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Spontaneous arteriovenous fistula of the superficial temporal artery: Diagnosis and treatment

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ABSTRACT

Objectives: Despite the increasing reports of traumatic and iatrogenic arteriovenous fistulas (AVFs) of the superficial temporal artery (STA), the spontaneous origination of fistulas is extremely rare, and very little is known about their natural development. Spontaneous AVFs of the STA have the characteristic findings of an expanding, painless pulsatile mass and a palpable thrill with or without a vascular murmur.

Patients and methods: This article describes five patients with AVFs of the STA with no history of a head injury. Four of them were treated successfully either by surgical resection or by endovascular embolization. These five illustrative cases with their medium-term follow-up results are reported.

Results: In two patients, we successfully used a single-balloon alone to occlude the fistula without any complications; the patients experienced no recurrences during the clinical follow-up. In the other two patients, we carefully identified and ligated all of the involved feeding arteries and draining veins, which was followed by an excision of the lesion. At the 6-month follow-up, the patients were doing very well, with no evidence of AVF recurrences or new neurological complaints.

Conclusions: AVFs of the STA can be detected via a computed tomography angiogram (CTA) or by intra-arterial angiography alone. Intra-arterial angiography, however, remains the definitive type of investigation. AVF may be treated either by surgical ligation and excision under a local or general anesthetic or by endovascular embolization. The former modality has been the most common method of treating the lesion in the vast majority of reports. However, endovascular embolization also appears to be suitable for treating this condition.

1. Introduction

An AVF is a direct and abnormal communication between the feeding arteries and the draining veins that bypasses the capillary network [1,2]. The feeding arteries and draining veins are typically grossly dilated and tortuous, resembling a varix. AVFs of the STA usually begin as a small, subcutaneous lump, which, over time, evolves into a painless swelling, with a continuous palpable thrill, pulsation and deforming mass. A machinery-like bruit can be detected on auscultation and eliminated by the compression of the STA proximal to the mass. AVFs of the STA have previously been reported to be a result of incidental trauma or iatrogenic manipulation [3–6]. Spontaneous AVFs of the STA that are not associated with trauma are extremely rare [7–9]. A recent study showed that approximately 36 patients with AVFs of the STA have been reported in the literature, and only three cases of AVF of the STA that were not acquired traumatically were described in this subset of patients [10]. Although intra-arterial angiography has been the standard criterion suggested by many authors, the appropriate choice of treatment for an AVF of the STA continues to be debated [3,11–16]. We present five cases of spontaneous arteriovenous fistulas of the STA that were detected via CTA or digital subtraction angiography (DSA) and treated either by

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surgical resection or endovascular occlusion without complications, along with a discussion of the diagnosis and management of this unusual lesion.

2. Patients and methods

2.1. Patient histories

Between October 2011 and August 2013, five patients were diagnosed with a spontaneous AVF of the STA in our department.

The patients' information, including age at presentation, principal clinical symptoms, signs of the lesion, treatments, and clinical outcomes, were collected (see Table 1). Radiographic evaluations, including CTA and DSA, were obtained preoperatively and postoperatively when possible. In two patients, histopathological examinations confirmed the diagnosis.

2.2. Patient 1

In July 2012, a 42-year-old male presented to our department with a tender, slowly growing, pulsatile mass noticeable on the right side of the fronto-temporal scalp. This mass had gradually expanded during the previous 8 years and had become not only a cosmetic problem but also an annoyance because of a discontinuous murmuring sound in the patient's right ear. The patient denied having had even a trivial head trauma to that region during the previous few years. In the clinical examination, there was an approximately 5×2 cm tender, painless and nonmobile pulsatile mass with a palpable thrill and a continuous machinery-like buzzing sound in synchrony with the heartbeat that disappeared upon compression of the STA near the external acoustic meatus. A CTA performed at another hospital revealed an AVF of the right STA (Fig. 1a). Both selective internal and external carotid angiography through the right transfemoral approach with a 6F sheath revealed a high-flow AVF shunt fed by a right enlarged tortuous STA and drained via the large superficial temporal vein and two dilated scalp veins (Fig. 1b and c). After discussion with the patient and his relatives, the decision to proceed with endovascular embolization of the AVF was made. An 8F sheath was placed in the left femoral artery, through which an 8F guiding catheter was placed in the cervical part of the right internal carotid artery. A detachable balloon (gold valve balloon, Nycomed, Paris, France) was mounted at the tip of the balloon catheter (Balt, France). Then, the balloon was advanced to the STA through the 8F guiding catheter. Finally, the AVF was completely occluded with one detachable balloon (Fig. 1d). The immediate postembolization angiography showed that the shunt had disappeared. The patient was discharged 4 days after the endovascular treatment without any neurological deficits. He was examined in the outpatient clinic eight weeks later, at which time he was asymptomatic. At the most recent follow-up performed by a telephone interview in December 2013, there was no recurrence of the AVF and he had no neurological complaints.

2.3. Patient 2

At the age of 47 years, patient 2 was hospitalized with a pulsatile tender swelling over the right temporal area, which had gradually expanded over the previous 13 years. For approximately 2 months, the patient had noticed a continuous buzzing sound in his right ear. A 320-row three-dimensional computed tomography angiogram (3D-CTA) and superselective catheterization of the right STA stump demonstrated a right STA arteriovenous malformation (AVM) associated with an AVF (Fig. 2a–d). Endovascular embolization was recommended because of the AVM. Following the determination of the appropriate route, a 15% mixture of Glubran 2 (Gem s.r.l. Laboratories, Viareggio, Italy) was slowly injected through the microcatheter under fluoroscopy after an injection of a 5% glucose solution. However, the immediate postembolization DSA revealed the continued filling of the AVM and AVF. We decided to perform an occlusion of the right STA stump with a balloon to treat the lesion. A detachable balloon (gold valve balloon, Nycomed, Paris, France) was advanced to the STA through an 8F guiding catheter. Then, the AVM and AVF were totally occluded with single detachable balloon (Fig. 2e and f). The patient made an uncomplicated and complete recovery and was discharged 8 days after the endovascular treatment. At the most recent follow-up performed by a telephone interview in December 2013, the patient reported no recurrence of the AVF and no neurological complaints.

2.4. Patient 3

Patient 3 was a 58-year-old man who was seen in our outpatient clinic with a 25-year history of facial asymmetry due to a left-sided protuberant temporal region. There was no history of trauma. In the left temporal region, there was a 15×10 cm nonmobile painless mass with pulsatile tinnitus, bruit and a palpable thrill, and a continuous machinery-like buzzing sound in synchrony with the heartbeat. Furthermore, the thrill and bruit decreased on compression of the left STA proximal to the mass. A CTA performed at another hospital showed an AVF of the left STA associated with the AVM (Fig. 3a–d). Unfortunately, this patient was not treated at our hospital and was lost to follow-up.

2.5. Patient 4

In February 2013, a 38-year-old patient was referred to our hospital with a 7-year history of a painless, pulsatile swelling over

Table 1
Summary of a series of 5 patients with AVF of the STA.

Case no.	Sex/age (years)	Lession location	Course of disease	Clinical presentation	Treatment modality	Follow-up outcome
1	M/42	Right STA	8 years	Tender, slowly growing, pulsatile mass with discontinuous murmur	Ballon occlusion	Stable at 17 months
2	M/47	Right STA	13 years	Gradually expanded pulsatile tender swelling with continuous buzzing	Ballon occlusion	Stable at 19 months
3	M/58	Left STA	25 years	Nonmobile painless mass with pulsatile tinnitus, bruit and continuous buzzing	None	Lost to follow-up
4	M/38	Left STA	7 years	Nonmobile regular pulsatile and painless soft mass with continuous murmur	Surgical ligation and resection	full recovery at 10 months
5	M/39	Left STA	13 years	Slowly growing, nonmobile and painless mass with a continuous murmur and pulsatile tinnitus	Surgical ligation and resection	Full recovery at 6 months

Note: M indicates male; STA, superficial temporal artery.