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CASE REPORT

Intra-arterial angiolymphoid hyperplasia with eosinophilia of the temporal artery: Report of two cases and review of the literature

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ABSTRACT

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a relatively common benign vascular disorder of uncertain etiology and has many synonyms, such as pseudopyogenic granuloma, atypical pyogenic granuloma, epithelioid hemangioma, and histiocytoid hemangioma. It usually develops on the head and neck, manifesting as reddish-to-dark purpuric papules or nodules. In this article, we report two patients who had uncommon intra-arterial ALHE that occurred in the temporal artery. In both cases, the ALHE presented as skin-colored subcutaneous nodules over the forehead, mimicking temporal arteritis. Histopathologically, intravascular epithelioid endothelial cell proliferation occurred, with lymphocyte and eosinophil infiltration in the stroma. One patient also showed typical findings of ALHE in the adjacent soft tissue. We know of seven similar cases that have previously been reported in the literature to date.

Introduction

Intravascular proliferative disorders may represent an extension or metastasis of a primary tumor, intravascular angiomatosis,[1] or reactive proliferation-associated repair processes. These repair processes include intravascular papillary endothelial hyperplasia,[2] intravascular pyogenic granuloma,[3] and intravascular fasciitis.[4,5] Angiolymphoid hyperplasia with eosinophilia (ALHE) is a vascular disease characterized by proliferation of capillaries and venules with plump endothelial cells and a variable inflammatory cell infiltration composed of lymphocytes and eosinophils.[6] It is most commonly seen on the head and neck,[7] and is rarely reported occurring intravascularly. We present here two cases of intra-arterial ALHE of the temporal artery and review the cases reported in the literature.

Case reports

Case 1

A 45-year-old man presented with two asymptomatic skin-colored subcutaneous nodules on the left temporal region (Figure 1A). With the impression that this was an epidermal cyst, an excisional biopsy was performed. Histopathologically, an occluded muscular blood vessel was found in the subcutaneous tissue (Figure 1B). An intravascular proliferation was present and was composed of epithelioid endothelial cells, lymphocytes, and many eosinophils (Figure 1C). Scattered CD68-positive histiocytes were seen. Neither granulomatous inflammation nor multinucleated giant cells were present. The Verhoeff–van Gieson stain demonstrated an internal elastic lamina (Figure 1D), indicating a medium-to-large-sized artery. No recurrence was noted 2 years after excision of the tumor.

Case 2

A 35-year-old man presented with one asymptomatic skin-colored subcutaneous nodule on his left forehead (Figure 2A). The mass was excised as it was believed to be an epidermal cyst. Histopathology showed a medium-sized artery with plump endothelial cells in the lumen (Figures 2B and 2C). The arterial wall was infiltrated by lymphocytes and eosinophils (Figure 2D). Common features of ALHE, such as a proliferation of capillary-sized vessels lined by epithelioid endothelial cells and infiltration of lymphocytes and eosinophils, were present in the peripheral stroma (Figure 2E). Verhoeff–van Gieson staining demonstrated the presence of an internal elastic lamina (Figure 2F). No recurrence was noted 1 year after surgical excision.

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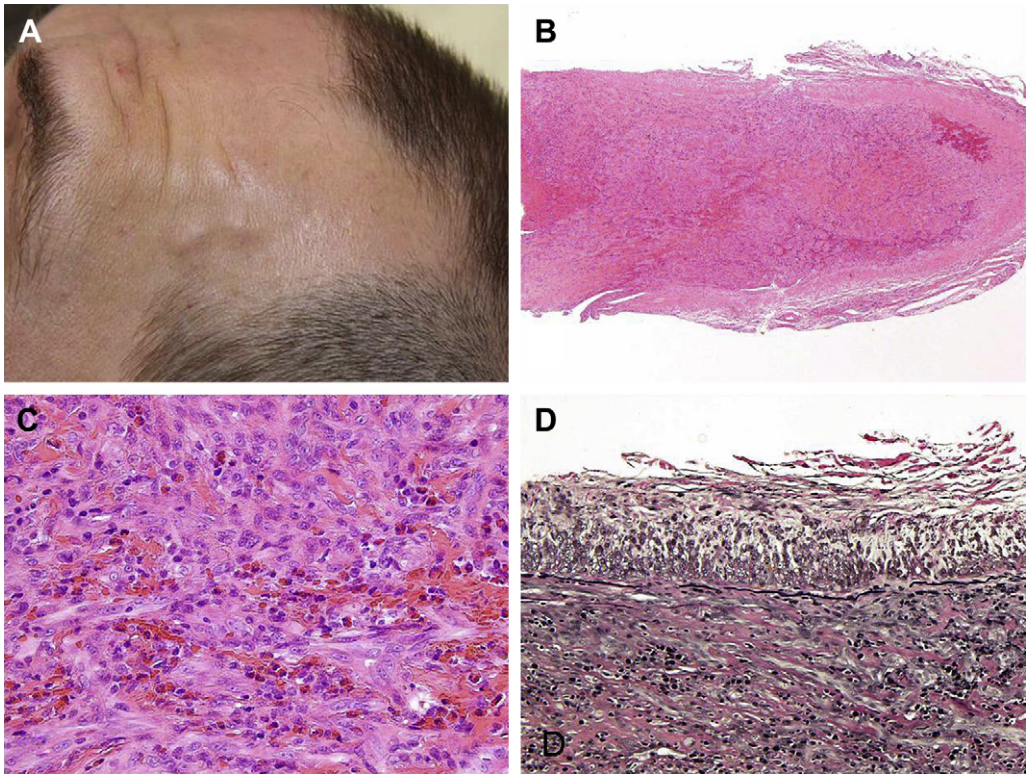

Figure 1 (A) Two skin-colored subcutaneous nodules on the left temporal area. (B) Skin biopsy showing an occluded vascular lumen. (C) Higher magnification showing intravascular epithelioid endothelial cell proliferation with eosinophil infiltration. (Hematoxylin and eosin; B, 40×; C, 400×.) (D) Verhoeff–van Gieson staining showing the internal elastic lamina (100×).

Discussion

The two cases we have reported have provided more evidence that ALHE may occur intravascularly. In Fetsch and Weiss's series, when ALHE occurred in the subcutis and deep soft tissue, a medium-sized vessel, such as an artery (37/96, 39%) or vein (23/96, 24%), was often observed located either in continuity with or close to the lesion.[6] Similarly, in Olsen and Helwig's study of 116 patients, 53 cases showed an association with the artery.[8]

Although the presence of a medium-sized artery near ALHE is not uncommon, the development of ALHE inside a muscular artery is rarely seen. About 19 cases have been reported, the involved arteries including the temporal artery,[9–15] radial artery,[16–18] facial artery,[19] post-auricular artery,[20] popliteal artery,[21] brachial artery,[22,23] occipital artery,[23] ulnar artery,[24,25]and axillary artery.[26] Among these, seven cases of ALHE developed within the temporal arteries (Table 1) and presented with narrowing or occlusion of the lumen. There are some pathological differences between these specimens. One case was diagnosed as a traumatic pseudoaneurysm and was considered to be an early lesion of ALHE.[9] One case did not have eosinophilic infiltration,[10] and two cases had typical ALHE findings in the peripheral stroma around the artery.[9, 11] Our two cases also showed ALHE developing in the temporal artery with obstruction of the lumen.

When the temporal artery is affected by ALHE, the most important differential diagnosis is temporal arteritis (Horton's disease). Temporal arteritis is an intra-arterial granulomatous inflammatory process involving the large and medium-sized arteries. The patient may experience tenderness, overlying skin necrosis, and systemic symptoms such as cyanosis of the extremities, retina ischemia, fever, weakness, myalgia and arthralgia, which have been infrequently reported in intra-arterial ALHE.[10,15]

Juvenile temporal arteritis should also be discussed here because of our two patients' young age. It is a localized form of temporal arteritis without a systemic inflammatory reaction. The histological features of temporal arteritis, such as the presence of mononuclear inflammatory cell and giant cell infiltration of the vascular media, are different from those of intra-arterial ALHE. The histopathology of juvenile temporal arteritis often reveals non-granulomatous panarteritis with lymphocyte and eosinophil infiltration. It shows some features that are similar to those of intra-arterial ALHE but lacks the characteristic of plump endothelial cell proliferation. The may be an association between temporal arteritis and intra-arterial ALHE as they may represent reactive vascular proliferation in the repair process for intravascular injuries.

In addition, Kimura disease and organized thrombosis should be put into the differential diagnosis of intra-arterial ALHE. Due to the similarities between Kimura disease and ALHE, they were previously considered to be part of a single disease spectrum, but now it is clear that they are two different entities. Kimura disease is usually located in the subcutaneous tissue of the head and neck, with systemic lymphadenopathy, marked eosinophilia, and elevated serum immunoglobulin E level. Histopathology shows eosinophil and lymphocyte infiltration with obvious lymphoid follicles and a lack of epithelioid endothelial cell proliferation. No intravascular growth has yet been reported.

Organized thrombosis implies a vessel that has been obstructed by connective tissue composed of numerous neovessels. Sometimes the thrombus shows recanalization with multiple, small, endothelium-lined papillary structures with hyaline stalks, which have been named intravascular papillary endothelial hyperplasia (Masson's tumor). However, our two cases lack the features of papillary structure and have large numbers of eosinophils with