Use of Color Doppler of Carotids in the Diagnosis of a Patient with Direct Carotid-Cavernous Fistula

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Abstract
An 18-year-old patient developed a direct carotid fistula after a mild craniocerebral trauma. The patient presented with tinnitus and exophthalmia, both pulsatile and on the left. A carotid Doppler study revealed high blood flow velocities and reduced resistance rates in the left internal and common carotid arteries compatible with a direct carotid fistula. Computed tomography angiography of the brain confirmed the carotid fistula. The patient was successfully referred for endovascular embolization treatment. This case demonstrates that carotid Doppler may play an important role in the diagnosis of direct carotid fistulae and the follow-up of patients undergoing endovascular therapy.

Introduction
A carotid-cavernous fistula (CCF) is an abnormal communication between arteries and veins inside the cavernous sinus\textsuperscript{1} (Figure 1). According to Barrow D (1985)\textsuperscript{5}, these fistulas are classified into types A to D: type A, a direct high flow deviation between the internal carotid artery (ICA) and the cavernous sinus; type B, a dural shunt between the meningeal branches of the ICA and the cavernous sinus; type C, a shunt between the meningeal branches of the external carotid artery (ECA) and the cavernous sinus; and type D, a shunt between the meningeal branches of the ICA, ECA, and cavernous sinus.\textsuperscript{2}

The most common cause of direct CCF is trauma, followed by less frequent causes such as rupture of an ICA aneurysm inside the cavernous sinus, Ehlers-Danlos syndrome type IV\textsuperscript{,1,4} or iatrogenic interventions\textsuperscript{5-11} including transarterial endovascular intervention, internal carotid endarterectomy, percutaneous trigeminal neuralgia treatment, transphenoidal resection of a hypophysis tumor, and maxillofacial surgery. Indirect CCF (Barrow types B, C, and D) are also called dural fistulas and usually present with low flow rates (Figure 2).

The largest arterial supply for indirect fistulae comes from branches of the internal maxillary, middle meningeal, accessory pharyngeal, and ascending pharyngeal ECA as well as from cavernous segments of the ICA.\textsuperscript{12} Indirect CCF tend to occur more frequently in postmenopausal women. The cause of these lesions remains uncertain, but pediatric cases of dural fistulae described in the literature evidence a possible congenital origin.\textsuperscript{13-15} Some factors that may predispose patients to the development of dural CCF include hypertension, diabetes, pregnancy, trauma, exertion, atherosclerosis, cavernous sinus thrombosis, sinusitis, and collagen vascular disease. Trauma is less commonly associated with indirect CCF.\textsuperscript{2,14,16}

Treatment modalities include transarterial or transvenous conservative, surgical, and endovascular management.\textsuperscript{17} Direct and indirect CCF have different mechanisms; thus, they present with different symptoms and severity levels.\textsuperscript{18}

Recent advances in endovascular technology have resulted in different CCF treatment options. As a result, the endovascular approach has become the main CCF treatment. Treatment choice varies according to type, exact anatomy, arterial defect size, and operator and institutional preferences.

In direct CCF, the aim of treatment is to occlude the rupture between the ICA and the cavernous sinus and preserve the unobstructed artery. These objectives can be achieved using transarterial obliteration of the fistula with a detachable balloon, transarterial or transvenous obliteration of the ipsilateral cavernous sinus with coils or other embolic material, or implantation of a covered stent through the fistula.\textsuperscript{1} Chi et al. studied 172 patients with direct CCF caused by trauma. They reported a medium-sized fistula predominance of 35.5% and large-sized fistula predominance of 51.7% in addition to a fistula occlusion success rate of 94% using endovascular embolization and 70% using carotid artery preservation.\textsuperscript{19}

In cases of indirect CCF, the objective is to stop fistulous communications and decrease the cavernous sinus pressure. This can be achieved by occluding the arterial branches supplying the fistula (transarterial embolization) or, more commonly, by occluding the cavernous sinus in which the fistulous communications occur (transvenous embolization).\textsuperscript{1}

Case report
An 18-year-old patient presented with the complaint of pulsatile tinnitus in the left ear. The patient mentioned that the symptoms had started after a recent car accident involving mild craniocerebral trauma. A physical examination detected discrete left eye proptosis. Carotid ultrasonography (Figures 3 and 4) showed healthy walls, high blood flow velocities, and decreased resistance index (RI) rates in the left common and internal carotid arteries. These findings were compatible with a diagnosis of a direct CCF. The velocities were normal on the right side.
Figure 1 – Direct carotid-cavernous (Barrow type A) fistula.

Figure 2 – Barrow classification of carotid-cavernous fistulae. Type A fistulae are characterized by a direct connection between the internal carotid artery and the cavernous sinus. Type B fistulae involve meningeal branches of the internal carotid artery. Type C fistulae involve external carotid branches. Type D fistulae include meningeal branches of the internal and external carotid arteries.

The patient underwent 256-channel cerebral computed tomography angiography (Figure 5), which showed the presence of CCF on the left, tumefaction of the respective cavernous sinus, and a large ipsilateral ophthalmic vein. He was successfully referred for endovascular embolization treatment.

Discussion

Carotid color Doppler is rarely reported in the literature as a diagnostic modality for CCF. Direct fistulae are characterized by a direct connection between the ICA and the cavernous sinus. Thus, direct CCF usually have a high flow that translates on a spectral Doppler study as high velocity. The reported patient showed typical direct CCF findings.

Lin et al. analyzed 2,600 consecutive carotid Doppler examinations performed from October 1987 to June 1992 and reported 14 CCF cases. Special emphasis was given to the RI and flow volume. Of the 11 patients with a direct CCF, the most common abnormal hemodynamic parameters included high systolic and diastolic peak velocities, increased flow volume, and decreased RI.

Dural CCF (Barrow types B, C, and D) often presents as recurrent arteriovenous communication after complete treatment. Tsai et al. compared carotid Doppler with cerebral angiography and reported that the ECA RI parameter (cutoff point of 0.72 on the right and of 0.71 on the left) resulted in better sensitivity (74%), specificity (89%), better positive
Figure 3 – Spectral Doppler (pulsed wave) of the left common carotid artery showing high velocities and spectrum oscillation (serrated).

Figure 4 – Spectral Doppler (pulsed wave) of the left internal carotid artery showing high velocities and spectrum oscillation (serrated).

Figure 5 – Image of 256-channel computed tomography angiography showing cavernous sinus tumefaction and a large ipsilateral ophthalmic vein.
predictive value (79%), better negative predictive value (86%), and higher accuracy (84%) for predicting dural CCF. Therefore, carotid Doppler can be used as an initial screening tool for diagnosing patients with symptoms related to dural CCF. In another study, the same authors reported that ECA carotid Doppler RI was correlated with treatment efficacy and sensitive to dural CCF clinical progression; therefore, they proposed that patients with a dural CCF should undergo carotid Doppler before and immediately after endovascular therapy to enable the evaluation of treatment efficacy and determination of the patient’s hemodynamic status before follow-up.23

Authors’ contributions
Research conception and design: Passos MD and Faria MG; data collection: Passos MD, Godoy-Gomes I, and Faria MG; data analysis and interpretation: Passos MD, Godoy-Gomes I, and Faria MG; manuscript writing: Passos MD, Godoy-Gomes I and Faria MG.

Conflict of interest
The authors have declared that they have no conflict of interest.

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