

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Spontaneous thrombosis of post-traumatic direct carotid-cavernous fistula [☆]

Anggriani Gita Fransiska*, Achmad Firdaus Sani, Dedy Kurniawan

Department of Neurology, Neurology, Airlangga University Faculty of Medicine, Dr. Soetomo General Hospital, Mayjend Prof. Dr. Moestopo No. 6–8, Surabaya 60264, Surabaya-Indonesia

ARTICLE INFO

Article history:

Received 25 March 2024

Revised 16 April 2024

Accepted 29 April 2024

Keywords:

Carotid-cavernous fistula

Spontaneous resolution

Embolization

ABSTRACT

Carotid cavernous fistula (CCF) is an abnormal communication between the carotid artery and the cavernous sinus, which is most commonly caused by trauma. Due to its high-flow nature and aggressive clinical course, spontaneous resolution of CCF is rare, making endovascular embolization necessary. This procedure aims to prevent potential vision loss, emphasizing the importance of timely intervention to preserve visual function. We report a case of Barrow type A carotid-cavernous fistula which resolved spontaneously. A 42-year-old male was referred to Emergency Room with a chief complaint of seizure 3 days before admission. The seizure was both arm stiff and jerking, the eyes gazing upward, bitten tongue, foamy mouth, no bed wetting. Meanwhile, the patient was unconscious during and after the seizure. Regarding the medical history, the patient experienced head trauma 8 months ago due to a traffic accident. Additionally, the patient reported symptoms of redness, swelling, double vision, and inability to look to the right eye. Initial cerebral angiography was performed, reporting a carotid-cavernous fistula of Barrow type A. Further endovascular treatment could not be carried out, but a second cerebral angiography was conducted after a year, demonstrating spontaneous thrombosis of the carotid-cavernous fistula of Barrow type A. CCF were classified by Barrow et al. into types A, B, C, and D. It has been proposed that spontaneous resolution of CCF Barrow type A may result from thrombosis of the cavernous sinus.

© 2024 The Authors. Published by Elsevier Inc. on behalf of University of Washington.

This is an open access article under the CC BY-NC-ND license

(<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

Introduction

A carotid-cavernous fistula (CCF) is an abnormal connection between the carotid arterial system and the cavernous sinus. It is classified as direct or indirect, high-flow or low-flow,

and can be referred to as Barrow types A, B, C, or D. Barrow type A is the connection between the internal carotid artery (ICA) and the cavernous sinus. Furthermore, all type A fistulas are direct communications and typically exhibit high flow. The majority of traumatic CCF cases are Barrow type A [1].

[☆] Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

* Corresponding author.

E-mail addresses: agfransiska@gmail.com (A.G. Fransiska), dedy_k2002@yahoo.com (D. Kurniawan).

<https://doi.org/10.1016/j.radcr.2024.04.089>

1930-0433/© 2024 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

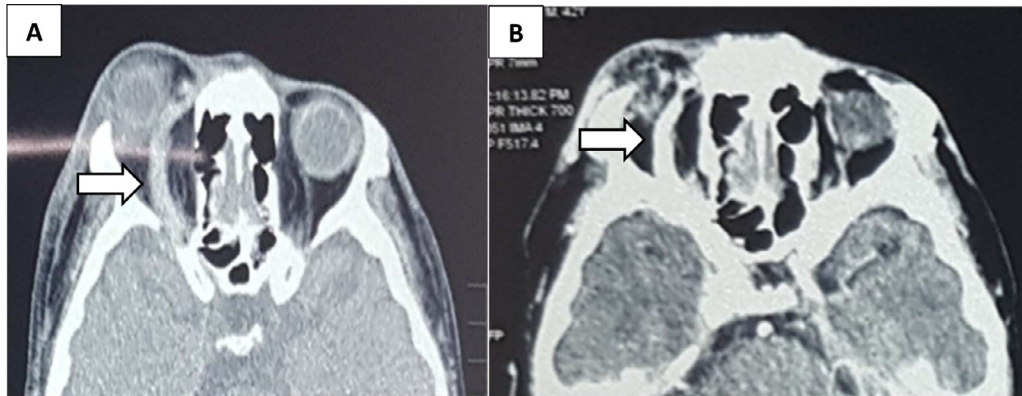


Fig. 1 – Axial Head CT on April 13th 2021. without contrast (A), Axial Head CT on April 13th 2021 with contrast. (B) showing dilated right superior ophthalmic vein (White arrow).

Types B, C, and D fistulas are considered “indirect” because they involve the connection between the cavernous sinus and the main branches of the ICA and external carotid artery (ECA) but not directly on the ICA or ECA. The cavernous sinus is connected to the meningeal branches of the ICA and ECA in the Barrow type B and C fistulas, as well as both the meningeal branches of the ICA and ECA in the Barrow type D fistula [1].

Venous congestion resulting from the blood flow in any type of fistula within the cavernous sinus can affect surrounding structures such as cranial nerves III, IV, V1, V2, and VI on the ipsilateral side. Meanwhile, clinical presentations are associated with ocular manifestations, including proptosis, chemosis, changes in visual acuity, diplopia, and eye pain, followed by headache and epistaxis [1].

Spontaneous resolution can be achieved in 20%-50% of cases with concurrent management of ocular symptoms with prism or patching therapy for diplopia, topical agents for glaucoma, lubrication for keratopathy, and/or systemic corticosteroids. However the patients who treated with a conservative approach should be followed carefully for worsening of ocular symptoms, which often warrants treatment to prevent serious complications including intracranial hemorrhage. In general, endovascular treatment is considered the primary treatment for CCFs. Transarterial obliteration is commonly utilized in the management of direct, high-flow CCFs [2].

Direct CCFs are always treated with endovascular methods. The goal is to occlude the fistula but preserve the patency of the internal carotid artery (ICA). Agents include detachable coils or liquid embolic agents delivered transarterially or transvenously. Arterial porous or covered stents are often used adjunctively. In rare cases, the ICA must be occluded [3].

terized by jerking movements of the hands, upward eye deviation, tongue biting, foaming at the mouth, and no urinary incontinence. Each seizure episode lasted 1 minute, occurring twice within a 5-minute interval, and remained unconscious during and after the seizures. In the past medical history, the patient had a traffic accident 8 months ago in Semarang while riding a motorcycle without wearing a helmet, suffered a head injury, and remained unconscious for 4 days. The right eye was red, swollen, experienced double vision, and had limited rightward gaze. There were no complaints of weakness on either side of the body, facial droop, or slurred speech. The patient denied a history of hypertension or diabetes and did not experience similar incidents before.

During the treatment at the Semarang Hospital, a cerebral angiography procedure was performed, showing a Barrow type A fistula (Figure 2). Computed tomography scan orbit showed proptosis of right eyeball with dilated tortuous superior ophthalmic vein (Figure 1). Cerebral angiography evaluation after 7 months shows complete resolution of carotid cavernous fistula with preservation of internal carotid artery blood flow (Figure 3).

Following a complete recovery of the initial condition, gradual onset of double vision was observed specifically while attempting lateral gaze. This double vision became persistent even in the absence of lateral eye movements. Consequently, the patient was readmitted to the hospital with complaints of recurring seizures. After the seizures were resolved, the patient complained of persistent double vision. Unfortunately, the previous hospital was unable to perform embolization due to the lack of modalities available at the site.

A second cerebral angiography was performed, but no direct right CCF was observed. Therefore, it was determined that the patient had experienced spontaneous thrombosis.

Case report

A 42-year-old male patient presented to the emergency department with the chief complaint of seizures. The patient experienced seizures for the past 3 days, which were charac-

Discussion

Spontaneous thrombosis in CCF is rare and often found in indirect-type fistulas. Seeger et al. reported 9 and 2 cases of this condition in indirect and direct CCFs. The mecha-

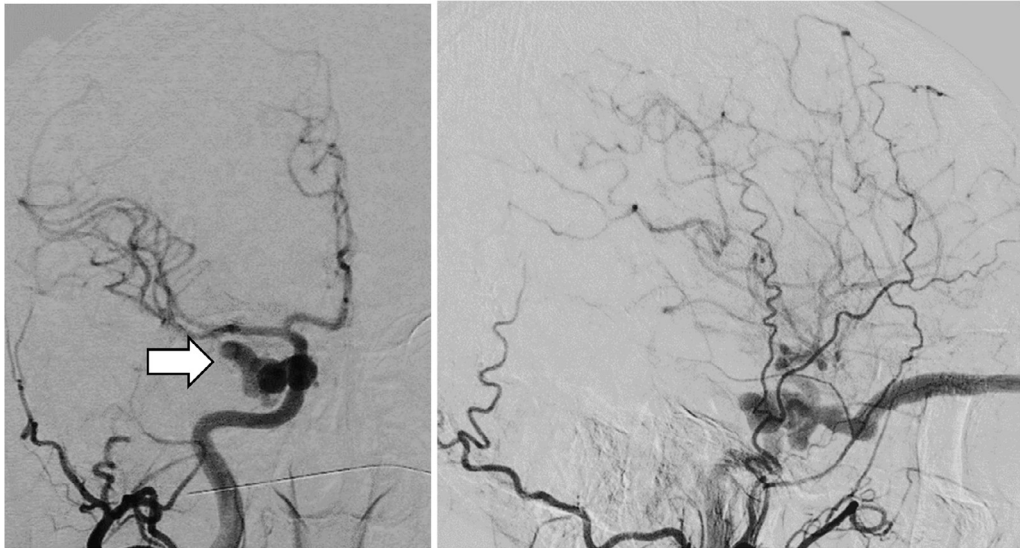


Fig. 2 – Cerebral angiography on May 24th, 2021 Right carotid angiogram showing traumatic carotid-cavernous fistula with dilated superior ophthalmic vein.

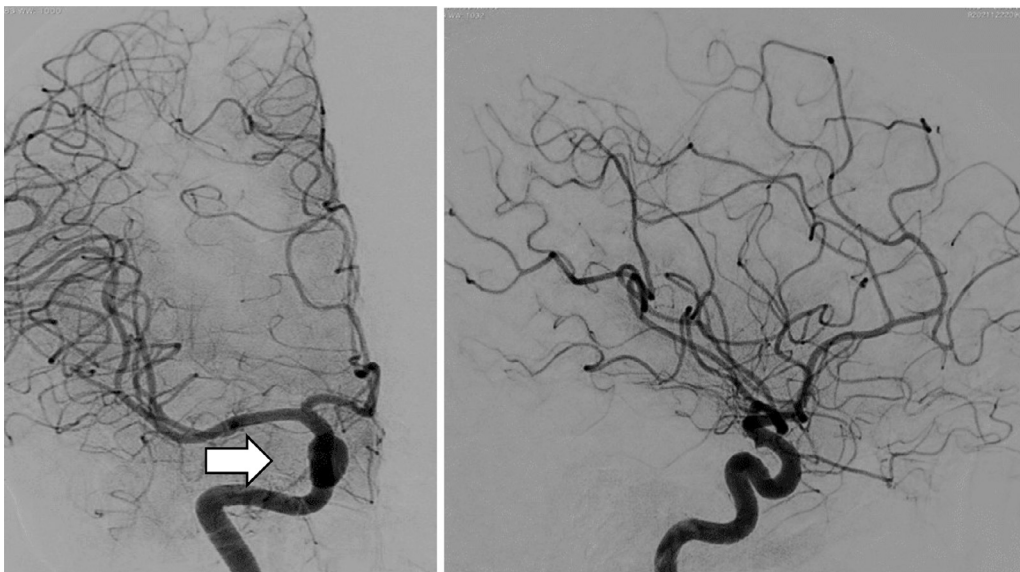


Fig. 3 – Cerebral angiography on December 22th, 2021 after 7 months shows complete resolution of carotid cavernous fistula with preservation of internal carotid artery blood flow.

nism behind spontaneous thrombosis in slow-flow CCFs involves venostasis and damage to the cavernous sinus due to venous hypertension [3,4]. Furthermore, the injection of iodine-containing contrast during angiography can lead to excessive leukocyte accumulation, clotting, and red blood cell aggregation, as well as directly affecting the endothelium of blood vessels, which can trigger spontaneous thrombosis events [5].

Non-invasive diagnostic methods such as ultrasound and magnetic resonance imaging can show plaque morphology and distinguish vulnerable plaques [6]. However, there are certain limitations as ultrasound waves are absorbed by the skull and brain structures are not presented in details. MRI or CT

cerebral images are of high quality, but rather expensive, time consuming, and impossible to repeat frequently. The ultrasound fusion imaging system gives new promising possibilities combining the benefits of both modalities [7].

Lewis et al. reported cases of spontaneous resolution in 3 cases of direct CCFs after failed attempts at balloon placement and 1 case of iatrogenic dissection of the cervical carotid artery. A similar spontaneous resolution was observed in experimental embolization attempts without balloon placement. Whang et al. also reported cases where inadvertent thrombus formation occurred during navigation with a microcatheter and micro guidewire through a small fistula, resulting in the closure of the fistula. Meanwhile, direct CCFs can

spontaneously close after failed transarterial approaches and during transvenous approaches [8].

Immediate spontaneous occlusion of traumatic CCFs following orbital venography was reported by Nishijima et al. The likely mechanism leading to the spontaneous closure of the fistula is a temporary reduction in the pressure gradient between the cavernous sinus and ICA, resulting in static blood flow and thrombus formation. Furthermore, direct cervical carotid angiography can lead to complications, including artery spasms and dissection. Proximal occlusion of the ICA and arterial dissection in the proximal segment of the fistula can be factors in spontaneous resolution due to decreased blood pressure, leading to fistula thrombosis [8].

In the case presented, a Barrow type A fistula was identified during cerebral angiography following a traffic accident. Meanwhile, CCF type A can be managed through endovascular embolization using balloon or coil placement. Transarterial balloon embolization is also the preferred procedure for CCF, with transvenous embolization as an alternative when the transarterial approach proves unsuccessful [9].

However, in this particular case, only cerebral angiography was performed without embolization, which completely improved the condition of the patient. After 8 months, a second cerebral angiography was conducted, showing spontaneous thrombosis. Direct high-flow CCF represents a rare complication in cases of head trauma and typically necessitates endovascular intervention. The occurrence of spontaneous occlusion after diagnostic cerebral angiography is an infrequent phenomenon [10].

Spontaneous closure of type A fistula is a rare phenomenon. The mechanism of spontaneous closure is unknown. Cerebral angiography might play a role in spontaneous resolution of CCF as reported in few cases. Mechanism thought to be responsible for spontaneous closure of direct CCF are slow flow due to ICA dissection leading to venous stasis and damage to vascular endothelium of cavernous sinus by venous hypertension [11].

Another possible mechanism is the use of iodinated contrast medium that exaggerates leukocytic accumulation, promotes red blood cell aggregation and direct effect on vascular endothelium leading to thrombosis. The absence of posterior drainage of CCF through superior and inferior petrosal sinus may also play a role in spontaneous closure of CCF, which was the most probable mechanism of closure of fistula in our case [11].

Nishijima et al. reported spontaneous occlusion of TCCF after orbital venography. Castillo et al. reported spontaneous thrombosis of a direct CCF, which was confirmed by Gadolinium-diethylene triamine pentaacetic acid (GD-DTPA) enhanced magnetic resonance [12].

Conclusion

In conclusion, direct high-flow CCF is a rare complication of head injury and usually requires endovascular intervention

for treatment. Spontaneous thrombosis after diagnostic cerebral angiography can also occur although it is a rare phenomenon.

Patient consent

I have received consent from the patients, and their legal representatives for use their radiology images and their case to publication in journal.

REFERENCES

- Hasan S, Pedro A-S, Avila MJ, El-Ghanem M, Dumont TM. Carotid cavernous fistula treatment via flow diversion: a systematic review of the literature. *World Neurosurg* 2021;149:e369–77. doi:10.1016/j.wneu.2021.02.015.
- Texakalidis P, Tzoumas A, Xenos D, Rivet DJ, Reavey-Cantwell J. Carotid cavernous fistula (CCF) treatment approaches: a systematic literature review and meta-analysis of transarterial and transvenous embolization for direct and indirect CCFs. *Clin Neurol Neurosurg* 2021. doi:10.1016/j.clineuro.2021.106601.
- Gemmete Joseph J, Ansari Sameer A, Gandhi Dheeraj M. Endovascular techniques for treatment of carotid-cavernous fistula. *J Neuroophthalmol* 2009;29(1):62–71. doi:10.1097/wno.0b013e3181989fc0.
- Newton TH, Hoyt WF. Dural arterio-venous shunts in the region of the cavernous sinus. *Neuroradiology* 1970;1:71–81.
- Sanders MD, Hoyt WF. Hypoxic ocular sequelae of carotid cavernous fistulae. *Br J Ophthalmol* 1969;53:82–97. doi:10.1136/bjo.53.2.82.
- Peycheva M, Deneva T, Zlatareva D, Zdravkova T, Chervenkov L, Harizanova Z. Image and laboratory aspects of Carotid atherosclerosis. *Experiment Appl Biomed Res (EABR)* 2023;24(2):135–44.
- Peycheva MV, Chervenkov L, Harizanova Z, Ahmed-Popova F, Zahariev ZI. Ultrasound fusion imaging system in neurology practice. *Folia Med (Plovdiv)* 2022;64(4):667–71.
- Ritchie WGM, Lynch PR, Stewart GJ. The effect of contrast media on normal and inflamed canine veins. A scanning and transmission electron microscopic study. *Invest Radiol* 1974;9:444–55. doi:10.1097/00004424-197411000-00005.
- Iampreechakul P, Tirakotai W, Tanpun A, Wattanasen Y, Lertbusayanukul P, Siriwimonmas S. Spontaneous resolution of direct carotid-cavernous fistulas: case series and literature review. *Interv Neuroradiol* 2019;25(1):71–89. doi:10.1177/1591019918800220.
- Halbach VV, Higashida RT, Hieshima GB, Hardin CW, Yang PJ. Transvenous embolization of direct carotid cavernous fistulas. *AJNR Am J Neuroradiol* 1988;9:741–7.
- Meena US, Gupta P, Shrivastava T, Purohit D. Spontaneous closure of post-traumatic high-flow carotid-cavernous cerebral angiography. *Asian J Neurosurg* 2016;11(2):172. doi:10.4103/1793-5482.175636.
- Nishijima M, Iwai R, Horie Y, Oka N, Takaku A. Spontaneous occlusion of traumatic carotid cavernous fistula after orbital venography. *Surg Neurol* 1985;23:48992.