CASE STUDY

CONTENTS

Dynamics of ophthalmological symptoms due to vascular pathology of the central nervous system

Valery D. Belyaev, Kateryna V. Bora, Alona V. Stoyka, Ivanna I. Mushak, Antonina V. Varvarynets
UZHHOROD NATIONAL UNIVERSITY, UZHHOROD, UKRAINE

ABSTRACT

An analysis of the patient's medical history with changes in the organ of vision due to TBI was performed. The difficulties in making the correct diagnosis in our case were associated with the delayed onset of the first symptoms affecting the organ of vision and their slow progression, the presence of other concomitant diseases, and the absence of classic complaints (such as constant noise in the head). More than two months passed from the first signs of the disease to the correct diagnosis.

With the start of the full-scale invasion of Ukraine by the Russian Federation, the number of traumatic brain injuries among the civilian population and military personnel in Ukraine has increased significantly, and the likelihood of KCS also increases. In all cases of traumatic exophthalmos, it is recommended to be vigilant in terms of the carotid-cavernous connection.

KEY WORDS: exophthalmos, carotid-cavernous fistula, ocular hypertension, angiography

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INTRODUCTION

The vagueness of the clinical picture in routine medical practice can lead to misdiagnosis and untimely treatment of patients. Diagnostic errors are more common in pathological conditions that occur at the borderline between two specialties. Carotid-cavernous fistula (CCF) is such a borderline disease between ophthalmology and neurosurgery. Carotid-cavernous fistula forms when a fistula develops in the cavernous part of the internal carotid artery (ICA). In this case, blood from the ICA enters the cavernous sinus cavity. Blood pressure in the sinus increases, which causes difficulty in the outflow of blood into the cavernous sinus from the ophthalmic vein and other veins. A characteristic clinical syndrome of pulsating exophthalmos develops.

The first observation of the carotid-cavernous fistula was made by Travers (1813). In 1856, Henry reported a patient with pulsating exophthalmos and suggested that the cause of the disease was a rupture of the ICA into the cavernous sinus cavity. After the death of this patient from nasal bleeding, the assumption was confirmed by autopsy. A complete rupture of the BCA in the cavernous sinus (after trauma) was found [1].

Different authors refer to this disease differently. The most commonly used terms are pulsating exophthalmos and arteriovenous carotid-cavernous (carotid-cavernous) fistula. Most often, CCS develops as a result of trauma in 75% of cases, but it can also occur spontaneously.

There are several working classifications of carotid-cavernous fistula (CCF). In 1985, Barrow et al. proposed a classification based on angiography data [2, 3]. The following types are distinguished: Type A (high-flow) CCS is defined as the presence of a direct connection between the cavernous segment of the internal carotid artery and the cavernous sinus. Type B (low flow) CCS is defined as the presence of an abnormal connection between the cavernous sinus and the meningeal branches of the internal carotid artery. Type C (low flow) CCS is defined as the presence of an abnormal connection between the cavernous sinus and the meningeal branches of the external carotid artery.

Type D (low flow) CCS is defined as the presence of an abnormal connection between the cavernous sinus and one or more meningeal branches of the internal carotid artery and external carotid artery.

The clinical picture of CCS is characterized by: pulsating exophthalmos; unilateral constant noise in the patient's head (mainly above the eye), synchronous with the pulse, of various nature; on auscultation, such noise

can be heard above the eye socket, temporal region, and mastoid process. Exophthalmos in CCS is most often unilateral, rarely bilateral. The degree of exophthalmos varies from slightly noticeable to 15-20 mm. Severe protrusion of the eyeball may be accompanied by its downward and outward displacement, since the largest orbital veins are located in the upper inner corner of the eye socket. [1]; chemosis, congestive injection of the conjunctival vessels, and eyelid edema occur as a result of impaired blood outflow; lagophthalmos, paralytic strabismus, and the loss of sensitivity are the result of compression by the enlarged cavernous sinus of the oculomotor, trochlear, abducens nerves, and the first branch of the trigeminal nerve (most often, damage to the abducens nerve is noted, followed by damage to the oculomotor nerve); Anisocoria may occur due to irritation of the sympathetic plexus; ocular hypertension is a common symptom.

Changes in the posterior segment of the eyeball in CCS can manifest as congestive neuro- and retinopathy (edema of the optic disc, retina, and dilation of the retinal veins), which are clearly recorded and tracked using optical coherence tomography (OCT) and angi-OCT-OCT, and photoregistration [4, 5]; OCT and angio-OCT also reveal an increase in choroidal thickness as a result of venous stasis [6, 7], possible central serous chorioretinopathy [8], glaucomatous or ischemic optic neuropathy; possible occlusion of the central retinal artery, Terson's syndrome (Terson's syndrome — the appearance of hemophthalmos due to subarachnoid hemorrhage. Hemophthalmos can also occur due to intracranial hemorrhage and increased intracranial pressure. Intraocular hemorrhage may be subretinal, retinal, preretinal, subhyaloid, or vitreous hemorrhage; combined retinal and choroidal detachment is possible; neovascular glaucoma [9].

According to the literature, the most common sign, regardless of the strength of blood flow in the CCS (low or high), was dilation of the retinal veins.

Alam, M.S., Jain, M., Mukherjee, B. et al., studying changes in the fundus in various types of CCS, identified the "3-point sign." Although none of the three signs (disc. hyperemia, retinal vein dilation, and intraretinal hemorrhage) individually predicted vision loss, their combined presence led to vision loss. The authors indicate that the "3-point sign" should be detected as early as possible to prevent functional deterioration [10].

The prognosis for spontaneous disease progression in CCS is poor. Recovery from spontaneous thrombosis of the choroid is observed in only 5-10% of cases, 10-15% of patients die from intracranial or nasal bleeding, and 50-60% are disabled due to vision loss and mental disorders [1].

Conservative and surgical methods are used to treat CCS. Conservative methods designed to clot CCS are not effective enough.

Until 1970, surgical treatment of CCS was performed as ligation or clipping of the BCA in the cranial cavity or on the neck. In 1971, the Burdenko Research Institute of Neurosurgery of the USSR Academy of Medical Sciences developed a new method for treating CCS—endovascular occlusion of the shunting opening in the cavernous section of the BCA with a balloon catheter (F.A. Serbinenko, 1971) [1].

Currently, direct CCS is treated using the endovascular method or with the use of detachable balloons or platinum coils, transarterially and transvenously. The most promising method is intravascular occlusion of the ostium using a balloon catheter, which is inserted into the internal carotid artery and advanced to the ostium. The balloon is filled with a rapidly hardening mass—silicone—and then deflated. In this way, the ostium is excluded from the blood circulation while maintaining the patency of the vessel [11, 12].

Early diagnosis and treatment of carotid-cavernous fistula increase the chances of complete regression of exophthalmos, restoration of eye muscle function, and visual function [1, 12].

To report a case of carotid-cavernous fistula (CCF) as an interesting clinical issue.

Purpose of the study

To report a case of carotid-cavernous fistula (CCF) as an interesting clinical issue.

CASE REPORT

Patient L., born in 1961, visited her local ophthalmologist on February 9, 2024, complaining of moderate pain and redness in her left eye. According to the patient, the symptoms appeared on February 6, 2024. The doctor diagnosed «acute conjunctivitis of the left eye» and prescribed local anti-inflammatory therapy. The treatment did not improve the condition, and during a follow-up examination (February 16, 2024), a significant increase in intraocular pressure (IOP) in the left eye (32 mm Hg) was detected. The doctor diagnosed «Open-angle I/C glaucoma of the left eye. Suspected glaucoma of the right eye, hyperopia of both eyes.» Hypotensive therapy (Lanotan, Mardozia) was prescribed. Against the background of hypotensive therapy, IOP was compensated, but the patient developed eyelid edema and an injection in the right eye. Due to the negative dynamics of the disease and the lack of effect from anti-inflammatory therapy, the patient was referred for consultation to the A. Novak Regional Clinical Hospital of the Zaporizhzhia Regional Council. On March 7, 2024, the patient

Fig. 1. Photo of the patient during examination on 03/28/2024 Bilateral exophthalmos, congestive injection, chemosis of the conjunctiva in both eyes
Picture taken by the authors

Fig. 2. Fundus photograph (March 28, 2024) of patient L., born in 1961 before surgery. Left eye: optic disc hyperemia, blurred contours, and dilated retinal veins
Picture taken by the authors

was consulted at the head and neck department with ophthalmology. At that time, the patient complained of redness in both eyes and swelling of the eyelids. Upon closer examination, it was found that on 06.12.2023, the patient had suffered a concussion and was treated on an outpatient basis. The patient also noted a history of thyroid disease. When collecting the medical history, it was noted that the patient was unable to concentrate on her answers and provide an accurate chronology of events. Visual acuity of the right eye = 0.3 with correction sph +2.0D=1.0, left eye = 0.3 with correction sph +2.0D=1.0. When measuring IOP with a pneumotonometer, 16 was determined in the right eye and 21 mm Hg in the left eye on hypotensive therapy. In kinetic perimetry, the boundaries of the visual field are within normal limits in both eyes. Objectively, on external examination, there is moderate swelling of the eyelids, congestive injection of both eyes, moderate chemosis in the left eye, moderate proptosis of the left eye, and full mobility of the eyeballs. Biomicroscopy showed that the cornea was transparent in both eyes, the anterior chamber was of medium depth, the iris was structural, the pupil reacted to light, the lenses were transparent, the vitreous body was transparent, the fundus of the optic disc was pale pink, the boundaries were clear, and in the right eye, there is hyperemia of the disc and significant dilation of the veins in the left eye. No hemorrhages or focal changes were found. The diagnosis was made:

Fig. 3. 11.04.2024 CT – angiography of the head and neck vessels with intravenous contrast, sagittal section. Signs of carotid-cavernous fistula
Picture taken by the authors

«congestive optic disc, exophthalmos, open-angle I C glaucoma of the left eye. Hyperopia of the second degree in both eyes. To clarify the diagnosis, an MRI of the orbits was prescribed to determine the size of the eye muscles (to rule out endocrine ophthalmopathy).

25.03.2024 — MRI of the orbits and eyeballs was performed on a SIMENS MAGNETOM AERA tomograph, magnetic field strength 1.5 T. Without contrast enhancement. The series of MRI tomograms of the orbits shows an enlargement of the lumen of the superior ophthalmic vein with a diameter of 0.47 cm on the right and 0.35 cm on the left, with patency preserved. The muscle cone of the right and left orbits - the extraocular muscles are not thickened, with no pathological changes in the MR signal, the fat- d tissue of the retrobulbar region shows no visible pathological changes. The optic nerves are not deformed or thinned, and visualization of the subarachnoid space along the optic nerves. Visually, on the left in the polar part of the temporal lobe, an area of cystic-gliotic changes measuring 5.8 by 2.1 cm is determined, which is probably an area of brain contusion as a result of a previous traumatic brain injury. Conclusion: MR signs of enlargement of the superior ophthalmic vein on the right. Cystic-gliotic changes in the left temporal lobe. Fig. 1.

At the next examination on March 28, 2024, the patient complained of a «protrusion» of the left eye and diplopia when looking to the left. Visual acuity of the right eye = 0.3 with correction sph +2.0D=1.0, left eye = 0.3 with correction sph +2.0D=1.0. Intraocular pressure on hypotensive therapy on a pneumotonometer was 15 in the right eye and 19 mm Hg in the left eye. The visual field boundaries were within normal limits on the kinetic perimeter. Objectively, on external examination, there is moderate swelling of the eyelids, congestive injection of both eyes, moderate chemosis in the left eye, insignificant exophthalmos of the right eye, and moderate exophthalmos of the left eye, with limitation of outward movement of the left eye. Biomicroscopy

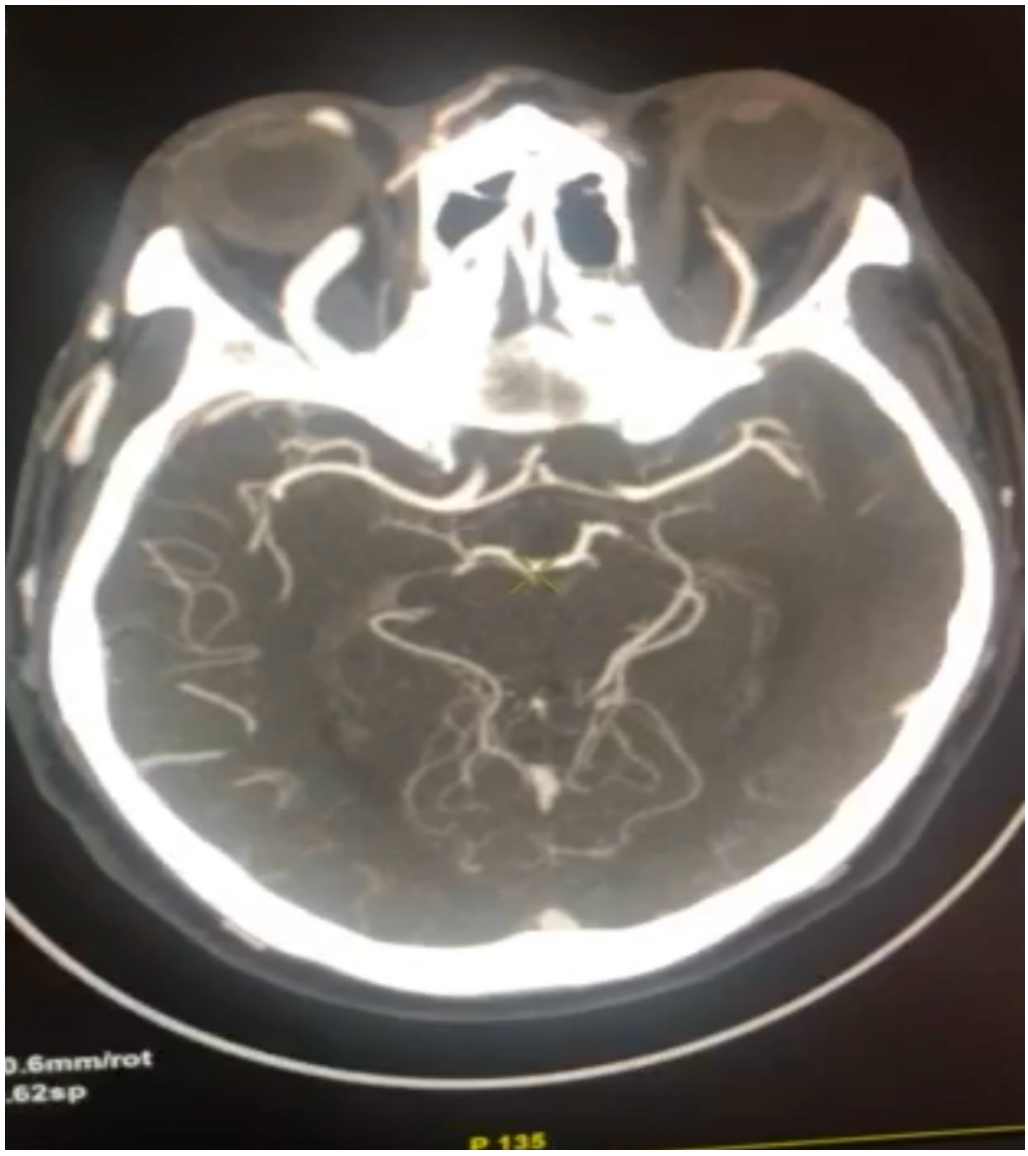

Fig. 4. 11.04.2024 CT angiography of the head and neck vessels with intravenous contrast, axial section. Signs of bilateral carotid-cavernous fistula include visualization of enlargement of the superior ophthalmic veins with a diameter of 4.8 mm and 2.4 mm on the left, which branch off from the enlarged cavernous sinus
Picture taken by the authors

of both eyes shows a transparent cornea, an anterior chamber of medium depth, a structural iris, a pupil reacting to light, transparent lenses, a transparent vitreous body, pale pink fundus, clear boundaries, dilated veins, and tortuous in the right eye. Noteworthy are hyperemia, blurred optic disc margins in the left eye, significantly dilated and twisted veins, clear macular reflex, no hemorrhages or focal changes (Fig. 2).

After ruling out a diagnosis of endocrine ophthalmopathy and worsening symptoms (increased exophthalmos in the left eye and its appearance in the right eye, stagnant phenomena in the fundus, diplopia, stagnant injection and chemosis of the conjunctiva in both eyes), MR signs of enlargement of the superior ophthalmic vein on the right, the patient was referred for angiography of the head and neck vessels to rule out the presence of CCS (Fig. 3, Fig. 4).

At place of residence 11.04.2024 Performed: CT angiography of the head and neck vessels with intravenous contrast Ultravist 370 80 ml. On a General Electric 64 tomograph, a series of MSCT scans showed no sharp narrowing of the lumen of the arterial vessels of the carotid and vertebral basins. In the bifurcation area of the right common carotid artery, a soft semi-concentric atherosclerotic plaque was detected, causing stenosis of the initial section of the right internal carotid artery within 20%. Both carotid arteries are visible throughout their entire length. The circle of Willis is closed. There is visualisation of the upper ophthalmic veins with a diameter of 4.8 mm and 2.4 mm on the left, which branch off from the enlarged cavernous sinus. CTA - no signs of organic pathological damage to other anatomical structures of the circulus arteriosum cerebri. Conclusion: MSCT shows signs of bilateral carotid-cavernous fistula.

It is recommended to continue treatment at the Scientific and Practical Center for Endovascular Neurosurgery of the National Academy of Medical Sciences of Ukraine, where the patient was hospitalized on 17.04.2025. At that

Fig. 5. Photo of patient L., born in 1961 (May 23, 2024), after successful surgery for CCS - eyes are calm, position is correct, no exophthalmos
Picture taken by the authors

time, the patient's general condition was relatively satisfactory. Consciousness — 15 points. No meningeal signs. Exophthalmos on both sides, photoreactions preserved. Tongue in the midline. Swallowing is not impaired. Complete blood count and biochemical analysis within normal limits. On the same day, total selective digital cerebral angiography was performed — a carotid-cavernous fistula on the left was diagnosed. No other pathological changes in the extra- and intracranial sections of the cerebral arteries were found. On 04/22/2024, endovascular total disconnection of the carotid-cavernous fistula on the left side was performed using microcoils with balloon assistance. There were no complications. In the early postoperative period, there were no negative dynamics. Significant regression of exophthalmos and chemosis. The pulsating noise in the head disappeared. Consciousness according to the Glasgow Coma Scale — 15 points.

April 25, 2024 — the patient's condition deteriorated sharply — complaints of sharp protrusion of the left eye and hyperemia of the sclera of the left eye, increased blood pressure.

April 25, 2024 — selective cerebral subtractive digital angiography — recurrence of carotid-cavernous fistula on the left. On the same day, surgery was performed: endovascular total separation of the carotid-cavernous fistula on the left using a micro-spiral with balloon assistance and an adhesive composition. There were no complications. Drug therapy was performed: Plasmovene, Asparkam, Ketolong, Co-Prenesa, Prenesa, Concor, Pangastro, Detralex, Azarga, Amlodipine. Against the background of drug therapy, the patient's condition showed positive dynamics: significant regression of exophthalmos, chemosis, and injection of both eyes. Consciousness according to the GCS — 15 points. The patient was discharged on 04/30/2024.

On May 23, 2024, the patient's condition is satisfactory. No complaints. Visual acuity of the right eye = 0.3 with correction sph +2.0D=1.0, left eye = 0.3 with correction sph +2.0D=1.0. Intraocular pressure without hypotensive therapy on a pneumotonometer is 13.5 in the right eye and 17.8 mm Hg in the left eye. Objectively, the position of the eyeballs is symmetrical, correct, there is no exophthalmos, full mobility of the eyeballs, no congestive injection of the eyeballs. (Fig. 5).

In both eyes, the cornea is transparent, the anterior chamber is of medium depth, the iris is structural, the pupil reacts to light, the lenses are transparent, the vitreous body is transparent, the fundus of the optic disc is pale pink, the borders are clear, the caliber and course of the vessels are normal, no hemorrhages or focal changes were found. The patient is under dispensary supervision at her place of residence (Fig. 6).

The difficulty in making a correct diagnosis was associated with the delayed onset of the first symptoms affecting the organ of vision and their slow progression, the presence of other concomitant diseases, and the absence of classic complaints (such as constant noise in the head). The first complaints of redness in the left eye appeared on February 6, 2024 (two months after the traumatic brain injury), and 10 days later (February 16, 2024), an increase in IOP in the left eye was recorded. On 07.03.2024, after another 3 weeks, swelling of the eyelids and injection of both eyes were recorded. After another 3 weeks, during the next examination on 28.03.2024, an increase in exophthalmos in the left eye and its appearance in the right eye, conjunctival chemosis and injection in both eyes, restricted outward movement of the left eye, and changes in the fundus during ophthalmoscopy—edema of the optic disc and significant dilation of the retinal veins. Considering

Fig. 6. Fundus photograph (May 23, 2024) of patient L., born in 1961, after surgery for CCS, left eye optic disc pale pink, clear borders, vein course and caliber within normal limits
Picture taken by the authors

the bilateral exophthalmos and the patient's history of thyroid disease, an MRI of the orbits was performed on March 25, 2024. Considering the study data, the eye muscles were not thickened without pathological changes in the MR signal, the retrobulbar area fat tissue had no visible pathological changes, and the presumed diagnosis of endocrine ophthalmopathy was rejected, but the MR signs of enlargement of the superior vena cava on the right were alarming.

11.04.2024 Performed: CT angiography of the head and neck vessels with intravenous contrast. Conclusion: MSCT — signs of carotid-cavernous fistula bilaterally. More than two months passed from the first signs of the disease to the correct diagnosis. 25.04.2024 Last surgical intervention performed — endovascular total disconnection of the carotid-cavernous fistula on the left side using microcoils with balloon assistance and an adhesive composition, which led to the patient's recovery with complete regression of ocular manifestations and preservation of visual functions.

CONCLUSIONS

1. In cases of atypical clinical presentation, unclear symptoms, and increasing dynamics of the pathological process, modern technologies should be used. In our case, diagnosis and targeted treatment yielded positive results in rehabilitation and restoration of visual functions.
2. With the start of the full-scale invasion of Ukraine by the Russian Federation, the number of traumatic brain injuries among the civilian population and military personnel in Ukraine has increased significantly, and the likelihood of TBI also increases. In all cases of traumatic exophthalmos, it is recommended to be vigilant in terms of carotid-cavernous connection.

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CONFLICT OF INTEREST

The Authors declare no conflict of interest

CORRESPONDING AUTHOR

Antonina V. Varvarynets
Uzhhorod National University
71 Minayska St, 88000 Uzhhorod, Ukraine
e-mail: Antonina.varvarynec@uzhnu.edu.ua

ORCID AND CONTRIBUTIONSHIP

Valery D. Belyaev: 0009-0003-1641-9076 A B
Antonina V. Varvarynets: 0000-0001-5859-1040 D F
Kateryna V. Bora : 0009-0009-6294-2523 A B
Alona V. Stoyka : 0009-0006-3031-685X E F
Ivanna I. Mushchak : 0009-0000-8871-9076 D E

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