CASE REPORT

A case of pulsatile scalp swelling in a child

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SUMMARY

Arteriovenous fistulas (AVF) are an entity most commonly seen in the head and neck region. An AVF is a direct connection between the arterial feeders and the draining veins without intervening capillary beds (unlike an arteriovenous malformation in which a nidus intervenes between the arteries and veins). We describe a case of traumatic AVF in a child aged 9 years, who presented with a pulsatile scalp swelling following blunt trauma to the head. It turned out to be an AVF between the superficial temporal artery and vein. Correct diagnosis of an AVF involves Doppler examination with digital subtraction angiography providing a good roadmap for treatment, which can be either endovascular or surgical.

BACKGROUND

Arteriovenous fistulas (AVFs) are an entity most commonly seen in the head and neck region. Traumatic AVFs in children have rarely been reported.

CASE PRESENTATION

Traumatic AVFs are a rare entity.¹ They can occur anywhere in the body, but are relatively commonly seen in the head and neck region probably due to a multitude of branching vessels coursing close to each other in this area. Here they can occur in relation to the superficial temporal artery (presenting as a pulsatile scalp swelling), vertebral artery (presenting due to cerebral arterial steal syndrome), cavernous internal carotid artery (presenting as ipsilateral pulsatile proptosis and chemosis) and the ophthalmic artery.

A girl aged 9 years presented to paediatrics outpatient department with a painless swelling over the left frontal region of the scalp. As per the history given by her father, this swelling had appeared gradually over a period of few months after an episode of blunt trauma sustained by the child about 8 months before presentation.

The swelling was 1.5 x 2 cm in size, located along the course of anterior branch of superficial temporal artery (STA) and was pulsatile in nature. The pulsations of the swelling decreased when the left STA was compressed anterior to the tragus. Few distended veins in the left frontal region extending to the right side were also noted (figure 1).

INVESTIGATIONS

The child was admitted to a hospital for a day following head trauma and CT scan of the brain was done which was normal. The child was discharged after a day of observation. Consequent to it when the swelling started appearing, an MRI of brain was

done outside which was unremarkable except for few flow voids in left frontal scalp region (figure 2) denoting high flow within them.

The child was referred to the Department of Radiology for evaluation of the swelling. Doppler of the lesion (Philips CV 550 scanner using a 9–12 MHz linear probe) revealed continuous low resistance waveform within it (figure 3). The left STA showed phasic low resistance arterial waveform suggesting a distal communication with a low resistance vascular bed. The dilated veins in the frontal region showed arterialised waveform suggesting proximal communication with arterial system. In light of the MRI and Doppler findings, a diagnosis of superficial temporal artery-superficial temporal vein (STA-STV) AVF seemed highly probable.

Since the exact location of AVF could not be revealed by Doppler examination, a selective angiography of the bilateral external carotid arteries (ECA) and internal carotid arteries (ICA) was performed on a single plane machine. It showed a single AVF being fed by a single branch from anterior division of left STA and drained through multiple veins in the bilateral frontal regions, presumably tributaries of left superficial temporal vein and right facial vein (figure 4).

TREATMENT

Since subtraction facility was not available and the fistula site was superficial and accessible, it was decided to treat the AVF by surgery. Preoperatively, the branches of the STA were marked out by colour Doppler and the site of distended veins were also demarcated (figure 5). To provide better cosmesis, a curved incision was given inside the hair bearing area of the scalp around the main trunk of the already marked out STA. The flap was elevated and the STA was identified, ligated and divided. All visible venous channels were also divided between ligatures. Smaller vessels were cauterised. After confirming the completeness of the procedure, the incision was closed in layers with an overlying pressure dressing.

OUTCOME AND FOLLOW-UP

In the postoperative period, the patient was discharged on the fourth postoperative day and followed-up in the paediatric surgery and paediatrics Out Patient Department. There was a gradual decrease in the swelling over a period of next 2 weeks (figure 6). The follow-up period has been 20 months and there has been no recurrence of the lesion.

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Figure 1 Clinical photograph at initial presentation showing the swelling in left frontal scalp region.

DISCUSSION

As far as the reason for occurrence of post-traumatic AVF of the scalp is concerned, two theories are proposed. The disruption theory suggests that endothelial cells transmigrate from vasa vasorum into the haematoma surrounding an injured vessel. This leads to formation of endothelial buds and multiple small vessels, which may eventually join an adjacent vein. There could be multiple fistulae in this case.¹ The other theory is known as the laceration theory, which says that simultaneous laceration of an artery and vein leads to formation of a single communication (fistula) between them.² In our case, the gradual appearance of the swelling suggests that the disruption and migration mechanism is more likely to have occurred.

The clinical manifestations of a STA-STV AVF are dependent on the site and size of the fistula. The swelling usually starts as a painless lump, which may progress slowly into a pulsatile, painless mass in the scalp. The symptoms may be of headache, dizziness, fatigue and pulsatile tinnitus (if the lesion is close to ear). The draining veins may become dilated due to transmission

Figure 2 Doppler trace of the lesion showing low resistance arterial waveform denoting vascular nature and arterial feeding to the lesion.

Figure 3 Axial T2-weighted MRI images showing multiple flow voids (arrows) in the subcutaneous region of left side of scalp denoting high flow vessels.

of high pressure arterial flow to them and in the past the term ‘cirsoid aneurysm’ has been used to describe them.³

Correct history and a good clinical examination form the basis of a correct diagnosis of a traumatic STA AVF. A well-performed Doppler study usually gives enough information so as to confirm the diagnosis, but delineation of the exact arterial anatomy requires CT angiography/MR angiography. Intra-arterial angiography remains the gold standard in confirming the exact site of fistula, flow dynamics (high flow vs slow flow), number of sites of fistulae (if multiple), demonstration of draining veins and excluding an intracranial component.

The indication for treating an AVF includes cosmetic reasons, prevention of bleeding, relief of headache and tinnitus. In our case, the parents were concerned about the presence of dilated veins over the scalp and forehead and hence wanted to get the child treated. The different methods used to treat an AVF include surgical excision and ligation of feeder arteries, endovascular embolisation, intralesional injection of sclerosants like polidocanol, sodium tetradecyl sulfate and absolute alcohol.⁴⁻⁶

Advent of improved hardware in the form of microcatheters and embolisation materials has made endovascular treatment a viable option for AVFs in the past decade. Effective endovascular treatment requires successful occlusion at the fistula site. However, it requires high-quality digital subtraction angiography facilities, which were not present at our institute at that time.

Figure 4 Angiogram of the left superficial temporal artery showing the superficial temporal artery (STA), superficial temporal vein (STV) and the fistula. The latter is identified as the point from which the vessel calibre changes from a smaller vessel (artery) to a larger vessel (vein).

Figure 5 The course of the artery and the fistula marked on the skin using Doppler for better intraoperative identification.

Also since the fistula site was superficial in location and accessible for surgery, it was decided to treat the lesion by surgery.

Surgical excision has been the conventional treatment for AVFs. For complete treatment, it is required that there is complete ligation of the feeding arteries otherwise there will be recurrence for channels that are mostly present between the STA and branches of ECA. It is important not to enter the lesion during surgery as it leads to torrential haemorrhage. A very good approach can be to inflate a balloon within the ECA (through endovascular route) during surgery so as to decrease the chances of intraoperative bleeding. This is an alternative to the surgical option of ligating the ECA before AVF surgery. The latter has the disadvantage of precluding any future chances of endovascular access to the nidus.

Guedes-Correa *et al*⁷ reported a case of a male child aged 9 years, diagnosed with AVF involving the frontal branch of

Figure 6 One month postoperative photograph of the child showing near complete resolution of the lesion.

the superficial temporal artery, secondary to blunt head trauma which occurred 3 years before diagnosis. Gupta and Kayal⁸ reported five cases of paediatric arteriovenous malformations of the scalp, out of which two were post-traumatic, one involving the anterior branch of the STA and the other involving the occipital artery.

We conclude that AVF of the superficial temporal artery should be considered in a case of a painless, pulsatile swelling in the scalp, appearing subsequent to minor or significant trauma.

Learning points

- ▶ Arteriovenous fistula (AVF) of the superficial temporal artery should be kept high in the differential diagnosis list of a painless, pulsatile swelling in the scalp, appearing subsequent to minor or significant trauma.
- ▶ The latent period between the trauma and appearance of the swelling is variable.
- ▶ Its diagnosis depends on a high clinical suspicion along with a good quality Doppler.
- ▶ The exact delineation of the vascular anatomy may require angiography, which can also be used for endovascular embolisation of the lesion.
- ▶ Surgical excision of a superficially located AVF also gives good result.

Contributors DK has contributed to the conception and design of the manuscript, drafting the article and revising it critically for important intellectual content and final approval of the version to be published and has agreed to be accountable for the article. PSK has contributed to the conception and design of the manuscript, drafting the article and revising it critically for important intellectual content and final approval of the version to be published and has agreed to be accountable for the article. AS has contributed to the drafting of the article and revising it critically for important intellectual content and final approval of the version to be published and has agreed to be accountable for the article. NM has contributed to the drafting of the article and revising it critically for important intellectual content and final approval of the version to be published and has agreed to be accountable for the article.

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