

Idiopathic necrotizing sialometaplasia of parotid gland

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ABSTRACT

Necrotizing sialometaplasia (NS) is an uncommon non-neoplastic, self-limiting inflammatory condition of the salivary glands. NS of major salivary glands is rare and simulates malignancy. If it is seen at this location, most of the cases are due to ischemia caused by vessel injury secondary to previous dental procedure or parotid gland surgery. We present a case of a parotid swelling that appeared as Warthin tumor on fine needle aspiration cytology (FNAC). On histology it turned out to be NS of parotid gland. The well known etiologies were absent in this case and hence it was labeled as idiopathic.

Key words: Necrotizing sialometaplasia, parotid, salivary gland

INTRODUCTION

Necrotizing sialometaplasia (NS) is an uncommon, non-neoplastic, self limiting inflammatory condition of the salivary glands. NS was first reported in 1973 by Abrams *et al.*^[1] as a reactive necrotizing inflammatory process involving minor salivary gland of the hard palate. In the World Health Organization (WHO) classification of salivary gland tumors, NS is classified under the group of tumor-like lesions. This condition shows a white predominance and could occur in any age group ranging from 17 to 80 years with a mean age of 50 years in men and 36 years in women with a male predominance of 2-1.^[2] Major salivary glands have been rarely reported as sites of NS, a disease primarily affecting minor salivary tissue, particularly that of the palate (80%).^[3] The clinical and histopathologic features of NS often simulate those of malignancies such as squamous cell carcinoma or salivary gland malignancy like mucoepidermoid carcinoma.^[4] Familiarity with NS and correct diagnosis are paramount in avoiding misdiagnosis

and inappropriate treatment. Ischemia of salivary gland tissue leading to infarction (trauma) is the most likely cause.^[4]

CASE REPORT

A 54 year old man presented with a history of progressively increasing swelling in pre-auricular region for the past 6-7 years. He developed pain in the swelling for the past two months only. On examination the parotid mass was firm and mildly tender. A clinical diagnosis of parotid tumor was kept and a fine needle aspiration cytology (FNAC) was conducted. FNAC showed abundant lymphocytes with few polygonal cells with dense eosinophilic cytoplasm. A cytological diagnosis of Warthin tumor was given. Patient was operated. Intraoperatively abscess was drained from the mass and superficial parotidectomy was done. The excised mass was sent for histopathological examination. A tan-brown colored mass measuring 4 × 3 × 1.5 cm was received. Cut section showed cystic space filled with necrotic material. Whole tissue was embedded. Microscopy showed coagulative necrosis of salivary gland lobules. There were cystic spaces lined by metaplastic squamous cells. Nests of squamous cells were also seen interspersed in the necrotic areas. The connective tissue frame work of necrotic glands was intact thereby preserving the lobular architecture of the salivary glands. Adjacent to the necrotic lobules were normal appearing salivary gland lobules. There were areas of fibrosis infiltrated by lymphoplasmacytic infiltrate with lymphoid follicles [Figures 1 and 2]. A diagnosis of NS-parotid was given.

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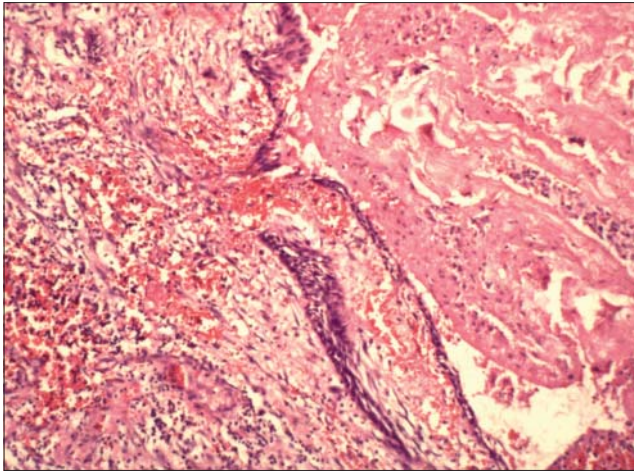


Figure 1: Nests of metaplastic squamous cells and a small cystic space lined by squamous epithelial cells. Surrounding them are areas of coagulative necrosis (H and E, $\times 100$)

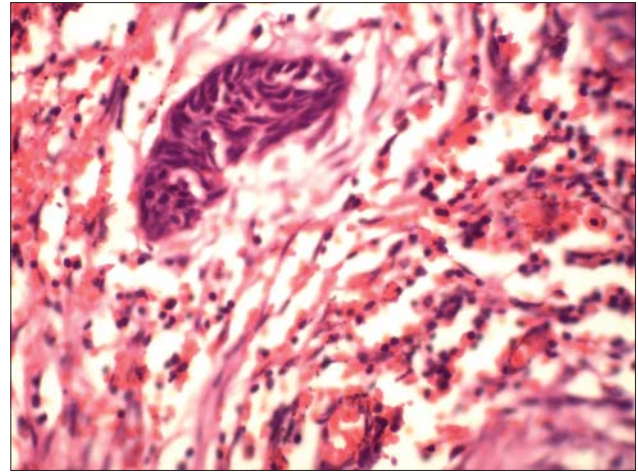


Figure 2: A nest of metaplastic squamous cells surrounded by necrotic tissue and lymphoplasmacytic infiltrate (H and E, $\times 400$)

DISCUSSION

NS of parotid is rare. Parotid masses are generally treated by surgery, the minimum diagnostic and therapeutic procedure is superficial parotidectomy.^[5] If the histopathological diagnosis of surgical specimen is benign, the treatment concludes and patient is followed up. If malignant, additional treatment is required. It is a self limiting benign lesion, which results in unnecessary surgery of parotid if FNAC is non-diagnostic as in our case. NS is not an easy diagnosis for an inexperienced pathologist.^[1,2] The result is unnecessary surgery, a high cost for a self limiting lesion. Diagnosis can be further supplemented via immunohistochemistry demonstrating focal to absent immunoreactivity for p53, low immunoreactivity for MIB1 (Ki-67), and the presence of 4A4/p63 and calponin positive myoepithelial cells. Nonetheless, to date, hematoxylin and eosin staining remains the gold standard.^[6]

Although squamous metaplasia and necrosis are non-specific findings observed in several inflammatory and neoplastic disorders of salivary glands, necrotizing metaplasia has distinctive histological features^[1] squamous metaplasia, lobular configuration of coagulative necrosis and preserved lobules of acini. The lobular arrangement is explained by separate blood supply to each lobule.

Compromise in blood supply leads to NS. The blood vessels pass through foramina that make them vulnerable. Standish and Shafer^[7] observed necrotizing sialometaplasia in salivary glands of rats by experimentally ligating the blood vessels.

The most common cause of NS of salivary glands is trauma caused by dental procedure. The parotid gland differs

from other sites as NS develops after excision of benign or malignant tumor.

Our case is unusual because of its site and absence of any predisposing factor, which is described in literature. NS is rarely encountered in the parotid gland. Sialometaplasia secondary to ischemia is known but idiopathic variety is largely unknown. Recognition of this entity in parotid is essential as it is a self limiting condition and patient does not require follow up once the parotid gland is removed as is the case in malignancies.

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