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Case Report

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Bilateral Palatal Necrotizing Sialometaplasia in Patient with Prurigo Nodularis: A Case Report

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Abstract

Necrotizing Sialometaplasia (NSM) is a rare, benign, self-limiting, inflammatory disease of mostly minor salivary gland origin.

NSM can resemble a malignant lesion in its early disease course, both clinically and pathologically. Bilateral involvement of the hard and soft palate by the lesion is a rare occurrence. NSM was first described as a reactive inflammatory process. Since then, over 200 cases have been reported and has been added to the WHO classification of salivary gland tumours under the tumour-like lesions. Here, we report a case of a 30-year-old male with bilateral NSM of both the hard and soft palate, on a background history of prurigo nodularis. The patient was initially referred to an Oral and Maxillofacial Department by his general practitioner for a palatal ulcer, concerning for oral malignancy. The ulcer was painless with a well-demarcated border and necrotic base. Subsequent biopsy and histological examination confirmed necrotizing sialometaplasia with healing occurring within 5 weeks.

Keywords: Palate, Necrotising Sialometaplasia (NSM), Bilateral, Prurigo Nodularis (PN)

Introduction

Necrotising Sialometaplasia (NSM) is a rare, benign, self-limiting, inflammatory disease of mostly minor salivary gland origin [1-3]. It was first reported by Abrams in 1973 [2] and despite almost 50 years passing since the first recording of the lesion, the aetiology is still not fully understood. It is believed that the injury is caused as result of a traumatic injury; either physical, chemical or biological, to the blood supply of the palate. This in turn results in ischaemic necrosis of minor salivary gland lobules [1, 2, 4].

The hard palate is the most common site affected however, it can also arise in areas with minor salivary glands such as labial mucosa [5, 6], floor of mouth [7], nasal cavity [6], maxillary sinus [6,8], parotid [6] and tongue [6,8].

NSM is more common in white men in the 5th decade, with a male to female ratio of 2:1. Despite its preponderance for middle age, it can also occur at any age. Most NSM lesions appear spontaneously with variable symptoms which can include swelling, fever, malaise with or without pain. Paraesthesia of the palate is prodromal in certain cases [9,10]. Typically, it presents as a shallow crateriform ulcer with everted edges. Two-thirds of cases are unilateral with the remainder being bilateral [6,9]. Prurigo Nodularis (PN) is a chronic disorder which presents as an area of itchiness, leading to excoriation and subsequent development of hyperkeratotic crusted nodules mostly affecting the extensor surfaces of the limbs. Epidemiological studies have shown that PN is more common in women of an older age [11,12].

Any part of the body amenable to scratching can be affected by PN and is often accompanied by the pathognomic "butterfly sign" on the back, due to the fact that central back is unreachable and therefore this site is spared [13].

Here we present the unusual case of NSM occurring in a 37-year-old male with PN.

Methodology

This was chosen due to its unusual presentation making the case a diagnostic dilemma. A literature search was conducted using the PUBMED, Web of Science and Google Scholar.

Case Report

A 37-year-old male was referred by his general practitioner to an Oral and Maxillofacial department due to ongoing oral ulceration, refractory to antibiotic therapy, of the right and left palatal region present for four weeks. The patient was also complaining of the presence of several skin lesions on the face and neck which predated the oral ulcers. The patient denied any contributing factors such as smoking or excessive alcohol consumption in addition to any causative factors for oral ulceration such as recent dental care requiring injection of a local anaesthetic to the area. This was the first occurrence of the palatal lesion which was painless with no swelling or altered sensation of the affected mucosa. The patient's medical history was significant for PN of 18-years duration for which he was extensively investigated for, with no underlying haematological, immunodeficient, hepatic or renal diseases.

Extra-oral examination revealed multiple, excoriating, crusted nodules on the skin of the face, trunk and limbs with associated scarring (Figure. 1 & 2).

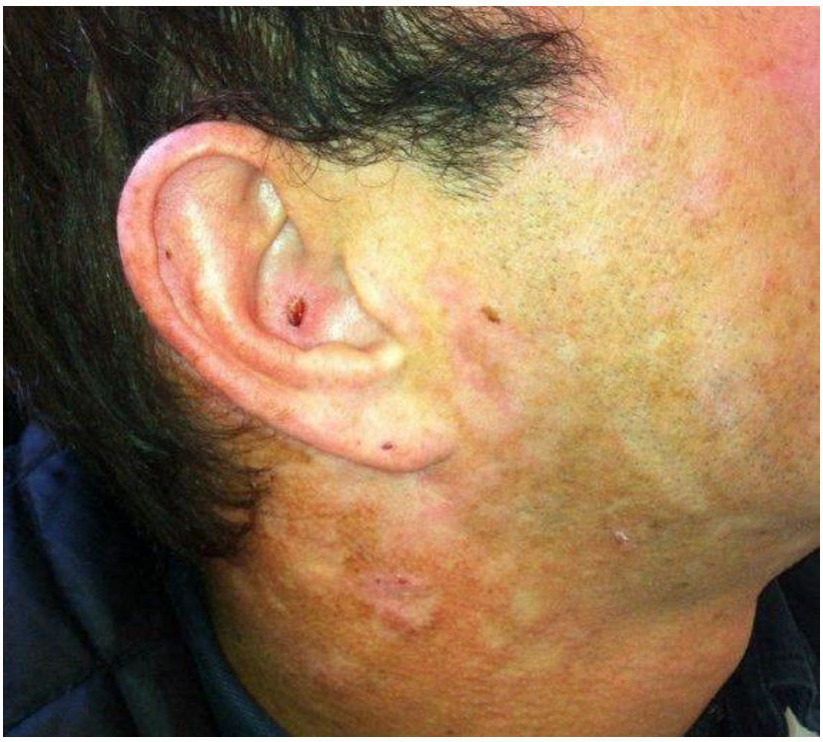


Figure 1: Right pre-auricular region showing cutaneous nodules and excoriations

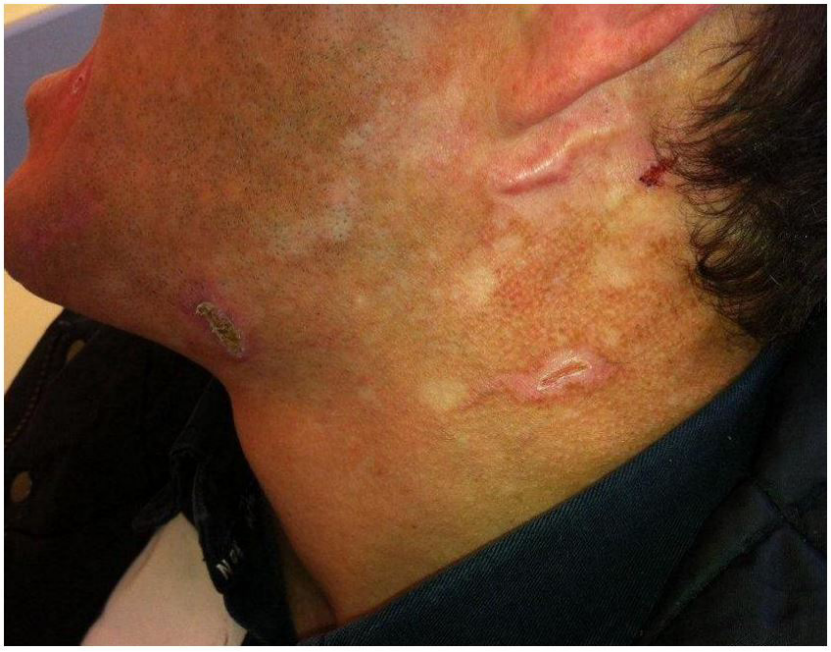


Figure 2: Left neck showing cutaneous nodules and excoriations

Intra-orally, an irregular area of sloughing ulceration was noted in the posterior hard palate bilaterally with erythematous raised margins. The ulcers measured 3.0 x 2.0cm on the right side involving the soft palate, crossing the midline with the left ulcer of 1.5 x 2.0cm (Figure. 3).

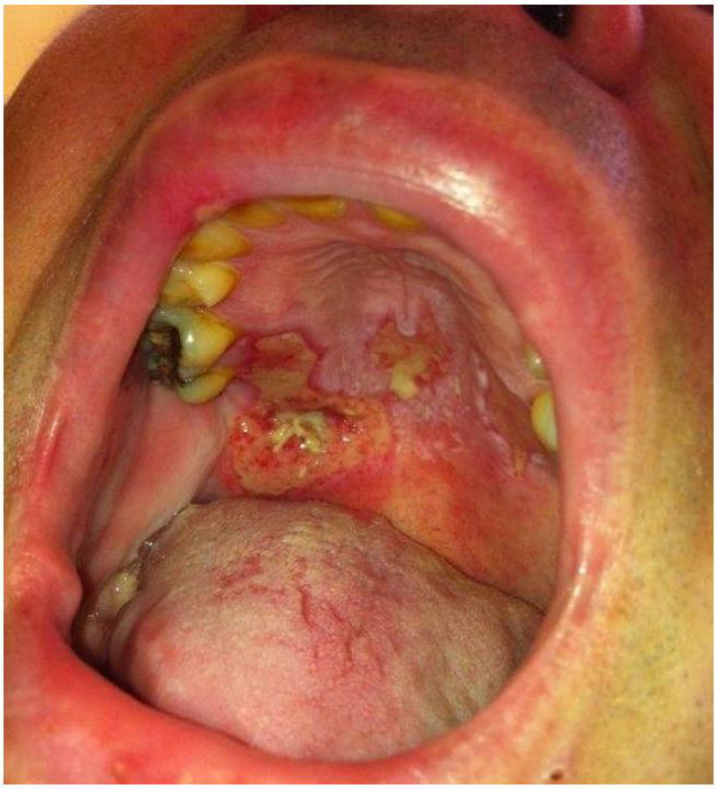
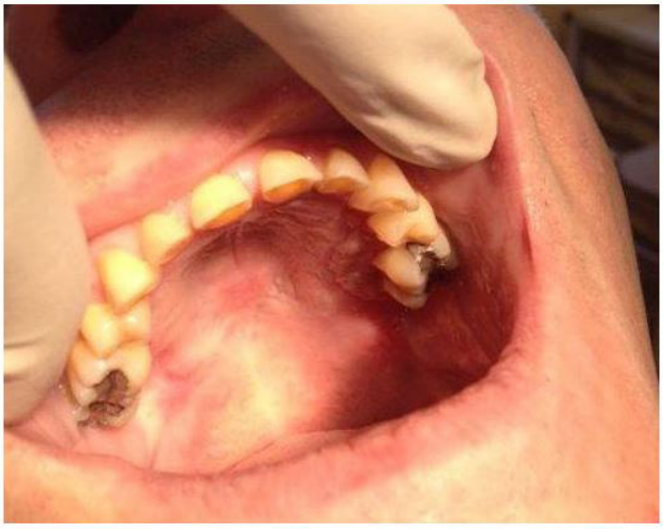


Figure 3: Intra-oral view of hard/soft palate displaying bilateral ulceration

Multiple incisional biopsies were performed of the ulcers which confirmed the diagnosis of NSM. The patient was managed with local oral hygiene measures with a chlorhexidine-based mouthwash. A follow up appointment at five weeks showed total remission of the oral lesion (Figure. 4 & 5)



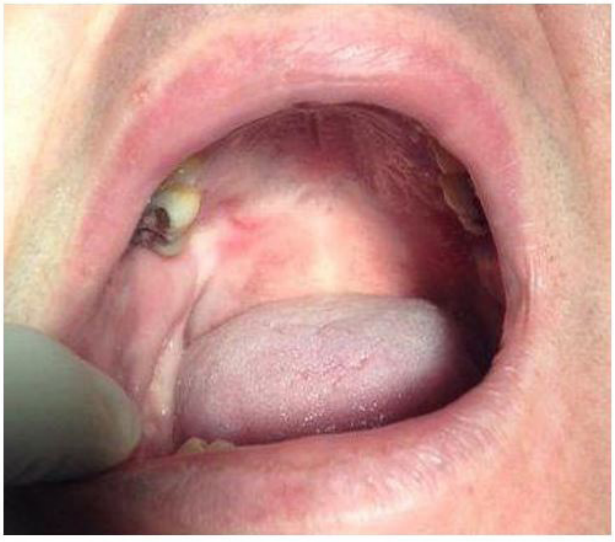


Figure 4 & 5: Intra-oral view of hard-soft palate showing resolution of ulceration

Discussion

This report describes a case of bilateral NSM in a patient with chronic PN. Although NSM has been reported with other conditions, this is the first recorded presentation of NSM arising on a background of PN. Since the first reporting of NSM by Abrams in 1973[2], the aetiology has still not been fully understood. It is believed to be due to an insult to the vasculature of the palate. Causes of the ischemic injury includes local trauma, local anaesthetic infiltration, alcohol, surgical procedures, smoking and cocaine usage [1], ill-fitting dental prostheses and upper respiratory infections. Microscopically, the ischemic changes lead to infarction of the salivary gland acini with associated necrosis followed by release of mucinous material. The subsequent inflammation and repair induce metaplastic changes in the mucosa.

Anneroth and Hansen [4] proposed five histological stages that are associated with NSM: i) infarction, ii) sequestration, iii) ulceration, iv) reparative and iv) healed stage.

Cases have also been reported to be associated with eating disorders [3], and systemic disease such as diabetes and HIV [14]. In this case, our patient had a history of chronic PN.

The presentation of NSM lesions vary in size from a few millimetres to centimetres and also in varying colours [15]. No particular causative factors identified in PN but substance P which is a major neurotransmitter or neuromodulator contribute in neurogenic inflammation, pain and pruritus [16].

NSM has a preponderance for white males in the 5th decade [10], with the patient in this case being below the average age of presentation. NSM usually presents as a unilateral, shallow crateriform ulcer with everted edges [6,9]. In this case, the whole of the posterior and midline palate bilaterally was affected by ulceration. An Exophytic form of NSM has been reported [17] and also non-ulcerated NSM as reported by Keogh et al [9] and Janner et al [3].

Any part of the body amenable to scratching can be affected by PN [13]. The patient denied any symptoms or itchiness or self-inflicted injury to the palate. PN has been associated with development of malignant disease especially lymphoproliferative such as Hodgkin lymphoma and leukaemia. The patient was investigated extensively for lymphoproliferative and malignant disorders. Due to the chronic and pruritic nature of PN, it has been associated with anxiety and depression [18].

Differential diagnoses for NSM should include: major aphthous ulcer, traumatic ulcerative granuloma with stromal eosinophilia (TUGSE), mucoepidermoid carcinoma, adenoid cystic carcinoma and squamous cell carcinoma. Granulomatous diseases including tuberculosis and syphilis should be ruled out. The diagnosis is confirmed following incisional biopsy and subsequent histopathological examination [17].

The following criteria aid in distinguishing NSM from a malignancy: (a) lobular morphological preservation, (b) bland appearance of squamous island or nest with no cytological evidence of malignancy and (c) no residual ductal lumina in any nest [16,19].

NSM can be distinguished from squamous cell and mucoepidermoid carcinoma by myoepithelial markers and cytokeratin subtype expressions as described by Rizkalla et al.,[19].

The first co-presentation of NSM and adenoid cystic carcinoma of the soft palate was described by Lee et al.,[20] for which, two theories were purported. First; the incisional biopsy or injection of local anaesthetic induced the formation of NSM and the second; the pressure effect of the tumour on the injuring the adjacent vasculature.

The NSM lesion is self-limiting and usually requires no treatment with healing seen in 4 to 10 weeks [19]. In this case, the lesion was healed in 5 weeks.

In conclusion, NSM is a self-limiting, benign condition of minor salivary gland origin. With proper history taking and good histopathological examination a misdiagnosis and inappropriate treatment can be avoided. The exact cause in this case could not be identified and as is the first case reported in a patient with PN, a causative relationship cannot be ruled out.

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