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Three Cases of Spontaneous Superficial Temporal Artery Aneurysm with Literature Review

Hiroshi KAWAI,[1] Tadashi HAMASAKI,[2] Junnichi IMAMURA,[3] Norio TOMONORI,[4] Takuya ODASHIRO,[5] Hitoshi YAMAHATA,[1] Hiroshi TOKIMURA,[1] Mitsuharu NOMOTO,[6] and Kazunori ARITA[1]

[1]*Department of Neurosurgery, Graduate School of Medical and Dental Sciences, Kagoshima University, Kagoshima, Kagoshima;*
[2]*Department of Neurosurgery, Kumamoto University, Kumamoto, Kumamoto;*
[3]*Department of Neurosurgery, National Hospital Organization Kagoshima Medical Center, Kagoshima, Kagoshima;*
[4]*Department of Surgery, Kokubu Seikyo Hospital, Kirishima, Kagoshima;*
[5]*Department of Surgery, Odashiro Commemoration Clinic, Minami Kyushu, Kagoshima;*
[6]*Department of Pathology, National Hospital Organization Kagoshima Medical Center, Kagoshima, Kagoshima*

Abstract

Spontaneous, nontraumatic, superficial temporal artery (STA) aneurysms have been rarely reported. We herewith report three cases of spontaneous and true STA aneurysms. All patients, a 65-year-old male, a 76-year-old female, and a 47-year-old female, had no history of head trauma that requires medical attention. Painless, pulsatile, and slowly growing calvarial lump was the symptom leading to image studies. All the lumps were preoperatively diagnosed as STA aneurysms by magnetic resonance angiography and/or three-dimensional computed tomographic angiography. One case was accompanied by anterior communicating aneurysm. And another case was associated with two more scalp aneurysms arising from occipital artery and contralateral STA. Pathologic studies showed that all three were true aneurysms, with intact media and adventitia but without organized hematoma. Literature review showed that 8% of all STA aneurysms comprised spontaneous STA aneurysms. We found 32 cases (19 males and 13 females) of well-described spontaneous STA aneurysms including ours. Twenty-eight cases (87.5%) were true aneurysms. Seven cases (21.9%) had coexisting vascular lesions. Five (15.6%) of these seven cases were diagnosed with cerebral or abdominal aneurysm. Multiple scalp aneurysms are quite rare; only two cases including ours have been reported. It seems important to know that spontaneous STA aneurysms may coexist with other vascular lesions including intracranial aneurysm.

Key words: superficial temporal artery, true aneurysm, intracranial aneurysm

Introduction

The first case of aneurysm of superficial temporal artery (STA) was reported in 1742 by Bartholin.[1)] Most of the STA aneurysms reported since then were post-traumatic pseudo-aneurysms because the STA passes through shallow and areolar coarse hypodermic connective tissue, so it is easily damaged by head injury.[2)] Spontaneous nontraumatic aneurysm of STA, which is extremely rare, constitutes 8% of all STA aneurysms.[3,4)] We encountered three patients having spontaneous and true STA aneurysm with intracranial aneurysm accompanying in one case. We present these three cases and the literature review for spontaneous STA aneurysms.

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Case Report

I. Patient 1

A 65-year-old Japanese male with hyperlipidemia under medical treatment consulted a clinic because of a painless, pulsatile mass in the right temporal region (Fig. 1A). He had first noticed this mass 10 years before, which had

gradually increased in size. He had no history of head or face trauma necessitating medical attention. It was diagnosed as aneurysm with ultrasonography. Magnetic resonance angiography (MRA) showed the aneurysm arising in right STA and revealed anterior communicating artery aneurysm also. Digital subtraction angiography showed that the STA aneurysm is 10 mm in diameter (Fig. 1B, C) and anterior communicating artery aneurysm is 6.5 mm × 9.7 mm in size (Fig. 1D, E). The right STA aneurysm was resected first (Fig. 2A) and the anterior communicating artery aneurysm was then clipped through right pterional approach. Photomicrographs of the resected STA aneurysm showed thinned but well-preserved intima, media, and adventitia (Fig. 2B, C). Areas of thickened intima and degradation of internal elastic lamina were observed (Fig.2D). There was no thrombus in aneurysm.

II. Patient 2

A 76-year-old female with hyperlipidemia, hypertension, abdominal aneurysm, and cerebral infarction under medical treatment but without a history of head injury consulted a physician because of a painless, pulsatile mass in the left temporal area (Fig. 3A). Ultrasonography and MRA revealed a left STA aneurysm. Three-dimensional computed tomographic angiography (3D-CTA) showed a STA aneurysm of 17 mm in diameter (Fig. 3B). The STA aneurysm was resected under local anesthesia. The aneurysm had no bleb and its surface was smooth (Fig. 4A, B). Hematoxylin and eosin stain of resected aneurysm revealed hyperplasia of intima and areas of indistinct internal elastic lamina, and multifocal mucoid degeneration in the media (Fig. 4C, D).

III. Patient 3

A 57-year-old female with aldosteronism and hypertension under medication consulted a neurosurgical clinic because of a pulsatile lump in her right temporal region which had grown gradually for the last three years. Ultrasonography and 3D-CTA revealed multiple aneurysms on branches of external carotid arteries, on bilateral STAs,

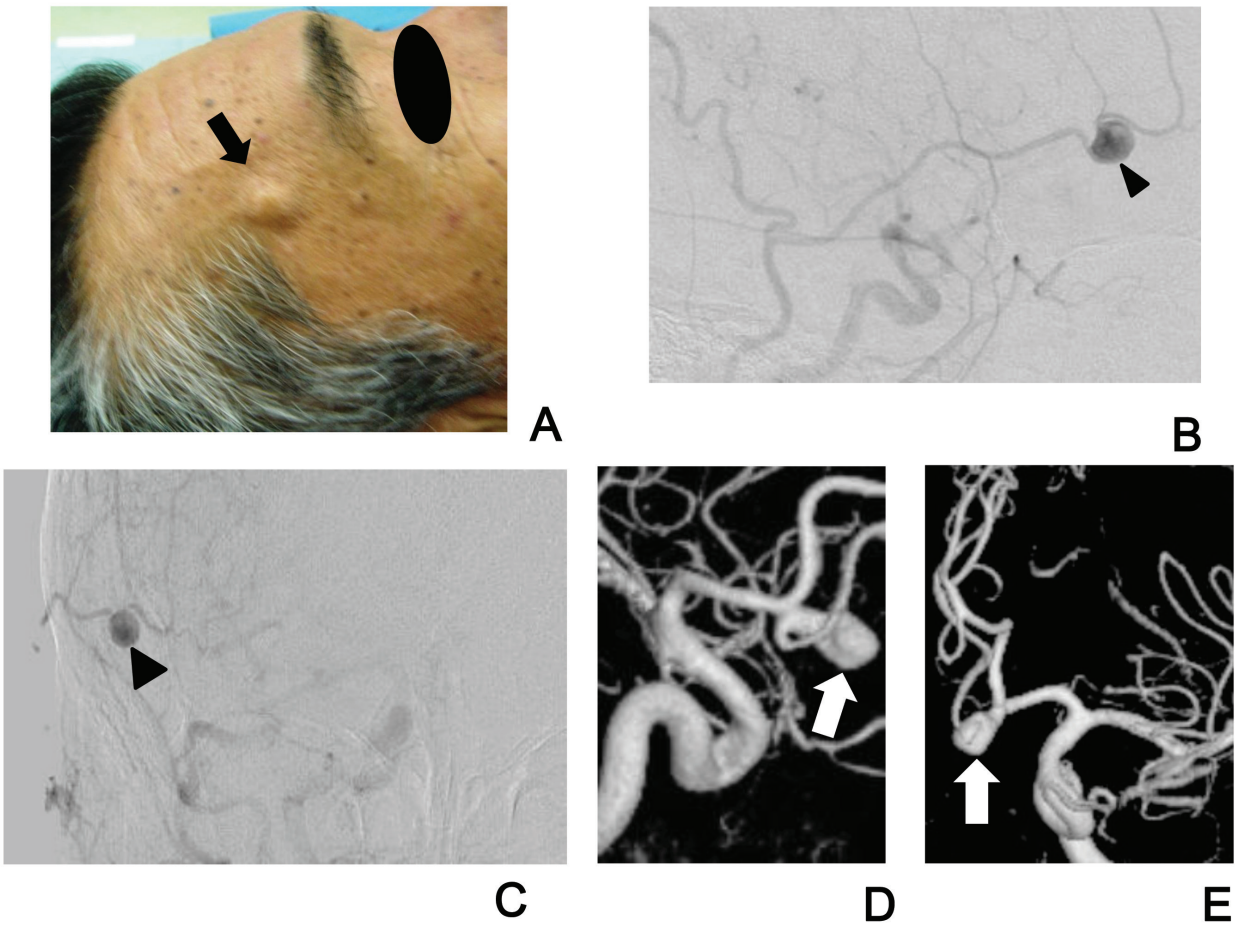

Fig. 1 A: Photograph of Patient 1 showing a lump in the right temporal region (*arrow*). B, C: Digital subtraction angiography showing a superficial temporal artery aneurysm (*arrowhead*). D, E: Three-dimensional digital subtraction angiography showing left carotid artery and anterior communicating artery aneurysm (*white arrow*). Lateral (D) and anterior–posterior (E) views.

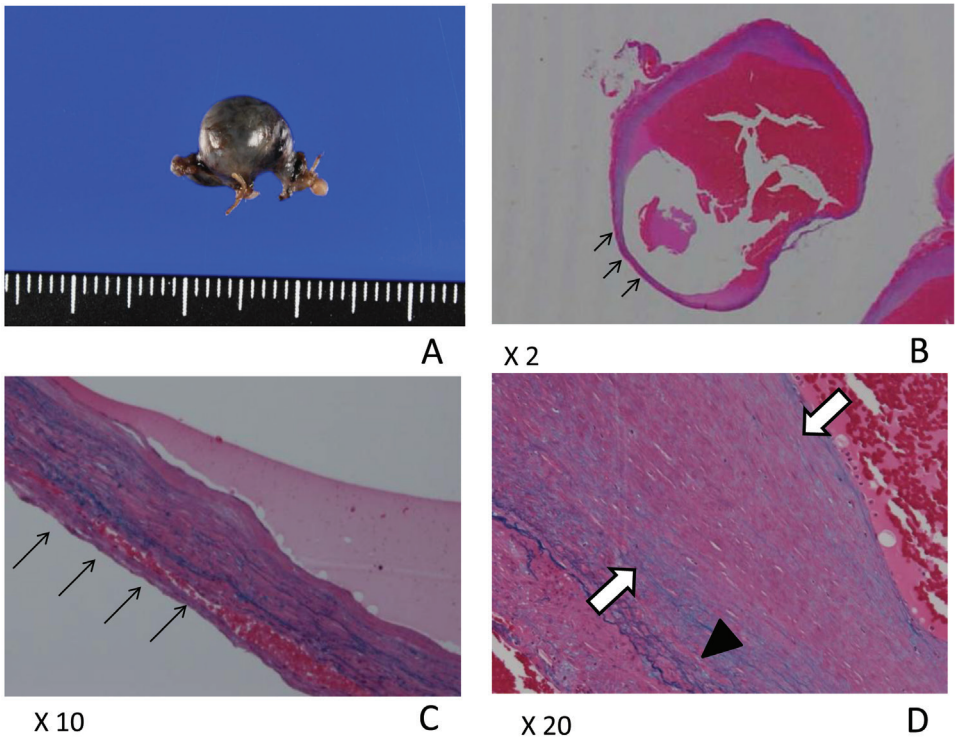

Fig. 2 A: Photograph of the resected aneurysm of Patient 1. B–D: Microscopic findings (hematoxylin and eosin stain). Photomicrographs of resected superficial temporal artery aneurysm showing thinned but well-preserved intima, media, and adventitia with thinner parts in them (B, C: *thin arrows*). Areas of thickening of the intima (D: *between white arrows*) and degradation of the internal elastic lamina (D: *arrowhead*) were observed. There was no organized thrombus in aneurysm.

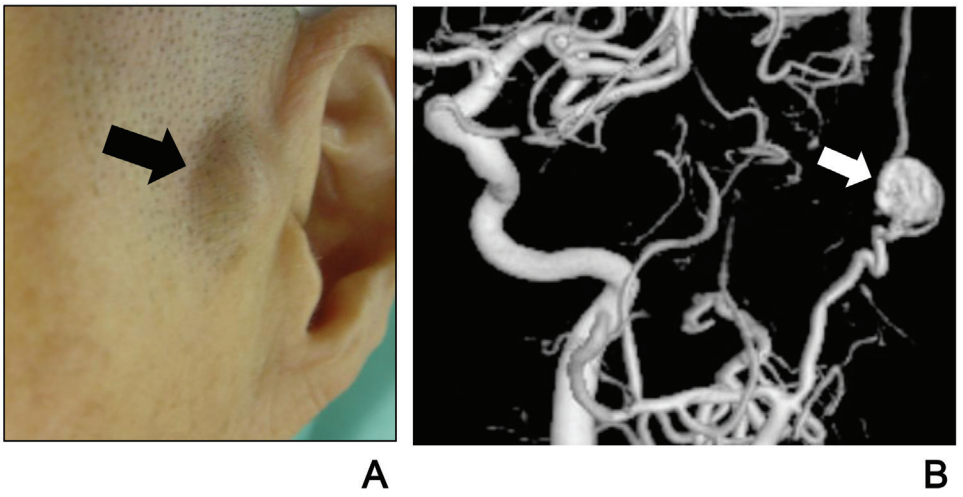

Fig. 3 A: Photograph of Patient 2 showing a lump of the left preauricular region (*arrow*). B: Left carotid angiogram (Lt. CAG) showing a superficial temporal artery aneurysm (*white arrow*).

and on right occipital arteries (Fig. 5A). Due to cosmetic reason, the right STA aneurysm was resected under local anesthesia. Resected aneurysm was 6 mm in diameter. Pathologically, intimal thickening with normally preserved media and adventitia was seen (Fig. 5B).

Literature Review

We found 32 patients, 19 males and 13 females, including ours with spontaneous STA aneurysm in the literature.[1,2,5–23] Patients' ages ranged from 13 years to 84 years with a mean age of 54.3 years. Patients generally visited the physician because of a pulsatile mass in the temporal or the parietal region. The pain surrounding the aneurysm was seen in seven cases (21.9%). The lag time between diagnosis and first awareness of symptoms widely ranged from 0 to 120 months with a mean of 21.7 ± 25.8 (standard deviation) months. The pathology included 28 true aneurysms (87.5%), two pseudo aneurysms, and not described in one. Thrombus inside the STA aneurysms was seen in 42.3% (11/26 cases). There were four cases (12.5%) with coexisting vascular lesion: two unruptured intracranial aneurysms and two aneurysms on contralateral STAs. Twenty-two cases (68.8%) increased in size

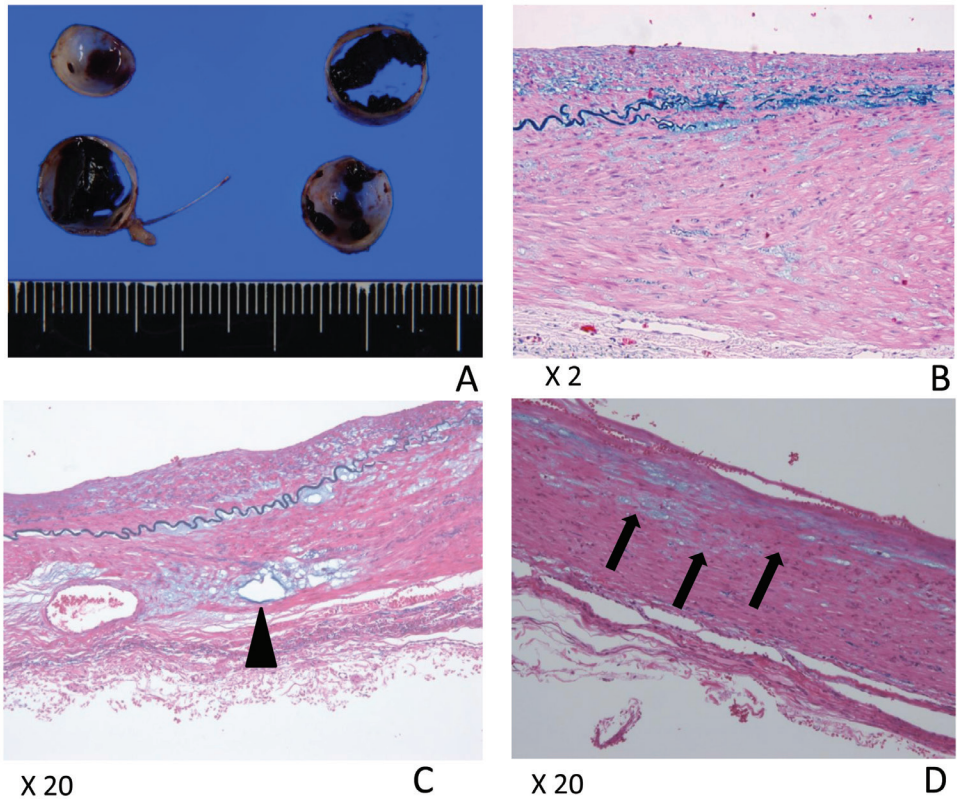

Fig. 4 A: Photograph of the resected aneurysm of Patient 2. B–D: Microscopic findings (hematoxylin and eosin stain). The thinned aneurysmal wall still generally maintained trilaminar arterial structure (B). The close observation with higher magnification showed multifocal cystic medial mucoid degeneration (C, *arrowhead*) and destruction or loss of elastic fiber (D, *arrows*).

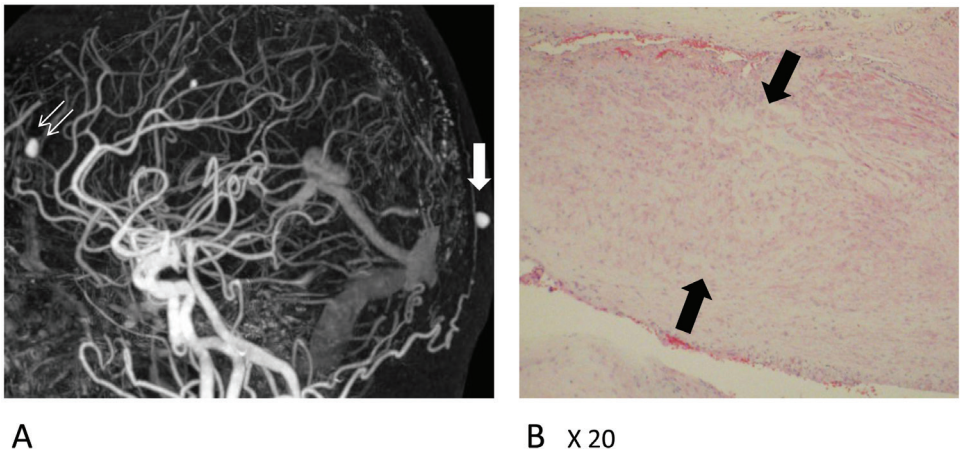

Fig. 5 A: Three-dimensional computed tomographic angiography showing a right superficial temporal artery aneurysm (*arrow*) and a right occipital artery aneurysm (*thin arrows*) in Patient 3. B: Microscopic findings (hematoxylin and eosin stain). Intimal thickening was seen in the background of normal media and adventitia (*between arrows*).

during 21.7 months long (mean) follow-up period since their first awareness of the mass (Table 1).

Discussion

The intracranial aneurysm is known to arise based on the combined background of congenital and acquired factors. On the other hand, the spontaneous STA aneurysm seems to be mainly caused by an acquired factor, such as the hypertension or the arteriosclerosis.[5,24] In our review, 7 cases (21.9%) had hypertension, and only 1 case had hyperlipidemia. But other cases had history of other vascular events such as abdominal aortic aneurysm, angina, and cerebral infarction. Pathologically, hyperplasia of the intima-media complex and adventitia as well as partial indistinctness of the internal elastic lamina was described in the previous reports including ours. Description of the detailed pathological findings was not obtained in all cases. When including our cases, Tamaki and other authors reported 3 cases (14.3%) with hyperplasia of the intima-media complex. Martin and other authors reported 13 cases (61.9%) with partial indistinctness of internal

Table 1 Reported cases of spontaneous superficial temporal artery aneurysms

Case	Author	Year	Age	Sex	Symptoms			Lag time to diagnosis (month)	Pathology (aneurysm)	Hyperplasia of the intima-media complex	Indistinctness of internal elastic lamina	Myxomatous changes of muscular layer	Thrombus	Complication of aneurysms in another location
					Pain	Pulsatility	Size increase							
1	Brown and Mehnert[6]	1942	34	M	–	–	+	60	True	n.d.	n.d	n.d	–	–
2	Martin and Shoemaker[16]	1955	60	M	–	+	+	60	True	–	+	–	–	–
3	Tamaki and Matsumoto[22]	1980	57	M	+	+	+	24	True	+	+	–	Unknown	–
4	Lozman	1982	78	F	–	+	+	24	Pseudo	n.d.	n.d	n.d	–	–
5	Inubusa[30]	1984	74	F	–	+	+	6	Unknown	n.d.	n.d	n.d	+	–
6	Buckspan and Rees[7]	1986	70	M	–	–	+	8	True	n.d.	n.d	n.d	+	–
7	Suzuki et al.[21]	1980	13	F	–	–	+	27	True	n.d.	n.d	n.d	Unknown	–
8	Locatelli et al.[14]	1982	10	M	–	–	–	Unknown	True	n.d.	n.d	n.d	Unknown	–
9	Nishioka et al.[18]	1988	14	M	–	–	+	24	True	–	+	–	+	–
10	Ezoe et al.[3]	1988	22	M	+	–	+	0.5	True	–	+	–	+	–
11	Yonetani et al.[29]	1995	63	F	–	+	+	36	True	n.d.	n.d	n.d	–	–
12	Fujii et al.[1]	1995	46	M	+	+	+	24	Pseudo	+	+	–	–	–
13	Ikeda and Watanabe[4]	1998	15	M	–	–	+	1	True	–	–	+	+	–
14	Uchida and Sakuma[25]	1999	34	M	–	+	+	3	True	–	+	–	+	–
15	Endo et al.[9]	2000	85	M	–	+	+	12	True	–	–	–	+	–
16	Porcellini et al.[20]	2001	24	F	–	–	–	0	True	n.d.	n.d	n.d	Unknown	–
17	Ohta et al.[19]	2003	55	M	–	–	–	0	True	–	–	–	–	Inrtacranial aneurysm
18	Riaz[2]	2004	65	M	–	–	+	24	True	–	–	–	–	–
19	Riaz[2]	2004	77	M	–	–	–	0	True	n.d.	n.d	n.d	–	–
20	Ysa et al.[23]	2008	59	F	+	+	+	8	True	–	–	–	Unknown	–
21	Kawabori et al.[13]	2009	78	F	–	+	+	24	True	–	+	–	–	–
22	Delis	2009	79	M	+	–	–	1	Pseudo	–	+	–	–	–
23	Piffaretti	2009	62	F	–	+	–	2	True	–	–	–	–	–
24	Piffaretti	2009	47	M	–	–	+	0.75	True	–	–	–	–	STA aneurysm
25	Karam et al.[11]	2010	34	F	–	+	+	24	True	–	+	–	–	–
26	Sakamoto	2011	77	F	–	+	+	2	True	–	+	–	+	–
27	Gokhan	2011	62	M	+	–	–	48	True	–	–	–	+	–
28	Bozkurt et al.[5]	2011	62	M	+	–	–	48	True	–	–	–	+	–
29	Nair et al.[17]	2011	84	F	–	+	–	24	True	n.d.	n.d	n.d	Unknown	–
30	Present case 1	2012	65	M	–	+	+	120	True	–	+	–	–	Inrtacranial aneurysm
31	Present case 2	2012	76	F	–	+	–	0	True	+	+	+	–	–
32	Present case 3	2012	57	F	–	+	+	36	True	+	+	–	+	STA and OA aneurysms

F: female, M: male, n.d.: not described, OA: occipital artery, STA: superficial temporal artery.

elastic lamina with our cases.[1,3,11,13,16,18,22,25] Focal myxomatous changes of the muscular layer in thickened aneurysm walls were also seen in two cases (9.5%) including our case.[4] Acquired factors, arteriosclerosis and hypertension, may be attributable of these pathologic changes.[5,24]

Spontaneous occipital artery aneurysms were also very rare. Five cases including our case have been reported so far,[26–28] which were less than those with STA aneurysms. Our Patient 3 had spontaneous occipital artery aneurysm; however, it has not been resected. Unfortunately, we could not confirm any pathological findings.

On the other hand, a considerable proportion of patients, i.e. 12.5% (4/32) were under 20 years of age. Given the fact that the young patients with same age bracket comprises only 1.3% in intracranial aneurysms and subarachnoid hemorrhage,[25] the higher ratio of the young segment of patients with spontaneous STA suggests some role of congenital factors in the genesis of this entity.

It is well known that the intracranial aneurysm preferentially occurs in patients with particular systemic diseases, such as Ehlers-Danlos syndrome, Marfan syndrome, and multicystic kidney. But these systemic diseases were not described in previous reports of spontaneous STA aneurysms including ours.

As for manifestations, pulsatile mass is the most common symptom; some of them are accompanied by pain. Facial nerve paralysis is rarely seen in this vascular pathology.[29]

The differential diagnoses for nontraumatic scalp lump include subcutaneous hematoma, dural arteriovenous fistula, inflammatory diseases as giant-cell arteritis, sebaceous cyst, dermoid cyst, neoplastic disease as facial nerve schwannoma and parotid gland tumor, meningocele, lymphadenopathy, subcutaneous abscess.[7,16,17,24,29] The pericranial sinus is one of the blood vessel masses, which also decreases in size by compression. Changes in mass size according to the fluctuation of venous pressure, accompanied by changes in posture and thoracic pressure, may clinically rule out the disease.

We found four patients having spontaneous STA aneurysm associated with aneurysms at other sites in the literature. Although these associations seem coincidental, the existence of common predisposing factors involving both intra- and extracranial aneurysms cannot be ruled out.

Ultrasonography, magnetic resonance imaging, and MRA are noninvasive imaging modalities for screening. Additionally, MRA and 3D-CTA are recommended to rule out the coexistence of intracranial vascular diseases.

Although there is a possibility of bleeding from spontaneous STA aneurysm as it runs through loose connective tissue, the subcutaneous hematoma has never been recorded; so cosmetic problem, pain, or discomfort is the reason for the treatment.

The main pillar behind the treatment is the direct removal of the aneurysm with ligation of inflow and outflow. It is a safe, effective, and simple method; in more than half of the reported cases, the patients underwent this procedure with local anesthesia.

Conclusion

We reported three cases of spontaneous and true STA aneurysms. All of those were resected under local anesthesia safely. Spontaneous STA aneurysm should be a differential diagnosis of pulsatile scalp mass. Literature review showed that this lesion might occur in younger people, suggesting congenital factors. It may be noteworthy that STA aneurysms may coexist with other vascular lesions such as intra- or extracranial aneurysm.

Conflicts of Interest Disclosure

None. All authors who are members of the Japan Neurosurgical Society (JNS) have registered Online Self-reported COI Disclosure Statement Forms through the website for JNS members.

References

1) Fujii S, Kajikawa H, Yamamura K, Wada M, Shimamoto F: [Spontaneous dissecting aneurysm of the superficial temporal artery: a case report]. *No Shinkei Geka* 23: 797–800, 1995 (Japanese)
2) Riaz AA, Ismail M, Sheikh N, Ahmed N, Atkin G, Richman P, Loh A: Spontaneously arising superficial temporal artery aneurysms: report of two cases and review of the literature. *Ann R Coll Surg Engl* 86: 861–863, 2004
3) Ezoe K, Shono M, Mizogutchi F, Hada S: [Superficial temporal artery aneurysm]. *Hifuka no Rinsho* 30: 1077–1081, 1988 (Japanese)
4) Ikeda S, Watanabe T: [Superficial temporal artery aneurysm associated with pathological changes mimicking cystic medial necrosis]. *Neurol Med Chir* 28: 1223–1227, 1998 (Japanese)
5) Bozkurt G, Ayhan S, Cakici N, Celik O, Ziyal IM: Spontaneous nonpulsatile aneurysm of the superficial temporal artery mimicking a subcutaneous mass lesion. *J Craniofac Surg* 22: 371–372, 2011
6) Brown RK, Mehnert RH: Aneurysm of the temporal artery. *Surgery* 12: 711–715, 1942
7) Buckspan RJ, Rees RS: Aneurysm of the superficial temporal artery presenting as a parotid mass. *Plast Reconstr Surg* 78: 515–517, 1986
8) Coscarón Blanco E, Benito JJ, Benito F, Gómez JL, del Cañizo Alvarez A, Antúnez P: [Non-pulsatile aneurysm of the superficial temporal artery: a report of a case]. *Acta Otorrinolaringol Esp* 54: 388–391, 2003 (Spanish)
9) Endo T, Mori K, Maeda M: [Multiple arteriosclerotic fusiform aneurysms of the superficial temporal artery—case report]. *Neurol Med Chir* 40: 321–323, 2000 (Japanese)
10) Harvey L, Moses N: Aneurysm of superficial temporal artery. *Am J Otolaryngol* 3: 376–378, 1982
11) Karam L, El Husseiny M, Abadjian G, Slaba S, Tabet G: Case report of a rare spontaneous superficial temporal artery

aneurysm. *J Mal Vasc* 35: 366–368, 2010
12) Kasai T, Jinbo M, Yamamono M, Ide M, Tanaka N, Watanabe E: [Traumatic aneurysm of the superficial temporal artery case report]. *J Tokyo Wom Med Univ* 55: 653–655, 1985 (Japanese)
13) Kawabori M, Kuroda S, Nakayama N, Kenmotsu Y, Shimizu H, Tanino M, Iwasaki Y: Spontaneous giant aneurysm of the superficial temporal artery: case report. *Neurol Med Chir* 49: 198–201, 2009
14) Locatelli D, Messina AL, Ricevuti G, Gajno TM: Superficial temporal artery aneurysm. *Am J Otolaryngol* 3: 376–378, 1982
15) Schechter MM, Gutstein RA: Aneurysms and arteriovenous fistulas of the superficial temporal vessels. *Radiology* 97: 549–557, 1970
16) Martin WL, Shoemaker WC: Temporal artery aneurysm. *Am J Surg* 89: 700–702, 1955
17) Nair R, Varsani D, Frecker P: An unusual bump on the head: spontaneous superficial temporal artery aneurysm. *Indian J Surg* 73: 240–241, 2011
18) Nishioka T, Kondo A, Aoyama I, Nin K, Shimotake K, Tashiro H, Takahashi J, Kusaka H: [A case of spontaneous superficial temporal artery aneurysm]. *No Shinkei Geka* 16: 1009–1012, 1988 (Japanese)
19) Ohta H, Sakai H, Nakahara I, Sakai N, Nagata I, Ishibashi-Ueda H: Spontaneous superficial temporal artery aneurysm associated with multiple intracranial cerebral aneurysms—does it segmental mediolytic arteriopathy of the intra- and extra-cranial arteries? *Acta Neurochir (Wien)* 145: 805–806, 2003
20) Porcellini M, Bernardo B, Spinetti F, Carbone F: Outpatient management of superficial temporal artery aneurysms. *J Cardiovasc Surg* 42: 233–236, 2001
21) Suzuki Y, Kobayashi S, Nagamine Y, Higuchi H: [A case of spontaneous superficial temporal artery aneurysm of child]. *Iwate Kenritsu Byoin Igakka Zasshi* 28: 85–88, 1980 (Japanese)
22) Tamaki N, Matsumoto S: [A case of superficial temporal artery aneurysm]. *Nihon Isikai Zasshi* 4: 629–631, 1980 (Japanese)
23) Ysa A, Arruabarrena A, Bustabad MR, Perez E, del Campo A, Garcia-Alonso J: Images in vascular medicine. True aneurysm of the superficial temporal artery. *Vasc Med*: 295–296, 2008
24) Matsuda F, Sugie A, Kajiwara H, Yamamura K, Komada O, Ogawa R, Wakabayashi S, Wakabayashi C, Shimamoto F: Traumatic pseudoaneurysm of superficial temporal artery: a case report. *Jpn Neurosurg* 10: 339–342, 2001
25) Uchida N, Sakuma M: Atherosclerotic superficial temporal artery aneurysm: report of a case. *Surg Today* 29: 575–578, 1999
26) El Shazly AA, Saoud KM: Results of surgical excision of cirsoid aneurysm of the scalp without preoperative interventions. *Asian J Neurosurg* 7: 191–196, 2012
27) Kim HS, Son BC, Lee SW, Kim IS: A rare case of spontaneous true aneurysm of the occipital artery. *J Korean Neurosurg Soc* 47: 310–312, 2010
28) Poncet E, Roulleau P: [Buzzing in the ears due to an aneurysm of the occipital artery]. *Ann Otolaryngol Chir Cervicofac* 87: 352–353, 1970 (French)
29) Yonetani M, Inoue H, Fushimi S, Hirayama A: [Two cases of superficial temporal artery aneurysm]. *Akita Ken Ishikai Zasshi* 46: 200, 1995 (Japanese)
30) Inubusa H, Yamauchi H, Wada E, Matsunami E: [A case of superficial temporal artery aneurysm]. *Gifu Daigaku Igakubu Kiyo* 32: 936, 1984 (Japanese)

Address reprint requests to: Hiroshi Tokimura, MD, PhD, Department of Neurosurgery, Graduate School of Medical and Dental Sciences, Kagoshima University, 8-35-1, Sakuragaoka, Kagoshima, Kagoshima 890-8520, Japan.
e-mail: tokimura@m3.kufm.kagoshima-u.ac.jp