



VERTEBROBASILAR INSUFFICIENCY DUE TO OSSEOUS ABNORMALITIES OF THE CRANIOVERTEBRAL JUNCTION

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Objective. To study pathogenetic significance of bone abnormalities of the craniovertebral junction (CVJ) for the insufficiency of the vertebrobasilar circulation by analyzing the long-term outcomes of surgical treatment and comparing them with preoperative data of patients.

Material and Methods. Long-term results of surgical treatment of 49 patients operated on for bone abnormalities of the CVJ in the period from 1985 to 2013 who had strongly proved clinical and instrumental signs of vertebrobasilar insufficiency (VBI) and responded the questionnaire. Decompression of the brain and vertebral arteries was performed through transpharyngeal approach developed by authors. Eleven patients with Kimmerle anomaly underwent resection of abnormal bone bridges on the arch of the atlas and arteriolysis of the vertebral artery.

Results. Surgery for craniovertebral abnormalities provided the improvement in 84.2 % of operated patients. Excellent and good results were observed in 12 patients (75.0 %) with unstable abnormalities and in 6 patients (27.3 %) with stable abnormalities ($p < 0.01$). All patients with Kimmerle anomaly got good to excellent treatment outcomes.

Conclusion. Decompression of the brain and vertebral arteries through transpharyngeal approach is a pathogenetically justified method of treatment of patients with vertebrobasilar insufficiency due to craniovertebral abnormalities.

Key Words: craniovertebral abnormalities, vertebrobasilar insufficiency, Kimmerle anomaly.

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Among all cerebrovascular disorders, ischemic stroke in the vertebrobasilar vascular pool accounts for 25–30 % of cases, and transient ischemic attacks – 70 % [2, 3, 5, 11, 13]. The latter are followed by ischemic stroke in half of the cases and therefore they are considered as potential stroke condition.

Recurrent acute cerebrovascular accidents occurs in 40 % of patients with the consequences of ischemic stroke in the vertebrobasilar system [4, 6, 13, 14]. Compression of the vertebral artery, its stenosis or occlusion can cause transient or persistent deficiency of blood supply to critical brain structures, such as brainstem, cerebellum, occipital and temporal lobes, and other important brain structures. It is easier to prevent brainstem stroke than to treat its consequences.

The possibilities of pathogenetic treatment of stenotic lesions of the vertebral arteries are less explored compared to

those of carotid arteries. The pathogenetic role of craniovertebral abnormalities in the development of vertebrobasilar insufficiency is also poorly understood.

The objective of this research is to study the pathogenetic relationship between the vertebrobasilar ischemic insufficiency and bone abnormalities of the craniovertebral junction by analyzing the long-term outcomes of surgical treatment and comparing them to preoperative data of patients.

Material and Methods

More than 600 patients with various forms of vertebrobasilar insufficiency were operated on at the Novokuznetsk neurosurgical clinic since 1965, including 202 patients with abnormalities of the vertebral arteries and surrounding anatomical structures. We obtained data of 49 patients, who were operated

on during the period from 1985 to 2013 for vertebrobasilar insufficiency due to bone abnormalities at the craniovertebral junction and responded to our questionnaire. Some of them (20.4 %) underwent on-site examination. Among the examined patients, there were 11 patients with Kimmerle anomaly, which has its characteristic clinical and pathogenetic features.

The following inclusion criteria were used in this study: patients with craniovertebral abnormalities necessarily accompanied by apparent clinical and instrumental signs of vertebrobasilar ischemic insufficiency, who required surgical treatment. The main exclusion criteria were as follows: no symptoms of vertebrobasilar insufficiency, only signs of anomalies alone and local cerebral symptoms depending on the location of spinal cord and medulla oblongata compression site.

We used clinical, neurological, neuro-ophthalmological, otoneurological, neurophysiological (EEG, SSEP, EMG), ultrasound (duplex scanning of the brain vessels, TCD), and radiodiagnosis (craniography, spondylography, SCT, SCT-angiography, MRI, digital selective angiography) survey methods. Functional status of patients and the results of treatment were quantitatively assessed using conventional scales and Nurick, Rankin, and Macnub questionnaires. Elements of these scales were included in the questionnaires. We used the common national classification of cerebrovascular ischemic diseases proposed by EV Schmidt [11]. We studied clinical and pathogenetic features of three main types of vertebrobasilar insufficiency (chronic vertebrobasilar insufficiency or discirculatory encephalopathy, transient ischemic attack, ischemic stroke) and their relationship with abnormalities in order to evaluate the differentiated approach to their diagnosis and pathogenetic treatment. Particular attention was paid to comparing neurological symptoms with angiographic data and description of intraoperative relationships between the vertebral artery and pathological substrate.

Types of craniovertebral abnormalities and age of patients at the time of surgical treatment are shown in Table. Thus, 30.6 % of patients were operated on at the age below 20, 65.3 % – below 30 years. This is indicative of early clin-

ical manifestation of craniovertebral abnormalities.

As can be seen from Table, there were twice as many males as females. Patients with stable craniovertebral abnormalities accounted for 63.3%, unstable – 36.7 % ($p < 0.05$).

The indications for surgery included progressive vertebrobasilar insufficiency due to craniovertebral abnormalities, especially recurrent episodes of transient ischemic attacks, and combination of ischemic and compression brainstem disorders with intracranial hypertension. In patients with Kimmerle anomaly, surgery was performed in the case of drop-attack episode, transient ischemic attacks, and syncopal attacks associated with rotation of the head, but only in the case of stenosis of the vertebral artery in the abnormal ring, arising from formation of the periarterial cicatricial knot, verified by functional angiography.

In the case of fixed craniovertebral anomalies, we performed radical elimination of the ventral compression of the brain and vertebral artery by resection of the *odontoid-clival* conglomerate or the odontoid process through the transpharyngeal approach (Fig. 1).

Resection of bone structures supporting the craniovertebral junction was preceded by reliable occipitospondylodesis using original nickel-titanium (Ti-Ni) structures having shape memory [7–9]. In the case of dynamic compression of the neurovascular structures by odontoid

process, we conducted fusion using Ti-Ni structures in position of atlantoaxial correction. In the case of Kimmerle anomaly, we resected abnormal bone bridges on the posterior arch of the atlas. Separation of the vertebral artery from the cicatricial tissue was an essential phase of intervention. Only after this procedure, the diameter of the artery increased and vessel pulsation improved.

Results

In patients with insufficiency of the anterior craniovertebral supporting complex (odontoid bone, agenesis of the odontoid process, agenesis of the transverse ligament of the atlas, atlas assimilation) we diagnosed dynamic (in 4 patients) or fixed (in 14 patients) atlas subluxation involving the head (Fig. 2), accompanied by compression of the medulla oblongata, spinal cord, and vertebral arteries by the odontoid process.

Persistent severe compression of the neurovascular structures located in the occipitocervical dural funnel was observed in the case of craniovertebral abnormalities (basilar invagination, basilar impression, and convexobasia), wherein the angle between the Blumenbach's clivus (body of the occipital bone) and the anterior wall of the spinal canal is decreased. This craniovertebral angle normally ranges 120–140°. In the case of

Table

Distribution of patients according to their sex, age and type of craniovertebral abnormality, n

Abnormality type	Sex		Age, years				
	M	F	under 20	21–30	31–40	41–50	Older than 50
Basilar impression and convexobasia	10	4	7	4	2	1	—
Basilar invagination	4	2	2	2	2	—	—
Assimilation of the atlas with head luxation	3	1	3	—	1	—	—
Aplasia of the odontoid process	2	1	1	2	—	—	—
Odontoid bone	5	4	2	5	1	1	—
Aplasia of the transverse ligament of the atlas	2	—	—	1	—	—	1
Kimmerle anomaly	7	4	—	3	4	3	1

basilar impression, craniovertebral angle may be reduced to 90° (Fig. 3).

These abnormalities are accompanied by bending of the brain over the craniovertebral angle (odontoid-clival complex). Along with this, intracranial portion of the vertebral arteries may be compressed near to the point, where they merge to form the main artery (Fig. 3).

Clinical symptoms consisted of two components: the symptoms of the craniovertebral abnormalities themselves and clinical signs of the vertebrobasilar insufficiency. Subjective symptoms of the craniovertebral abnormalities themselves were represented by occipitocervical pain, especially during head movements, general weakness, paresthesias in the limbs or the whole body, balance disorders, and dizziness. Dysraphic signs were detected in almost all patients.

Severe preoperative neurological disorders were observed. Severe tetraparesis on the edge of plegia, which required permanent patient care, was observed in 22.4 % of cases; four of them had severe respiratory failure, patients spoke in a whisper because of the weakness of the respiratory muscles and could not swallow liquid food. Moderate tetraparesis was observed in 46.9 % of patients, mild – in 30.7 %. Most patients had intermittent progressive course of the disease.

Vertebrobasilar insufficiency was characterized by a number of clinical signs that were indicative of dysfunction of the brain structures distant from the compression site. These are, above all, mid-brain symptoms detected in 77.6 % of patients, symptoms of ischemia of the occipital lobe (8.2 %) and nuclei of oral cranial nerves (28.6 %), as well as dysfunction of the superior ($n = 3$) and even medium ($n = 2$) cervical spinal segments. Remote brainstem signs included Parinaud ($n = 5$) and Hertwig – Magendie ($n = 2$) symptoms, insufficiency of the oculomotor nerve ($n = 7$), systemic supranuclear dizziness ($n = 26$), and Wallenberg – Zakharchenko syndrome ($n = 5$). The signs of ischemia of the brainstem also included blood pressure instability described by Unterharnscheidt [8], since it was eliminated in 11 of 12 operated patients. Patients suffered of

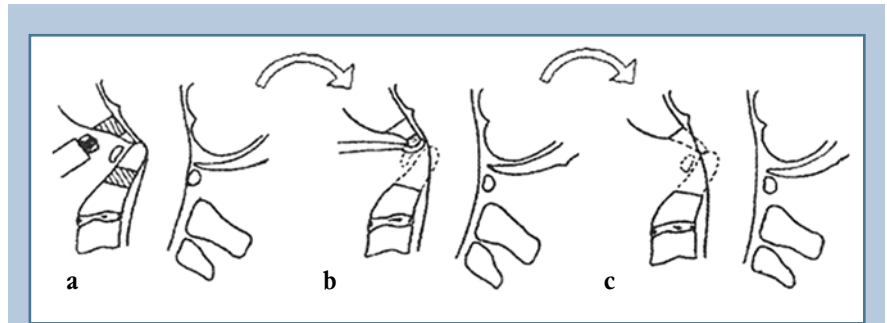


Fig. 1

Stages of resection of the odontoid-clival complex:

a – drilling holes in the base of the odontoid process and in the lower portions of the Blumenbach's clivus; **b** – resection of the remaining bony septum between these holes; **c** – excision of the cicatricial tissue, meningoysis followed by dural sac spread

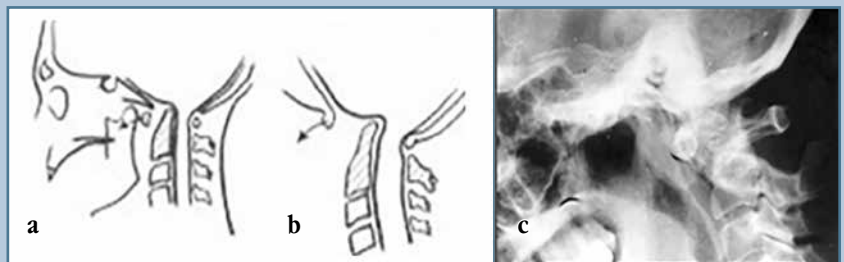


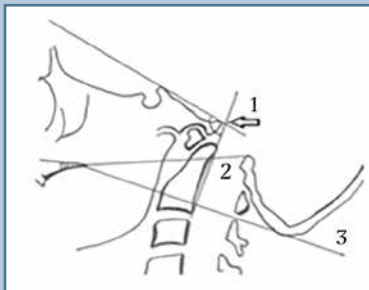
Fig. 2

Compression of the brain and its major vessels in patients with the anterior luxation of the atlas caused by aplasia of the transverse ligament of the atlas (**a**), assimilation of the atlas (**b**), and odontoid bone (**c**), when there is stable or dynamic compression of the neurovascular structures of the occipitocervical dural funnel

tinnitus, subjective hearing loss, and dizziness. They also had nystagmus, staggering, instability in the Romberg test. Bulbar paralysis symptoms were observed in 36.7 % of patients. In a half of cases, ischemia of mediobasal portions of the temporal lobes was accompanied by transient amnesia-like memory impairment. Ataxia dominated over other cerebellar disorders and was often accompanied by muscle tone dissociation along the cross-axis or predominated in the legs. A combination of the symptoms of spastic-atrophic or flaccid paresis of the hands with dissociative sensory disturbances resulted in formation of myelosingosis syndrome in two patients. In these patients, intracerebral cysts in the

superior and medial segments of the cervical spinal cord were detected. In 12.2 % of cases, severe deformity of the occipitocervical dural funnel was accompanied by intracranial hypertension syndrome.

The most pronounced dependence of transient ischemic attacks was observed in 11 patients with Kimmerle anomaly due to fixation of the vertebral artery in the abnormal bone ring on the atlas by periarterial cicatricial tissue. Nine patients had an isolated bilateral abnormality, two – unilateral. After separation of the vertebral artery from the abnormal bone rings and additional arteriolysis, good to excellent outcomes were obtained.

**Fig. 3**

Craniometric parameters in patients with basilar impression: craniovertebral angle (1) is reduced, the tip of the odontoid process is above the MacGregor's line (2) and particularly above the Chamberlain's line (3); odontoid-clival complex severely deforms the intracranial portion of the vertebral arteries

An analysis of medical records of operated patients was insufficient for detailed ranking of the immediate treatment outcomes according to specific criteria. Patients were divided into three groups depending on post-operative complications: 1) without postoperative complications or worsening of patient's state (63.3 %); 2) with temporary worsening of neurological signs and their regression within the next 4–6 weeks (32.6 %); 3) with persistent complications in the form of worsening of conduction and bulbar disorders (4.1 %).

Long-term outcomes of surgical treatment of these patients (on modified Macnub scale) are as follows:

1) excellent outcome (no symptoms of the diseases, restored working ability) was observed in 5 (13.2 %) patients with dynamic compression of the neurovascular structures;

2) good outcome (no subjective manifestations of the disease, decrease in neurological disorders, occasional transient symptoms, preserved working ability) – 13 (34.2 %);

3) satisfactory outcome (improved quality of life, slight decrease in the

symptoms of the disease, persisting neurological disorders, limited working ability of disability) – 14 (36.8 %);

4) no changes (stabilization of the pathological process, symptoms persist, patients are unable to work) – 4 (10.5 %), two patients need assistance;

5) progression of the disease could not be prevented in 2 (5.3 %) patients.

Excellent to good long-term outcomes were obtained in all 11 patients, who were operated on for Kimmerle anomaly. Long-term outcomes of treatment of 38 patients, who were operated on for abnormal conflicts at the occipitocervical dural funnel, are reported in accordance with Rankine scale: functional capacity score = 1 in 4 (10.5 %) patients; 2 – 21 (55.3 %); 3 – 6 (15.8 %); 4 – 4 (10.5 %); 5 (pronounced signs of disability, the patient is bedridden, incontinent and needs permanent assistance and care) – 3 (7.9 %).

No improvement of neurological status was observed in 6 of 9 patients with severe brainstem-related, supratentorial, and conduction neurological disorders ($p < 0.05$), long-term outcome of treatment was satisfactory in two patients, and good – in one patient. Five patients with unstable craniovertebral anomalies demonstrated excellent long-term outcomes; there were no excellent outcomes in patients with persistent compression of the brainstem and the vertebral artery. Excellent to good outcomes were obtained in a total of 12 (75.0 %) patients with unstable abnormalities and in 6 (27.3 %) patients with stable abnormalities; $p < 0.01$.

Discussion

Diagnosis of craniovertebral abnormalities is not always simple as evidenced by the late visits of patients to neurosurgeons (an average of 4.5 ± 2.4 years from the onset of the disease) and a large percentage of the erroneous diagnoses.

In the case of vertebrobasilar insufficiency due to craniovertebral abnormalities, diagnostic algorithm included the following criteria:

1) clinical and neurophysiological signs of common ischemic conditions of the brainstem and other brain structures within the vertebrobasilar vascular pool (occipital lobes, cerebellum, mediobasal portions of the temporal lobe);

2) clinical and radiographic correlation of vertebrobasilar insufficiency with craniovertebral abnormalities;

3) persistent or dynamic compression of the vertebral artery by a compressing substrate resulting from detected craniovertebral abnormalities;

4) exclusion of other causes of occlusion and stenosis of the vertebral arteries and their intracranial branches using transcranial Doppler ultrasound, duplex scanning, spiral CT angiography, and digital angiography. SCT angiography is an informative method for detecting the compression of the vertebral artery.

Digital angiography was found to be more reliable method to study the detailed pathogenesis of vertebrobasilar insufficiency, since it enables analysis of the arterial lumen, obtaining images of the vertebral artery in the oblique projections with various positions of the neck, which is highly important for the diagnosis of this pathology. The use of brainstem evoked potentials facilitates the evaluation of the effectiveness of decompression of the vertebral arteries and detection of high lesions of brainstem structures, which are characteristic of vertebrobasilar disorders.

Kimmerle anomalies (abnormal rings on the posterior arch of the atlas) should be discussed separately. They can restrain mobility of the vertebral artery loop at the site, where the abnormal ring is formed on the posterior arch of the atlas, and cause vertebrobasilar insufficiency [2, 6, 8, 10, 12].

In recent literature, there are many studies claiming clinical significance of common Kimmerle anomaly. Some authors [1, 12] diagnosed this abnormality in 37–80 % of patients. Cases of critical decrease in the vertebrobasilar blood supply accompanied by formation of macular ischemia of the brainstem have been reported. These findings are confirmed by angiographic studies with various head position and MRI [8].

Two clinical cases of ischemic stroke in patients with Kimmerle anomaly have been described by Barsukov et al. [3]. Our clinical observations with angiographic and intraoperative verification showed that Kimmerle anomaly is rarely accompanied by vertebrobasilar insufficiency (5.5 %), only when there is periarterial cicatricial knot located in the vicinity of the abnormal ring. Therefore, in most cases, Kimmerle anomaly does not cause vertebrobasilar ischemic insufficiency and may be asymptomatic.

Surgical treatment of patients with craniovertebral abnormalities is characterized by pathogenetically justified combination of anterior decompression of the brain and its vascular structures through the transoral approach with posterior decompressive and stabilizing interventions. Transpharyngeal approach provides optimal conditions for decompression of the neurovascular structures under good visual control without conflict with the brain due to an adequate surgical angle. Anterior compression of neurovascular structures can not be eliminated using the posterior approach. Thirty years-long prospective follow-up showed that stabilization of the craniovertebral junction using designed structures made of NiTi alloy with thermome-

chanical memory effect is characterized by primary reliability of the operated segment fixation and no need for external immobilization of the spine after surgery.

The anterior decompression of the brain and its vessel through the transoral approach in combination with the posterior stabilizing interventions provided stable positive effect in 28 (82.4 %) of 34 patients with abnormalities, distorting the occipitocervical dural funnel. They demonstrated decrease in the severity of cerebral symptoms, regression of cochleovestibular disorders, visual and oculomotor disturbances, cerebellar symptoms; termination of syncopal and drop-attacks. Treatment outcome was significantly better in the cases with shorter history and dynamic compression of the neurovascular structures as compared to stable compression. In patients with severe neurological disorders, long-term outcomes were worse. The most favorable treatment outcome was observed in patients with Kimmerle anomaly, where transient ischemic attacks were associated with rotational movements of the head. Stable good to excellent outcomes were observed in all 11 of the operated patients.

Conclusions

1. Bone craniovertebral abnormalities, along with other extravasal causes of compression of the vertebral artery, atherosclerotic and septal stenoses of the vertebral artery may cause vertebrobasilar insufficiency.

2. Extremely common Kimmerle anomaly causes vertebrobasilar insufficiency only when it occurs in combination with cicatricial knot formed around the vertebral artery due to chronic traumatization in the abnormal bone ring.

3. Decompression of the brain and vertebral arteries using transpharyngeal approach in combination with posterior stabilization, using preferably Ti-Ni structures with thermomechanical shape memory, is a pathogenetically justified method of treatment of patients with vertebrobasilar insufficiency due to persistent abnormal stenosis of the occipitocervical dural funnel. In the case of dynamic compression of the brain and vertebral arteries, occipitospondylodesis or atlantoaxial fusion is required (depending on the level of compression) in the position providing correction of occipitocervical structures.

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