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SHORT REPORT

A New Strategy for Treatment of a Congenital Arteriovenous Fistula of the Neck. Case Report

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Congenital arteriovenous fistulas (AVF) without associated vascular malformations are uncommon. Only a very few cases of AVF have been reported in the neck. We describe our findings in a patient with AVF treated by a combined vascular and endovascular approach.

Keywords: AVF; Carotid artery.

Introduction

Congenital arteriovenous fistulas (AVF) without associated vascular malformations are uncommon. Only a very few cases of AVF have been reported in the neck.^{1–12} We describe our findings in a patient with AVF treated by a combined vascular and endovascular approach.

Case Report

A 19-year-old female complained of the sensation of abnormal pulsation in the right side of her neck over the last 5 months. She had no history of trauma. On physical examination (PE) the right jugular vein was distended with an ovoidal swelling in the submandibular region. A thrill was felt near to the swelling, beneath the ear which decreased after carotid compression. A continuous murmur with a systolic bruit reinforcement was heard at auscultation. No signs of cardiac failure were present.

Duplex demonstrated that the right external carotid artery had a 10.9 mm calibre (twice the size of the internal carotid artery) with a continuous flow that was

greater than the ipsilateral internal carotid artery flow. Furthermore, the external jugular vein, had pulsatile flow. There was also an arteriovenous 'Nidus' of 2.9 cm diameter, near the submandibular region. The magnetic resonance imaging (MRI) showed a complex vascular malformation, that spread from the submandibular region to the infratemporal fossa, supplied by the external carotid artery, that appeared enlarged, extended and tortuous, with a precocious venous drainage into the external jugular vein (Fig. 1). The left vertebral artery, the internal carotid artery and the external carotid arteries were all normal at a selective angiography. There was also a high flow arteriovenous shunt with immediate transit of the contrast medium, between the right external carotid artery and the external jugular vein.

Two days after admission the patient was treated by platinum coil embolization. We decided to isolate and clamp the external jugular vein, in order to avoid pulmonary embolism and, in our case, was helpful to stop a coil, that migrated through the shunt during the procedure. The endovascular treatment was performed by using a microsystem Prowler Plus+ Terumo 12 followed by the release of number 25 DCS Cook (Fig. 2). On angiography (Fig. 3) and MRI, the arterio-venous malformation was occluded 7 days following the procedure. Duplex demonstrated that the external carotid artery regained its physiological

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Fig. 1. Complex vascular malformation that spread from the submandibular region to the infratemporal fossa, supplied by the external carotid artery, that appeared enlarged, extended and tortuous, with a precocious venous drain into the external jugular vein.

high resistance flow. Ten months later, the patient is well and has no signs of recurrence.

Discussion

Typically, congenital AVFs go undetected for many years. Signs and symptoms include pulsatile neck mass, thrill at palpation, systolic murmur and dilated superficial veins. Open surgical treatment may include direct repair, ligating afferent and efferent arteries and veins, or excision. Ligation of the feeding arteries is ineffective or can offer only a temporary improvement because of the recruitment of distal vasculature with persistence of the fistula.¹³ Primary surgical correction by ligation and resection give good results in very



Fig. 2. Complete exclusion of the AVF with coils.



Fig. 3. Post-procedure angiographic control: complete occlusion of the arterio-venous malformation and perfusion improvement of the external carotid artery and of the brain circulation.

selected cases with single communication. Facial nerve injury has, however, been reported.^{11,12}

Endovascular therapy, the least traumatic method, has increasingly been used in the treatment of vascular lesions in the head and neck over the last 15 years. Detachable balloons have been widely employed in treating external carotid–jugular vein fistulas,^{5,6} but caution is recommended when using this technique.^{5–10} Platinum coils and glues have also proved to be effective and durable,^{13,14} but considerable experience is required to safely use adhesives. Considering the high flow of this kind of fistula, we think that embolic materials such as gel foam and ethanol are at higher risk for pulmonary embolism. Moreover, we have extensively used platinum coils in transcatheter treatment of arterio-venous malformations. A combination of a covered stent and coil embolization has also proved to be effective in traumatic pseudoaneurysms and high flow AVFs involving internal jugular vein and common carotid artery.⁸ This attractive method has never been used in congenital AVF and we lack, till now, long term results of the above procedures even in adults. If arterial catheterization fails a transvenous route⁹ can be used, but navigating the balloon inside the vein may cause problems in presence of high flow. In order to avoid pulmonary complications we think that clamping of the external jugular vein with a short incision at the base of the neck is a simple and safe method. In conclusion, we present a technique for safely treating neck AVF.

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