Journal of Clinical & Experimental Ophthalmology

Ali et al., J Clin Exp Ophthalmol 2019, 10:5

Case Report Open Access

Spontaneous Non-Traumatic Bilateral Carotid Cavernous Fistula Type D

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Received date: November 17, 2019; **Accepted date:** December 03, 2019; **Published date:** December 10, 2019

Abstract

Aim: To help establishing a correct diagnosis of carotid cavernous fistula and appropriate management.

Carotid cavernous fistulas are usually accounted and caused by trauma. However, nontraumatic (spontaneous) carotid cavernous fistulas are uncommonly mentioned in the literature. Here, we are presenting a 59 years old gentleman known to have hypertension presented with binocular redness diagnosed as carotid cavernous fistula type D by Magnetic resonance angiogram and was successfully treated by coiling.

Keywords: Barrow classification; Carotid cavernous fistula; Corkscrew vessels; Superior ophthalmic vein; Vascular communication

Abbreviations: CCF: Carotid Cavernous Fistula; MRA: Magnetic Resonance Angiogram; DSA: Digital Subtraction Angiogram.

Introduction

Carotid Cavernous Fistula (CCF) can be classified as direct-which arise directly from internal carotid artery (or) indirect-mediated by small arteries feeding the sinus from internal or external carotid arteries.

As the cavernous sinus is a low-pressure system, arterial bood inflow leads to reversal of blood flow in the superior ophthalmic vein [1].

CCF most commonly are unilateral, but bilateral have been observed. In this report, we are presenting a rare case of bilateral spontaneous CCF diagnosed radiologically and was treated successfully through neuro-radiologic intervention.

Case Report

A 59-year old man presented with two months history of binocular redness. It was associated with right retrobulbar pain around the eye and the cheek, dizziness and tinnitus. There was no history of head or ocular trauma.

Best corrected visual acuity was 20/40 OD and 20/40 OS, Goldman applanation tonometry was 16 mm/Hg OD and 22 mm/Hg OS, and the pupils were bilaterally sluggish in reaction to light but there was relative afferent defect.

Color Vision testing using the pseudoischrmoatic plates was 15/15 OD and 1/15 OS. Extra ocular motility testing showed bilateral abduction deficit. On slit lamp examination, corkscrew dilated episcleral vessel and conjunctival injection were noticed in both eyes. Dilated fundus examination showed normal optic discs and maculas.

Topical anti-glaucoma medications (Brimonidine) were prescribed for the left eye and lubricants for both eyes. Magnetic Resonance Angiogram (MRA) of the brain and orbit revealed high flow fistula between the right external carotid artery and the cavernous sinus associated with a communication between the right ophthalmic artery and the cavernous sinus with dilatation of right superior ophthalmic vein.

MRA also established the presence of communication between meningeal branches of left internal carotid artery and cavernous sinus as well as a communication of the branches of left ascending pharyngeal artery and veins with inflow to inferior petrosal sinus Type D CCF.

Treatment was successfully done with trans-arterial embolization using platinum coils. Symptoms completely diminished after treatment.

Discussion

CCF is an abnormal vascular communication that allows blood to either directly or indirectly flow form the carotid arteries to the cavernous sinus. The fistula can be classified based on etiology; traumatic or spontaneous, velocity of the blood flow; high or low, or anatomical communication; direct or indirect.

It can also be further classified depending on the communication between the carotid artery and cavernous sinus as type A, B, C and D according to Barrow (Figure 1) [2].

Anatomically it is classified as direct that originates directly from carotid artery and indirect that originates from branches of carotid artery [3].

Most of the indirect CCFs are spontaneous and associated with increased age, pregnancy, and hypertension, diabetes and connection tissue disorders [4-6]. In our case report, our patient was 59 years old with positive history of hypertension. Traumatic CCFs compromise 75% of all CCFs, whereas spontaneous CCFs approximately 30% [3].

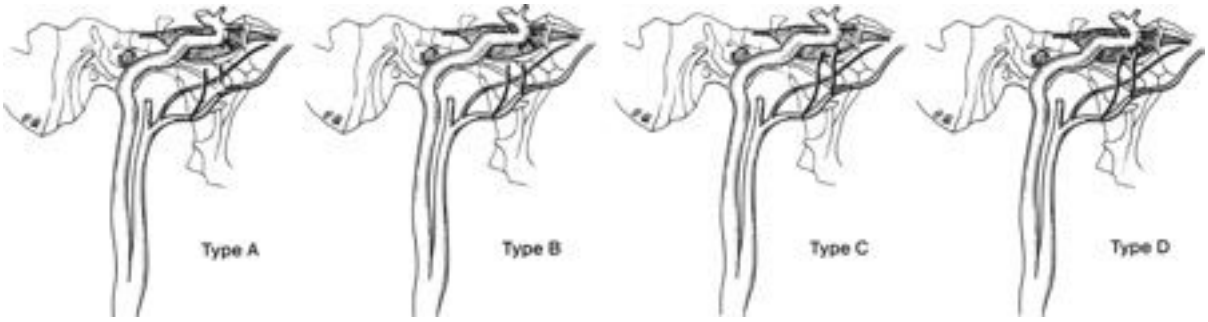

Figure 1: Barrows classification of carotid cavernous fistulas. Type A; fistulas are direct shunts between the internal carotid artery and the cavernous sinus. Type B and C are communications between meningeal branches of internal carotid artery and external carotid artery accordingly with the cavernous sinus. Type D is those between meningeal branches of the internal and external carotid artery and the cavernous sinus.

Most patients who have both direct and indirect CCFs share 3 important clinical signs, which are ocular congestion, ophthalmoplegia, and decreased visual acuity. Eighty eight percent of the patients reported having ocular pain, diplopia, eye redness and blurry vision [7]. Common clinical signs in patients with CCFs are proptosis (72%-98%), epibulbar loops (55%-100%), and orbital bruits (71%-80%) [7].

In the presence of previously mentioned signs and symptoms, the gold standard diagnostic modality for CCF is Digital Subtraction Angiogram (DSA) as it shows the exact anatomy of the pathology and helps to formulate a treatment plan [8]. However, usually no-invasive radiological investigations are carried out first such as CT and MRA to demonstrate baseline evaluation. Revealing findings include cavernous sinus enlargement, proptosis, extraocular muscle enlargement, superior ophthalmic vein dilation, or dilation of cortical or leptomeningeal vessels (Figure 2). Despite the fact that the non-invasive investigations might be negative, DSA must be conducted in cases of high clinical suspicion of CCF (Figure 3).

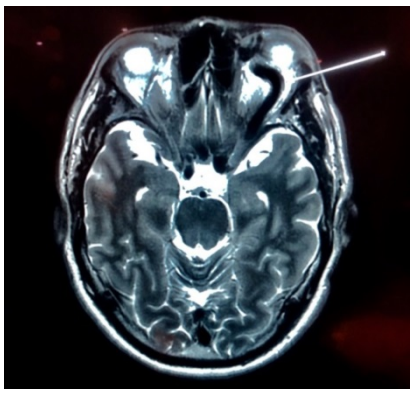

Figure 2: MRI brain and orbit axial view showing tortuous dilated Superior ophthalmic vein.

Figure 3: MRI Brain and Orbit coronal view post platinum coiling of left CCF by trans arterial catheterization after multiple attempts.

Conclusion

Treatment of CCFs might be challenging although the complications of not treating the pathology carries risk of only 3% of death, it may lead to life-long disabilities such as loss of vision and other ocular complications. The main purpose of CCFs treatment is intended to occlude the fistula and preserve the normal carotid artery flow. Currently, the best treatment modality for CCFs is endovascular approach replacing the surgical techniques.

Statement of Ethics

This report was published with the permission and informed consent of the patient.

Disclosure Statement

None of the authors has any conflicts of interest

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