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**Acquired carotid-cavernous fistula caused by traumatic intracavernous rupture of an embryonic anastomosis.**

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# Acquired Carotid-Cavernous Fistula Caused by Traumatic Intracavernous Rupture of an Embryonic Anastomosis

Angel Mironov

**Summary:** This case of traumatic carotid-cavernous fistula was caused by rupture of a peculiar ipsilateral intracavernous anastomosis between the accessory meningeal artery and the redundant deep recurrent ophthalmic artery. In the opposite cavernous sinus there was an obvious anastomosis between the accessory meningeal artery and the ophthalmic artery, probably located in the lateral part of the cavernous sinus. The patient was successfully treated with transarterial embolization followed by surgery of the cavernous sinus. In light of the vascular embryology, it is possible that unusual embryonic connections between the primitive dorsal ophthalmic artery and the accessory meningeal artery already existed in both cavernous sinus areas.

**Index terms:** Fistula, carotid-cavernous; Arteries, abnormalities and anomalies

The most common cause of an acquired non-spontaneous carotid-cavernous fistula is a traumatic (1–3) or iatrogenic (4–6) injury of the internal carotid artery. Carotid-cavernous fistulas caused by intracavernous rupture of an aneurysm arising from a persistent primitive trigeminal artery (7–10) or by traumatic rupture of a persistent primitive trigeminal artery (10–13) also have been proposed. We report the case of a patient with an uncommon carotid-cavernous fistula caused by traumatic rupture of a primitive intracavernous anastomosis between an accessory meningeal artery and a redundant deep recurrent ophthalmic artery.

## Case Report

Slowly progressive chemosis followed by proptosis and an audible bilateral bruit developed in a 30-year-old man a few weeks after head trauma with bilateral basal skull fractures. The left internal carotid artery angiogram (Fig 1A) and the right internal carotid artery angiogram (Fig 1B–C) showed the presence of a left indirect carotid-cavernous fistula filled from a left deep recurrent ophthalmic artery, which leaves the orbit through the superior orbital fissure, and from cavernous internal carotid branches. The

left external carotid artery angiogram (Fig 1D) demonstrated an extensive arterial contribution to the fistula from a dilated accessory meningeal artery, arising from the internal maxillary artery and from several dural branches of the artery of the foramen rotundum and also of the middle meningeal artery. The right external carotid artery angiogram (Fig 1E–F) revealed a peculiar arterial anastomosis between the right accessory meningeal artery, arising from the middle meningeal artery, and the right ophthalmic artery. This arterial connection seemed to be located in the right cavernous sinus. It led, first, to opacification of a redundant deep recurrent ophthalmic artery, entering the orbit through the superior orbital fissure, and second, to visibility of the whole right ophthalmic artery with particular opacification of the internal carotid artery. Although both the left and the right accessory meningeal arteries had a different origin, their intracranial portions show an identical angiomorphology. They entered the middle fossa through the foramen ovale and ran anteromedially through the cavernous sinus area toward the superior orbital fissure. There was a predominantly intracranial distribution of both the left and the right accessory meningeal arteries. Moreover, a traumatic arteriovenous fistula was visible at the level of the right jugular foramen with contributions from both the ascending pharyngeal artery and the stylo-mastoid artery on the right (Fig 1E–F).

As a first step, the patient was treated with transarterial embolization: the left accessory meningeal artery and the right ascending pharyngeal artery were embolized with Histoacryl (Braun; Melsungstn, Germany); the right stylo-mastoid artery was embolized with Ivalon (Nycomed; Paris, France) particles. The postembolization angiograms showed a remnant of the carotid-cavernous fistula filled by the left deep recurrent ophthalmic artery and by several dural branches of the artery of the foramen rotundum as well as the middle meningeal artery. There was complete closure of the traumatic arteriovenous fistula at the level of the right jugular foramen. As the next step we proposed to carry out direct venous embolization with coils. However, the patient preferred surgery. Therefore, a subsequent surgical obliteration with packing of the cavernous sinus with muscle was performed. A complete angiographic and clinical cure was obtained.

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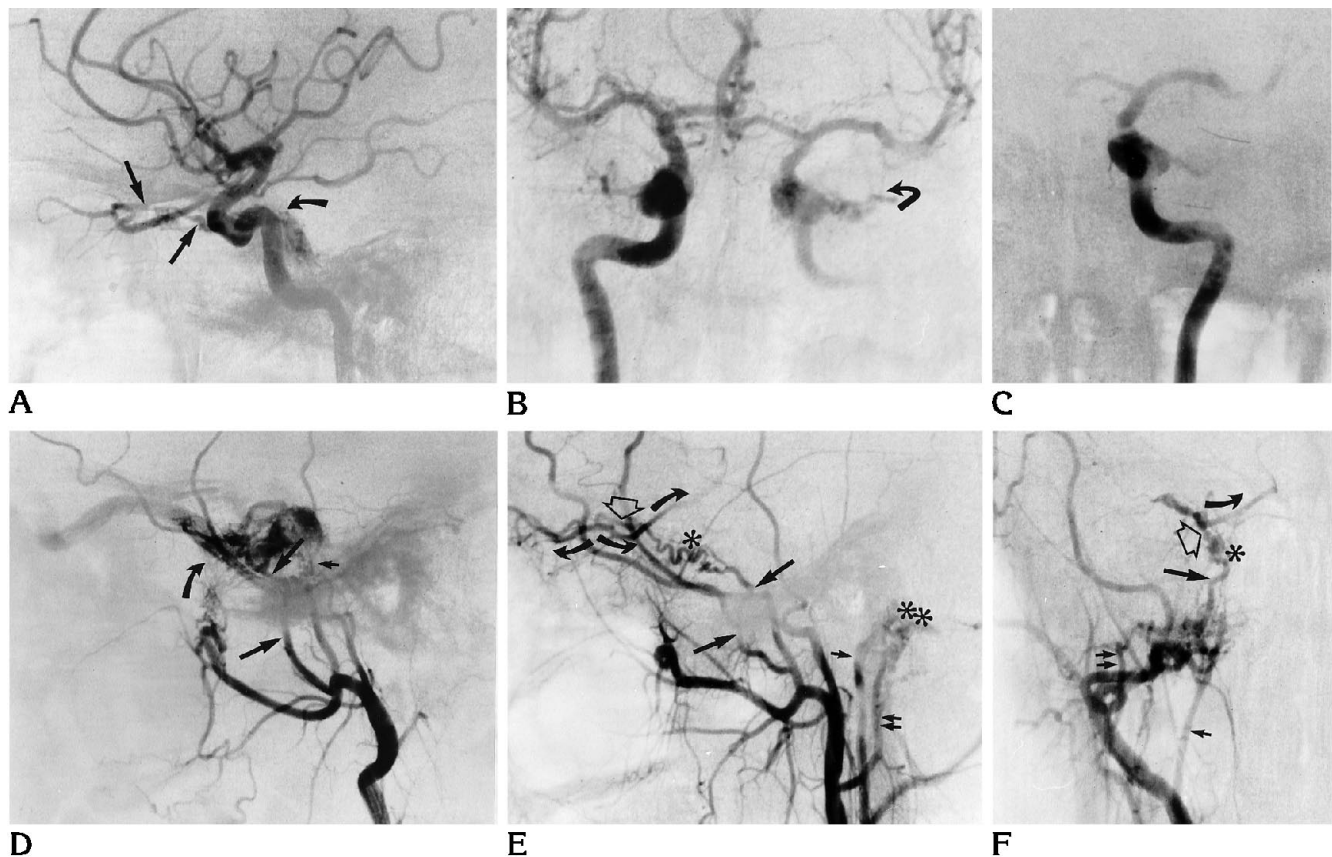


Fig 1. A, Left internal carotid angiogram demonstrates a carotid-cavernous fistula with main supply by the deep recurrent ophthalmic artery (*arrows*). There also is filling of the cavernous sinus through cavernous branches from the C-5 segment (*curved arrow*).

B, Right internal carotid angiogram during compression of the left internal carotid artery, and C, the early arterial phase of the left internal carotid angiogram show that the left ophthalmic artery fills primarily the deep recurrent ophthalmic artery (*curved arrow*), and then the fistula. The lesion therefore involves the deep recurrent ophthalmic artery and not the internal carotid artery. It is an indirect carotid-cavernous fistula.

D, Left external carotid artery angiogram demonstrates an extensive contribution to the fistula from a dilated accessory meningeal artery (*long arrows*) arising from the internal maxillary artery. There also is filling of the cavernous sinus through several dural cavernous branches of the artery of the foramen rotundum (*curved arrow*) and of the middle meningeal artery (*short arrow*).

E, Lateral and F, anteroposterior views of the right external carotid angiogram showing a peculiar arterial anastomosis (*asterisk*) between the right accessory meningeal artery (*arrows*), arising from the middle meningeal artery, and the right ophthalmic artery. This arterial connection is located in the right cavernous sinus. It leads first to opacification of a redundant deep recurrent ophthalmic artery (*open arrows*), which enters the orbit through the superior orbital fissure, and second to visibility of the whole right ophthalmic artery (*curved arrow*) with particular opacification of the internal carotid artery. Moreover, a traumatic arteriovenous fistula is seen at the level of the right jugular foramen (*double asterisk*) with contribution from both the ascending pharyngeal artery (*short arrow*) and the stylomastoid artery (*double short arrow*).

## Anatomy and Discussion

At the 8-mm stage in the embryo, there are two arterial supplies of the orbital region: the primitive ventral ophthalmic artery, which arises from the anterior cerebral artery and reaches the orbit through the optic canal, and the primitive dorsal ophthalmic artery, which arises from the carotid siphon and reaches the orbit through the superior orbital fissure (14). At the 39-mm embryo stage, both the proximal parts of the ventral ophthalmic artery and the

dorsal ophthalmic artery have usually disappeared. The ultimate adult configuration of the ophthalmic artery will arise from the supracavernous internal carotid syphon (14, 15). Variations in this developmental program result in numerous anatomic variants of the orbital arterial supply. If the embryonic dorsal ophthalmic artery has not disappeared at the superior orbital fissure, the orbit will be supplied by two branches arising from both the C-2 and the C-4 portions of the carotid siphon (the sections of

the internal carotid artery are categorized according to Kraysenbühl and Yasargil [16]), or by an ophthalmic artery with C-4 origin only, entering intraorbitally through the superior orbital fissure (17). The dorsal remnant of the primitive dorsal ophthalmic artery corresponds to the inferolateral trunk arising from the C-4 portion of the carotid siphon. The proximal remnant of the dorsal ophthalmic artery is called the "deep recurrent ophthalmic artery"; it unites with the intraorbital ophthalmic artery and, after coursing the superior orbital fissure, penetrates the cavernous sinus, where the deep recurrent ophthalmic artery connects cavernous branches (18).

The accessory meningeal artery is almost always present (96%) and arises about equally from the internal maxillary artery or the middle meningeal artery. Its origin varies with the anatomical variants of the internal maxillary artery (19, 20). In the deep variety (internal maxillary artery medial to the lateral pterigoid muscle), the accessory meningeal artery arises directly from internal maxillary artery (Fig 1D); in the superficial variation (internal maxillary artery lateral to the lateral pterigoid muscle), it originates from the middle meningeal artery (Fig 1E-F) (21). The greater part of the distribution of the accessory meningeal artery supplies structures outside the cranial cavity, divided into three regions: the lateral, the medial, and the interpterigoid. Only 10% of the blood carried by the accessory meningeal artery supplies structures of the intracranial territories by its ascending intracranial ramus, which usually enters the middle fossa through the foramen ovale or through the sphenoid emissary foramen (foramen Vesalius) (19). The caliber of this vessel varies widely and can sometimes be very large, even in the absence of vascular lesions, particularly when it constitutes the main supply for the cavernous area. In such cases, there are anastomoses to the four main territories of the inferolateral trunk: the superior or tentorial branch, the anteromedial branch, which passes through the superior orbital fissure, the anterolateral branch into the foramen rotundum, and the posterior branch under the trigeminal ganglion (18).

It is postulated that the anatomic variants in the blood supply of the cavernous area reproduce the major phylogenetic alternatives of the anatomic disposition in humans (17). Schematically, there exists a balance in the blood supply

to the cavernous region between the contribution of the internal maxillary artery, through its intracranial branches, and the contributions of the cavernous branches of the internal carotid artery (17). If the internal carotid branches are dominant, the C-4 segment gives off a prominent trunk, which gives rise to diverging branches in the four territories of the inferolateral trunk; these branches anastomose with branches of the internal maxillary artery, which are smaller but present. If the transcranial branches of the internal maxillary artery are dominant, it is the ascending intracranial branch of the accessory meningeal artery that enters this region via the foramen ovale (or foramen Vesalius), giving four branches identical to those described in the inferolateral trunk. In the intermediate forms, the cavernous region is supplied from two sources: one, the accessory meningeal artery entering through the foramen ovale (for the anterolateral and the posterior territories), and the other, the inferolateral trunk arising from the internal carotid artery (for the superior and the anteromedial territories).

In our patient, there were symmetric embryonic connections between the redundant deep recurrent ophthalmic artery and the accessory meningeal artery on both sides. These findings may be explained as the result of peculiarities of the vascular balance of the cavernous sinus areas in association with variations in the multiple steps during development as follows: First, although both ophthalmic arteries showed development of definitive adult type, both primitive dorsal ophthalmic arteries had obviously not disappeared at the superior orbital fissure, and each orbit was supplied by two branches—one entering through the optic canal (the definitive ophthalmic artery), and the other entering through the superior orbital fissure (the redundant deep recurrent ophthalmic artery). Second, there was an extreme situation of the vascular balance represented by the exclusive external carotid arterial supply of the cavernous areas via the cavernous branches of both accessory meningeal artery. This corresponds to an earlier morphologic arrangement identical to the rete mirabile of the earliest human embryo (17). Third, although neither inferolateral trunk was developed, both primitive dorsal ophthalmic arteries arose directly from the intracranial parts of both accessory meningeal arteries, and thus from the external carotid systems. In fact, there is at no stage of development any direct

connection between the dorsal ophthalmic artery and the accessory meningeal artery. Because a completely unambiguous embryologic explanation does not seem to be possible and the cause of the persistent redundant embryonal connections described in this patient is unknown, it seems feasible to postulate an abnormal embryonal connection between the primitive dorsal ophthalmic artery and the accessory meningeal artery.

From the severe trauma to the skull, the left embryonic connection was probably disrupted just at the cavernous sinus, leading to the development of an indirect carotid-cavernous fistula (Figs 1A, D). On the right, there is a complete embryonic anastomosis, probably located in the lateral part of the cavernous sinus (Fig 1E-F). The vascular contribution from the left artery of the foramen rotundum and the cavernous branches of both the left internal carotid artery and the external carotid artery might be explained either as a hemodynamic response to the high-flow lesion or as associated traumatic intracavernous disruptions of these branches.

It has been suggested that the transvenous embolization of the cavernous sinus is the treatment of choice in cases with symptomatic dural fistulas that fail to respond to carotid-jugular compression (22, 23) or in cases with nonspontaneous carotid-cavernous fistula that fail to respond to transarterial occlusion (24). The transarterial embolization in our patient resulted in complete closure of the traumatic arteriovenous fistula at the level of the right jugular foramen, but only a flow reduction of the left carotid-cavernous fistula. We concur that the treatment of choice for the remnant of the left carotid-cavernous fistula would have been a transvenous approach, but the patient did not agree to this procedure. However, the direct surgical approach was successful.

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