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Case Report

True Aneurysm of Superficial Temporal Artery Presenting as Subcutaneous Scalp Swelling: A Diagnostic Conundrum in Two Cases

Navanil Barua[1] Pragyan Sarma[2] Amit Kumar Sharma[2] Atul Agrawal[2] Sujata Chaturvedi[3] Ishita Pant[3]

[1] Department of Neuroscience, Guwahati Neurological Research Centre, Guwahati, India
[2] Department of Neurosurgery, Vardhman Mahavir Medical College and Safdarjung Hospital, New Delhi, India
[3] Department of Pathology, Institute of Human Behaviour and Allied Sciences, New Delhi, India

Address for correspondence Pragyan Sarma, MCh, Department of Neurosurgery, Vardhman Mahavir Medical College (VMMC) and Safdarjung Hospital, Delhi 110029, India (e-mail: pragyansarma@yahoo.com).

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Abstract

True aneurysms of the superficial temporal artery (STA) are very rarely encountered in clinical practice. Unlike pseudoaneurysms neither they have any preceding history of trauma nor are they associated with atherosclerosis or any other comorbid conditions. We hereby report two such cases, which according to our literature search are exceedingly rare. Two young patients presented with subcutaneous swelling of short duration in the scalp. They did not have any prior history of trauma or associated comorbidities. The only presenting symptom was headache with a pulsatile subcutaneous mass. Diagnostic workup revealed STA aneurysm in both the cases and they were managed with surgical excision of the aneurysm. Histopathology of the aneurysm wall revealed true aneurysm in both cases. True aneurysms of STA should be considered a differential diagnosis in patients with a scalp swelling along the course of the artery. Younger patients should be investigated further to look for any congenital predisposition or coexistent vascular lesion. Considering that it is a rare entity, a proper investigating protocol and surgery remains the key to their successful management.

Keywords
- aneurysm
- STA
- trauma
- true
- pseudoaneurysm

Introduction

The first case of superficial temporal artery (STA) aneurysm was reported by Bartholin way back in 1742. However, it was a pseudoaneurysm[1]. Since then, around 386 cases of STA pseudoaneurysms have been reported.[1] In more than three-fourths of these cases, there was a definite history of blunt trauma to the head; but penetrating injury, use of traction devices, hair transplantation, and previous craniotomy wound are also described in the literature as possible contributing factors.[2] The arterial course of STA makes it particularly vulnerable to trauma as it lacks bony protection and passes through mostly a subcutaneous connective tissue plane.[3] However unlike pseudoaneurysms, true aneurysms arising from the STA are extremely rare and till now only 34 such cases are reported. Most of them are reported in older age groups with an associated comorbidity like hypertension, atherosclerosis, hyperlipidemia, or a vascular event.[4] We hereby report two cases of true aneurysms of STA in patients younger than 20 years without any history of trauma or associated comorbidity, which is quite unique.

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Case Details

First Case

An 18-year-old male patient presented with complaints of a tiny swelling over the left side of the scalp along with headache for the past 14 days. There was no history of any joint pain or jaw claudication. On examination, the swelling was painless, pulsatile, and had no changes in the overlying skin. There was no history of trauma. On compression of the STA on the same side, pulsatility was eliminated. All blood investigations including erythrocyte sedimentation rate (ESR), C-reactive protein (CRP), and rheumatoid factor were normal. The patient neither had any clinical features suggestive of Marfan's syndrome, Pseudoxanthoma elasticum, or any other congenital disease nor had any features suggestive of vasculitis. Therefore, dedicated genetic tests including chromosomal studies to rule out congenital anomalies were not undertaken. Computed tomography (CT) of the head showed an extracranial swelling in the subcutaneous tissue. CT angiography (CTA) of the head revealed an STA aneurysm arising from the posterior branch of the STA, but no other vascular abnormality was detected in the intracranial circulation. Since we also considered endovascular management, dedicated CTA images of the abdominal aorta and aortic arch were also taken, which showed type 1 arch and no abnormal findings. The patient underwent surgery and the aneurysmal sac was excised with ligation of the STA. Biopsy of the sac revealed true aneurysm and there was no evidence of any vasculitis. During follow-up, CTA was repeated after a duration of 6 months, which was normal (►**Fig. 1**).

Second Case

A 16-year-old male patient presented with a subcutaneous swelling just above the right tragus for the past 2 weeks. The patient was also concerned about the cosmetic appearance. On examination, the swelling was of the size of a pea and was located along the course of the STA. The skin over the swelling was mobile and smooth. It was painless, pulsatile, and compressible in nature. There was no definite history of trauma. Diagnostic workup and its result were similar to the first case. Based on clinical findings, CTA was done, which revealed an aneurysm arising from the right STA. The patient underwent surgical excision of the aneurysmal sac. Histopathology of the wall of the sac showed a well-preserved tunica intima with hypertrophy of the muscular layer, suggesting that it was not a pseudoaneurysm. Repeat CTA at follow-up at 6 months did not show any vascular abnormality (►**Fig. 2**).

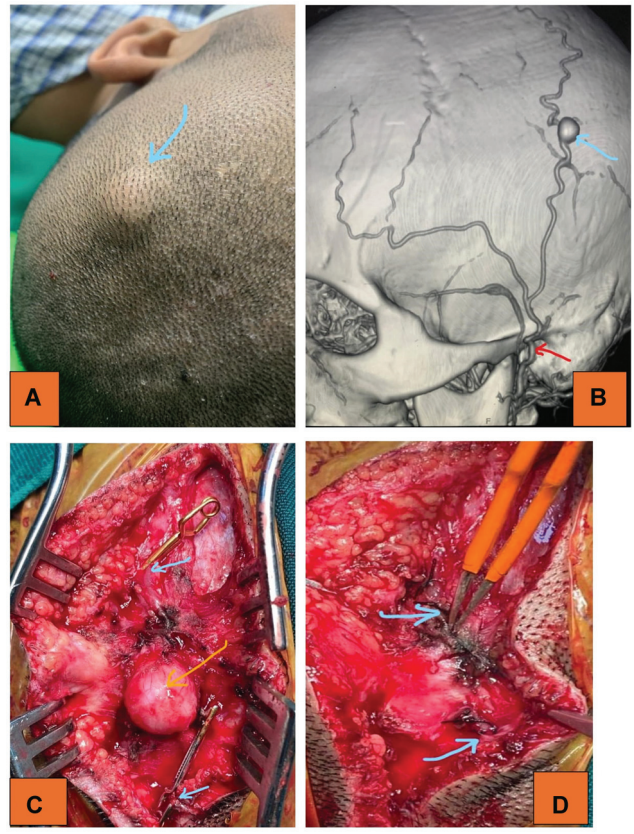

Fig. 1 (**A**) Subcutaneous swelling in the left temporal region. (**B**) Computed tomography (CT) angiography showing an aneurysm arising from the posterior branch of the superficial temporal artery (STA). (**C**) *Arrows* showing the aneurysm sac with both proximal and distal control. (**D**) Aneurysm is excised along with ligation of both proximal and distal segment of the STA.

Discussion

We could not find any large series of true STA aneurysms in published literature. Most of them are case reports of a single or few cases of pseudoaneurysms. Despite lack of any universally accepted classification, STA aneurysms can be broadly categorized into traumatic, iatrogenic, or spontaneous.[5] The most important feature in pseudoaneurysms is the disruption of the arterial wall leading to extravasation of blood. The accumulated blood ultimately forms a hematoma and the subsequent pseudo-capsule. However, this disruption of the arterial wall is not seen in true aneurysms. In the majority of the cases of true aneurysms, there is weakening of the vessel wall leading to its gradual dilatation. It can be clinically appreciated as a palpable pulsation or bruit.[6] However pulsatility of the mass is not always seen, more so when there is complete thrombosis inside the aneurysm cavity.[7,8] In such a scenario, they can be a cause of diagnostic conundrum as they can be confused with a subcutaneous lipoma, sebaceous cyst, hematoma, or abscess. If aspiration biopsy is mistakenly done in such cases, it can lead to profuse bleeding.[8] Although this pathology can occur in all age groups, out of 34 cases of true STA aneurysms reported till now, only 6 cases (17.64%) including our cases are seen in patients below 20 years of age. Although the exact etiological factors of true STA aneurysms are not known, considering the incidence rate of only 1.3% of other aneurysms in the same age group,[9] we can assume that hereditary factors definitely have a role to play. Kawabori et al reported that in younger population, congenital predisposition of the arterial wall may be an important factor in the genesis of true STA aneurysms rather than atherosclerosis in the elderly counterparts. They also suggested that the portion of STA where true aneurysms are commonly seen is not prone to trauma unlike false aneurysms as it runs inside a line from the auricle to the zygomatic arch.[10] These findings correlate with our cases too. The most common presenting symptom is a painless subcutaneous swelling of varied duration though it may not always be pulsatile.

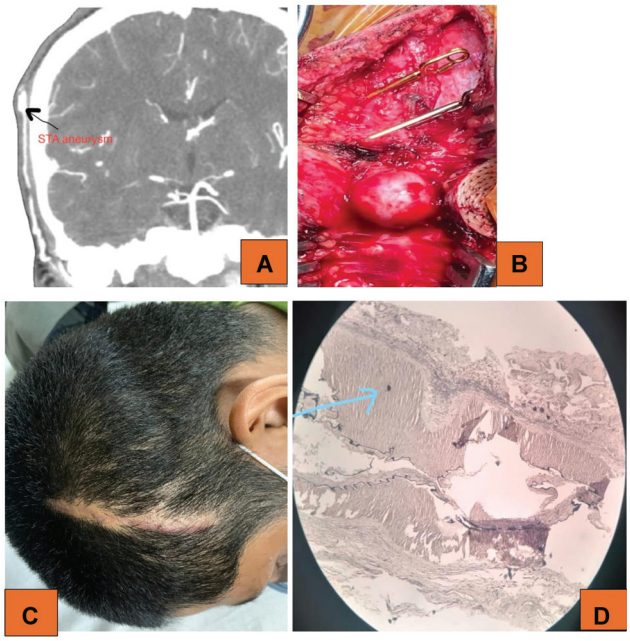

Fig. 2 (**A**) Computed tomography angiography (CTA) showing a right superficial temporal artery (STA) aneurysm. (**B**) Intra-op photograph with the aneurysm sac totally dissected out prior to excision. (**C**) Postoperative scar with the incision mark. (**D**) Elastic Van Gieson 20X: section showing intact tunica intima with hypertrophy in the muscular layer.

Headache, earache, cosmetic concern, or in rare cases cranial nerve deficits and vascular compromise are also described in the literature.[7–10] In pulsatile swellings, compression of the proximal STA causing disappearance of the pulsation in the aneurysm can be an important diagnostic clue. Apart from clinical examination, noninvasive imaging modalities like duplex ultrasound or CTA can confirm the diagnosis. Kawai et al reported four cases of spontaneous STA aneurysms, which also had associated intracranial aneurysms and concluded that although these associations seem coincidental, the existence of common predisposing factors involving both intra- and extracranial aneurysms cannot be ruled out.[4] Keeping this in mind, CTA can be utilized for confirmation of the diagnosis as well as to rule out other coexisting lesions if present. Also, it can define the patency of the vessel, calculate the size of the aneurysm, detect thrombosis inside the aneurysmal sac, and show luminal opacification. Diagnostic angiography can be utilized in difficult cases and for planning endovascular treatment whenever contemplated. However in our opinion, although endovascular treatment is not very challenging in these cases, it leaves a cosmetic deformity with a lobular mass with coils, which may not be acceptable for some patients. Further in a third world country like ours, taking into consideration that surgery provides a simple cure and involvement of high cost in endovascular treatment, it may not be a feasible option for a vast majority of the population. The treatment of choice in true aneurysms of the STA is surgical excision of the sac along with ligation of the proximal and distal ends of the vessel. If the aneurysm is in the proximal part of the STA, then dissection of the parotid gland and facial nerve may be required. The main goals of treatment are reduction of the risk of hemorrhage, relief of pain, and taking care of the cosmetic defect.[11] Although endovascular treatments are employed in the treatment of STA pseudoaneurysms,[12,13] the authors could not come across any similar literature for the management of true STA aneurysms.

Conclusion

Patients presenting with a pulsatile subcutaneous lesion or even a nonpulsatile lesion along the course of the STA should be evaluated properly. In these cases, true STA aneurysm should be considered a differential diagnosis. We hereby report two such cases, both seen in patients younger than 20 years without any history of existing comorbid conditions, which is exceedingly rare. Congenital predisposing factors in these cases and the presence of any coexistent vascular pathology should be evaluated further. Simple noninvasive imaging studies can establish the diagnosis. Surgery remains the treatment of choice and should be offered in all such cases.

Conflict of Interest

None declared.

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