

Vascular Brain Malformations In Combination With Extracranial Brachiocephalic Artery Lesions

Aleksey Khripun¹, Aleksandr Pryamikov^{1,2}, Aleksey Mironkov^{1,2}, Sarkis Asratyan², Viktor Suryakhin², Ruslan Loluev², Lyubov Gulina³

¹ - Chair of Surgery and Endoscopy of Advanced Medical Training Department, Pirogov Russian National Research Medical University, 117997, 1 Ostrovityanova Str., Moscow, Russian Federation

² - VM Buyanov City Clinical Hospital, 115516, 26 Bakinskaya Str., Moscow, Russian Federation

³ - Polyclinic N170, Moscow Department of Health, 117545 2 Podol'skikh kursantov Str., Moscow, Russian Federation

e-mail: pryamikov80@rambler.ru.

Abstract. *One of the unresolved problems in vascular surgery and neurosurgery is the internal carotid artery extra- and intracranial segments combined lesion, namely the combination of the arterial aneurysm or vascular brain malformation with extracranial brachiocephalic arteries lesions. Currently, there are no clear tactical approaches and recommendations for the brachiocephalic arteries' comorbid pathology treatment. This article shows the treatment experience of 55 patients with combined lesions of the internal carotid artery intra- and extracranial pool: a combination of the intracranial aneurysm or vascular brain malformation with brachiocephalic arteries stenotic lesions or asymptomatic internal carotid artery pathological deformation at the cervical level. Clinical cases were accumulated over 6 years (from 2013 to 2019). The incidence of comorbid pathology was 3.5%. The bigger part of brain vascular malformations (53 of 55 patients or 96%) were asymptomatic. The majority of patients with comorbidity were women (69% or 38 patients). Surgical activity among the target group of patients was 40%, 22 of 55 patients were operated on. Repeated ischemic stroke after asymptomatic internal carotid artery stenting was beared by one patient (4.5%) in the early postoperative period. Most aneurysms in the first group were localized in the anterior sections of the Willis circle (81% versus 11%, $p < 0.001$), saccular aneurysms prevailed (41 patients or 95%). There is an existing necessity of accumulation brachiocephalic vascular pool comorbid pathology surgical treatment experience and major studies conduction that should help in the development of therapeutic algorithms and optimization of surgical tactics for the brachiocephalic pool combined vascular pathology.*

Keywords: *carotid stenosis, intracranial aneurysm, comorbid pathology, treatment approaches, malformation, internal carotid artery lesions*

1. INTRODUCTION

Surgical tactics for symptomatic and asymptomatic internal carotid artery (ICA) stenosis, as well as various brain vascular malformations (VM), are defined by surgical guidelines and national recommendations [1, 2, 3]. A completely different situation arises

when these two pathologies - extracranial ICA lesions, and intracranial VM - are combined [4]. Nowadays, there are no unambiguous tactical approaches and recommendations in the treatment of the brachiocephalic artery pool comorbid pathology. This condition happens due to a rare incidence of such clinical cases, although the frequency of cerebral arterial aneurysms detection in patients with extracranial ICA stenosis, according to some authors, can reach 14% [5, 6, 7, 8]. Despite the infrequent occurrence, this problem remains controversial from tactical and surgical sides: the sizing and phasing of surgery and the timing between surgery stages are not defined [9, 10, 11, 12, 13]. On the one hand- endovascular reconstruction of the ICA can lead to the raising of volumetric and cerebral blood flow, which increases the risk of cerebral VM rupture. On the other- the brain arterial aneurysm intervention or other vascular malformation surgery on the background of the brachiocephalic arteries stenotic lesions and brain hypoperfusion may be accompanied by a high frequency of perioperative cerebral ischemic complications [5, 14, 15, 16].

2. PATIENTS AND METHODS

Among 55 patients-17 men and 38 women. The average age of the patients was 66 ± 7 years (from 48 to 83 years). There were no minor patients in this work.

In most cases (54 out of 55 patients), intracranial VM or arterial aneurysm were identified as a random finding while extracranial ICA lesions diagnostic (stenosis, occlusion, or pathological deformation). All patients had different comorbidities, diseases of the cardiovascular system prevailed. ICA lesion characteristics are presented in Table 1.

Table 1. Symptomatology of the ICA lesions at the cervical level.

Nature of the affection	Number of patients
Asymptomatic ICA stenosis	29 (53%)
Symptomatic ICA stenosis (history of stroke or transient ischemic attack)	12 (22%)
ICA deformation	7 (13%)
Occlusion or stenosis of the subclavian artery I segment with steal syndrome	4 (7%)
Asymptomatic ICA occlusion	3 (5%)
Total	55 (100%)

All brain vascular malformations and most aneurysms were asymptomatic (Table 2).

Table 2. Clinical symptoms of vascular malformations.

Nature of the affection	Number of patients
Asymptomatic intracranial arterial aneurysm	46 (83%)
Symptomatic intracranial arterial aneurysm (non-traumatic subarachnoid hemorrhage history)	2 (4%)
Asymptomatic intracranial arteriovenous fistula	3 (5%)
Asymptomatic arteriovenous malformation	2 (4%)
Asymptomatic venous angioma	1 (2%)
Symptomatic venous angioma	1 (2%)
Total	55 (100%)

Table 3 represents the aneurysm relation (vascular malformation) to extracranial ICA lesions.

Table 3. Relation of the extra- and intracranial parts of the internal carotid artery pathology.

Comorbidity	Number of patients
Occlusion (stenosis) of the brachiocephalic artery at the extracranial level and intracranial arterial aneurysm	43 (78%)
Pathological deformation of the extracranial brachiocephalic artery and intracranial arterial aneurysm	5 (9%)
Extracranial brachiocephalic artery stenosis and intracranial arteriovenous fistula	3 (5%)
Pathological deformation of the extracranial brachiocephalic artery and intracranial arteriovenous malformation	2 (4%)
Extracranial brachiocephalic artery stenosis and intracranial venous angioma	2 (4%)
Total	55 (100%)

Surgical intervention was provided in 22 of 55 patients with comorbidity. Patients had one- and two-stage surgery. On the first neurosurgical stage (6 cases) the intracranial arterial aneurysm was clipped (n=5), and ones (a single case) a torn aneurysm was operated in the volume of enveloping aneurysm of the anterior connecting artery. The second vascular surgery stage was provided on average for 5.7 ± 3.4 months, where 6 patients underwent ICA resection with further redressment (n=3), carotid endarterectomy (n=2), and left subclavian artery I segment stenting (n=1).

One-staged surgery had 16 patients. Cerebral arterial aneurysm clipping was performed in 3 patients. Remaining 13 patients had carotid endarterectomy (n=6), ICA stenting (n = 5), ICA resection with its reduction (n = 1) and stenting of the I subclavian artery segment (n=1).

All procedures (manipulations, operating aids) used in the research-work followed to the ethical standards of the responsible institutional committee on human experiments and to the 1975 Helsinki Declaration, revised in 2000. All patients agreed to participate in the experiment and did not deny the results of the experiment, which will be presented in the given research paper.

3. RESULTS

The incidence of comorbid pathology was 3.5%. The bigger part of the brain VM (53 from 55 patients, or 96%) was asymptomatic. The same situation we can watch about the pathology of the extracranial ICA: 78% of cases (43 out of 55 patients) with stenosis, occlusion or pathological deformation of the ICA were asymptomatic; the remaining 12 patients had a history of various ischemic cerebral events (transient ischemic attack or ischemic stroke).

Most of the patients with combined pathology were women: 69% or 38 patients. Surgical activity among the target group of patients was 40%, 22 of 55 patients were operated on. From 22 operated patients, 8 patients had an ipsilateral location of stenosis or deformation of the extracranial ICA and cerebral aneurysm, a contralateral position was revealed in 9 cases, and 3 patients had combined pathology of extracranial ICA with aneurysm of the anterior communicating artery. 2 patients had a combination of critical ICA stenosis with

ipsilateral cerebral venous angioma. The average diameter of the brain aneurysm was 9.1 mm. There were no intra- and postoperative vascular malformation ruptures, hemorrhagic wound complications, and no lethal cases among the surgery group of patients.

Repeated “minor” ischemic stroke after ICA stenting was beared by one patient (4.5%) in the early postoperative period. Characteristics of cerebral aneurysms in patients with brachiocephalic arteries atherosclerosis and extracranial ICA pathological deformation analyzed separately (Table 4).

Table 4. Cerebral aneurysms localization versus the type of extracranial internal carotid artery lesion.

	Group 1 (patients with brachiocephalic arteries' atherosclerosis, n=43)	Group 2 (patients with ICA deformations, n=5)
Arterial pool of aneurysm		
Willis circle anterior sections' aneurysms	35 (81%)	
Internal carotid artery	16 (37%)	
Middle cerebral artery	12 (28%)	1 (20%)
Anterior communicative artery	5 (11%)	1 (20%)
Anterior cerebral artery	2 (5%)	2 (40%)
Willis circle posterior sections' aneurysm	5 (11%)	
Basilar artery	3 (7%)	
Posterior communicating artery	2 (5%)	
Posterior cerebral artery		1 (20%)
Multiple aneurysms	3 (7%)	
The average size of aneurysm (mm)	5.7±3,8	5.2±1.8
Shapes of aneurysms		
Saccular	41 (95%)	4 (80%)
Fusiform	2 (5%)	1 (20%)
History of intracranial hemorrhage	1 (2%)	1 (20%)

Cerebral aneurysm in combination with brachiocephalic arteries atherosclerosis (first group) was diagnosed more often in women: 70% or more than 2/3 of patients. Most aneurysms in the first group were localized in the anterior sections of the Willis circle (81% versus 11%, $p<0.001$), saccular aneurysms prevailed (41 patients or 95%). The sizes of aneurysms in both groups were almost the same and most of the aneurysms in the second group also were located in the Willis circle anterior sections and were saccular-shaped.

4. DISCUSSION

A combination of vascular BM and extracranial ICA lesions is rare but still not completely resolved the problem of vascular surgery and neurosurgery [17, 18]. The real frequency of such comorbidity fully unknown, it ranges from 0.5 to 14% [5, 6, 14, 19, 20, 21]. Our work presents the frequency of cerebral VM with a combination of ICA

stenosis/occlusion/deformation at a 3.5% level. Some evidence of cerebral aneurysms higher frequency in those patients who are suffering from brachiocephalic arteries atherosclerosis are existing [22]. The impact of ICA stenosis in the formation and development of intracranial aneurysms is not proved [7]. Linkage of cervical ICA stenosis and brain aneurysms was presented [22]. The incidence of asymptomatic aneurysms in 247 patients with more than 30% ICA stenosis was 6.5% (16 patients). Among women, aneurysms were detected more often comparing with men: 15.6% vs. 4.5% ($p < 0.05$). Less often, aneurysms were detected in patients with ICA C1 segment stenosis (4.4%), while localization of ICA stenosis in other segments, the aneurysm rate was 28.6% ($p < 0.05$). Independent risk factors of intracranial aneurysms for these patients were: gender and the ICA stenosis localization [22]. Other authors also indicate a high incidence of comorbidity (aneurysm and ICA stenosis) in women [14].

Among patients with brachiocephalic arteries atherosclerosis and intracranial aneurysms 70% of the patients were women. Other authors indicate connection of cerebral arterial aneurysms and extracranial pathological ICA deformation with systemic dysmorphism and connective tissue dysplasia of the vascular wall [23].

The frequency of intracranial hemorrhages after carotid endarterectomy ranges from 0.4 to 5.4%, and one of the complication reasons is undiagnosed cerebral vascular malformation [3, 24]. Believed, that revascularization of the brain does not increase the risk of intracranial aneurysms rupture with not more than 10 mm diameter [14, 21].

There are no clear recommendations for surgical tactics in patients with comorbidity of the extra- and intracranial ICA pool. The main problem of the cerebral arterial malformation and extracranial ICA lesions surgical treatment is the lack of balance between the risks of ischemic events and intracranial hemorrhage [12, 14, 25]. In the previous article, we described the current world experience and results of given pathology treatment, the frequency of its diagnostic, and depending on the types of interventions surgical approaches (open and endovascular surgery) [26].

Some authors use a two-stage approach in ICA stenosis (pathological deformation), or arterial aneurysms treatment while other articles describe a simultaneous approach. The recommended intraoperative period in case of a staged surgical treatment vary, the nature of the surgical intervention (open and/or endovascular) is different [23, 27, 28, 29, 30].

In the world literature, the plenty of works are devoted to patients with a combination of arterial intracranial aneurysms and ICA stenosis at the cervical level [5, 6, 31, 32], however, there are articles on approaches of treating patients with aneurysms and with extracranial ICA pathological deformation. The influence of each other and the connection between these two states is discussed [23].

Usachev D.Y. presented a clinical case of successful combined surgical treatment of comorbid ICA lesions. The patient had a giant aneurysm of the left internal carotid artery cavernous segment in combination with the inflection of the ipsilateral common carotid artery. In the first stage, to ensure adequate intracranial aneurysm access, the authors performed a common carotid artery resection. Then, after 3 months, a flow diverter stent into the left ICA siphon was implanted. According to angiography, a slowdown of blood flow in the aneurysm has already been observed intraoperatively. The patient released from the hospital in satisfactory condition [23]. Cho Y.D. and coauthors (2013) examined 606 patients with more than 50% ICA stenosis, while aneurysms (120 aneurysms) were detected in 86 (14.2%) patients. After analyzing the interplay between aneurysms and ICA stenosis, the authors haven't found any regularity in their localization. The authors underscored that with an increase of ICA stenosis degree, there is a tendency towards more frequent detection of cerebral aneurysms which are contralateral to stenosis. In the remote period (28 ± 14.3 months), ICA stenosis surgical correction did not have affection on the size and stability of

the aneurysms. Only one case showed increasing in aneurysm size during the observation period (41 months). The size stability of intracranial aneurysms in patients with ICA stenosis may indicate that there is no need for urgent interventions on vascular malformation of the brain [6]. Jou L.D. and coauthors (2010) came to similar results by analyzing the data of 209 patients survey with cerebral aneurysms, found that aneurysms size on the contralateral to ICA stenosis are bigger then ipsilateral aneurysms (13.6 versus 6.6 mm, $p < 0.01$). The risk of rupture of aneurysms is higher in patients with aneurysms and contralateral extracranial stenosis, as well as in patients with aneurysm and ipsilateral intracranial stenosis of the ICA [7].

Currently, we are trying to adhere to the following tactic in patients with such comorbidity who have indications for surgery. In the case of the ipsilateral localization of asymptomatic aneurysm and asymptomatic ICA stenosis on the first stage is the aneurysm surgery; on the second stage is the carotid endarterectomy or ICA resection. If one of the ICA pool lesions (extra- or intracranial) is symptomatic, then regardless of location – ipsi- or contralateral - we start from the symptomatic lesion. Tactically, in our opinion, the most difficult is the situation when both lesions are asymptomatic and have a contralateral localization.

5. CONCLUSION

Comorbid pathology frequency (a combination of ICA stenosis or pathological deformation with cerebral vascular malformation), according to our data, was 3.5%. The frequency of ischemic cerebral events surgery group was 4.5%. There were no hemorrhagic complications and fatal outcomes among patients during surgical interventions. There is an existing necessity of accumulation brachiocephalic vascular pool comorbid pathology surgical treatment experience and major studies conduction that should help in the development of therapeutic algorithms and optimization of surgical tactics for the brachiocephalic pool combined vascular pathology.

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