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Address for correspondence

Dr. Dipak Ranjan Nayak MS, FUICC, FICS
Professor & Head of unit II,
Department of Otolaryngology - Head & Neck Surgery,
Kasturba Medical College & Hospital,
Manipal 576104, Karnataka, India.
email: drment@rediffmail.com
Phone: 0820- 2922143 (o)
Fax No: 0820-2570061

Short Communications

A RARE CASE OF INFLAMMATORY PSEUDOTUMOUR OF THE SUBMANDIBULAR LYMPHNODE.

Anjana Trivedi,* Jitendra Patel.* *

Abstract: Inflammatory pseudotumour (IPT), also known as inflammatory myofibroblastic tumour, is a rare cause of benign cervical lymphadenopathy. In its acute clinical presentation it mimics malignant and infective causes of cervical lymphadenopathy. The combined clinical history, histopathological findings, and imaging features of an infiltrating soft-tissue mass should lead to the possibility of IPT. Radical surgery should be avoided before there is histological proof of a malignant tumor.

Keywords: Inflammatory pseudotumour, submandibular lymphnode, radio-patho findings, steroids.

INTRODUCTION

Inflammatory pseudotumor (IPT) is a rarely occurring lesion. The condition was first described in 1905 by Birch-Hirschfeld. Because an inflammatory pseudotumor has an abundance of polyclonal plasma cells, its etiology (postinflammatory vs immunologic) is still being debated. The term pseudotumor was coined because these lesions mimic expansive, invasive, malignant tumors, both clinically and radiologically. IPT most commonly involves the lung and orbit. It is rarely found in the head and neck outside the orbits. The condition may mimic a malignant lesion, and may occur at sites that hinder or preclude biopsy, possibly exposing the patient to the risk of unnecessary and potentially mutilating surgery. The combined clinical history, histopathological and radiological features of a soft-tissue mass can be helpful in the diagnosis of IPT. In confirmed cases of IPT, conservative steroid therapy may cure the disease.

CASE REPORT

An 20 year old female patient was admitted with a 5 months history of swelling of the left submandibular region. Clinically it was like a submandibular gland tumour. CT scan of neck showed a poorly defined, enhancing soft-tissue mass lying against the enlarged and enhancing left submandibular salivary gland, extending into the masticator space (Fig 1). An MR study confirmed the mass lesion in the right masticator space, which appeared slightly hyperintense relative to muscle on T2-weighted SE images, and clearly enhancing on postcontrast T1-weighted SE images. The differential diagnosis included neoplastic lesion of submandibular lymphnode with inflammation of the submandibular salivary gland. FNAC revealed reactive inflammatory tissue with fibrous tissue and inflammatory cells. A lymphoid infiltrate and multiple vascular structures were recognized. No malignant cells seen. Higher antibiotic (Tab.

Fig 1. Axial contrast-enhanced CT scan shows a poorly defined, enhancing soft-tissue mass (L symbol) around an enlarged right submandibular salivary gland (S symbol).

Fig.2 Microphotograph showing a fibrous background with spindle (myo)fibroblast like cells admixed with a lymphoplasmocytic infiltrate (H and E, magnification x325).

Cephalexin 375 mg twice a day) and anti-inflammatory (Ibuprofen and Paracetamol combination thrice a day) drugs were given for 7 days. Three months later, a follow-up CT study showed no changes. Because a neoplastic lesion could not be excluded with certainty, partial excision of the mass was performed. Histopathological examination showed a fibrous background with spindle (myo)fibroblast like cells admixed with a lymphoplasmocytic infiltrate (Fig 2). The patient's clinical condition gradually improved with steroid treatment. Five months after this treatment, the patient is asymptomatic.

DISCUSSION

IPT was first described in the lung. It is most commonly found in the orbits. Submandibular lymphnodes are uncommon locations for an inflammatory pseudotumor. Even so, isolated cases have been reported in the parapharyngeal space and maxillary sinuses^[1]. One third of patients present with asymptomatic nodal enlargement. The rest have pain or constitutional symptoms such as fever and weight loss^[2]. An inflammatory pseudotumor can be defined as a localized mass made up of a fibrous stroma and chronic inflammatory infiltrates with a predominance of plasma cells or histiocytes and an absence of anaplasia and mitotic figures^[3]. Inflammatory pseudotumour of the soft tissues is a benign and self-limiting condition which although rare is important because it mimics neoplasia clinically.

Several triggers for its development have been described, including smoking, minor trauma, post-adenoidectomy and infection, however its exact aetiology is not known^[4]. It is suggested that inflammatory mediators such as cytokines and interleukin-1 are released in response to an insult, causing proliferation of fibroblasts, leaky and procoagulant endothelium and extravasation of polymorphous cellular infiltrate into the extracellular space^[2]. The key histological findings in establishing the diagnosis of IPT are the co-existence of variable numbers of inflammatory cells and spindle cells, consisting of fibroblasts and myofibroblasts and with varying degrees of

fibrosis. The wide spectrum of histological appearances has led to a number of terms for this condition being used synonymously, including plasma cell granuloma, inflammatory myofibroblastic tumour, inflammatory myofibrohistiocytic proliferation and inflammatory fibromyxoid tumour. The imaging appearances in this patient were of confluent enlargement of the lymph nodes, peripheral rim enhancement and multiple areas of central cavitation corresponding to necrosis seen on histology^[5]. The radiological differential of confluent cavitating lymphadenopathy includes metastatic spread from a head and neck squamous primary neoplasm, tuberculosis and atypical mycobacterial infection, other granulomatous diseases and rarer causes such as Kikuchi's disease. It is important to make an early histological diagnosis, not only to exclude malignant and other treatable causes, but also to start early treatment with steroids. A dramatic response to steroids has been demonstrated for acute IPT occurring in extranodal head and neck locations^[6]. Similar to cases of orbital pseudotumor, long term systemic steroid therapy proved effective in the management of non-orbital pseudotumor^[7].

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Address for correspondence:

Dr. Jitendra Patel,
Raj ENT Hospital,
20/25, New Jaganath, "Pratik",
Rajkot: 360001, Gujarat.
Phone: (Hosp.) 0281-2466797
Mobile: 09824224272
E-mail: doctorjitendra@yahoo.com