoral decompression and craniocervical fixation is the most appropriate approach for surgical treatment of nonspecific spondylitis of the CVJ, provided that the patient's condition allows it. Preservation of mobility in the cervical spine facilitates the transoral stage; therefore, occipitospondylodesis is advisable to be performed at the second stage.

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The study was approved by the local ethics committees of the institutions. All authors contributed significantly to the research and preparation of the article, read and approved the final version before publication.

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