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Inflammatory pseudotumour in the submandibular region

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Abstract
We present a rare case of inflammatory pseudotumour in the submandibular region. A review of the literature revealed that this is only the second case of inflammatory pseudotumour in the submandibular region to be reported. Clinical presentation and management of this condition are discussed.

Key words: Plasma Cell Granuloma; Submandibular Gland

Introduction

Inflammatory pseudotumour is a pathological term that has been used to describe an inflammatory fibrosing tumour-forming process that involves a variety of organs in the body.[1–5] It is a non-neoplastic lesion and is rarely seen in the major salivary glands.[6] The aetiology and pathogenesis of these lesions remain controversial and it is generally accepted that they are secondary to an unusual tissue response to injury. The clinical presentation and management of an inflammatory pseudotumour in the submandibular region are discussed.

Case report

A 72-year-old man presented with a three-month history of a rapidly enlarging swelling in the left submandibular region. This caused mild discomfort but he was otherwise symptom-free. At presentation there was an 8 × 5 cm hard and fixed swelling in the left submandibular region. The skin over the swelling was indurated and hyperaemic. Examination of the ears, nose and throat were normal. Jaw movements were normal. There was no other cervical lymphadenopathy. The patient was afebrile and had a normal white cell count. He gave a past medical history of a ventricular aneurysm with an endocardial thrombus and was on oral anticoagulants. The patient underwent sialography, an ultrasound scan and computed tomography (CT) scan of the neck. The sialogram showed no obstructing calculus. The ultrasound scan showed the lesion to be a solid mass invoving the left submandibular gland. CT scan showed an enlarged left submandibular gland with some residual contrast and the swelling was not confined to the gland alone. There was haziness of the subdermal fat overlying the submandibular region. Overall the appearances on the left suggested an inflammatory rather than a malignant aetiology.

In view of the induration and skin involvement an infective element was thought to be present. The patient was therefore admitted and treated with IV cefuroxime and IV metronidazole. Over the next few days the swelling decreased in size but a hard ill-defined swelling was still palpable. The clinical picture was very suggestive of a malignant lesion. Fine needle aspiration cytology showed a picture of chronic inflammatory cells. In view of the patient's poor medical status and the opinion that if the mass were malignant, it would be inoperable, an ultrasound guided trucut needle biopsy was performed. Histology was in keeping with inflammatory pseudotumour. In view of the patient's poor anaesthetic risk a conservative line of management was adopted. The swelling has slowly regressed over a period of two months and is no longer discretely palpable except for a smooth submandibular gland. A repeat CT scan wsa normal. The patient has been followed up for the last two years and there has been no evidence of recurrence.

Pathology

We received two needle cores up to 8 mm in length and each showed similar histopathological features. At one pole salivary parenchyma was separated by a thin fibrous pseudocapsule from a central inflammatory mass. This central zone was composed of bland spindle shaped cells in a loose collagenous stroma interspersed with a xanthomatous and lymphoplasmacytic inflammatory response. Immunohistochemical studies showed the spindle cells within this lesion to stain positively with vimentin and smooth muscle actin and negatively for desmin and cytokeratin. This conforms with the histochemical profile described in previous papers.[5]

Discussion

Inflammatory pseudotumour is a tumour-like lesion of unknown aetiology. Varying terminology has been used to describe this non-neoplastic lesion, such as plasma cell granuloma, xanthomatous pseudotumour, post-inflammatory pseudotumour, fibrous xanthoma and inflammatory myofibroblastic tumour.[5] The preferred pathological term is inflammatory pseudotumour.[5] The common sites of presentation are in the lungs, lymph nodes, liver, spleen and breast.[1–5] It rarely occurs in major salivary glands. However, most reported cases in this area are in the parotid gland.[5] There is one reported case in the submandibular region.[6]

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Weisman and Osguthorpe reported that orbital CT, MRI and ultrasound helped in differentiating inflammatory pseudotumour from inflammatory and malignant lesions around the orbit. Many other reports suggest that CT and MRI are unable to differentiate between pseudotumour and a malignant tumour.[6,7] In our case the CT findings were highly suggestive of an inflammatory lesion. Davis *et al*. have discussed the differentiation of inflammatory pseudotumour from other proliferative neoplastic and inflammatory lesions.[1] Williams *et al*. have applied similar considerations to major salivary glands.[5] The differential diagnosis includes obstructive sialadenitis, non-specific sialadenitis and myoepithelioma.[5]

Obstructive and non specific sialadenitis are characterized by ductal lithiasis and squamous metaplasia, fibrosis, and preservation of lobular architecture. Myoepithelioma and spindle cell epithelial neoplasms are usually encapsulated and are more homogeneous, with virtually no signs of inflammation. Myoepitheliomas are also differentiated by immunostaining, by the presence of cytokeratin S-100 protein and the smooth muscle actin phenotype.[5]

In none of the series has a neoplastic change been noticed but there is always a risk of masked malignancy. In our case the swelling has regressed in size and there are no obvious clinical signs of malignant transformation. The changes observed are all secondary to a functional adapative transformation by the phagocytic cells.[5] A specific cause is yet to be found, but whatever the cause a localised derangement in immune response after the initial injury may represent the underlying mechanism.

Inflammatory pseudotumours are rarely associated with recurrence.[5] A few cases have been aggressive, and patient death has been reported with liver inflammatory pseudotumour.[7]

Treatment options include surgical excision, radiotherapy and steroids. Most cases are treated by surgical excision in view of the misdiagnoses of malignancy. In the above case surgical excision was not embarked on as the tumour was thought to be inoperable even if malignant with regard to the patient's poor general health. We have not proceeded with radiotherapy as the lesion has regressed in size. The patient's poor general condition and the ensuing conservative line of management that has been adopted have given us an opportunity to watch the course of the disease.

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