Angiolymphoid hyperplasia with eosinophilia involving the lacrimal gland: case report

H T COOK¹ and N D STAFFORD²

From the Departments of ¹Histopathology, and ²Otologygology, St Mary's Hospital, London

SUMMARY A case is described of a 25-year-old male who developed angiolymphoid hyperplasia with eosinophilia involving the tissue over the left parotid gland and subsequently involving the right lacrimal gland with proptosis. Treatment was by excision.

Angiolymphoid hyperplasia with eosinophilia (ALHE) is an uncommon lesion which presents as one or more nodules involving the dermis or subcutaneous tissues mainly of the head or neck but occasionally at other sites. It is characterised by lymphoid aggregates with germinal centres, tissue eosinophilia, vascular proliferation with typical plump endothelial cells, and variable fibrosis. In some cases blood eosinophilia is present. We report a case in which the initial presentation was with a swelling over one parotid gland with subsequent involvement of the contralateral lacrimal gland.

Case report

A 25-year-old male of Asian descent, but resident in the UK since birth, presented with a three-month history of a soft diffuse swelling in the region of the left parotid gland. The swelling was painless, and there were no other abnormalities on clinical examination of the head or neck. A parotid sialogram was normal, and a diagnosis of a lipoma overlying the parotid gland was made. The swelling slowly enlarged over the following 10 months, and therefore a superficial parotidectomy was undertaken. By this time the patient had begun to develop slight swelling in the region of the right lacrimal gland. A differential white blood cell count at this time showed 66% neutrophils, 23% lymphocytes, and 11% eosinophils.

Over the next nine months the swelling above the right eye increased and a marked proptosis developed (Fig. 1). There was no ophthalmoplegia, but the patient experienced considerable discomfort. A CT scan demonstrated the lacrimal gland swelling and also swelling of the medial, lateral, and inferior rectus muscles of the right orbit (Fig. 2). Excision of the lacrimal gland was carried out. At operation it

Fig. 1 There is downward displacement of the globe by swelling in the lacrimal region.

Fig. 2 CT scan demonstrating mild proptosis and a mass above and lateral to the right globe.

Correspondence to Dr H T Cook, Department of Histopathology, St Mary's Hospital, Praed Street, London W2 1NY.
Angiolymphoid hyperplasia with eosinophilia

was found to be infiltrated by a mass arising from its anterior part. Some of the apparently normal gland was left in order to facilitate tear production, and the operation was followed by rapid resolution of the proptosis and associated symptoms.

The patient was last seen 10 months after his second operation and had no evidence of any other swellings, either in the head and neck region or elsewhere over the body.

PATHOLOGY
The specimen from the parotid region consisted of part of the parotid gland and superficial fibrous tissue measuring 9.0×4.5×1.5 cm. The lacrimal gland specimen was two pieces of light brown tissue measuring in total 3.0×2.0×0.6 cm. Histologically the two specimens showed similar features which in the parotid specimen were predominantly in the tissue superficial to the parotid gland, while the deeper part of the gland was unaffected. In the second specimen the changes were seen within the lacrimal gland with extension into the surrounding connective tissue. In both specimens there was marked fibrosis and prominent lymphoid follicles, most of which contained germinal centres (Fig. 3). Within the fibrous stroma were many lymphocytes, eosinophils, and mast cells and con-

Fig. 3 Photomicrograph of lacrimal gland showing fibrosis and prominent lymphoid follicles with germinal centre formation. Haematoxylin and eosin, ×45.

Fig. 4 Photomicrograph of an area of vascular proliferation within the lacrimal gland. Note the vessels lined by plump endothelial cells (arrows) and the lymphocytic infiltration. Haematoxylin and eosin, ×190.
siderable capillary proliferation. Many of these pro-
lerating vessels were lined by plump endothelial
cells with large vesicular nuclei (Fig. 4). In occasional
areas cells of this type were forming small aggregates
with no apparent lumen. The appearances in both
specimens were those of angiolymphoid hyperplasia
with eosinophilia.

Discussion

ALHE was first reported in 1969.2 It is closely related
to Kimura’s disease,3 an entity which has been mainly
described in China and Japan, and it has been argued
that Kimura’s disease is part of the spectrum of
ALHE.145 Others, however, have emphasised the
differences, pointing out that Kimura’s disease has
different clinical and pathological characteristics.1 In
particular Kimura’s disease is said to affect predomi-
nantly young males with a high incidence of blood
eosinophilia and histologically to show more fibrosis
and fewer distinctive endothelial cells. It seems
probable that the degree of fibrosis may depend on
the duration of the disease before biopsy.

In a report of 116 patients with ALHE submitted to
the Armed Forces Institute of Pathology1 the
anatomical sites of the lesions were face and scalp in
100, trunk 4, extremities 12, and miscellaneous 2. In
only one case was the diagnosis made preoperatively.
Hidayat et al.4 have reported eight cases of ALHE
involving the orbit; in none of these cases was the
malignant to involve the lacrimal gland. Eisenberg
and Lowlicht4 have reported a case similar to ours in
a 34-year-old female who presented with ALHE
involving the right cheek and subsequently
developed lacrimal gland involvement.

The consensus is that ALHE is a reactive inflam-
matory process, and the presence of eosinophils and
occasional blood eosinophilia has led to speculation
that it represents in an infectious or allergic process
or a response to parasitic infection. No parasites or
micro-organisms have ever been described within the
lesions. Fernandez et al.7 have demonstrated the
presence of renin-containing cells in some cases of
ALHE and speculate that it may have an angiogenic
role. Staining with a polyclonal antibody to human
renin was negative in the present case.

ALHE can be successfully treated by local
surgery,8 though complete excision may be difficult.4
In addition radiation therapy has been effective4 and
intralesional injections of corticosteroids have been
successful in some cases.

Professor E Wilson Jones and Dr R W Cox are thanked for advice on the
histology of this case.

References
1 Olsen TG, Helwig EB. Angiolymphoid hyperplasia with eosino-
philia: a clinicopathological study of 116 patients. J Am Acad
2 Wells GC, Whimster I W. Subcutaneous angiolymphoid hyper-
3 Kimura T, Yoshimura S, Ishikawa E. On the unusual granulation
combined with hyperplastic changes of lymphatic tissue. Trans
4 Eisenberg E, Lowlicht R. Angiolymphoid hyperplasia with
5 Reed RJ, Terazakis N. Subcutaneous angiolymphoid hyperplasia
6 Hidayat AA, Cameron JD, Font RL, Zimmerman LE. Angio-
lymphoid hyperplasia with eosinophilia (Kimura’s disease) of the
7 Fernandez LA, Olsen TG, Barwick KW, Sanders M, Kaliszewski
C, Inagami T. Renin in angiolymphoid hyperplasia with eosino-
8 Tham K-T, Leung P-C, Saw D, Gwi E. Kimura’s disease with
9 Iguchi Y, Inoue T, Shimono M, Yamamura T, Shigematsu T,
Takahashi S. Kimura’s disease and its relation to angiolymphoid
hyperplasia with eosinophilia: report of three cases and review of

Accepted for publication 2 July 1987.
Angiolympoid hyperplasia with eosinophilia involving the lacrimal gland: case report.
H T Cook and N D Stafford

*Br J Ophthalmo* 1988 72: 710-712
doi: 10.1136/bjo.72.9.710

Updated information and services can be found at:
http://bjo.bmj.com/content/72/9/710

**Email alerting service**
Receive free email alerts when new articles cite this article. Sign up in the box at the top right corner of the online article.

**Notes**

To request permissions go to:
http://group.bmj.com/group/rights-licensing/permissions

To order reprints go to:
http://journals.bmj.com/cgi/reprintform

To subscribe to BMJ go to:
http://group.bmj.com/subscribe/