

Case Report

Carotid cavernous fistula with retrograde flow to cortical veins successfully treated with endovascular detachable balloon

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ABSTRACT

A carotid-cavernous fistula (CCF) is an abnormal vascular connection, which allows blood to flow either directly or indirectly from the carotid artery into the cavernous sinus. The classic triad of ocular symptoms mostly seen in direct CCFs but less common without ocular symptoms. This difference in symptoms depends on the draining venous flow from the fistula, whether it affects the ophthalmic veins or affects the cortical veins. We presented a case of a 32-year-old man with decreased consciousness, seizures, vomiting, a history of long-standing headache, ringing in the ears and double vision, without ptosis, ophthalmoplegia and without proptosis. Head CT showed a sulcal and perimesencephalic type of subarachnoid hemorrhage. Digital subtraction angiography (DSA) results obtained arterial vein fistula from the right internal carotid artery to the cavernous sinus with the dominant vein draining into the cortical vein, accompanied by dilatation and elongation of the cortical veins. No disturbances in ophthalmic venous outflow were found. Endovascular therapy was performed to close the fistula with detachable balloon embolization modality, with the result of complete fistula closure. The symptom pattern of CCF depended on flow velocity, location of CCF venous drainage, inflammation and pressure within the venous sinuses. Cerebral catheter angiography is the gold standard imaging modality used in the diagnosis and classification of CCF and embolization using a detachable balloon is one of the treatment options for direct CCF.

Keywords: Carotid cavernous fistula, Detachable balloon embolization, Cortical vein retrograde drainage

INTRODUCTION

A CCF is an abnormal vascular connection, which allows blood to flow either directly or indirectly from the carotid artery into the cavernous sinus. CCF is commonly associated with cortical venous reflux. Obliteration or stenosis of the venous drainage route causes convergent venous outflow that progresses to cortical venous reflux and results in cerebral parenchymal venous congestion or intracerebral hemorrhage.¹

CCFs are classified according to the hemodynamic nature, etiology or anatomy of the fistula. Hemodynamic

classification separates CCF into high-flow and low-flow fistulas.

The etiologic classification distinguishes spontaneous lesions from those that occur due to trauma. Anatomical classification defines direct CCF, which is a direct connection with the carotid artery, whereas CCF does not originate directly from the carotid artery branch vessels.¹

There is one classification that facilitates the clinician in planning the endovascular approach namely the venous drainage-based classification system proposed by Thomas et al complements Barrow's classification, which characterizes CCFs based on their arterial supply.²

Traumatic CCF is the most common type, accounting for up to 75% of all CCFs.

Traumatic CCF is reported to occur in 0.2% of patients with craniocerebral trauma and up to 4% of patients who have skull base fractures with demographics associated with traumatic injury, traumatic CCF is most commonly seen in young male patients. The incidence of spontaneous CCF is about 30% of all CCFs, usually found in older female patients. Ruptured ICA cavernous aneurysm is often cited as a direct and spontaneous cause of CCF.¹

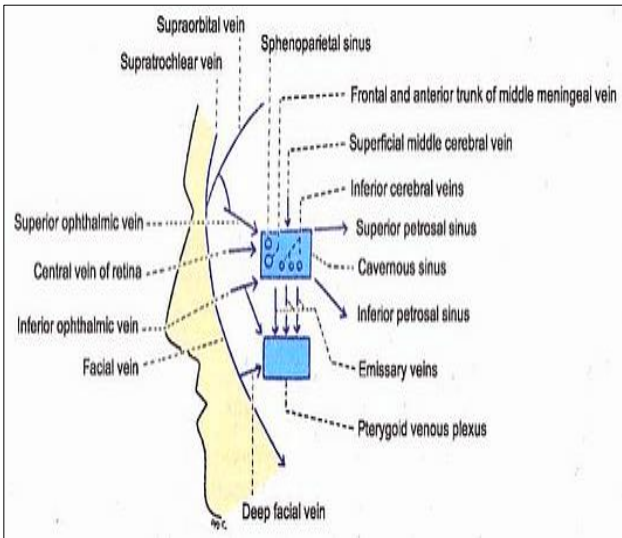


Figure 1: Cavernous sinus flow.³

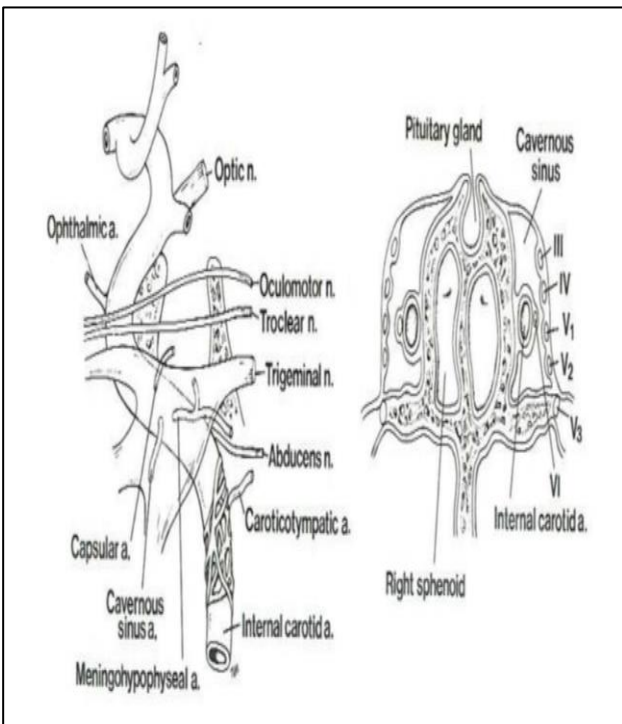


Figure 2: Sinus cavernous.³

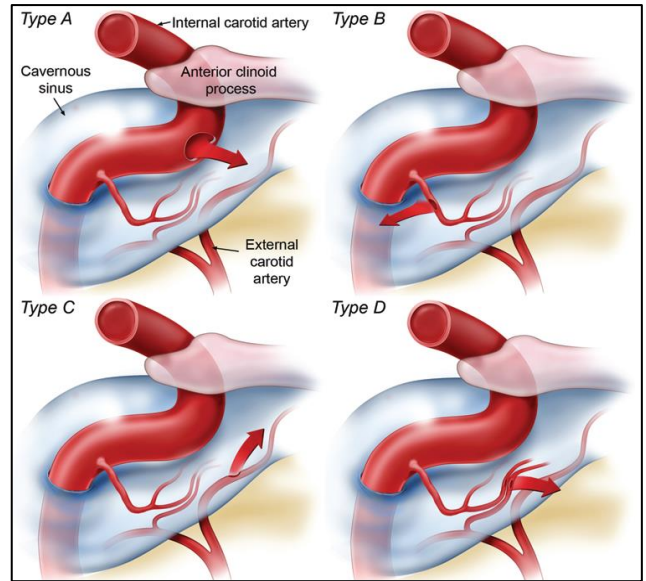


Figure 3: Barrow classification of CCF. Type A fistulas are characterized by direct shunting of blood flow from the ICA into the cavernous sinus. Type B and C fistulas are shunts to the cavernous sinus from branches of the ICA and ECA, respectively. Type D fistulas have shunts from both the ICA and ECA simultaneously.¹

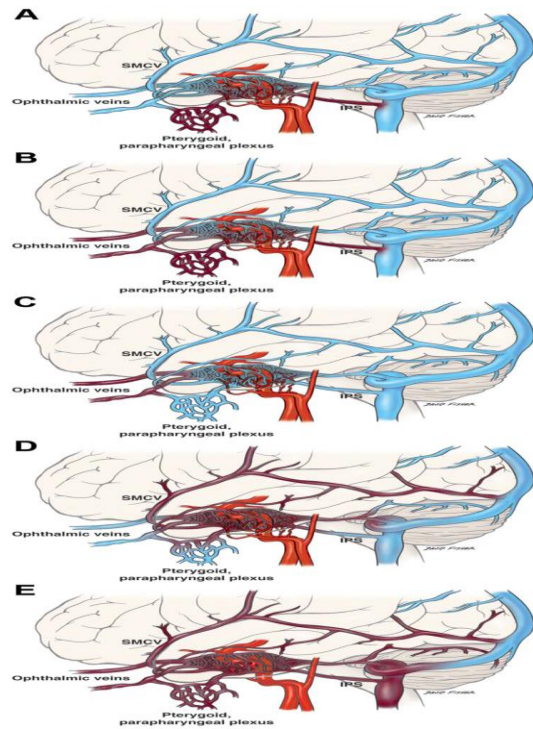


Figure 4: Venous drainage-based classification system for carotid cavernous fistulae. The venous drainage-based classification system for carotid cavernous fistulae. Normal venous anatomy is colored light blue. Preferential drainage of the individual fistula types are colored dark red. Type A, type IB, type IIC, type IIID, type IVE, type V- IPS (inferior petrosal sinus); SMCV (superficial middle cerebral vein).²

Cerebral catheter angiography is the gold standard imaging modality used in the diagnosis and classification of CCF.

Although historically difficult to treat, these lesions are now routinely managed with low morbidity and mortality. Endovascular intervention with the goal of complete fistula occlusion while maintaining normal blood flow through the internal carotid artery is the treatment of choice. In certain cases, surgery, radiosurgery, or conservative management are also treatment options. Resolution of symptoms with a low recurrence rate can be expected in most cases after appropriate therapy.³

Flow of the venous system into the cavernous sinus: from orbitals were superior ophthalmic vein; inferior ophthalmic vein and retinal central vein. From the brain were superficial middle cerebral vein and inferior cerebral vein. From the meninges were sphenoparietal sinus and middle meningeal sinus (venous).

Classification

Based on the nomenclature Barrow et al defined 4 types (type A-D) CCF. Type A CCF is a direct high-flow lesion that connects the internal carotid artery (ICA) directly to the cavernous sinus. Type A CCF often resulted from a single tear in the carotid artery wall, caused by trauma or aneurysm rupture. This was the most common type of CCF, accounting for about 75-80% of all CCFs. CCF types B, C and D were all indirect, low-flow lesions that arose from the meningeal branches of either the ICA or the external carotid artery (ECA). Type B CCFs arose from the meningeal branches of the ICA, type C CCFs arose from the meningeal branches of the ECA and type D CCFs arose from the meningeal branches of the ICA and ECA. In a study of 132 patients with CCF, 100 CCF (75.8%) were classified as type A, none were classified as type B, 4 (3%) were classified as type C and 28 (21.6%).¹

Today, CCF venous drainage was more important for the management of these lesions and should be a key component of any contemporary classification system. Thus, a classification system based on venous drainage presented by Thomas et al complemented Barrow's classification, which characterized CCF based on their arterial supply.

Venous drainage-based classification system for carotid cavernous fistula

Type 1

Type 1 is drainage only posterior/inferior.

Type 2

Type 2 is posterior/inferior and anterior drainage.

Type 3

Type 3 is drainage only to the anterior.

Type 4

Type 4 is retrograde drainage to cortical veins and other venous drainage routes.

Type 5

Type 5 is high-flow direct shunt between the internal carotid cavernous and cavernous sinuses (Barrow type A) and other multiple venous drainage routes.²

CASE REPORT

A 32-year-old man experienced a sudden loss of consciousness at rest 4 days before being admitted to the hospital. Previously there was a headache, throbbing pain on the right side spreading to the entire back of the head, severe intensity, continuous frequency, tinnitus, projectile vomiting, tonic seizure in once a time, stiff all over, duration ±3 min, eyes glaring upwards, there was a bitten tongue, there was no foaming at the mouth, pee one's pants. Before, during and after the seizure the patient was unconscious. Weakness on both sides of the body, the mouth was stuck to the right.

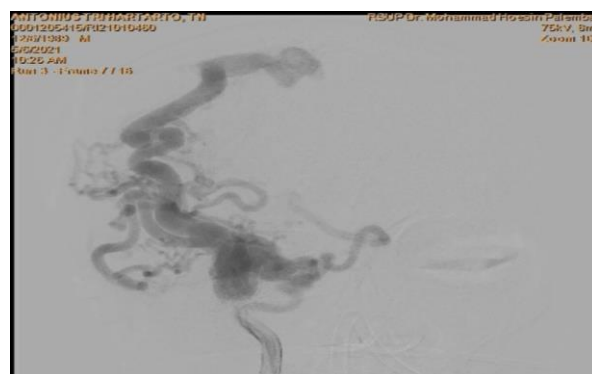


Figure 5: Cortical drainage of right ICA cavernous fistula.

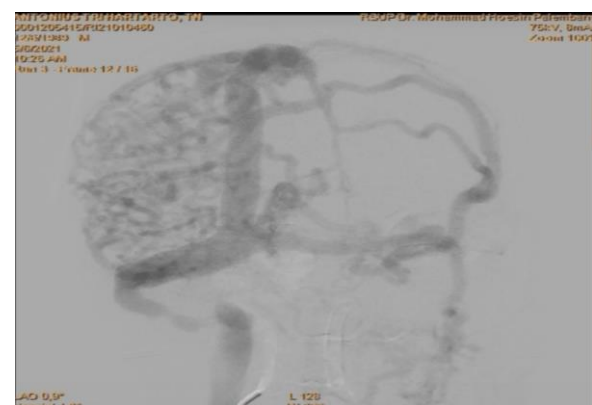


Figure 6: Dilatation of cortical superficial vein.

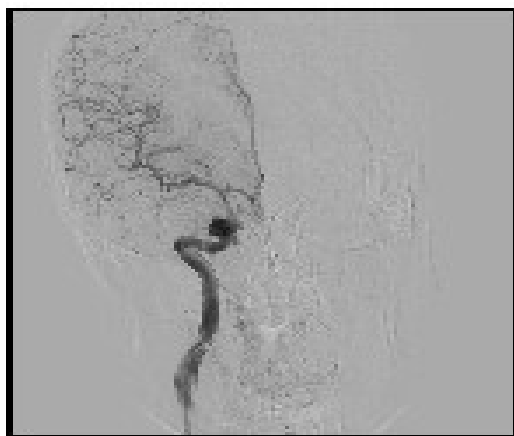


Figure 7: After treatment with detachable balloon.

There was no history of hypertension, no history of diabetes, no history of heart disease, no history of kidney disease. History of long-standing headache since March 2021, intermittent, throbbing pain on the rightside that spreads to all parts of the head, moderate to severe intensity, tinnitus, no hearing loss, no red eyes, no double vision. The patient had been treated at the previous regional hospital for 1 week but there was no change and was sent home. Then the patient's headache was getting worse accompanied by tinnitus, so the patient was referred to another hospital in Palembang and treated for 1 week, the patient was sent home because the headache improved. There was a history of head trauma 12 years ago, said it was light bleeding, was treated for about 1 month, sequelae of double vision were presented (lasted 1 month), the mouth was stuck to the right, the right eye was difficult to close and the ears were tinnitus. Previous medical history at the hospital were tranexamic acid 3×500 mg intravenously, nimotop 4×30 mg orally, tramadol 3×100 mg intravenously, dexamethasone 3×5 mg intravenously.

The results of the physical examination showed sensorium, quadriparesis with increased tone in all four extremities, lagging right corner of the mouth, lagophthalmus, flat right forehead wrinkles. Decreased consciousness, positive Babinski group pathological reflex on both legs. The CT scan results showed suggestive images of arteriovenous malformation (AVM) in the right frontoparietotemporal and left frontotemporal regions and bilateral cerebral edema.

The patient was then performed on the first DSA. Arterial vein fistula image was obtained from the right internal carotid artery to the cavernous sinus with dominant venous drainage to the cortical vein, accompanied by dilatation and elongation of the veins, with collateral from the left anterior and right posterior circulation to both the right middle carotid artery (R MCA) and right anterior cerebral artery (R ACA). Then it was suggested that the right internal cerebral artery (R ICA) sacrifice be preceded by a balloon occlusion test. While the

administration of tranexamic acid was continued with carotid massage.

In the second DSA, an arterial vein fistula image from the right internal carotid artery to the cortical vein accompanied by dilatation and elongation of the vein, with collateral from the left anterior and right posterior circulation both to the R-MCA and R-ACA post embolization using a gold ball that produces blood vessels patented R ICA, R MCA, and R ACA.

DISCUSSION

In this case report, a 32-year-old man, private work, experienced a sudden decline in consciousness. Previously, the patient had a headache, severe intensity, convulsions and projectile vomiting. From the anamnesis, the first thought was an acute condition, namely vascular disorders or trauma. For now, the most likely cerebrovascular causes in this case, which were close intracerebral hemorrhage and subarachnoid hemorrhage. This patient had a history of trauma 12 years ago, at that time the patient experienced a loss of consciousness and was admitted to a local hospital where he said there was bleeding in the head from the CT scan results, then the patient experienced double vision which lasted for 1 month. The double vision felt by the patient may be due to vascular abnormalities due to trauma such as CCF, because in CCF there can be disturbances in N III (N III compression due to increased pressure in the cavernous sinus) which caused accommodation disorders. It was estimated that type D CCF initially occurred in this patient, in this patient categorized as mild symptoms because there was good compensation from the collateral system, this was also influenced by arteriovenal malformations will be symptomatic depending on angioarchitecture. Since 12 years, there had been sequelae in the form of a tight mouth to the right and difficulty in closing the right eye, which was suspected as a sequel to Bell's palsy. Significant symptoms towards CCF were complaints of tinnitus that had been felt since 12 years ago until now. Currently, the sequelae of Bell's palsy in the patient cannot be associated with the vascular causes of the patient at this time.

On physical examination, the NPRS value on admission was 9. Clinical values that can be obtained in conditions of increased intracranial pressure, especially in SAH. The presence of GRM in this case also confirmed the diagnosis towards subarachnoid hemorrhage. However, other investigations were needed to prove the cause of SAH in this patient, whether it was due to aneurysm rupture, AVM rupture or CCF rupture. This patient did not have paresis N III, IV, VI, red eye symptoms or swelling in the eye area, this meant that the ophthalmic vein was not involved in this CCF case.

Laboratory investigations showed values within normal limits. On the ECG examination with the impression junctional PVC. Anteroseptal ischemia V1-V4 and

diagnosed as compensatory coronary artery disease (CAD), but in this case the effect of heart disease experienced by the patient as a risk factor for CCF had not been linked. A CT scan of the head showed suggestive images of the arterial venous malformation (AVM) in the right frontoparietotemporal and left frontotemporal regions and angiography was suggested.

On angiography examination through the right internal carotid artery, RICA flow was limited to the carvesnosus segment, there was no flow to R-MCA and RACA. The early venous phase appears from the cavernous segment of the RICA to the right cavernous sinus with the draining direction to the medial superficial cerebral vein continues to Troland (superior anastomonic vein) to the superior sagittal sinus. Draining into the superior cerebral vein and inferior superficial cerebral vein was also seen. There was also drainage from the cavernous sinus to the inferior petrosal vein to the transverse sinus. The angular veins were dilated to the fascial veins. No nidus phase was seen. There was dilatation of the right transverse sinus with stenosis of the right sigmoid sinus. The appearance of slow draining veins at the time of injection of contrast from left internal carotid artery (LICA) indicated high pressure in the cerebral sinuses. On LICA angiography, good collateral appears from LACA to RACA and RMCA via anterior comunnican artery (ACom).

And collateral from right posterior cerebral artery (R PCA) to R-MCA via posterior comunicans artery (Pcom). Thus obtained the impression of an arterial vein fistula from the right internal carotid artery to the cavernous sinus with dominant venous drainage to the cortical vein. accompanied by dilatation and elongation of the veins, with collateral from the left anterior and right posterior circulation to both the R-MCA and R-ACA.

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At the next angiography, embolization using a gold balloon size 4 with the aim of closing the fistula, assisted navigation using a MABDTE magic microcatheter. Post embolization, balloons appeared to close the fistula, RICA flow seemed patent to R-MCA and R-ACA. And collateral from L ACA to R-MCA and R-ACA via Acom was no longer visible.

Flow from the arterial phase to the sinus phase as contrast injection through the LICA improves, indicating normal cerebral sinus pressure. The impression obtained was that the arterial vein fistula from the right internal carotid artery to the cortical vein accompanied by dilatation and elongation of the vein, with collateral from the left anterior and right posterior circulation both to the R-MCA and R-ACA post embolization using a gold balloon that produces R ICA R-MCA blood vessels and the patented RACA.

Other investigations carried out were EEG examination because the patient had a history of seizures, the results obtained on EEG II were bilateral and bilateral temporal centroparieto-occipital deceleration and it was suggested for an EEG of sleep deprivation after the neuro intervention was completed. Then an EEG examination of sleep deprivation was performed. The results were normal EEG, this happened because of the impact of decreased intracranial pressure due to fistula closure which had been carried out with balloon embolization techniques and the seizure assessment in this patient was acute symptomatic seizure. Seizure was one manifestation of vein congestion in drainage of ICA in CCF.^{4,5}

Thus, from the results of the history, physical examination and supporting examinations, this patient was suspected of having CCF for a long time (12 years ago) characterized by symptoms of double vision that lasted for 1 month and the sensation of ringing in the ears (pulsatile tinnitus) that had been felt until now. Symptoms during MRS, the patient previously complained of severe headache, seizures, projectile vomiting, and decreased consciousness, it was thought that CCF rupture would cause subarachnoid hemorrhage, which was amplified by positive meningeal excitatory movements. In the present patient there was no N III, IV, VI paresis and double vision suggested that at this time the ophthalmic vein was not involved in the CCF drainage pattern. CCF was thought to have a type 4 pattern in the CCF classification according to Thomas et al retrograde drainage to the cortical veins (middle superficial cerebral vein, superior anastomonic vein, superior and inferior sagittal sinus superficial cerebral vein).² This assumption was clarified by the DSA examination.

CCF generally involved ophthalmic symptoms but may not be dominant in type 4 CCF, in addition CCF is generally acute after trauma.¹ However, in this patient CCF had occurred since 12 years ago with mild symptoms, but well compensated. This was related to the different angioarchitecture of each patient which determines the onset of symptoms and signs in an arteriovenal malformation. An example of a venous dam occurs when the draining disturbance from a malformation occurs. Likewise, in fistulas that were getting wider due to sigmoid stenosis so that the flow that had been formed was blocked causing a dam in the

draining fistula which caused elongation of the vein as a space-occupying lesion. SAH in this case was suspected as PMSAH (perimencephalic SAH) like the other case where there was seepage from dilated veins that occurred in CCF.⁶ The choice of therapy in this patient was carotid artery massage while preparing for definitive action in the form of fistula closure. Carotid massage was performed in the hope that a thrombus will form in the fistula area so that the fistula can close spontaneously.¹

However, carotid massage had not been successful because the patient experienced bradycardia and hypotension during this procedure. So that the definitive action planned was closure of the fistula with balloon embolization technique, intraoperative if it failed, a second option will be carried out as a recommendation, namely carotid vessel sacrifice.⁷ Balloon embolization was chosen because it was economical, elegant and simple.^{1,7} However, the use of this material was highly dependent on the skill of the operator to place the balloon right on the fistula.

CONCLUSION

All type of direct CCF is not always with ophthalmic sign, some cases are only accompanied by headache or other high intracranial pressure symptoms depend on the type of drainage. It will be accepted that vein drainage of CCF can't influence ophthalmic vein, but only influence cortical veins.

Detachable balloon embolization can be considered as a choice for management in direct CCF cases. This method is effective, elegant and efficient, but need endovascular technic in good skill of operator.

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