Post-traumatic arteriovenous malformation of the superficial temporal artery

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ABSTRACT
An arteriovenous malformation is a rare vascular anomaly composed of a complex network of interconnected arteries and veins of the scalp. It is usually congenital, but infrequently occurs after trauma. Over the years, several terms have been used to describe these lesions, such as cirsoid/rasemose/arteriovenous aneurysm, plexiform angioma and aneurysma serpentinum, or arteriovenous fistula when a single connection exists. Head and neck malformations occur in 0.1% of the population. Involvement of the superficial temporal artery is rare, occurring in about 0.5% to 2.0% of cases. They are diagnosed by angiography and can be managed by endovascular or open resection. The case of a 23-year-old man who presented with a pulsatile head mass after blunt trauma 5 years prior is presented. This entity was diagnosed as an arteriovenous malformation supplied by the superficial temporal arteries. He subsequently underwent successful open exploration and resection. The information is presented with the patient’s consent. (J Vasc Surg Cases and Innovative Techniques 2020;6:50-4.)

Keywords: Trauma; Arteriovenous fistula; Superficial temporal artery; Vascular abnormalities; Cirsoid aneurysm

CASE REPORT
A 23-year-old man was referred from his local clinic with a history of a painless, pulsatile scalp mass. On history, he had an incident in 2011, where his head was hit against a window resulting in blunt injury. There was no history of loss of consciousness or amnesia. He presented to the local clinic where he was noted to have a mild hematoma. He was reassured, given analgesics, and discharged without further imaging. Four years later, he noticed a small mass on his scalp in the region of the old injury. He did not seek medical care at this time but noted that the mass gradually increased in size. He had no neurologic symptoms, but was concerned about the aesthetics of the mass. On further enquiry, he had no significant medical history or previous surgeries of note. He did however have a history of smoking and occasional ethanol ingestion.

On examination, he had an approximately 3- x 6-cm soft, pulsatile mass on the left superior-posterior aspect of the parietal region of his scalp (Fig 1). This mass was immobile, nontender, and had no superficial skin changes. It did, however, have a thrill on palpation. He was hemodynamically stable and the rest of the systemic examination was unremarkable, with no neurologic deficits.

Fig 1. External view of the “scalp mass” with an underlying arteriovenous malformation (AVM) of the superficial temporal artery.

His hematologic full blood count and coagulation studies were normal. A computed tomography angiogram of the head was done which demonstrated a superficial mass comprised of a high-flow vascular malformation located in the high parietal region of the scalp. It was noted to be supplied predominantly by vessels from the bilateral superficial temporal artery (Figs 2-5). There was no intracranial extension and no focal parenchymal
lesions were seen. The basal cisterns were patent and the sinuses were clear.

Owing to the large mass lesion, lack of endovascular facilities, and the fact that the aneurysm was located within the scalp region permitting a cosmetically acceptable result, a decision was made to take him for open exploration and resection. During the procedure, a lateral incision was made over the left temporal region to identify the feeder vessels. These were traced distally toward the vascular bed by incising the galea, after which they were cauterized, ligated, and divided to achieve proximal control. The mass was accessed through a posterior incision, after which it was excised and removed en bloc. Careful attention was made to ensure complete removal and the judicious cauterization and ligation of the surrounding vessels, to decrease the risk of recurrence. The wound was closed, a porto-vac drain was inserted, and a dressing applied.

The specimen submitted for histopathologic examination revealed a 65-mm x 35-mm x 16-mm irregular mass weighing 81.6 g macroscopically. Microscopic examination demonstrated fibro-fatty connective tissue composed of veins and arteries in close proximity to each other, surrounded by a small vessel component. The specimen had patchy chronic inflammation and surrounding fibrosis. The histopathologic features were compatible with an arteriovenous malformation (AVM).

Postoperatively, the patient remained stable and there was good hemostasis. The drain was removed and he was discharged 3 days later. He was stepped down to his referral facility for further management and follow-up. On assessment 3 years later, he had no clinical features suggestive of complications or recurrence and no clinical indications for further investigation or management. He was satisfied with the cosmetic outcome. He was counselled to return should any symptoms recur, for more definitive investigation and management at that stage. Ethics approval has been obtained by the University of Witwatersrand Ethics Committee (M190481).

**DISCUSSION**

AVMs are rare vascular anomalies with a prevalence of 18 in 100,000. Although these are generally considered to be congenital, approximately 10% to 20% involving
the scalp are associated with a history of trauma as was the case in the patient presented.\textsuperscript{1,2} Chowdhury\textsuperscript{1} presented a case series in 2013 in which a history of trauma was found in 18\% of patients. The involvement of the superficial temporal artery, being vulnerable to injury owing to its superficial and long course, occurs in 75\% of cases of scalp AVMs but 0.5\% to 2.0\% of the general population.\textsuperscript{4-6} Complications include hemorrhage, thrombosis, ulceration and aesthetic complications.\textsuperscript{1,4,5}

The mass is formed by a system of feeding arteries, which drain directly into veins via arteriovenous fistulae, without an interposed capillary network.\textsuperscript{1,4,7} AVMs develop when the pathways responsible for the stability of the vasculature is disrupted.\textsuperscript{8} Congenital and post-traumatic forms have been described in the literature, but controversy exists about whether the trauma acts as an independent cause or rather a trigger for the disintegration of preexisting fistulous embryonic connections.\textsuperscript{8} Regardless, proposed mechanisms include (1) simultaneous direct trauma resulting in communication between the walls of veins and arteries in proximity, and (2) the development of small bridging vessels in the arterial vasa vasorum as a result of proliferation of endothelial cells and neovascularization in the healing process.\textsuperscript{5-8} Vascular malformations may be classified based on their components and properties into high and low flow. With AVMs, the direct relationship between the arteries and veins with an absence of a capillary bed tends to result in a high flow lesion.\textsuperscript{9} This explains the clinical findings of a thrill and bruit noted in this patient. More definitive characteristics are demonstrated on imaging, particularly angiography, in which abnormal vessel caliber, shunting, and fistulization are clearly demonstrable.\textsuperscript{1,9} The patient described here presented with an esthetically displeasing mass. Other symptoms associated with scalp AVMs include headache, local pain, paraesthesia, tinnitus, ischemic necrosis, ulceration, and hemorrhage of the nidus.\textsuperscript{1,4}

Conventional angiography is the investigation of choice however, computed tomography or magnetic resonance angiogram are the noninvasive alternatives for outlining anatomy and excluding organ involvement.\textsuperscript{1,2,6} These can also be diagnosed less definitively by ultrasound with Doppler. Magnetic resonance imaging offers a more distinct analysis of the adjacent soft tissue structures such as muscles, nerves, tendons, bones, and solid organs.\textsuperscript{10,11}

The indications for surgery are cosmetic, symptomatic relief and prevention of haemorrhage.\textsuperscript{1,4,7,12} A study by Visser et al\textsuperscript{13} in 2011 showed an overall recurrence rate of 8.7\%, the greatest risk factor of which is incomplete excision.\textsuperscript{7} The gold standard of management has traditionally involved surgical excision of the lesion followed by reconstructive surgery as indicated.\textsuperscript{3,11} However, less invasive interventions such as endovascular coil embolization, sclerotherapy, and electrothrombosis and ligation of the feeder vessels may be preferred.\textsuperscript{1,5,8,10,11,13-16} Endovascular treatment can be used as definitive therapy or as an adjunct to open interventions to preoperatively reduce the flow and subsequent hemorrhage, thus facilitating complete removal of high flow lesions.\textsuperscript{10,11} During this procedure, the fistula is accessed via the transarterial, transvenous, and direct percutaneous routes.\textsuperscript{1,3,4,15}

**Fig 3.** Left lateral view of the computed tomography angiogram three-dimensional reconstruction demonstrating the superficial temporal artery feeder vessels forming a large arteriovenous nidus.

**Fig 4.** Superior view of complex network of the scalp cir- soid aneurysm as seen on three-dimensional computed tomography angiogram reconstruction.
Although highly effective, limitations of these endovascular techniques include recanalization or recruitment of other external carotid branches; delayed resolution of the mass systemic; thrombin effects with thrombin embolization; and ischemia with endovascular coiling. A retrospective study by Do et al evaluated ethanol embolization in 175 patients and demonstrated a 40% complete cure, 18% persistence, and 2% worsening of symptoms. This was associated with a 52% overall complication rate. In cases where there is a significant high flow mass, skin changes or complex architecture open resection may in fact have better cosmetic outcomes.

CONCLUSIONS

AVMs are vascular malformations that are typically congenital in origin, but infrequently have a temporal association with trauma. They rarely involve the superficial head and neck structures and the diagnosis is confirmed by angiography as the gold standard. There are many
different treatment options (open or minimally invasive) but cosmesis becomes an important consideration when involving head and neck structures. The choice of modality is based on the resources and surgical capabilities available as well as patient factors and preference. As seen in this case, open resection remains a viable definitive option with suitable outcomes.

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