Necrotizing Sialometaplasia: A Case Report

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Abstract

Necrotizing sialometaplasia is a benign, self-limiting lesion of both major & minor salivary glands; it mimics mucoepidermoid carcinoma or squamous cell carcinoma in its clinical and histological features. The presentation of such an ulcer, mimicking malignancy should be diagnosed correctly to avoid mental and surgical trauma to the patient. We present diagnosis and treatment of ulcers on the palate and in oral vestibule in a 26-year-old, Indian female. The base of ulcer on the left posterior aspect of the palate rested on the palatine bone and was covered with necrotic debris. The ulcer in the vestibule was deep and a fungating mass was seen at its margin. Computed tomography and histopathological examination was done. Following the treatment an improvement in the patient's condition was observed; the ulcer had healed both at the vestibule and palate within 10 weeks.

Introduction

Several cases of necrotising sialometaplasia, a benign condition have been reported in the oral cavity as well as in other sites in the body that contains elements of the salivary gland, from the paranasal sinuses to the lung.1,2,3 Correct clinical diagnosis of necrotizing sialometaplasia is important because this lesion mimics the appearance of malignant disease, both clinically and microscopically.4 Inability to make the correct diagnosis may result in unnecessary surgery leading to physical and mental trauma to the patient subsequent to wrong diagnosis of squamous cell carcinoma and mucoepidermoid carcinoma.5 A case of necrotising sialometaplasia of the hard palate in a 26-year-old woman, following local anaesthesia is reported.

Case Report

A 26 year old Indian female from Bihar, India, was referred to Faculty of Dental Sciences, Institute of Medical Sciences, Banaras Hindu University, Varanasi. She presented herself with a diffuse swelling and numbness extending from her lower left eyelid to the lower border of the mandible (Fig 1). Oral examination revealed a unilateral large ulcer on left side of her hard palate, 20mm in antero posterior dimension and 10mm in transverse diameter. Its margins were raised, inflamed and irregular. The base had extended to the bone. Another ulcer in the buccal mucosa with a fungating mass and raised margin in the vestibule on the left side, around the first and second molar was observed (Fig 2 A and B). The ulcer had developed following her visit to a private local dentist for extraction of her first and second molar 26 and 27, which were badly decayed and causing her severe pain. During earlier treatment the dentist had injected lignocaine, to anaesthetise the area for teeth extraction but could not extract the teeth due to severe pain. Subsequently, she developed ulcers within 24 hours. She was a non-smoker. Her medical history in the past revealed no relevant medical problem. For the said lesion a week course of amoxicillin was taken with no improvement before. A differential diagnosis was made which included:

Necrotizing Sialometaplasia
Another Infective lesion

Computed tomography was done which did not reveal any abnormality. Biopsy from the ulcer was taken for histopathological examination. The incisional biopsy showed no evidence of any neoplastic changes; the histological examination of the lesion showed severe inflammatory infiltrate, coagulative necrosis and partial necrosis of salivary gland (Fig 1 C and D). It was consistent with early necrotizing sialometaplasia. Patient was kept on observation for six weeks and prescribed painkillers for occasional pain. Chlorhexidine mouthwash was used vigorously after every meal three times daily. Patient underwent root canal treatment (RCT) on 26 and 27 with crowns fitted on teeth. An improvement in the patient’s condition was observed, the ulcer had healed both at the vestibule and palate within 10 weeks (Fig 2 C and D).

Discussion

Necrotizing sialometaplasia was first described by Abrams et al., (1973). It may arise in any area containing salivary gland tissue. Classically, it involves the mucoserosous glands of the hard palate. The lesion is usually painful and presents as a sharply circumscribed ulcer, frequently 1 to 3 cm in diameter. Palate involvement usually appears as a single,
unilateral ulcer on the posterior hard palate or at the junction of the hard and soft palates. The ulcer borders are often erythematous and may be raised. There is a wide age range (1.5-83 years), although most patients are older than 40 years.6,7,8. The incidence appears to be 2 to 3 times greater in males than females.8. The most widely accepted theory regarding the development of necrotizing sialometaplasia is the ischemia of the vessels that supplies the salivary gland lobules.1,2. In our case, the possible etiologic factor appears to be either the use of an expired local anaesthetic or the prolonged use of anaesthetic medication. In an experimental study in a rat model, local anesthetic injections induced necrotizing sialometaplasia.9 A range of histologic findings, ranging from coagulation necrosis of salivary gland acini in early lesions to squamous metaplasia of ducts and reactive fibrosis in late lesions can be seen. Usually there is vascular proliferation, prominent inflammatory infiltrate, and partial necrosis of salivary glands, associated with regeneration and squamous metaplasia of the adjacent duct and acini.3,10 The two most important differential diagnosis includes squamous cell carcinoma and mucoepidermoid carcinoma. The benign, although focally atypical, cytologic appearance of the cells and, more importantly, the maintenance of the acinar architecture distinguishes necrotizing sialometaplasia from either squamous cell carcinoma or mucoepidermoid carcinoma.10 Management involves adequate biopsy, observation and reassurance. The lesions undergo spontaneous healing within 2-3 months. No treatment is required other than an analgesic for a patient whose lesion is painful. Necrotizing sialometaplasia does not usually recur.8,10 Repeat biopsy is indicated for a patient whose lesion fails to resolve. Awareness of this potential diagnostic pitfall is of great importance because an inaccurate histopathological diagnosis can result in inappropriate or unnecessary treatment, ranging from conservative excision to maxillectomy.3. This case report shows the importance of a careful clinical examination combined with an adequate histopathological examination in order to avoid misdiagnosis as well avoiding mutilating surgery after wrong diagnosis.

References

Illustrations

Illustration 1

Fig 1. A and B. Patient before and after the treatment. C and D. Microphotographs showing increased infiltration of the inflammatory cells, necrotising salivary glands coagulative necrosis (*) and associated large hemorrhagic areas (bold arrow)
Illustration 2

Fig.2. A and B. Ulcers on the posterior part of the palate (arrow) and in the vestibule (arrow head). C and D. Healed ulcer with healthy mucosa on the palate and in the vestibular area.
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