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View from Beneath: Pathology in Focus

Cystic lymphangioma in the adult parotid

J. R. Livesey, M.A., F.R.C.S., F.R.C.S.Ed., J. V. Soames, Ph.D., F.D.S., F.R.C.Path. (Newcastle upon Tyne)

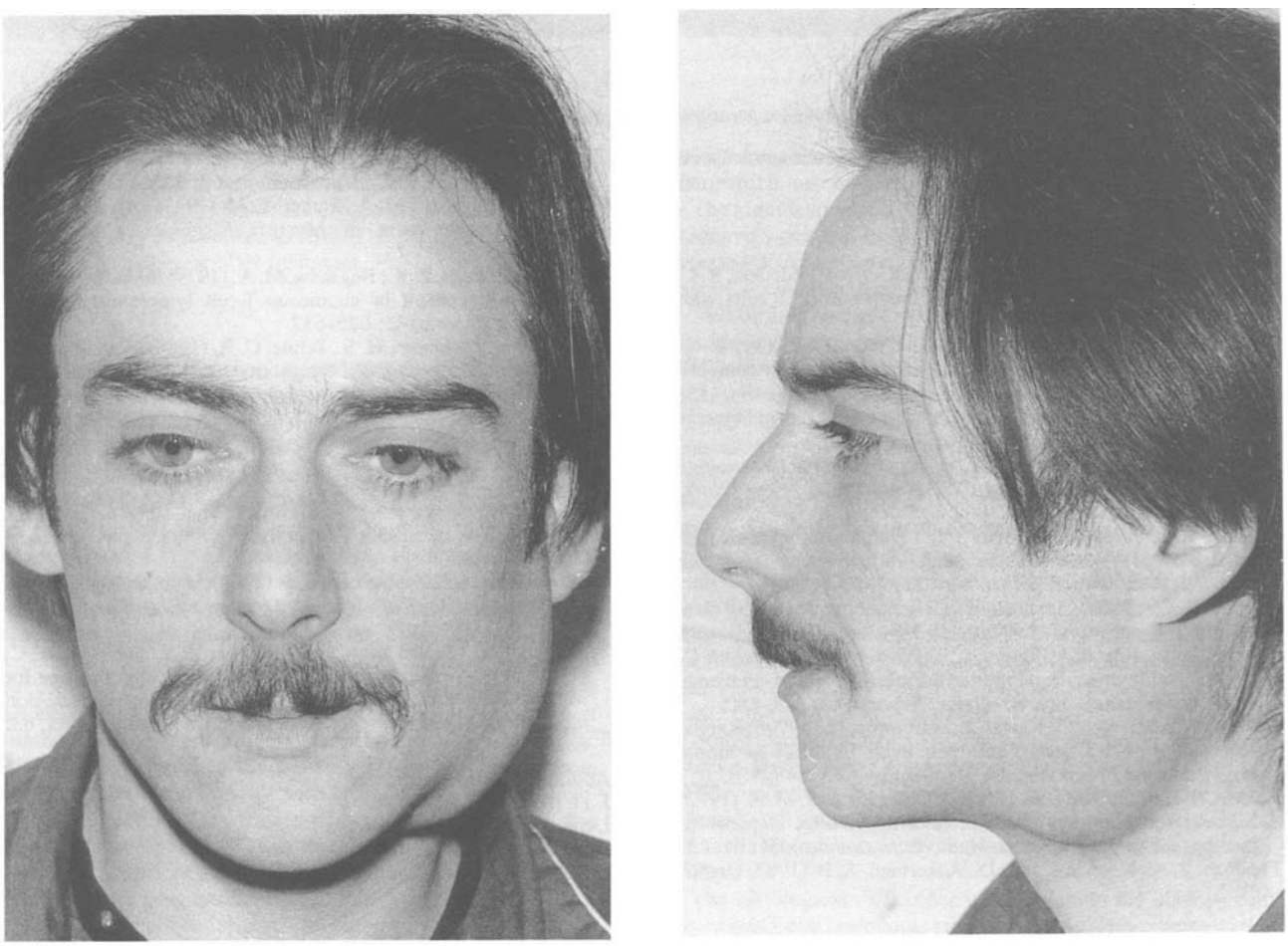
Abstract
Cystic lymphangioma is a congenital lesion which rarely presents in adult life and even less commonly arises within salivary tissue. We report the sudden appearance of a large cystic lymphangioma within the parotid gland of a young man and discuss the management of this rare condition.

Introduction

A cystic lymphangioma or hygroma is a congenital hamartomatous malformation arising from the same primordia as normal lymph vessels. Found in different tissues these benign lesions occur frequently in the head and neck region, although very rarely within the parotid (Kornblut *et al.*, 1973). Lymphangioma are usually noticed at or shortly after birth (Batsakis, 1979). We report a case in an adult which appeared suddenly in the region of the parotid gland.

Case report

A 26-year-old Causcasian male presented with a one week's history of a progressive non-tender swelling in the region of the left parotid gland. There was no precipitating cause and no



(a) (b)

Fig. 1
The fluctuant non-tender swelling in the region of the left parotid measured 10 × 6 cm.

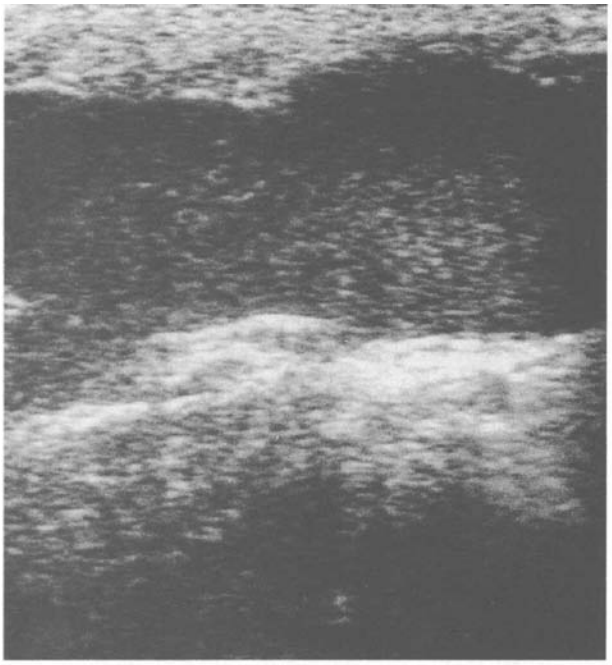


Fig. 2

Ultrasonography showing a homogeneous hypoechoic large soft tissue mass in the region of the left parotid.

change associated with eating. Aged three he had a kick to the left side of his face and subsequently had always been aware of a small firm, non-tender mass in the region of the left angle of the mandible. He had no other past nor ongoing medical history of note.

On examination, a fluctuant non-tender swelling measuring 10 × 6 cm was noted in the region of the left parotid (Fig. 1). The overlying skin was normal and the swelling did not transilluminate. There was no palpable left submandibular mass nor associated lymphadenopathy and the oral cavity and parotid duct were unremarkable. Clinically this was thought to be cystic and fine needle aspiration produced a small amount of blood-stained fluid which was reported to show heavy blood-staining with some macrophages, plasma cells, polymorphs and a large number of lymphocytes. Epithelial cells were absent and the sample was felt to be inconclusive.

An ultrasound scan showed a homogeneous, hypoechoic, large, soft tissue mass in the region of the left parotid (Fig. 2). The margins were lobulated with extensions into the surrounding soft tissue planes. A small linear anechoic area was visible behind the ear and adjacent to the postero-inferior aspect of the main mass. The parotid gland was not visible, being either replaced by the mass or grossly displaced. The radiologist thought the lesion was probably a tumour, either from a parotid primary or a lymph node mass.

At surgical exploration, a multi-loculated cyst filled with altered blood was found lying within the parotid gland both deep and superficial to the plane of the facial nerve. A meticulous excision with preservation of the facial nerve was performed and a satisfactory post-operative recovery was made. The patient continues to attend for review in Out Patients.

Histopathological examination showed the parotid gland permeated by numerous thin walled and cystically dilated, endothelial lined, vascular channels consistent with a diffuse cystic lymphangioma. Some of the channels contained organized thrombus and there was widespread interstitial inflammation with abundant neutrophils suggesting recent infection of the lesion which may have brought it to the patient's attention. There was no evidence of malignancy (Fig. 3).

Discussion

Traditionally lymphangioma has been classified as capillary, cavernous and cystic. However, a unified concept relating morphological differences to anatomical locations has been proposed (Bill and Sumner, 1965). The smaller lymphatic lesions occur in the lips, cheek and tongue whilst cystic lymphangiomas occur in regions in which there are clearly definable tissue planes or loose areolar tissue permitting the expansion of the endothelial lined spaces (Batsakis, 1979). In all cases lymphangiomas insinuate throughout tissues and around vital structures.

Cystic lymphangiomas are more common in the posterior triangle of the neck and tend to reach up into the cheek and parotid region or down towards the mediastinum or axilla. Large lymphangiomas may extend into the anterior triangle and even across the mid-line. The natural history of cervical lymphangiomas varies according to their histological configuration and may be progressive, static or intermittent in growth whilst regression or spontaneous disappearance may occur. Usually they grow slowly with the infant, thus only becoming gradually prominent (Batsakis, 1979).

Cystic lymphangiomas are usually a honeycomb of multiple loculated, dilated, lymphatic channels lined by a single layer of flattened endothelium. They are compressible, lie deep to platysma and infiltrate tissues causing compression or stretching. The overlying skin is usually normal. Rupture of one locule can cause collapse of the whole system at the time of surgery. The fluid content is usually watery, serous, clear or straw coloured. Occasionally chylous, the fluid does not coagulate and contains cholesterol, leucocytes and lymphocytes (Batsakis, 1979). Transillumination is usually very good except following haemorrhage or infection when the fluid becomes discoloured (Kornblut *et al.*, 1973). Lymphangiomas are extremely rare intra-salivary lesions, more frequently salivary glands are incorporated by lymphangiomas of adjacent tissue though the parotid is the commonest site for such intra salivary lesions when they do occur (Kornblut *et al.*, 1973).

This congenital lesion usually presents at birth although most have appeared by the second year of life. Apart from appearance, the signs and symptoms associated with lymphangioma include: stridor with cyanosis due to compression or mediastinal extension, branchial plexus compression with pain or hyperaesthesia (Batsakis, 1979) or rapid changes in size due to haemorrhage within the lesion or supervening infection, which may be fatal (Ward *et al.*, 1970). When presenting at a later age a lymphangioma may become evident only after trauma (Ward *et al.*, 1970) and this as well as infection may bave been relevant in this case.

The diagnosis of lymphangioma relies upon the signs, symptoms and fine needle aspiration although ultrasonography is useful in demonstrating the internal structure and extent. Lymphangiomas appear as large anechoic communicating cavities whilst haemangiomas have hypoechoic areas separated by

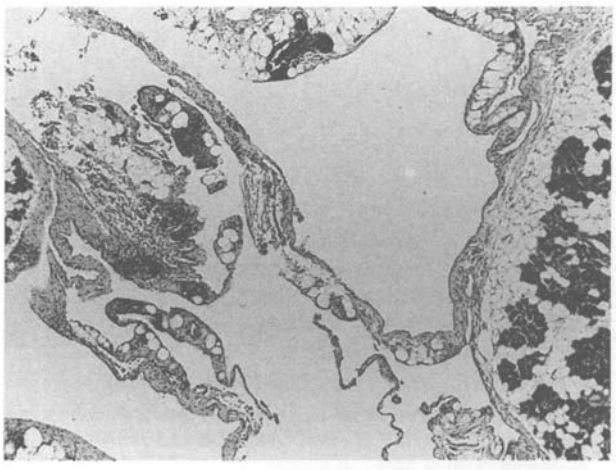


Fig.3

Cavernous and cystically dilated lymphatic channels within the parotid gland.

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thin hyperechoic septa (Betti *et al.*, 1990). In this patient, the small linear anechoic area behind the ear probably represented a portion of the cystic lymphangioma which did not communicate freely with the main part and thus still contained clear fluid. The main part of the system contained heavily blood-stained fluid which gave the hypoechoic appearance whose interpretation did not confirm the clinical impression of a cystic lymphangioma.

Surgery is the treatment of choice for a lymphangioma within the parotid and for confirming the histology. However, a subtotal excision may be necessary to preserve structures of functional or cosmetic importance *i.e.* the facial nerve (Lack and Upton, 1988) and therefore revision surgery may be required particularly in children. Cellulitis has been reported from residual lymphangiomatous lesions following incomplete excision (Bill and Sumner, 1965). Radiotherapy should not be used since lymphangiomas are radio resistant and irradiation can cause maldevelopment of the jaws and hypodontia (Gingrass and Gingrass, 1971), in addition to the potential for malignant induction. Sclerosing solutions are also ineffective (Bauer and Hardman, 1976).

Long-term follow-up for this rare condition in adults is important for management of any residual disease.

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Key words: Lymphangioma, cystic; Parotid gland

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Address for correspondence:
J. R. Livesey,
ENT Department,
Freeman Hospital,
Newcastle upon Tyne,
NE7 7DN.