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Angiolymphoid hyperplasia with eosinophilia of the parapharyngeal space

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Abstract
Angiolymphoid hyperplasia with eosinophilia (ALHE) is an uncommon benign condition characterized by cutaneous nodules with a predeliction for the head and neck region. Extracutaneous involvement is rare. We report a 44-year-old woman who had a large submucosal ALHE tumour in the parapharyngeal space. Our patient is of interest because of the unusual, and as far as we are aware from the literature, unique site and presentation of her lesion.

Key words: Angiolymphoid hyperplasia with eosinophilia; Neck

Case report

A 44-year-old woman presented to clinic when after nine months of increasingly severe snoring she inspected her throat to find a large left-sided pharyngeal swelling. She had no other symptoms. She smoked 10 cigarettes per day and had no previous medical history.

Examination revealed a non-tender longitudinally fusiform submucosal swelling of the left posterior pharyngeal wall, extending from the nasopharynx to the pyriform fossa and protruding to the mid-lumen of the oropharynx. The overlying mucosa was intact and of normal appearance. There was no palpable lymphadenopathy. There were no further abnormal clinical findings.

Computed tomography (CT) scan of the patient's neck (Figure 1) showed a vascular mass in the parapharyngeal space. Full blood count and white cell differential, urea and electrolytes, liver function tests, bone marrow aspirate, chest radiograph and thoraco-abdominal computed tomography (CT) scan were all normal.

Despite two biopsies (first transcutaneously and second transorally) histological diagnosis remained inconclusive although initially suggestive of low grade B-cell non-Hodgkins lymphoma. The patient started to develop dysphagia. The mass was then excised via a paramedian mandible-splitting approach to the parapharyngeal space. The patient had no further treatment and has had no recurrence after one year of follow-up.

Pathological examination of the excised specimen revealed a $60 \times 50 \times 40$ mm well-defined but not encapsulated mass. Histopathological and immunocytochemical examination revealed the tumour to consist of angiolymphoid hyperplasia with eosinophilia (Figure 2). The composition was of a loose collagenous connective tissue with a rich vascular supply and associated lymphocytic infiltrate with follicle formation and interspersed eosinophils.

The endothelial markers (Factor 8, Q BEND 10 (CD 34)) showed the numerous blood vessels and their varying and irregular luminal calibres from muscular arterioles 1 mm in diameter to capillary size vessels which permeated the lymphoid tissue. The majority of these vascular channels were lined by prominent endothelial cells with large nuclei and frequent cytoplasmic vacuolations.

The lymphocytic infiltrate with follicle formation was intimately associated with the vascular channels (Figure 3). Immunocytochemistry for B- and T-cell markers showed a diffuse T cell infiltrate with prominent B cell follicle centres staining strongly with BCL-2 and MT2. Equal numbers of kappa and lambda positive lymphocytes were seen. The B cell lymphoid staining pattern for BCL-2 and MT2 was similar to that of an adjacent piece of normal tonsillar tissue and this finding emphasized the benign reactive nature of the lymphocytic infiltrate.

Small numbers of eosinophils were seen in vascular lumens and interspersed throughout the specimen but there was no infiltration of the lymphoid follicles or folliculolysis. There was no evidence of vasculitis, no fibrin

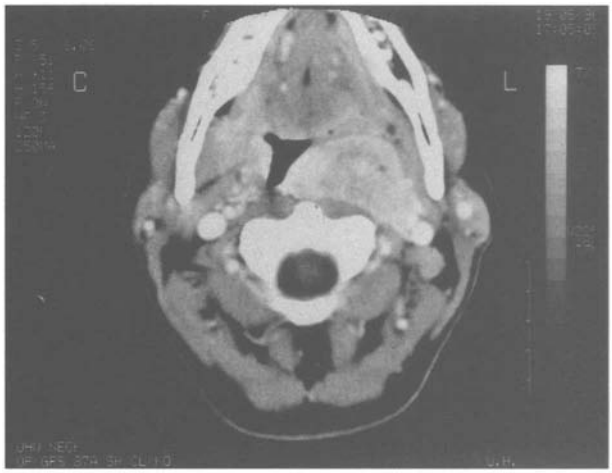

Fig. 1
CT scan showing parapharyngeal mass.

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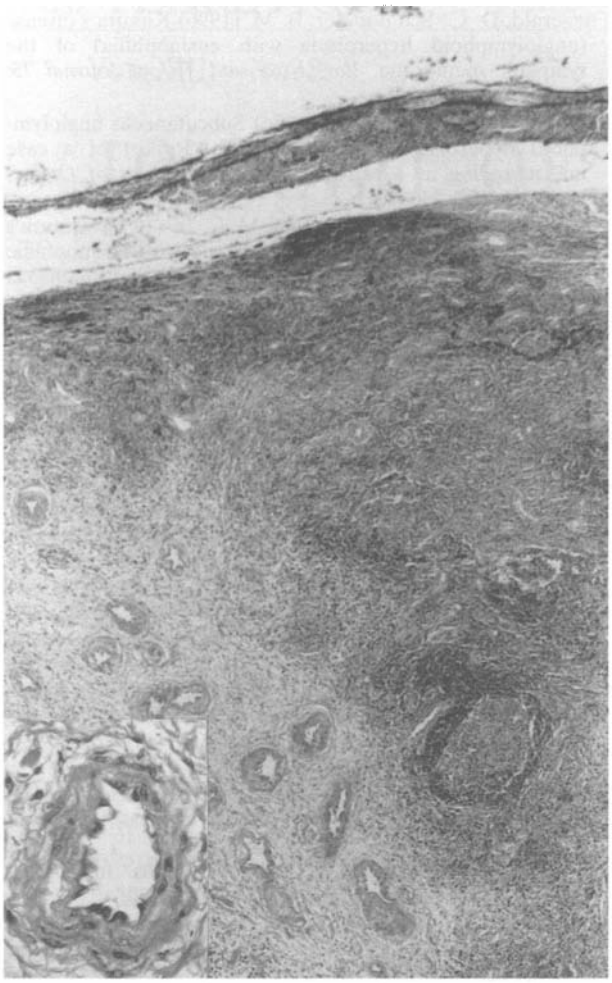

FIG. 2

Photomicrograph showing well-defined mass of lymphoid tissue containing follicles and prominent thick-walled blood vessels. Insert: the vessels are lined by prominent endothelial cells some of which contain intracytoplasmic clear vacuoles.

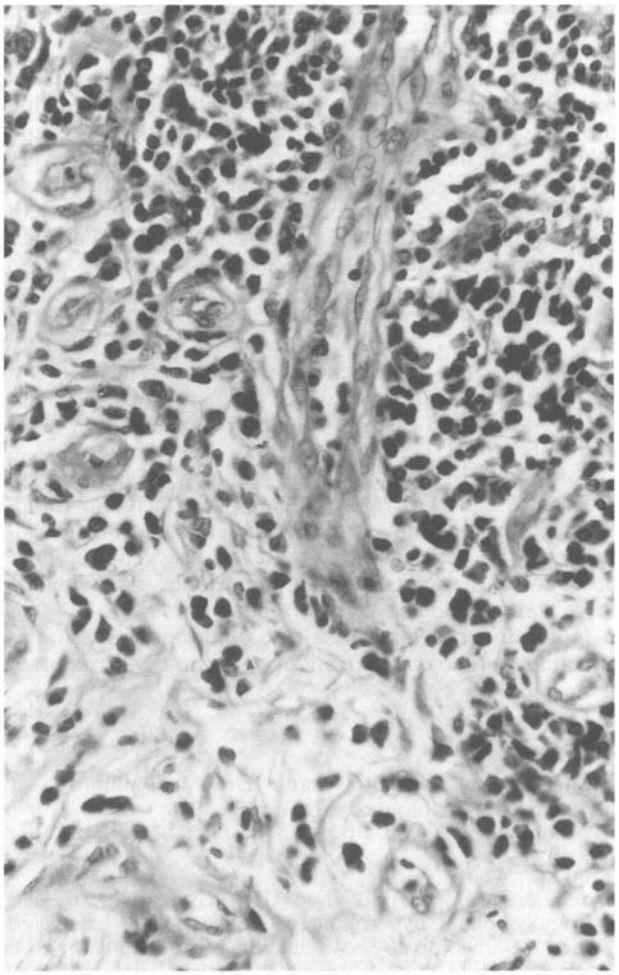

FIG. 3

Photomicrograph showing vascular channel lined by swollen endothelial cells with a few intraluminal eosinophils and surrounded by a lymphoid infiltrate.

in the vascular lumen of the vessel walls and no infiltration of vessel walls by lymphocytes. There was no evidence of haemorrhage or necrosis and no significant mitotic activity in either the endothelial cells or in the lymphocytic component.

The diagnosis of ALHE was made from the above findings.

Discussion

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rare benign lesion which has had numerous other titles (Chang and Ch'en, 1962; Kawada *et al.*, 1966; Dannaker *et al.*, 1989). Kandii (1970) and Wells and Whimster (1969) were the first to call it ALHE.

ALHE was commonly confused with Kimura's disease but it is now considered to be a separate and distinct clinical pathological entity (Kung *et al.*, 1984; Googe *et al.*, 1987).

ALHE usually appears in Caucasian females in the third and fourth decades and is characterized clinically by unilateral asymptomatic superficial nodules with a predeliction for the head and neck region, often on the ear and pre-auricular area (Olsen and Helwig, 1985). In ALHE regional reactive hyperplastic lymphadenopathy is uncommon (19 per cent), and there is mild peripheral eosinophilia in 20 per cent (Olsen and Helwig, 1985). The histological features of ALHE are a vascular component with thick-walled vessels showing concentric rings of perivascular fibrosis and prominent swollen endothelial cells with cytoplasmic vacuolation, and an inflammatory component composed of mature small lymphocytes and variable numbers of eosinophils. The lymphocytic infiltrate in ALHE is T cell rich but follicles of B lymphocytes may be present.

Kimura's disease by contrast most commonly presents as a pre-auricular or salivary gland swelling which may be bilateral in young males, usually with associated regional lymphadenopathy and blood eosinophilia. Histologically Kimura's disease contains blood vessels and lymphocytes but by contrast with ALHE the blood vessels are thin-walled and do not show enlarged vacuolated endothelial cells. The eosinophilic infiltrate in Kimura's disease is more consistent and conspicuous than in ALHE. B cell lymphoid follicles are a consistent feature of Kimura's disease with infiltration of germinal centres by eosinophils (so called folliculolysis) which is not seen in ALHE (Chun and Ji, 1992).

ALHE may rarely involve extracutaneous sites including the nasal mucosa (Bosnic *et al.*, 1987), muscle (Saxe and Kahn, 1977), bone (Rosai, 1978), salivary glands (Goldman and Klen, 1976), tympanic membrane (Fitzgerald and Schmookler, 1996), oral mucosa (Bartralot *et al.*, 1996), orbit (Smith *et al.*, 1988), and lacrimal gland (Bartralot *et al.*, 1996).

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We are aware of no previous reported case of pharyngeal involvement.

The pathological process of ALHE has been the subject of speculation. Olsen and Helwig (1985) contended that the presence of arteriovenous shunts was the first event; others suggested that the arteriovenous shunts were related to a benign localized form of vasculitis (Buckerfield and Edwards, 1979) or an atopic reaction to a variety of agents (Hallam *et al.*, 1989; Akosa *et al.*, 1990). A further theory suggests that ALHE is a true vascular neoplasm perhaps representing a stage of histiocytoid or epithelioid haemangioma (Rosai *et al.*, 1979; Googe *et al.*, 1987; Urabe *et al.*, 1987).

Whatever the pathogenesis, ALHE is considered a benign process with no tendency to metastasize and there are even reported cases of spontaneous remission (Olsen and Helwig, 1985).

Adjuvant therapy following surgery or after local recurrence has been variously employed, including radiotherapy, cryotherapy, laser treatment, injection corticotherapy and cytotoxic drugs. In the case reported here no adjuvant therapy was given and at one year follow-up there was no evidence of recurrence.

In conclusion, we report a case of an unusual lesion of uncertain aetiology occurring at a previously unreported site.

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