Necrotizing Sialometaplasia: A Case Report

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Sir,

Necrotizing sialometaplasia is a benign, self-limiting, reactive, inflammatory process affecting the major and minor salivary glands throughout the upper aerodigestive tract (1). The aetiology is unknown. It was first described in 1973 by Abrams et al. (2), and is a relatively rare condition. In a recent publication by Fowler & Brannon (3), reviewing all cases previously published in English language medical literature, a total number of 184 cases were described. Because of the rareness and localization of the disease, it is a rather unknown condition among dermatologists. Most cases have been reported in the otorhinolaryngic literature; however, it is also an important disease for dermatologists to recognize.

Necrotizing sialometaplasia can be confused clinically and histologically with malignancy (4), but it is a self-limiting disease and only conservative treatment is necessary. We here present a case of bilateral necrotizing sialometaplasia in a woman.

CASE REPORT

A 22-year-old woman experienced abrupt onset of swelling and pain of the hard palate. Within 3–4 days two ulcers developed, proceeding over the next 2 weeks to two crater-like lesions. No fever was present. In the 6 months before onset of her oral symptoms she had had a weight loss of approximately 12 kg. She denied having any eating disorders and attributed stress as the cause of her weight loss.

The patient was a student at the school of dentistry. A few days before the onset of symptoms she was the subject in practical demonstration of a routine clinical oral examination. She was a non-smoker.

Two weeks after onset of symptoms the patient was referred to the Department of Dermatology. At physiological examination the hard palate presented with two symmetrically deep ulcers (Fig. 1a); the ulcers were 1×1 cm and 1×1.5 cm in size. Weak bilateral adenitis was present at the angulus mandibulae. No affection of the remaining mouth, genitalia or skin was seen. HIV, hepatitis, Epstein–Barr virus, cytomegalovirus, syphilis and antinuclear antibody screens were negative. Cultures from the ulcers were normal. X-ray of the chest and ultrasound examination of the abdomen were normal.

A biopsy specimen from the palate ulcer revealed the diagnosis necrotizing sialometaplasia (Fig. 2).

The patient was treated conservatively with the non-steroidal anti-inflammatory fluid benzydamine (Andolex®, 3M Pharma) five times a day and obtained spontaneous recovery within 5 weeks (Fig. 1b). Furthermore, she was referred to a psychologist because of a possible eating disorder.

DISCUSSION

Necrotizing sialometaplasia is a rare disease. Among the 184 cases reviewed by Fowler & Brannon (3) in the year 2000 the average age was 45.9 years. The male:female ratio was 1.9:1; 58% occurred on the hard palate but only 12% occurred bilaterally (3). Most cases presented with deep ulcers, with a lesion size from 0.7 to 5.0 cm, but non-ulcerated swelling was also seen. The duration of the disorders ranged from 4 days to 3

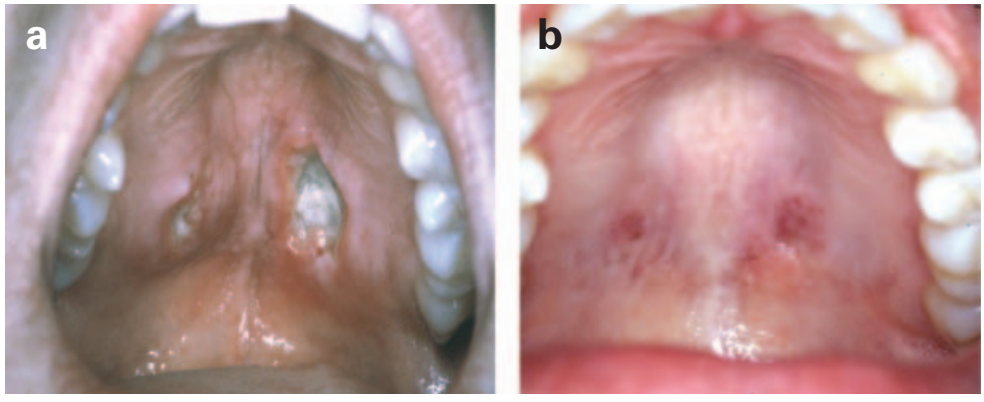

Fig. 1. (a) Necrotizing sialometaplasia of the hard palate 2 weeks after onset of symptoms. (b) Spontaneous recovery 7 weeks after onset of symptoms.

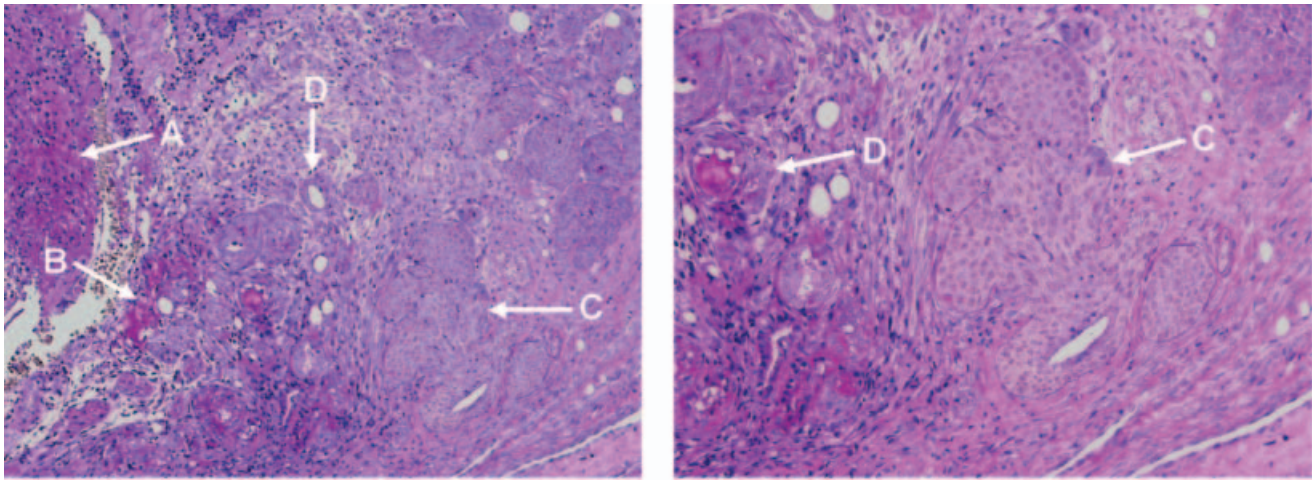

Fig. 2. Biopsy from the hard plate with necrotizing sailometaplasia (PAS staining) showing (left) necrosis (arrow A), and extravasation of mucus (B), and (left and right) squamous metaplasia of ducts and acini (C) and remnant of intact glandular structures (D).

months. Seventy-one of the 184 cases were smokers. Recently, bone involvement has been described (5).

The pathogenesis is most often ischaemia of the salivary gland leading to necrosis and ulceration followed by a spontaneous recovery. Necrotizing sialometaplasia has been produced in rats by ligating the blood vessels supplying the submandibular and sublingual glands (6). Traumatic injury such as dental instrumentation has been described as a possible initiating event (3), but local mucosal trauma in patients suffering from bulimia and chronic self-induced vomiting have also been described as possible aetiological factors (7, 8).

Our patient presented two possible triggering factors for necrotizing sialometaplasia. Firstly, she had participated in practical demonstrations at the school of dentistry a few days before the onset of symptoms. Secondly, she suffered from a possible eating disorder although she denied that possibility. Eating disorders should always be considered as a pathogenic factor in younger patients with necrotizing sialometaplasia.

Histologically, necrotizing sialometaplasia may simulate squamous cell carcinoma or mucoepidermoid carcinoma. It is recognized by the presence of squamous metaplasia of ducts and acini of seromucinous glands accompanied by necrosis, sometimes with ulceration (Fig. 2). Demonstration of intact lobular architecture is very important in the differential diagnosis (9, 10).

Necrotizing sialometaplasia often evolves very aggressively in the first few weeks to months and may therefore be mistaken for a malignant disease. In several cases patients have been treated with unnecessary surgery ranging from conservative excision to total maxillectomy (1). Because necrotizing sialometaplasia is a benign and self-limiting disease which only requires conservative treatment it is important to recognize this disease before more aggressive and often disabling treatment regimes are carried out.

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