Necrotizing sialometaplasia: Report of 2 cases

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ABSTRACT

Necrotizing sialometaplasia (NS) was defined by Abrams et al. in 1973 as a reactive necrotizing inflammatory process involving minor salivary glands of the hard palate. Before that recognition, many patients with this condition had been improperly treated because of its clinical and histologic resemblance to malignancy such as mucoepidermoid carcinoma and squamous cell carcinoma. We report two cases of necrotizing sialometaplasia. One case involved a 58-year-old male who had an ulcerative palatal lesion exposing underlying bone which has the typical features of the above mentioned condition. Another case involved a 59-year-old male who developed a necrotizing sialometaplasia in association with a dome-shaped palatal swelling which was proved as an adenoid cystic carcinoma after operation biopsy. (***Korean J Oral Maxillofac Radiol 2006; 36 : 207-9***)

KEY WORDS : Sialometaplaisa, Necrotizing; Palate; Salivary Gland Disease

Necrotizing sialometaplasia (NS) was defined by Abrams et al.[1] in 1973 as a reactive necrotizing inflammatory process involving minor salivary glands of the hard palate. It is a benign, self-limiting ulcerative disease that histologically and clinically simulates squamous cell carcinoma or mucoepidermoid carcinoma. Before the recognition of NS, many patients with this condition had been improperly treated because of its resemblance to malignancy.

Brannon et al.[2] reviewed 69 cases and reported NS were found at age ranged from 1.5 to 75 years (average 45.5 years) showing male and white predominance. Majority of cases occur on the posterior hard palate and the junction of the hard and soft palate was the second most common site. About two thirds of the palatal lesions were unilateral, however, bilateral[3,4] and midline locations were also noted. The size ranged from 0.7 to 5.0 cm (average 1.8 cm). Painful lesions are more than twice as common as asymptomatic lesions.

From the histomorphologic findings, ischemia involving the glandular tissue is probably the underlying cause of NS. However, the nature and pathophysiology of the ischemia remain unclear in many cases.[2]

Though it is not hard to find reports of NS in the literature, NS is still a diagnostic challenge. Mesa et al.[5] suggested that NS occurs in only 0.03% of all biopsied oral lesions. We report two cases which had ischemia as the underlying cause of NS. One case involved a 58-year-old man who noticed an ulcerative palatal lesion 15 days after denture irritation. The other case involved a 59-year-old man who developed a red ulcerative lesion 9 days after operation of a dome-shaped palatal swelling which proved to be an adenoid cystic carcinoma.

*This work was supported for two years by Pusan National University Research Grant.

Received August 23, 2006; accepted September 27, 2006

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Case reports

Case 1

A 58-year-old man presented with an ulcerative palatal lesion measuring 3 × 2 cm. The margin of the ulcer was slightly elevated and the underlying bone was exposed (Fig. 1). The patient noticed this condition 15 days ago after denture irritation. There were no pain or any discharge associated with the ulcer. His past medical history was non-specific. He smokes one pack a day and drinks. The radiographic features were non-contributory. Incisional biopsy under local anesthesia was done and the result reported as NS. The histologic feature (Fig. 2) showed pseudoepitheliomatous hyperplasia of the overlying epithelium and squamous metaplasia of salivary ducts.

Case 2

A 59-year-old male presented with a dome-shaped swelling at the right junction of hard and soft palate. The size of the

Fig. 1. Ulcerative palatal lesion measuring 3 × 2 cm. The margin of the ulcer was slightly elevated and the underlying bone was exposed.

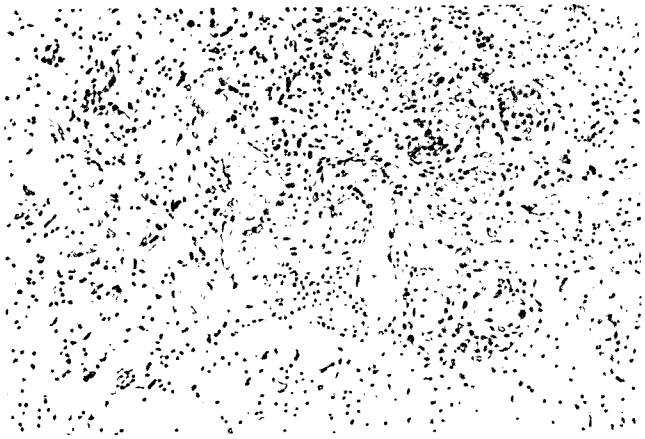

Fig. 2. The histologic feature showed squamous metaplasia of salivary ducts (H & E stain, × 200).

Fig. 3. A dome-shaped swelling measuring 2 × 2 cm at the right junction of hard and soft palate.

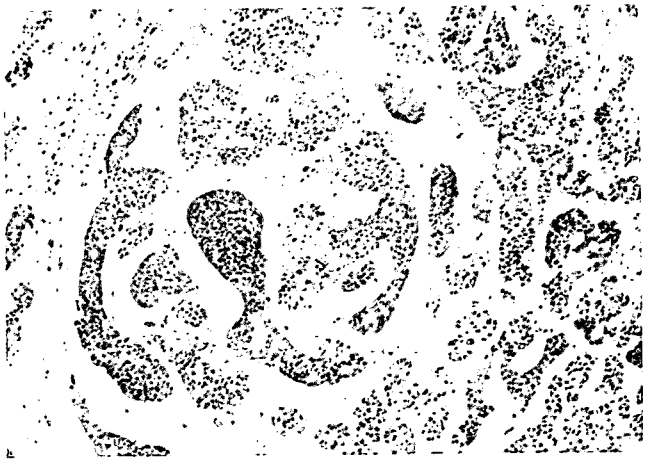

Fig. 4. Small ductal elements composed of basoloid cells and tubuloductal componensts were arranged with an intervening hyalinized stroma. The histologic diagnosis was adenoid cystic carcinoma (H & E stain, × 100).

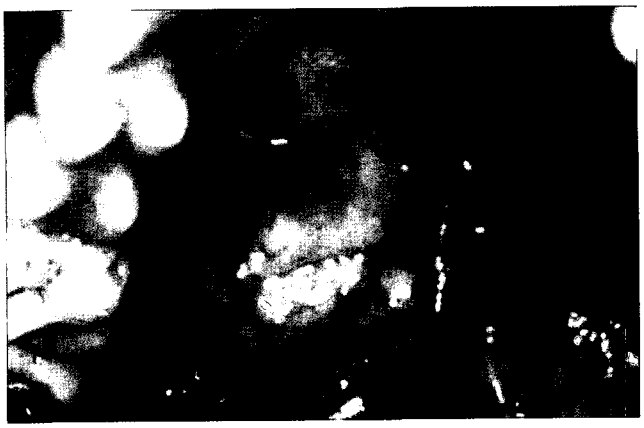

Fig. 5. Red ulcerative lesion developed at the posterior margin 9 days after operation and rebiopsy was done.

lesion was 2 × 2 cm and the overlying mucosa had normal appearance with no pain or erosive change (Fig. 3). The patient noticed this condition one year ago and he had a cerebrovascular accident at that time. The computed tomographic image showed no bone involvement with slightly elevated soft tissue mass at the right palate. Excisional biopsy under general anesthesia was done under the impression of pleomorphic adenoma. But the histologic diagnosis was adenoid cystic carcinoma. Small ductal elements composed of basoloid cells were observed and these tubuloductal componensts were arranged with an intervening hyalinized stroma (Fig. 4). Healing was uneventful but red ulcerative lesion developed at the posterior margin 9 days after operation (Fig. 5) and rebiopsy was done. The histologic diagnosis was NS.

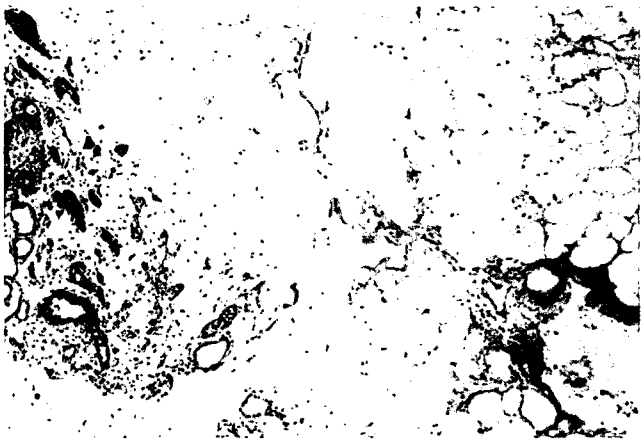

Fig. 6. There were necrosis of acini and ductal squamous metaplasia showing the histologic features of NS (H & E stain, ×40).

There were necrosis of acini and ductal squamous metaplasia (Fig. 6).

Discussion

Clinically, NS most often presented as a deep-seated ulcer,[1,3,6-9] however, a number of cases manifested as a nonulcerated swelling or mass[10-12] and may develop into an ulcerative lesion.[11,12] In some cases unilateral or bilateral anaesthesia of the greater palatine nerve preceded the appearance of the typical swelling and ulceration of NS.[4,8]

Some reported cases of NS[2,10,13] followed an operative procedure for another primary tumor of the major or minor salivary gland. In each such instance, the clinical manifestation of a mass, suggesting recurrence, presented within a period of 6 to 53 days (mean 18 days).[2]

From the histomorphologic findings, ischemia involving the glandular tissue is probably the underlying cause of NS. However, the nature and pathophysiology of the ischemia remain unclear in many cases. Possible predisposing factors suggested are traumatic injuries such as occlusal trauma, burn injury, a severe blow to the lip or face, laceration, ill-fitting dentures, smoking and/or alcohol habits, upper respiratory infection,[14] and even the palatal injection of local anesthesia for dental procedure.[10] Some cases[13] were found in association with another lesion and in these cases, the tumor or cyst itself likely compromised the blood supply to the involved glands, resulting in ischemic necrosis and sialometaplasia. In this report, case 1 could have ischemic condition because of denture irritation, and case 2 of surgical procedure.

Radiographically most of NS show no bony involvement but few cases demonstrated an associated saucerization of the underlying palatal bone.[2]

Histologically classical features of NS involves acinar necrosis, squamous metaplasia of ducts, pseudoepitheliomatous hyperplasia of surface epithelium, and mucus liberation with an associated inflammatory response. In early lesions, coagulation necrosis of glandular acini predominates, whereas for later lesions, extensive squamous metaplasia and reactive fibrosis dominate the histologic features. In virtually every case where adequate tissue samples were available, preservation of the overall lobular architecture of the involved gland was evident.

Usually, no treatment is required and the lesion heals by secondary intention within 4 to 10 weeks (average 5.2 weeks).[2] Even a full-thickness palatal lesion communicating with nasal cavity[3] resolved completely in six months.

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