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Research Article

Comparative characteristics of ophthalmological symptoms of orbitocranial and orbital cavernous venous malformation

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ABSTRACT
BACKGROUND: Cavernous venous malformation of the orbit, formerly known as cavernous hemangioma of the orbit, refers to vascular malformations with slow blood flow, and is a frequent lesion of the orbit in adults. The spread of cavernous venous malformation of the orbit into the cranial cavity is extremely rare.
AIM: The aim of this study is a comparative analysis of ophthalmological manifestations of orbitocranial and orbital cavernous venous malformation.
MATERIALS AND METHODS: The analysis was performed on 50 patients operated in N.N. Burdenko National Scientific and Practical Center for Neurosurgery from 2004 till 2023. Two groups of patients with cavernous venous malformation of the orbit were identified: group 1 — with malformation spreading into the cranial cavity, group 2 —with malformation localized in the orbit only.
RESULTS: In 29 patients, cavernous venous malformation of the orbit spread into the cranial cavity through superior orbital fissure, inferior orbital fissure, optic canal and/or through combinations thereof; in 21 patients, the malformation was located in the orbit only. Females prevailed in both groups; the average age was 44 years. The first group included patients with the following features: visual impairments were more frequent and more prominent, oculomotor disorders caused mainly by the involvement of oculomotor and abducens nerves; optic nerve atrophy, slight proptosis. The second group was identified by more prominent proptosis, more mild visual impairments, oculomotor disorders caused by the presence of the malformation in the orbit, at the eye fundus, optic nerve head edema prevailed.
CONCLUSIONS: Ophthalmic symptoms due to topographic and anatomical variants of a cavernous malformation.

Keywords: cavernous venous malformation of the orbit; orbitocranial cavernous venous malformation; vascular mass formation.

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Научная статья

Сравнительная характеристика офтальмологической симптоматики орбитокраниальной и орбитальной кавернозной венозной мальформации

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АННОТАЦИЯ
Актуальность. Кавернозная венозная мальформация орбиты, ранее известная как кавернозная гемангиома орбиты, относится к сосудистым мальформациям с медленной скоростью кровотока и является частым поражением орбиты у взрослых. Распространение кавернозной венозной мальформации орбиты в полость черепа встречается крайне редко.
Цель — сравнительный анализ офтальмологической симптоматики орбитокраниальной и орбитальной кавернозной венозной мальформации.
Материалы и методы. Исследованию подверглось 50 пациентов, оперированных в ФГАУ «НМИЦ нейрохирургии им. акад. Н.Н. Бурденко» с 2004 по 2023 г. Было выделено две группы пациентов с кавернозной венозной мальформацией орбиты: группа 1 — распространяющейся в полость черепа, группа 2 — локализующейся только в орбите.
Результаты. У 29 пациентов кавернозная венозная мальформация орбиты распространялась в полость черепа через верхнюю, нижнюю глазничную щель, зрительный канал и/или их сочетания; у 21 пациента объёмное образование находилось только в орбите. В обеих группах превалировали лица женского пола; средний возраст составил 44 года. Отличительными особенностями пациентов группы 1 были чаще встречающиеся и более выраженные зрительные нарушения, глазодвигательные нарушения, обусловленные преимущественно поражением глазодвигательного и отводящего нервов, атрофия зрительного нерва, нерезко выраженный экзофтальм. У пациентов группы 2 экзофтальм был более выраженным, зрительные нарушения были негрубыми, глазодвигательные нарушения обусловлены наличием объёмного образования в орбите, на глазном дне превалировал отёк диска зрительного нерва.
Заключение. Офтальмологическая симптоматика обусловлена топографо-анатомическими вариантами кавернозной мальформации.

Ключевые слова: кавернозная венозная мальформация орбиты; орбитокраниальная кавернозная венозная мальформация; сосудистое объёмное образование.

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BACKGROUND

Cavernous venous malformation (CVM) of the orbit, formerly known as cavernous hemangioma (CH) of the orbit, is a common orbital condition in adults [1].

Formerly, this space-occupying lesion was commonly recognized as benign tumor. In accordance with the classification of the International Society for the Study of Vascular Anomalies (ISSVA) of 2018, CVM of the orbit is equal to vascular malformations with slow blood flow velocity. In our study, we refer the CBM of the orbit to in accordance to the data of this classification. However, currently the term "cavernous hemangioma" (CH) is used by many authors, that is why in this review of literature, we shall follow the terminology used by the author.

CH of the orbit occurs 2.5 times more frequently in women as in men [2, 3].

The clinical picture depends on the localization of the orbital CVM. Most prevalent localization becomes the muscle cone [2]. Clinical signs of this localization are: slowly progressing axial nonpulsatile proptosis (60–70%), visual acuity decrease (41%), hypermetropic shift due to the compression of the posterior pole of the eyeball, oculomotor disorders (20–30%) are the result of the action of the space-occupying mass on extraocular muscles. If the orbital CH is positioned outside of the muscle cone, there is a proptosis with the eyeball dislocation and the limitation of eye movements towards the space-occupying lesion. More rarely, such symptoms as diplopia, lid edema, chemosis of the bulbar conjunctiva are registered [1–6].

CVM localized at the orbital apex and with intracranial spreading, as a rule, through the superior orbital fissure (SOF) and the optic canal (OC), — is an extremely rare condition.

Most frequent clinical signs of the CH localized at the orbital apex according to several authors [7–10] are: visual acuity decrease, appearance of visual field defects, development of optic disc edema or of primary optic atrophy, presence of oculomotor disorders due to compression of cranial nerves, diplopia, facial *hypoesthesia*, slowly progressing proptosis.

The literature analysis showed that in a small number of publications, a comparison is made between ophthalmic signs of the orbital apex CVM spreading intracranially and the CVM localized only in the orbit.

Thus, *the aim of our study* became a comparative analysis of ophthalmic signs of the orbitocranial CVM and of the orbital one.

MATERIALS AND METHODS

To solve the problem, 50 patients with the CVM of the orbit were included into the study treated at the National Medical Research Center for Neurosurgery named after Academician N.N.Burdenko during the period from 2004 to 2023. There were 39 female and 11 male patients (4 : 1). The age of patients was 17–69 years, median 43.5 years (Fig. 1).

All patients underwent an ophthalmologic examination, which included: visual acuity testing, perimetry, exophthalmometry using the Hertel exophthalmometer, testing of oculomotor, pupillary motor function, direct and indirect ophthalmoscopy, biomicroscopy of the anterior segment.

According to our tentative classification, the degree of proptosis was estimated as follows: 1–2 mm — degree I, 3–4 mm —degree II, 5–6 mm — degree III, 7 mm and more — degree IV.

The visual acuity testing was performed using Roth's device by Golovin–Sivtsev, Landolt optotypes.

The visual field was tested by static automated perimetry using the Humphrey Field Analyzer II (Carl Zeiss Meditec Inc., Germany), Threshold test Central30–2 program, and by kinetic manual perimetry using the Förster perimeter.

Following grades of visual disturbances were formed: visual acuity from 0.9 to 1.0 and absence of visual field defects were considered as norm; visual acuity from 0.8 to 0.5 and initial visual field defects (<1/4 to 1/4 of the surface)

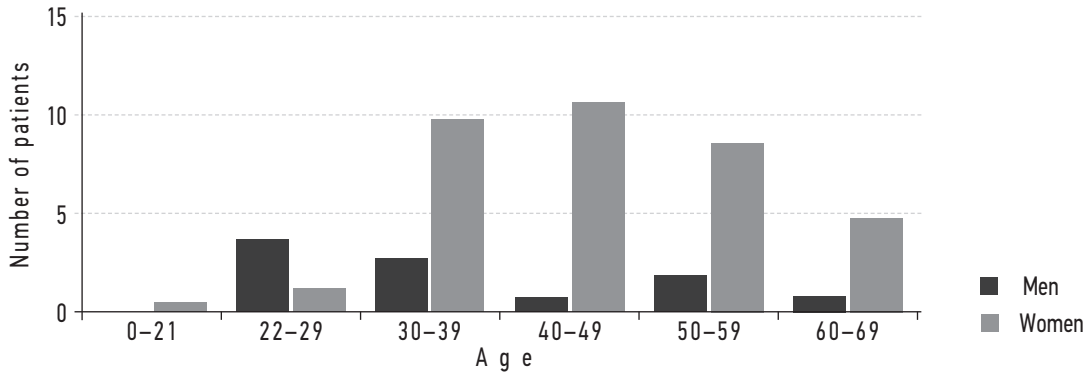


Fig. 1. Distribution of patients by gender and age
Рис. 1. Распределение пациентов по полу и возрасту

corresponded to initial visual disturbances; visual acuity decrease between 0.4 and 0.1 and visual field defects >1/4 to 3/4 of the surface were considered as moderate visual disturbances; visual acuity less than 0.1 and down to practical blindness with severe visual field defects (>3/4 of the surface), or with a central scotoma, were taken as significant visual disturbances.

According to the degree of visual disturbances and the state of the optic disc, two stages of visual disturbances were differentiated: early and late. Categorized to the early stage were high visual functions or moderate visual disturbances, normal fundus or initial optic disc pallor with edema or without it; the late stage was characterized by low visual functions and sever optic disc pallor.

At examination, attention was driven to such indices as symmetrical palpebral fissures, presence of semiptosis/ptosis, lid edema, presence and direction of eyeball deviation in the orbit. Magnetic-resonance imaging was performed in 31 patients, in 19 patients — computed tomography. The presence of a mass in the upper orbital fissure and optic canal area evidenced the spread of cavernous malformation into the intracranial cavity. Apart from neurovisualization methods, the intracranial spread of cavernous malformation was confirmed by the protocol of surgical procedure.

All patients were operated at the Center for Neurosurgery named after Academician N.N. Burdenko. To remove the CVM, the following approaches were used: osteoplastic lateral orbitotomy — in 32 patients, supraorbital approach — in 13, orbitozygomatic approach — in 3, endoscopic transnasal approach — in 1, simple subperiostal orbitotomy — in 1 patient.

Morphologic examination confirmed the CVM in all cases.

RESULTS

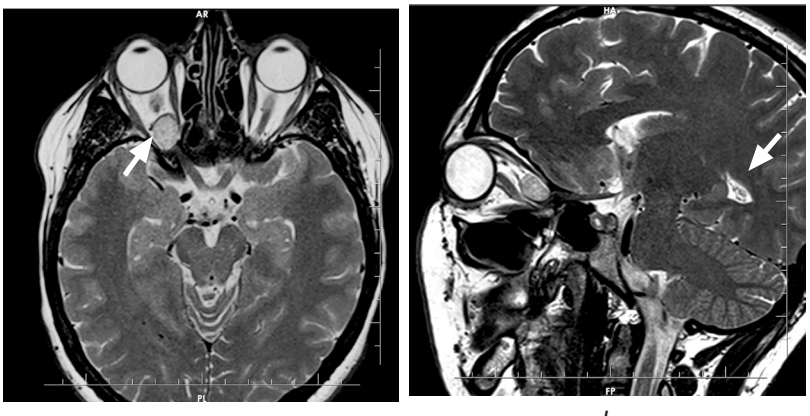
The disease history from the appearance of patient's complaint was from 1 month to 10 years (in average 2.5 years).

Based on the neurovisualization results, two groups of patients were differentiated: group 1–29 patients with CVM of the orbit spreading intracranially, 15 out of them — with spread into the upper orbital fissure and optic canal, in 8 patients — into the upper orbital fissure, 2 — into the optic canal, 3 — into the upper orbital fissure and lower orbital fissure, in 1 patient, the orbital CVM spreaded intracranially destroying the roof of the orbit; into the group 2, patients with only orbital CVM localization were included — 21 patient (Fig. 2, 3).

Ophthalmic symptoms and signs in group 1 patients (*n* = 29)

Upon examination, 13 patients presented complaints on pain in the area of the orbit/frontal region on the lesion's side, most often exercise-induced. In 7 patients, there was lid swelling, in 3 — semiptosis. Eyeball dystopia was noted in 6 patients.

In 27 patients, proptosis was diagnosed: in 8 — degree I, in 16 — degree II, in 1 — degree III, in 2 patients — degree IV. Proptosis variability was from 1 to 8 mm, in average 3 mm. Visual disturbances were noted in 16 patients (55.2%): initial ones — in 7 patients, moderate — in 5, practical blindness — in 4. In a majority of them (13 patients), there was a CVM spread into the cranial cavity through the optic canal and the upper orbital fissure. The duration of visual disturbances was from 1 month to 10 years (in average 2.5 years).



a *b*

Fig. 2. Magnetic resonance tomography. Cavernous venous malformation of the orbit, spread into the cranial cavity through the superior orbital fissure and the optic canal: *a* — axial projection, T2 mode; *b* — sagittal projection, T2 mode

Рис. 2. Магнитно-резонансная томография. Кавернозная венозная мальформация орбиты, распространяющаяся в полость черепа через верхнюю глазничную щель и зрительный канал: *a* — аксиальная проекция, режим Т2; *b* — сагиттальная проекция, режим Т2

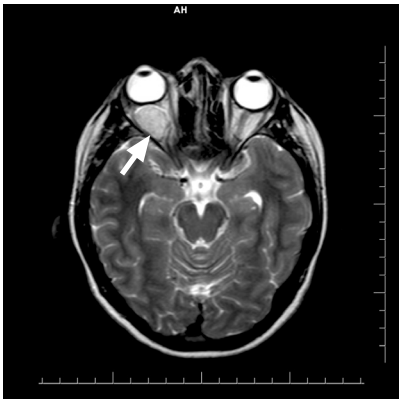


Fig. 3. Magnetic resonance tomography. Cavernous venous malformation localized only in the orbit, axial view, T2 mode

Рис. 3. Магнитно-резонансная томография. Кавернозная венозная мальформация, локализующаяся только в орбите, аксиальная проекция, режим Т2

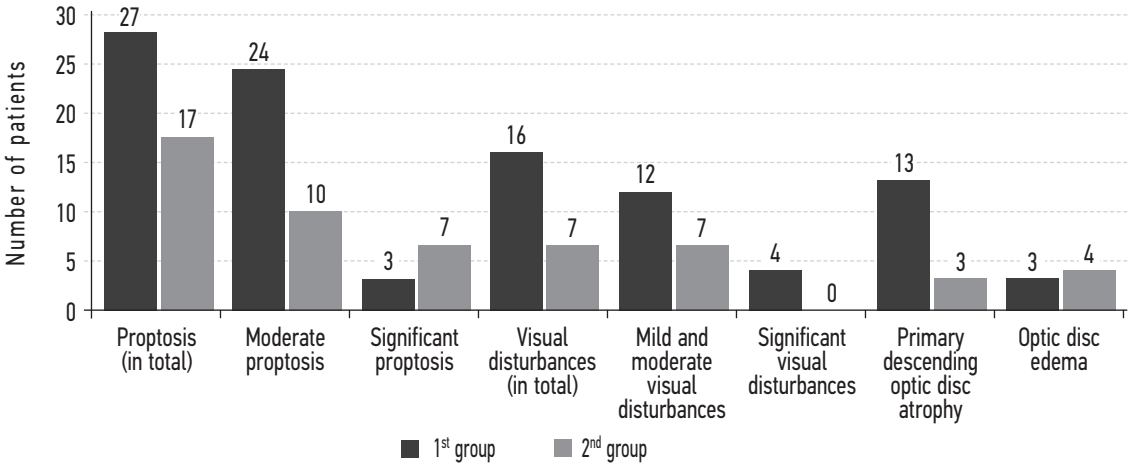


Fig. 4. Comparative characteristics of ophthalmological manifestations
Рис. 4. Сравнительная характеристика офтальмологической симптоматики

Normal ophthalmoscopic picture was observed in 13 patients (44.8%). In 9 patients, initial signs of primary optic disc atrophy were revealed, in 4 the atrophy was significant, in 3 patients, optic disc edema was present.

The early stage of visual disturbances was diagnosed in 12, late one — in 4 patients.

Oculomotor disorders were found in 7 patients (24.1%). In 4, they were the result of oculomotor and abducens nerves involvement in spread of the CVM from the orbit into the cranial cavity through the upper orbital fissure and manifested by the limitation of eye movements by 1/3 from the norm and more; in 3 patients, oculomotor disorders became the result of the mass lesion action on extraocular muscles.

Ophthalmic symptoms and signs in group 2 patients (*n* = 21)

A complaint on pain in the area of the orbit/frontal region on the mass lesion's side presented 7 patients. Upon examination, mild lid edema was noted in 5 patients.

In patients of this group, same as in group 1, among clinical signs, proptosis from 1 to 10 mm prevailed and was found in 17 patients: in 5 — degree I, in 5 — degree II, in 4 — degree III, and in 3 patients — degree IV.

Normal ophthalmoscopic picture was observed in 14 patients (61.9%), in 4 patients, optic disc edema was revealed, 3 patients had initial signs of primary optic disc atrophy. Visual disturbances were present in 7 patients (33.3%): in 5 — initial ones, in 2 — moderate. Thus, in all 7 people, there was an early stage of visual disturbancies.

Oculomotor disturbances were present in 7 patients (33.3%), in 5 out of them, malformation was located within the muscle cone, the ocular motility disorder was due to its action on extraocular muscles, manifesting by the limitation of lateral eye movements no more than 1–2 mm. In 2 patients, the malformation was localized outside the muscle cone, the limitation of eye movement was towards the localization of the space-occupying lesion.

Thus, as the result of the investigation, definitive aspects were revealed of ophthalmic symptoms and signs in patients with malformation localization only in the orbit and of those with its intracranial spread. A comparative characteristic of ophthalmic symptoms and signs in groups is presented in the Fig. 4.

In both groups, proptosis was a frequent sign, but in the group 1, it was mostly moderate, and in group 2 — significant. Visual disturbances of different degree were noted predominantly in the group 1, whereas in the group 2, there were initial visual disturbances; on the eye fundus in group 1 patients, more often there was a primary optic nerve atrophy, in group 2 — predominantly optic disc edema.

DISCUSSION

Since 2018, CVM of the orbit according to the classification of the International Society for the Study of Vascular Anomalies (ISSVA) are ranked among malformations with slow blood flow [11]. However, currently the term "cavernous hemangioma" may be encountered in the literature.

The growth of the orbital CH is extremely slow, and the first sign is a slowly progressing indolent proptosis. However, after its spread into the cranial cavity the first symptom of the disease is a not uncommon visual acuity decrease and an appearance of visual field defects [5–8].

The orbital CVM is more prevalent in women than in men. In publications of G.J. Harris and F.A. Jakobiec [3], the female/male rate was 7 : 3, whereas J.W. Henderson et al. [12] reported a fairly equal rate (8 : 7). In our study, female patients prevailed as well — 4 : 1, what is in accordance with the results of the other study [3].

The disease is usually diagnosed, according to data of different authors, at the age of 30–50 years [1, 2, 4, 13, 14]. Among our patients, the age was 17–69 years (mean age 44 years).

The most frequent localization of the orbital CH is the muscle cone [2]. CVM localized at the orbital apex with an intracranial spread, as a rule, through the upper orbital fissure and the optic canal, is an extremely rare condition. The analysis of the literature showed that there are no sufficient data concerning the comparison of the ophthalmic signs and symptoms of the CVM localized at the orbital apex and spreading intracranially, and malformation localized in the orbit only.

Thus, in the reviews of literature, the information is presented on ophthalmic signs and symptoms of the orbital CH [5, 15]; on malformation localized at the orbital apex [7, 9, 13], or the ophthalmic signs and symptoms of the CH of the orbit and of that at the orbital apex is estimated, without paying attention to their distinctions [6, 16].

After a comparative estimation of two groups of patients, we revealed that in patients with the spread of the malformation into the cranial cavity visual disturbances are encountered much more often (55.2%) and are more significant, up to practical blindness. This is caused by compression of the optic nerve at the orbital apex and in the optic canal. Our results are in line with the literature data [5, 7].

In patients with CVM localized only in the orbit normal visual functions were noted more often, or mild and moderate visual disturbances (33.3%).

In our series, on the fundus of group 1 patients primary optic nerve atrophy prevailed (44.8%), rather than optic disc edema. Edema was found predominantly in group 2 patients (19.1%), and was related to the impairment of venous outflow from the orbit and to the disturbance of axonal flow in the optic nerve.

Oculomotor disturbances in group 1 patients were caused mostly by the action of the space-occupying lesion on the oculomotor and the abducens cranial nerves, this is consistent to the literature data [13]. In group 2 patients, oculomotor disturbances were more often caused by the action of the space-occupying mass on the extraocular muscles.

According to A. Suri [13], J.D. Osguthorpe et al. [17], in patients with CH of the orbital apex spreading into the cranial cavity, the proptosis is rare and progresses slowly. Among our patients, the proptosis was found practically with equal frequency in group 1 patients (93.1%) and in group 2 patients (81%), however in the group 1 it was less significant — predominantly of the 2nd degree.

CONCLUSION

Thus, ophthalmic signs and symptoms are induced by topographic and anatomic variants of the space-occupying lesion. In both groups, the main ophthalmic signs and symptoms were: proptosis, visual disturbances, oculomotor disorders, fundus changes. However, when comparing both groups, there are characteristic differences between patients with the CVM of the orbital apex with intracranial spread (group 1) and patients with the space-occupying lesion localized only in the orbit (group 2): visual disturbances and oculomotor disorders were more pronounced in group 1 patients, on the fundus, signs of the primary optic disc atrophy prevailed; in group 2 patients, the degree of proptosis was more significant, at ophthalmoscopic examination, the optic disc edema is encountered more often.

ADDITIONAL INFORMATION

Authors' contribution. Thereby, all authors have made a significant contribution to the development of the concept, research, and preparation of the article, as well as read and approved the final version before its publication. Personal contribution of the authors: N.K. Serova — collection and processing of materials, analysis of obtained data, concept and design of the study, text writing; A.P. Trunova — collection and processing of materials, analysis of obtained data, concept and design of the study, text writing, literature review; N.N. Grigorieva — collection and processing of materials, analysis of obtained data; N.V. Lasunin — surgical treatment, editing.

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Competing interests. The authors declare that they have no competing interests.

Ethics approval. The protocol of the study was approved by the local Ethics Committee of the N.N. Burdenko National Scientific and Practical Center for Neurosurgery (No. 11/2020, 25/11/2020).

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