CASE REPORT

The ultrasound halo sign in angiolymphoid hyperplasia of the temporal artery

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ABSTRACT. An ultrasound halo sign surrounding the temporal artery is a well recognized feature associated with giant cell arteritis. We report a previously unreported case of this halo sign being present around the temporal artery due to angiolymphoid hyperplasia with eosinophilia (ALHE) in a young female patient.

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Angiolymphoid hyperplasia with eosinophilia (ALHE) is a vascular proliferative disease with an unknown aetiology. It is an uncommon cause of a forehead or temporal region nodule [1]. Typically, ALHE arises in the head and neck region of young women, especially around the ear, presenting as raised sometimes coloured nodules which are often itchy. It rarely regresses spontaneously, but there are no reports of metastatic spread [2].

ALHE is most often found in vessels of the subcutis or dermis, but can affect larger muscular arteries, such as the facial artery and the temporal artery, and rarer cases have also been reported on the hand and in the mouth [3–6].

Microscopic features include vessels lined by atypical endothelial cells, which are plump and epithelioid. There is a characteristic eosinophilic infiltration in the perivascular tissue, associated with lymphocytic infiltration. 20% of cases also have generalized eosinophilia, and occasionally raised serum IgE levels [3, 7].

Being uncommon, it is easily misdiagnosed; nodules have been mistaken for more common pathologies such as lipoma, sebaceous cyst, Dupuytrens disease or giant cell arteritis – in some cases leading to unexpected severe haemorrhage at operation [5, 8, 9].

We present a case in which angiolymphoid hyperplasia mimicked giant cell arteritis on preoperative Doppler ultrasound due to the presence of a halo sign.

Case report

A 31-year-old female patient presented to clinic with an 18 month history of a solitary painless but itchy nodule in her left temporal region. There had been no history of trauma, and it had not bruised or bled. She did not report migraines, jaw claudication, muscle cramps or visual symptoms.

On examination she was found to have a fixed, pulsatile mass in the left temporal region with no overlying skin changes. A vascular lesion was suspected and pre-operative Doppler ultrasound was performed. Routine haematology, erythrocyte sedimentation rate (ESR) and biochemistry tests were all within normal reference ranges.

Pre-operative ultrasound scanning was performed using a 13 MHz linear transducer (Sonoline Antares; Siemens, Erlangen, Germany). The ultrasound study demonstrated circumferential but asymmetric thickening of the left superficial temporal artery and its branches, with wall thickening of between 1 mm and 2 mm. The changes were purely arterial and there was no abnormal flow pattern seen within the lumen. There was a halo effect seen around the affected vessels over a length of 20 mm. These ultrasound appearances were strongly suggestive of a diagnosis of giant cell arteritis (Figure 1).

Subsequent excision (Figure 2) and histological analysis (Figure 3) of the lesion revealed a diagnosis of angiolymphoid hyperplasia with eosinophilia involving branches of the superficial temporal artery.

Discussion

As previously stated, the aetiology of ALHE is unknown. Some argue ALHE represents a reaction to previous microtrauma to vessels as there is an association with evidence of previous vessel damage such as arteriovenous shunting and vascular wall repair [7, 10]. Others propose ALHE is a benign vascular neoplasm [11] which has been shown to be associated with hyperoestrogenism [2, 12]. Even though ALHE has some similar features to Kimura's disease, it is considered a different entity [13].

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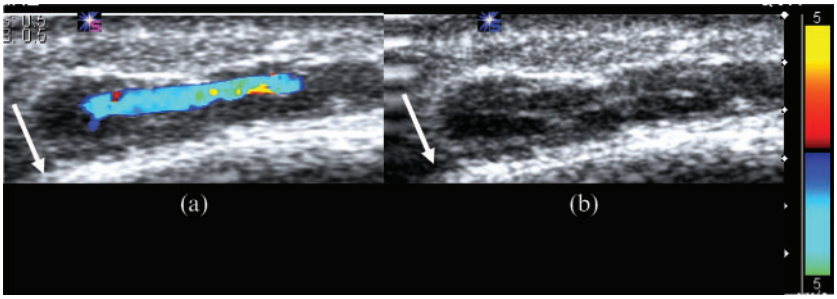

Figure 1. Ultrasound image of a branch of left superficial temporal artery showing marked asymmetric thickening of the vessel wall, producing a ''halo'' around the lumen. (a) With colour Doppler. (b) Same image without colour. Arrow indicates the outer table of skull.

A variety of treatments for the nodules have been attempted, most of which are based on small case numbers. Modalities have included cryosurgery [14], corticosteroids [15], lasers [16, 17], intralesional interferon injections [18] and Mohs micrographic surgery [19], but overall up to a third of cases are thought to recur. Complete surgical excision has been shown to have lower recurrence rates [20].

Diagnostic imaging of ALHE has included angiography, FDG PET scanning, CT and nuclear magnetic resonance scanning (MR), with Doppler ultrasound being mentioned in some reports [3, 5, 21, 22].

CT of ALHE usually shows an enhancing mass [3, 23]. Angiographic appearances demonstrate a thickened enhancing arterial wall or mass, often with fusiform dilatation which can lead to the misdiagnosis of aneurysm. This enhancement begins in the late arterial phase and becomes most intense during the capillary and venous phases [23]. At MR and PET scanning, the lesion has been described as being vascular [21]. Some of these imaging features are also seen in giant cell arteritis, which is characterized by a hypoechoic halo at ultrasound [24] and a thickened enhancing wall at MR [22].

The principal unusual finding in this case was the asymmetric ultrasound halo seen around the temporal artery affected by ALHE. In previous reports of a halo sign around the temporal artery, the diagnosis has almost always been that of giant cell arteritis. Rare reports have associated the halo effect with other diagnoses including polymyalgia rheumatica, infection, malignancy and osteoarthritis [25].

Published meta-analysis has shown that the halo sign is only predictive of giant cell arteritis if the pre-test probability of giant cell arteritis is moderate to high, based on clinical criteria including age (over 50 years) and ESR (raised). If the pre-test probability of giant cell arteritis is low, such as in this case, then the post-test probability of giant cell arteritis is approximately 30–50% [26].

Ultrasound features of ALHE that have been previously reported include a vascular mass and low resistance arterial waveforms seen on Doppler [3, 27]. A halo sign at ultrasound has not previously been reported in AHLE. In giant cell arteritis it is thought that the halo sign is due to oedema rather than inflammatory infiltrate, as the halo is not always associated with an infiltrative process [24]. We are uncertain of the exact

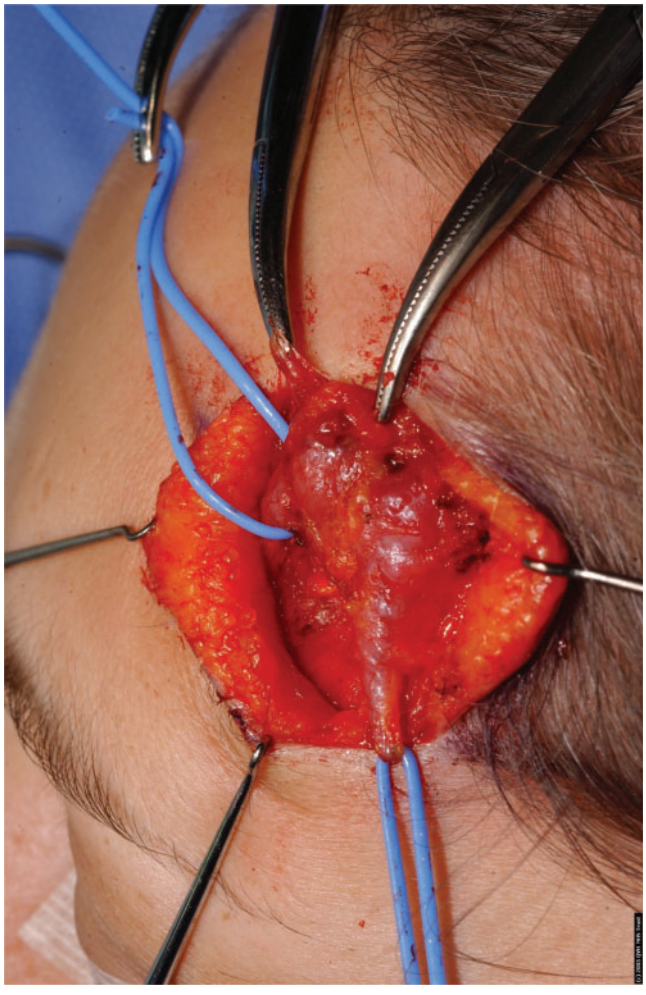

Figure 2. Macroscopic appearance of lesion at surgery.

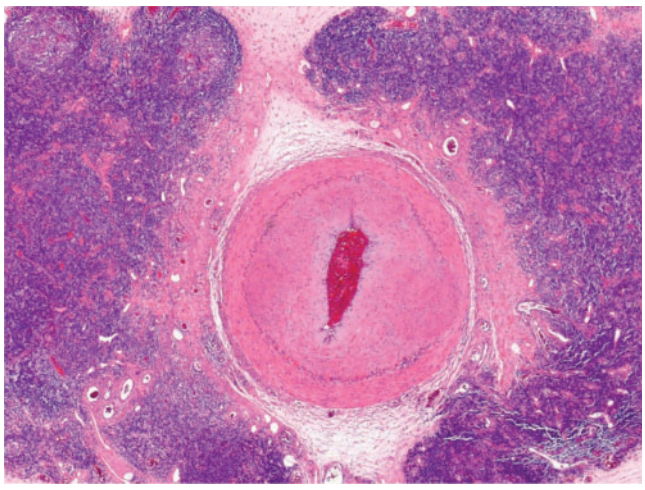

Figure 3. Central artery surrounded by florid lymphoid tissue. Haematoxylin and eosin, field width 3.4 mm.

histological correlate of the halo in ALHE, but it is likely to be due to the perivascular infiltrate and, provided the lumen has not been completely obliterated, the authors feel that the halo is not an unexpected finding in view of the marked thickening of the arterial wall that is associated with ALHE.

We report this case to acquaint radiologists with this rare vascular lesion, and to encourage pre-operative ultrasound imaging of any prominent itchy skin nodule prior to excision, especially in the region of the head and neck.

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