CASE REPORT

Necrotizing sialometaplasia: A diagnostic dilemma!

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INTRODUCTION

Necrotizing sialometaplasia (NS) is a benign, self-limiting inflammatory reaction of salivary gland tissue which may mimic squamous cell carcinoma or mucoepidermoid carcinoma, both clinically and histologically, that creates diagnostic dilemma leading to unwarranted aggressive surgery. This pathological lesion was first reported in 1973 by Abrams et al. as a reactive necrotizing inflammatory process involving minor salivary glands of the hard palate.

NS occurs in only 0.03% of all biopsied oral lesions with predominance in Whites. The age prevalence of NS ranges from 17 to 80 years with male predominance of 2:1. Most commonly affected site is the minor salivary glands of the palate. The pathogenesis is unknown but is believed to be due to ischemia of vasculature supplying the salivary gland lobules. A simple incisional biopsy is required to confirm the histological diagnosis and to rule out more serious disease processes. It is a self-limiting disease process and requires no treatment. It will be prudent to do repeat biopsy in case if the lesion does not heal within 3 months.

CASE REPORT

A 35-year-old male reported with the history of non-healing ulcer on left side of the palatal region from 15 days with fever and malaise. He had habit of gutkha and tobacco chewing since 10 years.

On clinical examination there was single ulcerative lesion on left side of the palate measuring about 1.5 cm in diameter. Ulcer had indurated margins giving clinical picture of malignant lesion. There were no palpable neck nodes.

Incisional biopsy was performed and minimal bleeding was noticed during the biopsy procedure. Histopathological examination showed pseudoepitheliomatous hyperplasia of the overlying epithelium. The underlying connective tissue showed areas of necrosis, salivary glands showed clear acinar lumina with squamous metaplasia of the ductal epithelium. Inflammatory infiltrate was also seen. All these features lead to the diagnosis of necrotizing sialometaplasia.

The ulcerative lesion had healed completely after period of 5 weeks.

DISCUSSION

NS is squamous metaplasia of the salivary gland ducts and acini with ischemic necrosis of the salivary gland lobules which occurs most frequently on hard palate. The pathogenesis is unknown but it is believed to be due to ischemia of vasculature supplying the salivary gland lobules. There are variety of factors causing ischemia like direct trauma, administration of...
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Figure 1: Intraoral view revealing the lesion on the left posterior part of the palate

Figure 2: Histological photomicrograph showing pseudoepitheliomatous hyperplasia (H&E stain, x100)

Figure 3: Histological photomicrograph showing squamous metaplasia, necrosis and inflammatory cells. (H&E stain, x100)

Figure 4: Histological photomicrograph showing ductal metaplasia. (H&E stain, x400)

local anesthetic, ill-fitting dentures, alcohol, smoking, cocaine use, radiation, intubation, surgical procedures and upper respiratory tract infections. The long-term use of salbutamol might lead to dehydration and thinning of the mucosa, making it more susceptible to local trauma.[5]

NS has wide range of differential diagnosis which includes primary adenocarcinoma of the palate, squamous cell carcinoma, subacute necrotizing sialadenitis (SANS), major aphthous ulcer, mucoepidermoid carcinoma, secondaries from adenocarcinoma of rectum, secondary syphilis and tuberculous ulcer.

NS occurs spontaneously and the initial symptoms may include fever, chills, malaise or swelling.[3] The posterior hard palate is the most common site to be affected by NS and junction of the hard and soft palate being the second most common site. About two-thirds of the palatal lesions are unilateral; however, bilateral synchronous and metachronous lesions are not uncommon. Also the lesion could occur in the midline.[6] The size may range from 0.7 to 5.0 cm (average 1.8 cm).[7]

The diagnosis of NS is challenging and is based on a complete clinical history and a well-oriented biopsy section. Biopsy taken from base of the ulcer and the edge that is most indurated and raised, yields the most representative sample. A combination of histopathological and clinical findings is often helpful in establishing the confirmatory diagnosis.

The 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. Hematoxylin-eosin staining remains the gold standard in histopathological diagnosis of NS.[8]

Necrotizing sialometaplasia is characterized by lobular necrosis and associated squamous metaplasia of ducts and acini, with the preservation of lobular architecture. The areas of necrosis consist of small pools of mucin surrounded by neutrophils located within or adjacent to metaplastic ducts. An inflammatory background is typically present.
The mucosal surface is often ulcerated and pseudoepitheliomatous hyperplasia and squamous metaplasia of the excretory ducts are seen. The pseudoepitheliomatous hyperplasia as well as the deeply seated islands of metaplastic squamous epithelium often lead to mistaken diagnosis of squamous cell carcinoma. In addition, the squamous metaplasia of the ducts and acini juxtaposed with residual mucous cells may suggest a mucoepidermoid carcinoma.

In most cases, however, the lobular architecture, areas of necrosis and mixed inflammatory background, together with the distinctive epithelial nests, distinguish NS.\(^9\)

As described by Anneroth and Hansen, the histopathogenesis of NS has five histological stages: infarction, sequestration, ulceration, repair and healing. Histological features exhibit a spectrum ranging from ulceration, lobular necrosis, sequestration of necrotic acini, pseudoepitheliomatous hyperplasia of adjacent epithelium, squamous metaplasia of ductal epithelium and inflammatory changes.\(^{10}\) As these diagnostic criteria are quite distinctive, proper care should be taken in diagnosis of this lesion, so that misdiagnosis and unnecessary radical treatment can be avoided.

Usually no treatment is required and the lesion heals by secondary intention within 4-10 weeks (average 5.2 weeks). Even a full-thickness palatal lesion communicating with nasal cavity resolves completely in 6 months.\(^3\)

**CONCLUSIONS**

NS is a self-limiting disorder of salivary glands mostly affecting the hard palate giving clinical presentation of malignant neoplasm. Unfortunately it has been misdiagnosed clinically and microscopically as a malignant neoplasm, resulting in inappropriate and aggressive treatment. A simple incisional biopsy is required to confirm the histological diagnosis and to rule out more serious disease processes, hence, the role of oral pathologist is of paramount importance. It is a self-limiting disease process and requires no treatment. It will be prudent to do repeat biopsy in case if the lesion does not heal within 3 months.

**REFERENCES**