Case Report

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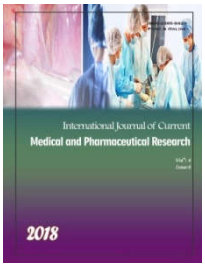

NECROTISING SIALOMETAPLASIA – A RARE CASE REPORT IN PAEDIARTIC PATIENT

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ABSTRACT

Necrotising sialometaplasia is an uncommon benign, self-limiting inflammatory disease of the minor salivary glands. It always gives a diagnostic dilemma to the clinician because of its close resemblance to malignancy both clinically and histopathologically. It has wide range of age of occurrence, but mostly occurs at middle age group. Here we report a case of necrotising sialometaplasia which is very rare to occur at paediatric patients.

INTRODUCTION

Necrotising sialometaplasia represents only <1% of all biopsied oral lesions [1]. It is a self-limiting lesion mostly involves the hard palate which will be mistaken for either mucoepidermoid carcinoma or squamous cell carcinoma. Some of the cases had been reported in major salivary glands also [2]. It can occur at any age group but incidence of this lesion in children has been not reported much.

Case presentation

A 2 years-old boy with his parents presented at our institution with chief complaint of ulcer in his palate since 1 week. History of trauma in the palate while he was playing with pen in the mouth 2 to 3 weeks prior to the onset of lesion. Initially it was started as a smaller erythematous lesion and within a week become an ulcer to the present size, after that there was no tendency for progression in the size of the lesion. History of mild discomfort and pain associated with ulcer. On intraoral examination an irregularly shaped ulcer noted at the right posterolateral aspect of palate around the junction of hard and soft palate measuring around 1.5 cm x 1 cm. Margins of the ulcer were normal but the surrounding mucosa was pale, greyish white and erythematous in few places. Floor of the ulcer showed through and through opening with partially exposed peripheral bone. On palpation the ulcer was mild-tender without any blanching and bleeding tendency. OPG revealed no bony destruction. Patient was referred to paediatric department to rule out any other systemic conditions.

Based on the clinical features it was provisionally diagnosed as necrotising sialometaplasia. Differential diagnosis of traumatic ulcer was considered and malignant ulcer was excluded considering age of the patient which is uncommon to occur. Incisional biopsy was not done considering the age of the patient. The parents were counselled about the self-limiting nature of the lesion and it had to be wait and watch for the regression of the lesion. No interventional treatment was required for this lesion unless it fails to heal. The patient was advised to apply surface anaesthetic and antibiotic (mucopain) 2-3 times a day with cotton andmaintain good oral hygiene with saline rinse 3-4 times a day. After 1 month of period, parents gave the history of completely healed ulcer but unfortunately the patient didn't turn up for the follow up review due to their abroad job transfer.

Based on the history, clinical presentation and the self-healed nature of this ulcer we presumed this case as necrotising sialometaplasia, although only histopathological diagnosis will give confirmatory diagnosis which we excluded for this patient considering risk and age of patient.

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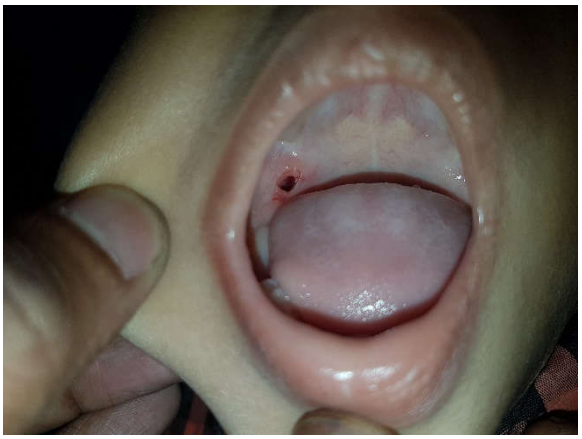

Fig 1 Intraoral photograph depicting a reddish erythematous ulcer involving right posterolateral region of palate around the junction of hard and soft palates

DISCUSSION

Necrotising sialometaplasia is an uncommon non neoplastic inflammatory lesion and it can be diagnostically challengable because of its overlaping features with other neoplastric lesions like squamous cell carcinoma and mucoepidermoid carcinoma. The probable etiopathogenesis for this lesion could be ischemia due to any blunt trauma, palatal local anaesthesia, surgical procedures, denture wear, alcohol and tobacco use [3]. Histopathologicallythere are proposed to be five stages in the development of necrotizing sialometaplasia:infarction, sequestration, ulceration, repair, andhealing[4].

Clinically it often present as an ulcer in the hard palate which will be complained by the patient as, their palate itself fell off.Few cases can present as a swelling follwed by ulcer formation. The ulcer size ranged from 0.7 to 5.0cm (average 1.8 cm)[5]. Clinical appearnce always gives impression of a maligant lesion due to its irreugular shaped crater form. Ususally asymptomatic in nature but our case had been reported with mild pain. Like our case most of the lesion doesn't undergo bony invovement , but few cases noted with mild saucerisation of underlying bone.

Clinical differential diagnosis for nicrotising metaplasia are squamous cell carcinoma,mucoepidermoid carcinoma, traumatic ulcer, major apthous ulcer, tuberculous ulcer and syphillitic ulcer. In our case all the differntial diagnosis were ruled out based on Chest X ray, OPG findings where both didn't show any abnormalities and routine blood investigationwas within the normal limits. Considering the possibilities of other malignacies which are all very uncommon at very early age, incisional biopsy was not performed in this case and only based on the history and clinical features we suggested the diagnosis of Necrotising sialometaplasia.

But ideally the diagnosis of Necrotising sialometaplasia should be based on histopatholgy only. Histopathologically it shows Pseudoepitheliomatous hyperplasia, Squamous metaplasia of ducts and acini, Preservation of lobular architecture, Lobular infarction with or without mucin spillag, Inflammation secondary to extravasation of mucin. The key for diagnosis is presevation of lobular architecture which will be absent in squamous cell carcinoma and mucoepidermoid carcinoma. On further differtial diagnosis can be done with immunohistochemistry, where cytokertain 7 positivity will be only present in necrotising sialometaplasia and mucoepidermoid casrcinoma. To diffrerentialte mucoepidermoid carcinoma and necrotising sialometaplasia, numerous tumour markers like ki-67 and p53 will be helpful where both will show intense staining in mucoepidermoid carcinoma unlike necrotising sialometaplasia [6].

Because of its self limiting nature no interventional treatment required for this lesion, with symptomatic treatment will heal spontaneously within 1 or 2 months. In our case ulcer was healed completely within one month duration. Recurrence is very rare.

In conclusion necrotising sialometaplasia is an uncommom lesion to occur in paediatric patients and clinical diagnosis is very challenging for the clinicinas. It can be esaily mistaken for malignancy so sound knoweldege about this lesion is very essential.

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