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Case Report

Recurrence of partially thrombosed superficial temporal artery aneurysm after endovascular trapping: A case report and literature reviews

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ABSTRACT

Background: Superficial temporal artery (STA) aneurysms are relatively rare diseases, and the treatment approach is based on factors such as the location of the aneurysm, curability, complications, and patient request. However, the detailed outcome of STA aneurysms treated with endovascular surgery remains unknown.

Case Description: A 75-year-old woman found a pulsatile mass lesion at a preauricular region with gradual enlargement. Angiography showed a thrombosed aneurysm originating from STA. Endovascular trapping was selected because she rejected open surgery for cosmetic reasons. Intra-aneurysmal blood flow disappeared on postoperative angiography. However, the aneurysm had enlarged for 8 months after the initial embolization. A recurrent aneurysm was resected, and pathological examination revealed neovascularization within the thrombotic lesion, suggesting neovascularization and re-canalization.

Conclusion: Endovascular trapping for thrombosed STA aneurysm might include recurrent risk, and direct resection should be considered as the first-line treatment.

Keywords: Angiogenesis, Channel formation, Superficial temporal artery aneurysms, Thrombosed aneurysm, Vasa vasorum

INTRODUCTION

The superficial temporal artery STA aneurysm is a relatively rare disease, with approximately 400 cases reported since 1740 and about 150 cases reported in Japan.[24,26] Various factors involve aneurysm formation, such as arteriosclerosis or connective tissue disorders and traumatic injury.[23,24,26] Aneurysms originating from STA are classified into true aneurysms, where the vessel wall structure is maintained circumferentially, and false aneurysms, where the vessel wall structure is not maintained due to trauma or other causes.[11,19] Direct resection, ligation of STA, endovascular embolization, thrombin injection embolization, and long-term compression

promote thrombosis.[5,16,19,20,23,24,26] The detailed outcome of STA aneurysms, especially thrombosed STA aneurysms treated with endovascular surgery, remains unknown. We experienced a rare case of a recurrent STA aneurysm after endovascular trapping with coil embolization and reported the clinical course and pathological findings with previous literature reviews.

CASE PRESENTATION

A 75-year-old woman noticed a pulsatile preauricular mass, which had gradually enlarged without tenderness for 5 years. She had no history of trauma, vasculitis, or other relevant diseases and consulted a dermatology clinic. Ultrasound examination revealed blood flow signals within the subcutaneous pulsatile mass, leading to a referral to our department. Three-dimensional computed tomography angiography revealed a 25 mm × 20 mm aneurysm of the STA in front of the right ear and above the zygomatic arch, associated with intra-aneurysmal heterogenous contrast enhancement on the dorsal side of the aneurysm, suggesting thrombotic formation [Figures 1a and b]. Magnetic resonance images (MRI) showed blood flow signals in the anterior part of the aneurysm and the heterogenous contrast enhancement in the posterior thrombotic lesion [Figures 1c and d].

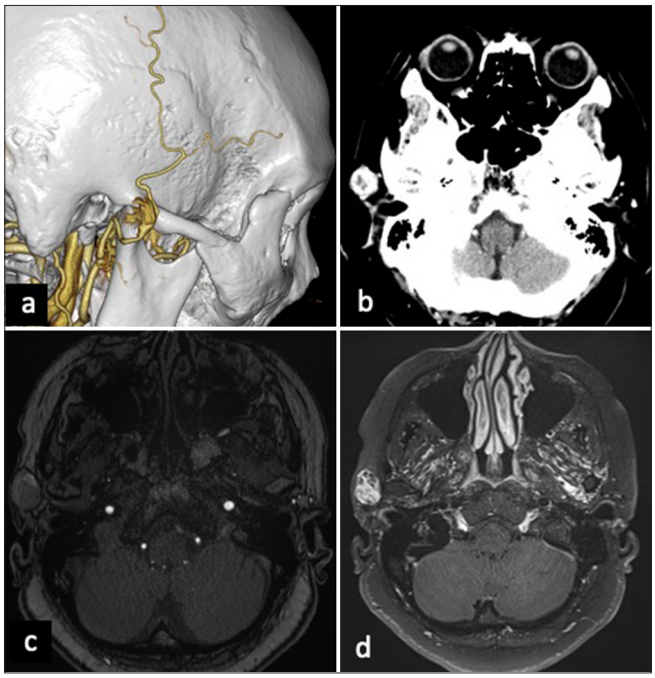

Figure 1: (a) Three-dimensional computed tomography angiography revealed an aneurysm of the right main trunk superficial temporal artery and (b) the dorsal side of the aneurysm showed thrombosis with heterogenous enhancement. (c) Magnetic resonance images showed a signal of a superficial temporal artery on the ventral side of the aneurysm and (d) the thrombosed area showed heterogenous enhancement same as computed tomography.

The total resection of the STA enlarged aneurysm was recommended, considering the dominance of the curability. However, the patient worried about the risks of cosmetic deformity, facial nerve injury, and parotid gland injury caused by direct surgery and strongly wished for the disappearance of the mass lesion with endovascular treatment. We planned endovascular trapping with coil embolization at the proximal and distal side of the STA aneurysm. Thorough coil embolization inside of the aneurysm was not planned to avoid the expansion of the coil mass.

Operative findings of the first endovascular surgery

Under local anesthesia, a 6Fr Roadmaster (Goodman, Aichi, Japan) and TACTICS (Technocrat Corporation, Aichi, Japan) were introduced into the right external carotid artery for diagnostic imaging. Subsequently, an Excelsior XT-17 (Stryker Neurovascular, Fremont, CA, USA) and TENROU (Kaneka Medical Products, Osaka, Japan) were introduced to the distal part of the STA from the aneurysm. A total of 15 Guglielmi Detachable Coils (GDC) were used for embolization: five coils in the distal part of STA from the aneurysm, seven coils inside of the aneurysm, and three coils in the proximal part of STA from the aneurysm. Post-embolization imaging showed the disappearance of the intra-aneurysmal blood flow from the proximal STA [Figure 2].

Postoperative course of the first endovascular surgery

There was an uneventful postoperative course. The pulsation of the mass disappeared, and the patient was discharged 7 days after the surgery. No new clinical symptoms occurred, and the size of the mass lesion had not changed. Follow-up MRI at 1 and 4 months after surgery showed no blood flow signal at the embolization site [Figures 3a and b]. However, the aneurysmal wall and intra-aneurysmal thrombosis were markedly enhanced, and the size of the aneurysm

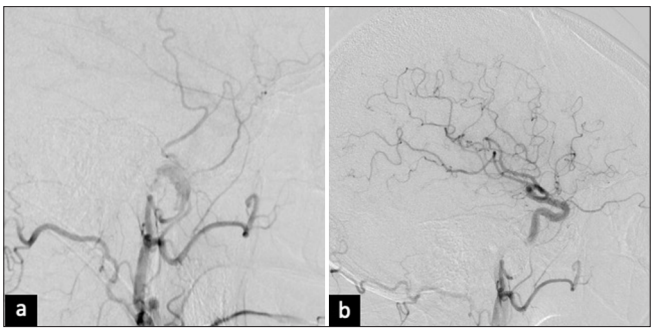

Figure 2: (a) An angiograph of the right external carotid artery showing a thrombosed aneurysm of the main trunk superficial temporal artery. (b) Post-embolization imaging showed the absence of blood flow from the proximal superficial temporal artery.

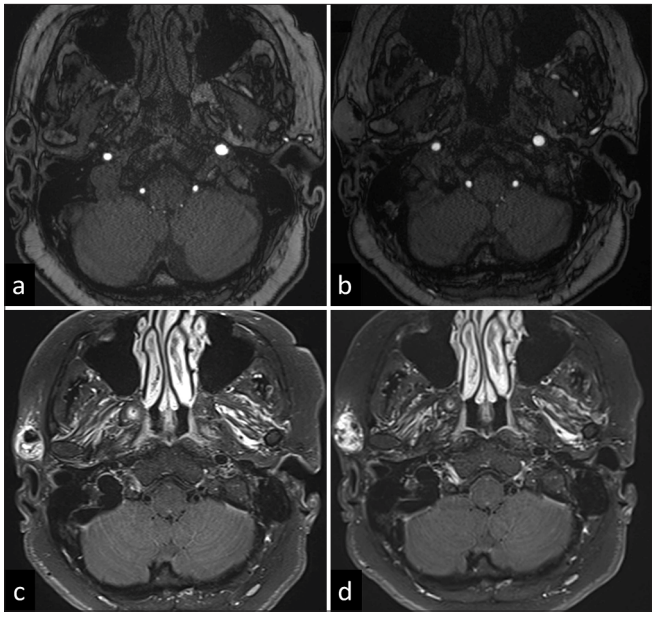

Figure 3: (a and c) Follow-up magnetic resonance images at 1 and (b and d) 4 months after surgery showed (a and b) no blood flow signal at the embolization site, but (c and d) enhanced thrombosed area was increased. The size of the aneurysm increased from (c) 15.25 mm to (d) 16.11 mm.

gradually enlarged [Figure 3c and d]. The total resection was recommended for the recurrent thrombosed aneurysm. The patient agreed to our treatment plan, and STA ligation and aneurysm resection were performed 8 months after the initial surgery.

Operative findings of the second direct surgery

Under general anesthesia, an approximately 8 cm skin incision was made over the right preauricular aneurysm [Figure 4]. The proximal and distal ends of the STA were identified, and temporary clips were applied before ligating the artery. The STA was coagulated and exposed, and the aneurysm was carefully dissected from the surrounding connective tissue and resected. The aneurysm had marked adhesion with surrounding tissues and a bleeding tendency during dissecting. The resected aneurysm showed coils filling both the proximal and distal parts of the STA [Figure 5].

Postoperative course of the second direct surgery

Pathological examinations showed that the vessel wall structure, including elastic fibers and smooth muscle layers, was preserved circumferentially, confirming a diagnosis of true aneurysm. Within the thrombus, an expanded lumen was observed, and the inner surface was lined with CD34-positive endothelial cells, indicating

Figure 4: An approximately 8 cm skin incision was made at the site corresponding to the aneurysm in front of the right ear (black circle).

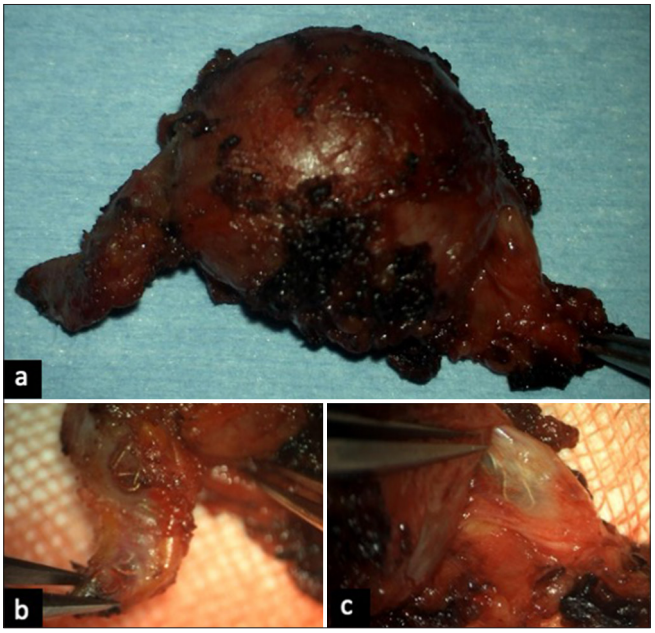

Figure 5: (a) Resected superficial temporal artery aneurysm and (b) its distal vessel and (c) proximal vessel were filled with coils.

neovascularization. Fibroblasts and CD68-positive macrophage infiltration were also noted, suggesting the organization of the thrombus [Figure 6]. Postoperative MRI showed complete removal of the aneurysm. The patient had an uneventful recovery, was discharged, and has shown no recurrence.

DISCUSSION

True STA aneurysms account for only 5–18.8% of all cases, making them relatively rare. Congenital factors, atherosclerosis, connective tissue disorders, segmental medio-

lysis arteriopathy, and other diseases caused them.[2,4,22,23,26] According to Itani *et al*.,[8] the most common site for STA aneurysms is the main trunk (48.2%), followed by the frontal branch (33.9%). Treatment for STA aneurysms is aimed at cosmetic improvement, reducing pulsation and pain, and preventing rupture or bleeding.[3,19,26] Reported treatments include STA ligation and aneurysm resection, endovascular embolization, thrombin injection embolization,[20] and long-term compression to induce thrombosis.[14] In general, STA ligation and aneurysm resection are preferred due to their high curative potential,[8,10] although endovascular embolization may be chosen depending on the aneurysm location.[11]

To date, six cases of STA aneurysm treated with coil embolization are shown [Table 1],[3,6,8,12,23] one of which experienced recurrence.[3] Coenegrachts *et al*. reported the recurrent case of a giant STA aneurysm, and the regrowth was detected 4 months after the endovascular occlusion with particles and coil embolization.[3] According to reports by Hong *et al*. and Komiyama *et al*., four cases without recurrence involved coil embolization of both the aneurysm and the proximal and distal segments of the STA.[6,12] One of these cases, reported by Saguchi *et al*., involved performing coil embolization under ultrasound guidance, confirming the absence of blood flow.[23] These reports suggest that endovascular trapping by coil embolization of the inside of the aneurysm and both the proximal and distal parts of the main stem of the STA was an effective and safe treatment method for non-thrombotic STA aneurysm.

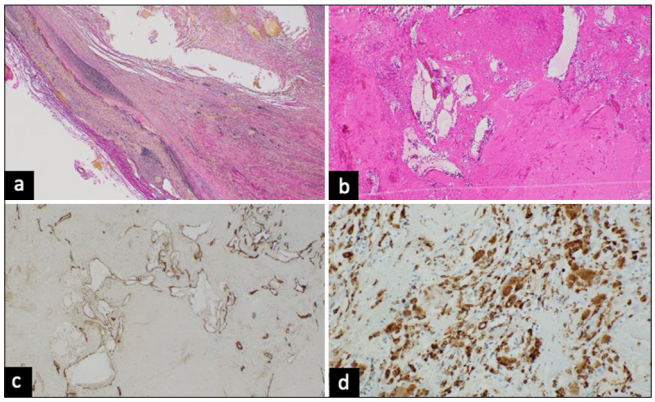

Figure 6: (a: Elastica van Gieson stain, magnification ×40) In the histological findings of a superficial temporal artery aneurysm, the vessel wall structures such as Elastic fibers and smooth muscle layer were preserved circumferentially. (b: hematoxylin and eosin stain, ×100, c: CD34 immunostain, ×100, d: CD68 immunostain, ×100) In the thrombus, there were many dilated small vessels with endothelial cells and macrophage infiltrates.

Regarding thrombosed aneurysms, recurrence mechanisms have been reported in intracranial aneurysms.[9,17,18,25] Mechanisms of recurrence of thrombotic aneurysm include increase of intra-aneurysmal blood flow due to recanalization, neovascularization through vasa vasorum, wall hemorrhage from vasa vasorum and inflammatory reaction through aneurysmal wall.[1,9,13,17,18,25] After aneurysm coil embolization, a channel may form from the parent artery into the thrombus, and the development of new blood vessels can lead to blood flow into the thrombus, causing it to expand and resulting in recurrence.[17] This mechanism is also seen when the parent artery is embolized, where neovascularization within the thrombus leads to enlargement and recurrence of the aneurysm.[17] This may be part of a response involving macrophages removing the thrombus, as seen in the present case, where numerous CD68-positive macrophages were observed within the thrombus. The formation of endothelial progenitor cells, which differentiate into endothelial cells, is involved in CD34-positive cells. These progenitor cells play a key role in vascular repair and vascularization. The presence of CD34-positive endothelial cells, an undifferentiated cell associated with neovascularization, within the thrombus after coil embolization suggests that angiogenesis, which contributes to thrombus enlargement, has occurred.[21] In addition, extra-cranial arteries have vasa vasorum, which supplies nutrients and oxygen to the vessel walls,[25] and vasa vasorum involvement has been reported in the recurrence of thrombosed aneurysms.[18] When the aneurysm wall in contact with the thrombus undergoes ischemia due to

Table 1: Summary of superficial temporal artery aneurysm treated with endovascular surgery.

Author (year)	Age	Sex	Size of aneurysm (mm)	Location of aneurysm	Thrombus	Position of embolization	Follow-up periods	Recurrence
Komiyama *et al*. (1997)[12]	21	M	15	Main trunk	–	Proximal and distal	10 months	–
Komiyama *et al*. (1997)[12]	78	M	30	Frontal branch	–	Proximal and distal	4 months	–
Coenegrachts *et al*. (2005)[3]	59	M	27	Main trunk	–	Proximal only	4 months	+
Hong *et al*. (2006)[6]	25	M	15	Frontal branch	–	Proximal and distal	/	–
Itani *et al*. (2022)[8]	/	/	/	/	/	/	/	/
Saguchi *et al*. (2016)[23]	69	M	/	Main trunk	–	Proximal and distal	5 years	–
Present case	75	W	25	Main trunk	+	Proximal and distal	8 months	+

M: Man, W: Woman, /: Details unknown, Position of embolization: Location of superficial temporal artery from aneurysm

embolization, neovascularization from the vasa vasorum occurs into the aneurysm, leading to increased blood flow and potential aneurysm growth.[7,9]

In our present case, there were characteristic points as follows: first, an aneurysm occurred in the main trunk. Second, endovascular trapping was selected to reduce the mass volume through a progression of thrombosis, but the effectiveness was not enough to cure it. Total resection was required. Third, enlargement of intra-aneurysmal thrombosis occurred without revival of blood flow from apparent recanalization of the embolized STA. Fourth, pathological findings showed neovascularization of the intra-aneurysmal thrombus and inflammatory reaction of the aneurysmal wall. Although the marked development of vasa vasorum could not be detected histologically, micro-circulation from vasa vasorum might contribute to neovascularization inside the thrombus. Hypoxic conditions in the thrombus after trapping of STA might also cause neovascularization and thrombotic progression. Furthermore, Murakami's report suggests that recurrent bleeding within a thrombus may contribute to aneurysm enlargement.[15]

Treatment selection for preauricular STA aneurysms includes direct surgery of ligation and aneurysm resection, endovascular trapping, or intra-aneurysmal embolization. Endovascular treatment is minimally invasive and has cosmetic dominancy, but it might be inferior to direct surgery in curability for thrombotic STA aneurysm. Further clinical experiences and careful treatment selection are essential.

CONCLUSION

Endovascular trapping for thrombotic STA aneurysm has a potential risk for regrowth of intra-aneurysmal thrombosis through neovascularization and inflammatory reactions from both intra- and extravascular sources. Direct surgery with the aneurysm resection might be the first choice for curative treatment.

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