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Case report

A case of necrotizing sialometaplasia with bone resorption at the hard palate

Tomoko Fujii[a,*], Yuichi Ohnishi[a], Yuki Minamino[a], Masahiro Watanabe[a], Motohiro Gotoh[a], Masahiro Wato[b], Kenji Kakudo[a]

[a] Second Department of Oral and Maxillofacial Surgery, Osaka Dental University, 1-5-17 Otemae, Chuo-ku, Osaka 540-0008, Japan
[b] Department of Oral Pathology, Osaka Dental University, 8-1, Kuzuhahanazono-cho, Hirakata-shi, Osaka 573-1121, Japan

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ABSTRACT

Necrotizing sialometaplasia (NS) is a relatively uncommon benign disease of the salivary glands that most commonly occurs in the palate. It is often confused clinically and histopathologically with malignancies, such as squamous cell carcinoma or mucoepidermoid carcinoma. We report a rare case of NS with bone resorption at the hard palate.

A 35-year-old woman was referred to our hospital complaining of swelling of the left hard palate. The lesion was 30 mm × 22 mm in diameter, with the ulceration. Magnetic resonance imaging showed a well-defined mass with bone resorption of the left hard palate. The patient underwent an incisional biopsy and histopathological diagnosis of NS was made. After 10 weeks, the lesion healed and no evidence of recurrence was noted.

1. Introduction

Necrotizing sialometaplasia (NS) is a benign, self-limiting and rare inflammatory disease of the minor salivary glands [1–5], which may mimic squamous cell carcinoma or mucoepidermoid carcinoma and can lead to unnecessary surgical approach [6]. NS was first reported in 1973 by Abrams et al. [7] as a reactive necrotizing inflammatory process involving minor salivary gland of the hard palate. In most cases, bony involvement is not generally described or to be expected in NS [8]. We report the clinical and histopathological features of a rare case of NS with bone resorption at the hard palate.

2. Case report

A 35-year-old woman was referred by her dentist to our hospital with a swelling of the left hard palate in October 2009.This swelling had been discovered by her dentist about 2 weeks earlier; the patient had not noticed it until that time. After the dental examination, the patient was given a diagnosis of alveolar abscess, caused by dental infection. Then the patient was made an incision and medication. But this is not effective. Clinical examination showed a relatively circumscribed mass with the ulceration, 30 mm × 22 mm in diameter, in the left hard palate. On palpation, the swelling was elastic hard with no fluctuation elicited (Fig. 1). Hematological and biochemical parameters were with in normal limits. An orthopantomogram showed a cloudiness in the left sinus floor. MRI findings consisted of submucosal lesions in left hard palate with hypointense on T1 signal and hyperintensity on T2 (Fig. 2). They revealed a fluidfilled polypoid mucous membrane thickening of the left sinus floor and the bone resorption at the hard palate. The first diagnostic hypothesis was salivary gland tumor; most likely pleomorphic adenoma. An incisional biopsy was performed under local anesthesia. The diagnosis of NS was performed. So, the patient was advised to maintain good oral hygiene with saline rinse. The ulceration showed remission after 6 weeks of follow-up except for an area of erythema. The lesion had healed after 10 weeks (Fig. 3). The 2-year follow up there has been no evidence of recurrent disease.

Histopathologic findings: The histologic sections revealed mucosa composed of acanthotic and hyperkeratotic stratified squamous epithelium. The connective tissue was composed of young fibroblastic tissue, infiltrated by chronic inflammatory cells. In the submucosa, squamous metaplasia of residual acinar and ductal elements was found. Some nests showed evidence of residual lumina (Fig. 4A and B). Lobular necrosis was noted with through the maintenance of the architecture of salivary glands (Fig. 4C). Immunohistochemical examination revealed that the

☆ AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

* Corresponding author. Tel.: +81 6 6910 1510; fax: +81 6 6910 1030.
E-mail addresses: tomoko.fujii@mac.com (T. Fujii), onishi_yu_ku@msn.com (Y. Ohnishi), minamino@cc.osaka-dent.ac.jp (Y. Minamino), watana-m@cc.osaka-dent.ac.jp (M. Watanabe), goto-m@cc.osaka-dent.ac.jp (M. Gotoh), wato@cc.osaka-dent.ac.jp (M. Wato), kakudo@cc.osaka-dent.ac.jp (K. Kakudo).

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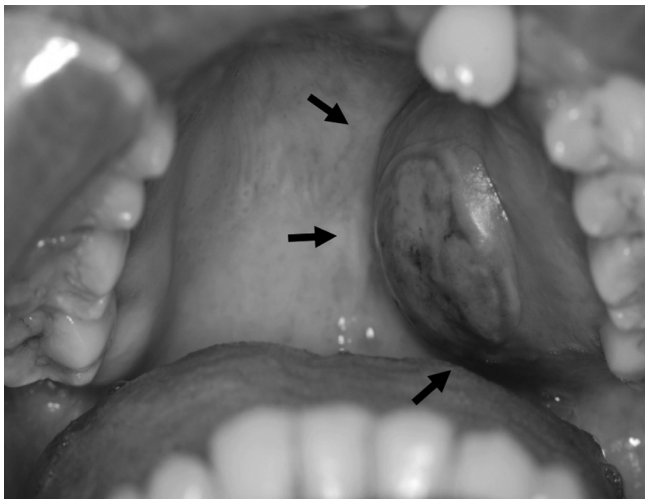

Fig. 1. Photograph of patient at initial presentation showing an ulcerated swelling of the left hard palate.

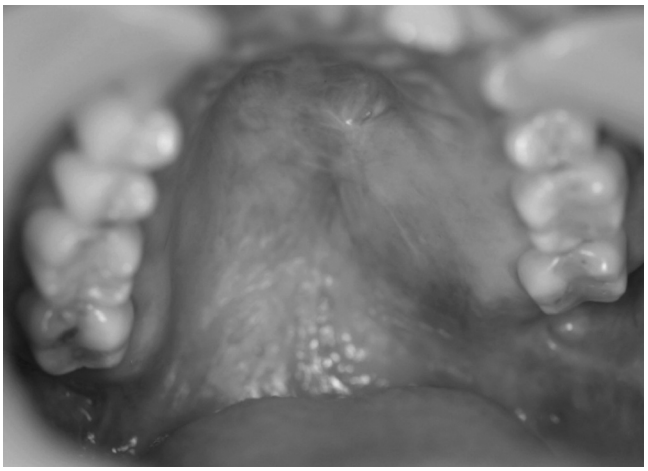

Fig. 3. The follow-up photograph after 15 weeks shows remission of the lesion.

squamous metaplasia cells were moderately positive for cytokeratine AE1/AE3 (Fig. 4D), weekly positive for p53 and negative for PAS and Mucicarmine. The result of SMA was focally positivity in the periphery of the squamous metaplasia (Fig. 4E). The pathological diagnosis was NS.

3. Discussion

NS is defined as squamous metaplasia of the salivary gland ducts and acini with ischemic necrosis of the salivary gland lobules and it occurs at hard palate most frequently. The pathogenesis is unknown but it is believed to be due to ischemia of vasculature supplying the salivary gland lobules. Etiological factors for ischemia can be local trauma, local anesthesia, ill-fitting dentures, smoking, alcohol, radiation, allergies, upper respiratory tract infection, intubation, surgical procedures involving the area [1,2,4,9], cocaine use [10], and chronic vomiting [4,5,11]. It was hypothesized that in our patient, damaging with the toothbrush on the palate, lead to ischemia, which resulted in infarction and ulcer formation of the salivary tissues.

The most common clinical manifestation of this condition is swelling and/or ulceration of the hard palate, which may be associated with pain [8]. Paresthesia in the affected area is rare [1–3]. The size ranged from 0.7 to 5.0 cm (average 1.8 cm) [8]. In the present case, it was a relatively large mass with the ulceration, 30 mm × 22 mm in diameter, and the differential diagnosis was salivary gland tumor, most likely pleomorphic adenoma or mucoepidermoid carcinoma.

Radiographically, most of NSs showed no bony involvement except for few cases [8]. Brannon et al. [8] reported a case of unusual NS which demonstrated an associated saucerization of the underlying palatal bone. So, it is not because of NS. In the present case, MRI findings revealed bone resorption in the left hard palate but no treatment performed. Therefore, we consider this case is very rare. Farina et al. [12] reported the MRI findings of necrotizing sialometaplasia in their study. They said that a fluid-like appearance of a mass lesion and the absence of contrast enhancement could help to exclude salivary gland neoplasm and this might be the MRI findings suggestive of NS. A hyperintense T2 signal is quite common in pleomorphic [12]. But in our case, no fluid-like appearance and bone resorption was found in MRI. That is the reason why we thought it might be pleomorphic adenoma or maybe mucoepidermoid carcinoma. We think that the MRI findings might be different for each stage of NS.

Anneroth and Hansen [11] described the histopathogenesis of NS by proposing five histological stages: infarction, sequestration, ulceration, repair stage, and healed stage. During infarction, necrosis of the glandular acini predominates and culminates in the formation of the ulcer. At the beginning of the healing stage, proliferation of the overlying epithelium is observed, which is demonstrated microscopically by pseudoepitheliomatous hyperplasia. If infarction is limited, no sequestration occurs. Healing

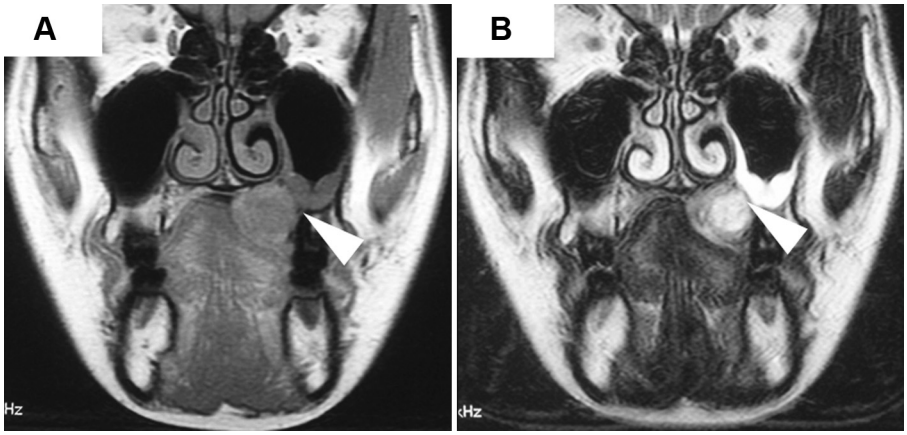

Fig. 2. The enhanced T1-weighted (A) and T2-weighted (B) coronal images demonstrated diffuse enhancement in left hard palate. Arrows: the bone resorption at the hard palate.