ANGIOLYMPHOID HYPERPLASIA WITH EOSINOPHILIA OF THE ARM

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ABSTRACT

A case of angiolymphoid hyperplasia with eosinophilia (ALHE) was shown in a 25-year-old female patient. The lesion was at the arm. CT scan showed a soft tissue mass with density 41-51 H.U. at the fat plane of the medial aspect of the lower one third of the arm. The lesion had infiltrated border with encasement of the vessels. Faint homogeneous enhancement was observed in the lesion. Peripheral eosinophil was mildly elevated. This is the first reported case of ALHE at the arm by CT scan

INTRODUCTION

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rare condition of uncertain etiology (1). It is thought by some to be a neoplasm of epithelioid endothelial cells (2). There are wide racial differences in presentation, with oriental patients being predominantly male and young in contrast to western cases which tend to occur in females and older patients (3). It causes papular or nodular angiomatous lesions in the dermis, subcutaneous tissues and adjacent lymph nodes, which average 1 cm in diameter (4). Distribution is almost entirely restricted to the head and neck (5) and there seems to be a predilection for area around the external ear and external auditory canal (6,7,8). There are reports involving the orbit (9), lacrimal gland (10), oral mucosa (11) and the arm (12).

We present a case of ALHE in a young female patient at her right arm. The images were of CT scan.

CASE REPORT

A 25-year-old single female patient from Nontaburi province had a mass at right arm for 1 year. The mass has increased size slowly and was not tender. The pain at the mass was observed only when she overused her right arm. There was no previous history of trauma. The soft tissue mass was palpated at the medial aspect of right upper arm, size 6 X 12 cm. The physical examination otherwise was normal. Complete white cell count showed total WBC 12.61 X 10^3/ul, Neu 59, Lym 29, Mono 5, Eos 7. CT scan at the mass showed a soft tissue mass, (density=41-51 H.U.) in the fat plane of lower medial part of the distal one third of the arm. The mass had irregular border. Faint and homogeneous enhancement in the mass was seen. Multiple linear soft tissue density was seen around the mass (Fig. 1).

At surgery, the mass with fibrofatty component, size 3 x 5 x 10 cm. was seen. The mass had ill defined border, infiltrating around the cephalic vein and sensory branch of the musculocutaneous nerve. The mass adhered to the brachial artery below and skin above. The CT scan also showed the vascular encasement (Fig. 2).

The mass was dissected with preservation of the vasculature. At pathology, section of the soft tissue of the arm revealed angiolymphoid hyperplasia with eosinophilia. Section of the lymph nodes of the arm showed angiolymphoid hyperplasia with mild eosinophilia, clusters of atypical lymphoid cells. Section of the skin revealed nonspecific perivascular lymphoid cells infiltration in the dermis.

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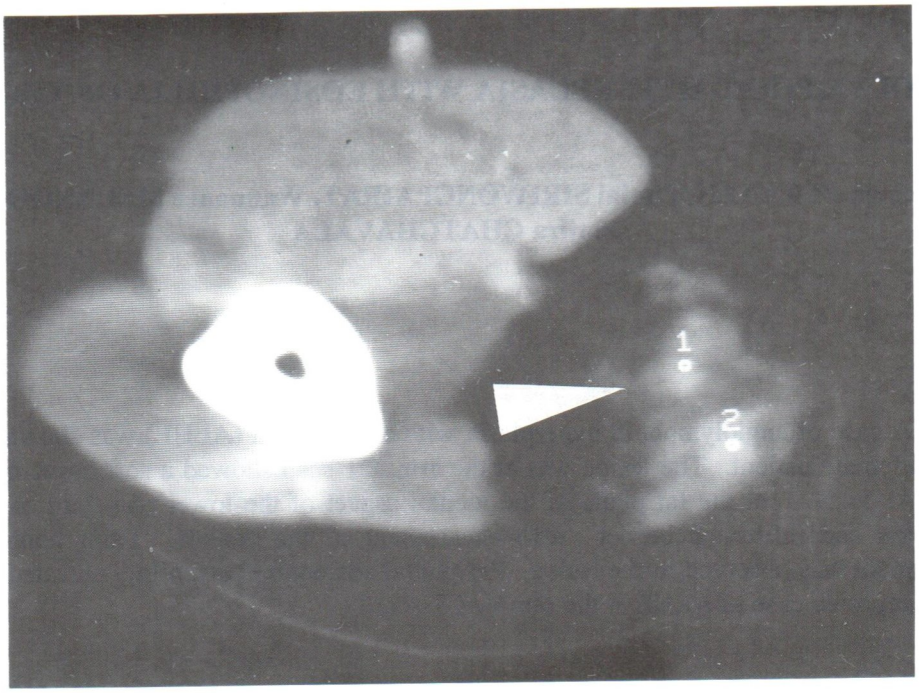

Fig. 1A. Non enhanced axial CT scan of the right arm showed an infiltrative border solid mass in the subcutaneous fat plane.

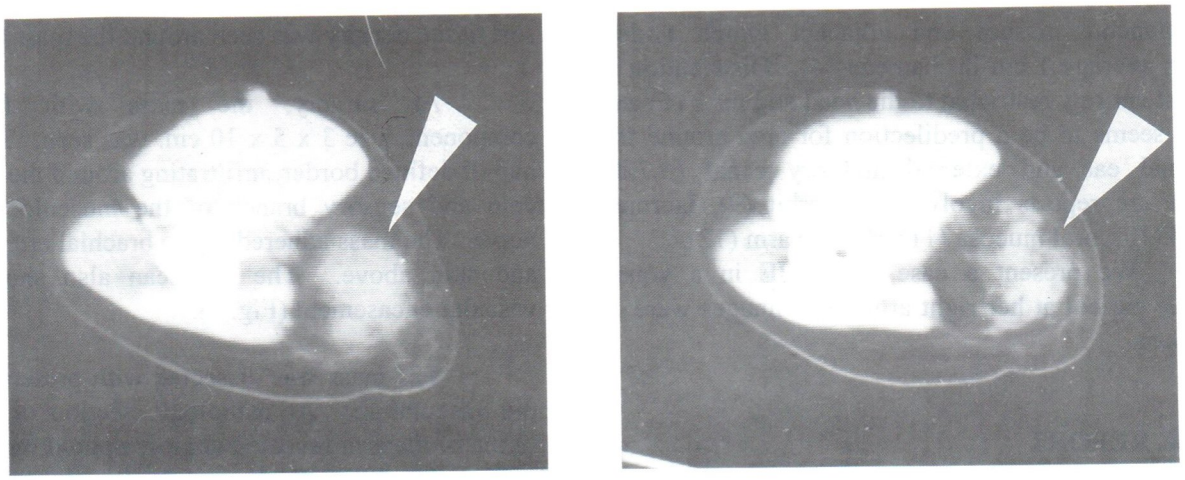

Fig. 1B Enhanced study showed faint homogenous enhancement in the lesion.

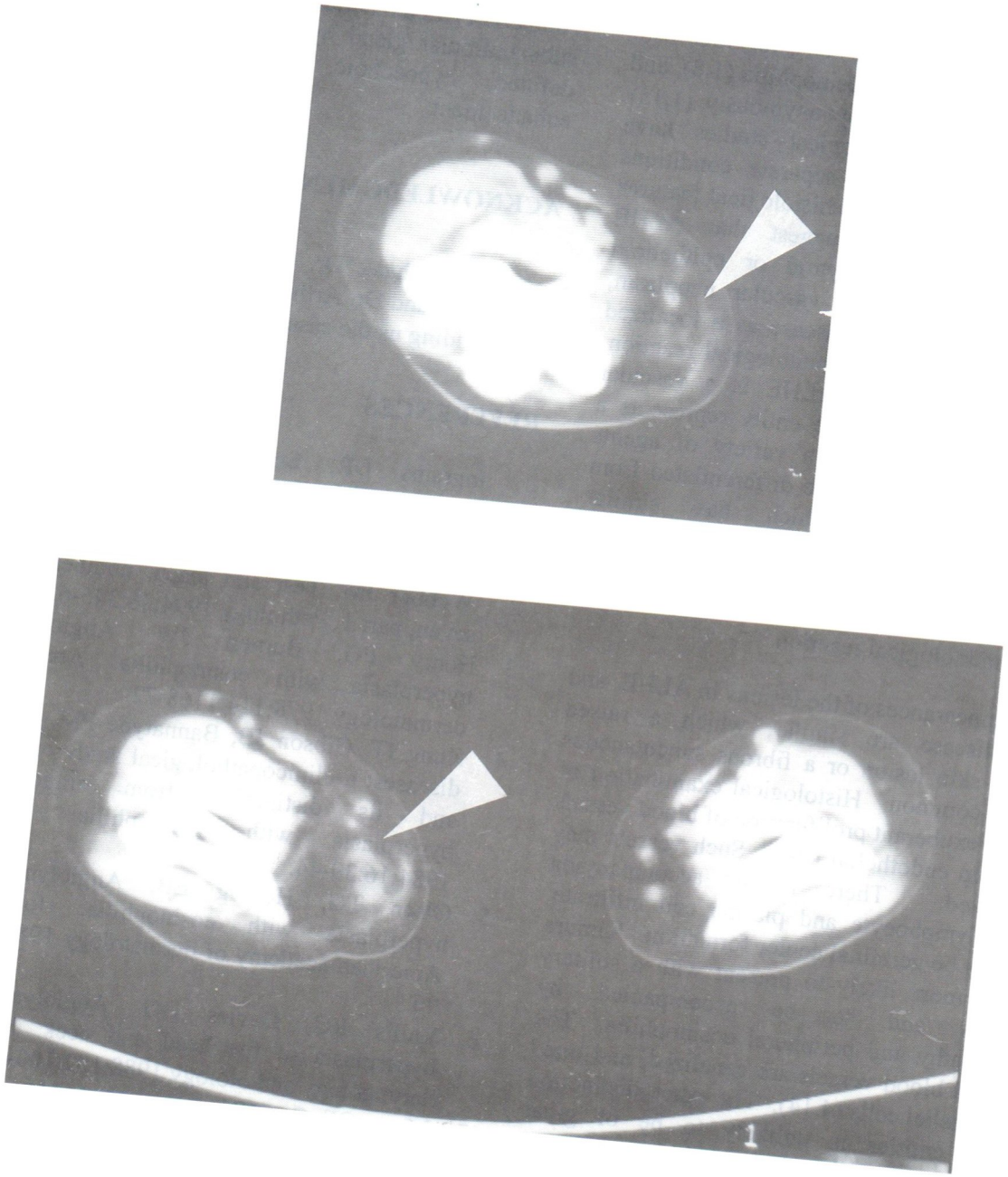

Fig. 2 Vascular encasement was also demonstrated by images, corresponding to the surgical findings.

DISCUSSION

Originally ALHE was thought to be equivalent to Kimura's diseases, a condition prevalent in Japan, China and South-east Asia which also causes angiomatous skin lesions, together with lymphadenopathy and marked eosinophilia (1,8), and the terms have often been used synonymously (1,15). However recent clinicopathological studies have suggested that these are two separate conditions which have different clinical and histological features (1,14,15). These authors suggest that ALHE represents a stage of histiocytoid or epithelioid hemangioma, which is a true vascular neoplasm, whereas they view Kimura's disease as a localized manifestation of a systemic immunological reaction. Other authors disagree that ALHE is a vascular neoplasm; they believe that the entity represents a localized atrophic reaction to a variety of agents (12,16). It therefore needs to be differentiated from epithelioid hemangioma, which has similar histological features to ALHE regarding endothelial cell morphology, but is without the eosinophilic infiltrate or formation of germinal centers which suggest an immunological reaction.

The appearances of the lesions in ALHE and Kimura's disease are similar, which a raised erythematous skin lesion or a fibrous subcutaneous nodule being common. Histological examination of ALHE shows exuberant proliferation of blood vessels lined by plump endothelial cells. Such vessels may be uncanalized. There is an accompanying perivascular lymphocyte and plasma cell infiltrate, and there may be germinal center formation. Kimura 's disease is more likely to present with a solitary larger mass and to be accompanied by lymphadenopathy and peripheral eosinophilia. The newly formed blood vessels are canalized, and lined by flat endothelial cells (4,14). The exact distinction of the two entities is unlikely to be of great importance, as these diseases probably represent a similar reactive process in the tissue, with the minor histological differences depending on whether the insult is localized or systemic.

ALHE is benign, and may regress spontaneously, but the majority of masses persist as slow growing tumours (17). There are no report of malignant change. The appearances of the lesion have led it to be mistaken for Kaposi's sarcoma, malignant lymphoma and angiosarcoma, as well as pyogenic granuloma, hemangioma and dermatofibroma (18,19).

Imaging study of ALHE was not reported before, according to our knowledge. Smith (20) reported CT of Kimura disease of the parotid gland. Ahuja (21) demonstrated Kimura's disease of the submandibular gland by ultrasonography as a well defined, hypoechoic, oval mass with distal enhancement.

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