Title：

Intravascular Papillary Endothelial Hyperplasia in an Aneurysm of the Superficial Temporal Artery: Report of a Case

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Abstract

We herein report a rare case of an intravascular papillary endothelial hyperplasia in an aneurysm of the superficial temporal artery. The patient was a 67-year-old Japanese female. She noticed a throbbing swelling in her left forehead, which had gradually been increasing in size. She had no previous history of head trauma. The ultrasound and three-dimensional computed tomography angiography revealed an aneurysm with a mural thrombus measuring 10mm in diameter fed by the frontal branch of the left superficial temporal artery. The aneurysm of the superficial temporal artery was dissected from the surrounding tissues, and was resected after ligation of feeding vessels. A microscopic examination revealed papillary endothelial hyperplasia in a true aneurysm. Non-traumatic aneurysms of the superficial temporal artery are quite rare. In the previous English literature, there have only been a few reports of papillary endothelial hyperplasia in an artery, and none in an aneurysm of the superficial temporal artery.

(151 words)

Key Words: superficial temporal artery aneurysm, true aneurysm, intravascular papillary endothelial hyperplasia

Introduction

Traumatic aneurysm of the superficial temporal artery (STA) is not unusual, with more than 400 reported cases.[1] However, spontaneous true STA aneurysms are quite rare, with only 14 cases described.[2-9] In addition, intravascular papillary endothelial hyperplasia (IPEH) is a well recognized but not frequently reported histological diagnosis, first described by Masson in 1923.[10] Most commonly, it appears in thrombosed dilated veins or vascular tumors. There have been a few reports of its presence in peripheral artery aneurysms.[11-13] We herein report a case of IPEH associated with a true aneurysm of a STA.

Case report

A 67-year-old Japanese woman first recognized a small mass in her left lateral forehead region two years previously, but did not consult a physician because of the absence of pain or discomfort. However, the mass gradually increased in size over two years, so she consulted a clinic and was referred to our hospital. She had no evidence of head or face injury.

The physical examination was entirely normal except for a 10 mm pulsatile swelling in the left forehead (Fig.1A), which had well demarcated edges and no bruit over it. Compression of the proximal artery diminished the pulsation but did not reduce the size. She had a history of hypertension and had no family history of any connective tissue disorders.

Laboratory studies showed no abnormalities. Computed tomography (CT) with contrast medium showed homogeneous enhanced mass with thrombus in the forehead subcutaneous region. Three-dimensional volume-rendered CT angiograms revealed an aneurysm arising from the frontal branch of the left STA, and were also helpful for better understanding the spatial relationship between the aneurysm and the STA (Fig.1B). Ultrasonography showed turbulent flow within the aneurysm with mural thrombus. Based on these physical and radiological findings, our diagnosis was a non-traumatic aneurysm of STA.

She underwent surgical excision under general anesthsesia. After exposure of the proximal and distal STA, the aneurysm was carefully dissected from the surrounding tissue. The aneurysm was resected after ligation of the afferent and efferent arteries. The aneurysm was 10 x 10 mm with thickened wall. The histological examination revealed that three layers of the aneurysmal wall were seen to be intact and that the papillary structures confined within aneurysmal space. The papillary structures were covered by single layer of endothelial cells. The stalk of papillary structures contained hyalinized fibrin with sinusoidal capillary (Fig.2). In addition, there was no evidence of any giant cells, inflammation or vasculitis. The recovery period was uneventful and the patient was discharged without complications on the third postoperative day.

Discussion

The majority of STA aneurysms are false aneurysms arising secondarily to trauma. Peick et al.[14] reviewed a total of 174 aneurysms of STA as of 1988. Of these 90% were caused by trauma, generally a blunt injury. The STA is vulnerable to injury because the temporal muscle is the only protective tissue between the STA and the skull. In 1942, Brown and Mehnert[2] described the first histologically confirmed case of a true aneurysm. A histological distinction is made between true aneurysma, in which all three layers of the arterial wall are intact, and pseudoaneurysms, in which there is a partial break in the arterial wall. In the present case, all three layers were intact. In the English-language literature, spontaneous true aneurysm of STA without history of head injury have been found in only 15 cases including the present case (Table 1).[2-9] The nine male and six female patients were aged between 10 and 85 years old. The aneurysm arose in the main trunk of STA in seven cases and in distal branch of STA in eight cases. All cases were safely underwent surgical excision. The age range suggests that atherosclerosis is not the common and only reason, although atherosclerotic change of STA and/or hemodynamic stress to the arterial wall might be important in the development of true aneurysm of STA.[5]

The natural history of STA aneurysm is not well documented. Treatment includes conservative treatment, surgery, selective catheterization with embolization, or percutaneous thrombin injection. [14-16] In the literature, there are no indications as to the optimum size of the aneurysm of STA to initiate surgical excision. The recognized indications for surgery include pain, increasing size, cosmesis, changes to the overlying skin or adjacent structures.

Aneurysm formation has been described in temporal arteritis. [17] The histological features of classical temporal arteritis generally involve granulomatous arteritis with prominent Langerhans giant cells, predominantly at the media of the artery with smooth muscle necrosis; nonspecific inflammation reaction, with lymphocytes, and eosinophils concentrated at the arterial wall; and intimal fibrosis with occlusion of the vessel lumen.[18] Accumulation of eosinophils along the entire vascular wall is more often related to hypersensitive angitis and might provoke lymphoid granulomatous inflammation with eosinophilic infiltration inducing the prominence.[19] Eosinophilic accumulation might be involved in temporal arteritis. Aneurysm formation may be caused by such inflammatory changes of vascular wall. Histological examination in the present case revealed no giant cells or fibrinoid necrosis, and no infiltration of inflammatory cells were observed along whole arterial wall. In addition, present case had no major clinical symptoms of temporal arteritis such as headache or blurred vision. Therefore, in the present case, the pathogenesis seemed to be different from typical temporal arteritis.

In addition, in the present case, histological examination revealed the papillary structures,

which contained hyalinized fibrin with sinusoidal capillary, covered single layer of endothelial cells confined within aneurysmal space. The present histological findings were met the definition of IPEH or otherwise known as Masson tumor. **IPEH**, a non-neoplastic reactive process, was first described by Masson, [10] who observed this peculiar endothelial proliferation in a thrombosed hemorrhoidal vein. He regarded it as a benign neoplastic process and coined the term vegetant intravascular hemangioendothelioma. **IPEH** is a benign but is a form of abnormal organization for thrombus formation.[20] **IPEH** can mimic vascular neoplasms such as angiosarcoma, which virtually never occurs within the lumen of vessel. Typical IPEH arises within and remains confined to the lumen of vessel, such as a vein in setting of inflammation or vascular stasis.

The pathogenesis of IPEH remains contentious. It is presumably related to an unusual form of thrombus organization. The exuberant endothelial cell proliferation may be stimulated by an autocrine loop of endothelial basic fibroblast growth factor (bFGF) secretion. The proposed mechanism is that IPEH development is triggered by the release of bFGF from macrophages attracted to a site of trauma. The proliferating endothelial cells release more bFGF, setting up a positive feedback loop of endothelial proliferation.[21]

Including the present case, only four cases of histologically verified IPEH arising from within the peripheral artery aneurysm have been reported (Table 2).[11-13] The one male and three female patients were aged between 44 and 67 years old. All of these four cases were preoperatively diagnosed with thrombosed peripheral artery aneurysm or aneurysm with mural thrombus. Furthermore, in these cases definitive diagnosis were made postoperatively, after histological reviews of resected aneurysms.

IPEH generally has good prognosis but there are cases of IPEH recurring locally, should the excision be incomplete.[22] Ther**e**fore, if IPEH is observed within an aneurysm, complete resection should be performed and long term follow up is necessary.

To our knowledge, this is the first reported case of an **IPEH** in an aneurysm of the STA. We selected surgical resection because the aneurysm had gradually increased in size. Surgical resection can prevent rupture or recurrence.

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Figure legend

Fig. 1. **A** Preoperative photograph of the patient showing a mass in the forehead. **B** Three-dimensional volume-rendered computed tomography angiogram revealing an aneurysm arising from the frontal branch of the left superficial temporal artery.

Fig.2. **A** Photomicrograph showing intact three layers of the aneurysmal wall and the complex intraluminal papillary structures. **B** Photomicrograph showing the stalk of papillary structures, which coverd by single layer of endothelial cells, contained hyalinized fibrin with sinusoidal capillary.