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Early-Stage Non-Ulcerated Necrotizing Sialometaplasia Mimicking Malignancy in a Pediatric Patient: A Case Report

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Abstract Necrotizing sialometaplasia (NS) is a rare, benign, self-limiting inflammatory condition that primarily affects the minor salivary glands of the hard palate. It often presents as a nodule that may ulcerate over time. The most commonly known trigger is ischemia. Diagnosing non-ulcerated NS can be challenging, particularly because its clinical features may resemble malignant salivary gland tumours. We report a case involving a 9-year-old female who presented with a painful, slightly raised erythematous lesion on the hard palate. An incisional biopsy and histopathological analysis confirmed the diagnosis of NS. Awareness of non-ulcerated presentations is essential to avoid misdiagnosis and unnecessary aggressive treatment.

Key Words Necrotizing Sialometaplasia, Non-Ulcerated Lesion, Minor Salivary Glands, Salivary Gland Tumour Mimic, Paediatric Oral Pathology

INTRODUCTION

The primary structures affected by pathological changes in the palate are the numerous minor salivary glands located within the hard palate. Among the most significant alterations are neoplastic and inflammatory conditions. A well-known inflammatory condition, necrotizing sialometaplasia (NS), typically involves the minor salivary glands of the hard palate. Clinically, NS is usually described as a nodule that develops into a central ulcer approximately 1 cm in diameter. Although ischemic events are the most commonly recognized triggers, other potential causes include local trauma, substance abuse and eating disorders [1,2]. It is important to emphasize that ulcerated NS may mimic mucoepidermoid carcinoma the most common malignant salivary gland tumour and its clinical features can resemble those of other malignant salivary gland tumours [3]. In rare instances, NS may present as a non-ulcerated lesion. In such cases, when a bluish-red swelling is observed, malignancy should also be considered in the differential diagnosis [1,4,5]. Other granulomatous conditions that can resemble NS include deep fungal infections and syphilitic gum disease [6].

Aim

The purpose of this report is to present the clinical and histological characteristics of a case of non-ulcerated necrotizing sialometaplasia (NS), described as an eroded area beginning to develop ulceration and to emphasize the importance of including malignant salivary gland tumours in the differential diagnosis.

CASE REPORT

A 9-year-old girl, accompanied by her father, presented with acute, radiating pain on the right side of her jaw lasting approximately three minutes. Her medical history included multiple episodes of sublingual aphthous ulcers. At presentation, she did not report any persistent painful trigger points. Clinical examination revealed a crossbite, attributed to a slight leftward deviation of the mandible.

During a follow-up appointment, the patient reported similar pain episodes. She was prescribed antibiotics (amoxicillin 10 mg/kg) and analgesics (paracetamol 15 mg/kg). Intraoral examination revealed pain and swelling across the palatal mucosa, along with signs of pericoronitis around teeth #16 and #17 as shown in Figure 1.

CBCT imaging showed significant thickening of the maxillary sinus mucosa, with the mucosa extending superiorly and nearly filling the antrum (Figure 2). Based on the suspicion of a dentoalveolar abscess, fungal infection or maxillary sinus pathology, hospitalization was recommended.

The patient was referred for otorhinolaryngology (ORL) evaluation, including a CT scan of the facial bones and a biopsy of the antral lesion (Figure 3). She was admitted for intravenous treatment and underwent an exploration and biopsy under general anaesthesia. A biopsy was obtained and referred to the pathologist for examination. The lesion showed progressive healing and achieved complete resolution within a few days following the biopsy (Figure 4). One week later, she returned with a complaint of a periapical abscess associated with the upper right second primary molar #55 as seen in (Figure 5). A 5-day course of amoxicillin (10 mg/kg) was prescribed, followed by extraction of the affected tooth (Figure 6).

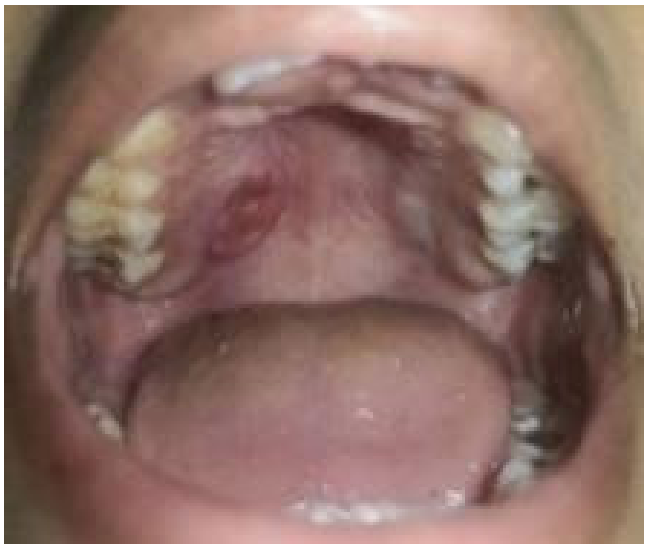

Figure 1: Clinical image showing a discrete, elevated, erythematous area on the right hard palate, indicative of a non-ulcerated NS lesion.

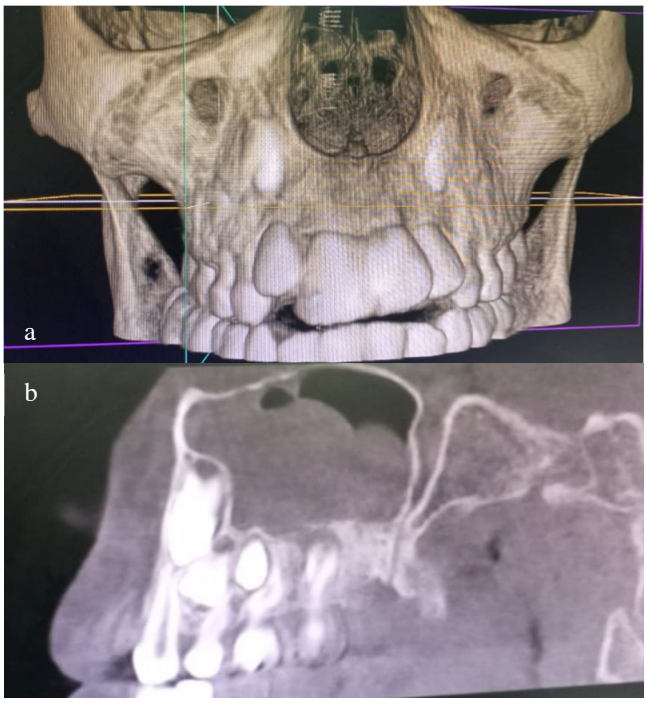

Figure 2(a,b): Cone Beam Computed Tomography imaging revealed pronounced mucosal thickening in the maxillary sinus, with the mucosa extending upward and almost completely occupying the antrum.

The Histopathological Analysis

The histopathological examination revealed a section of stratified oral mucosa without surface ulceration. The adjacent connective tissue contained salivary ducts exhibiting squamous metaplasia (Figures 7c and 7d). As necrotizing sialometaplasia (NS) is a self-limiting condition,

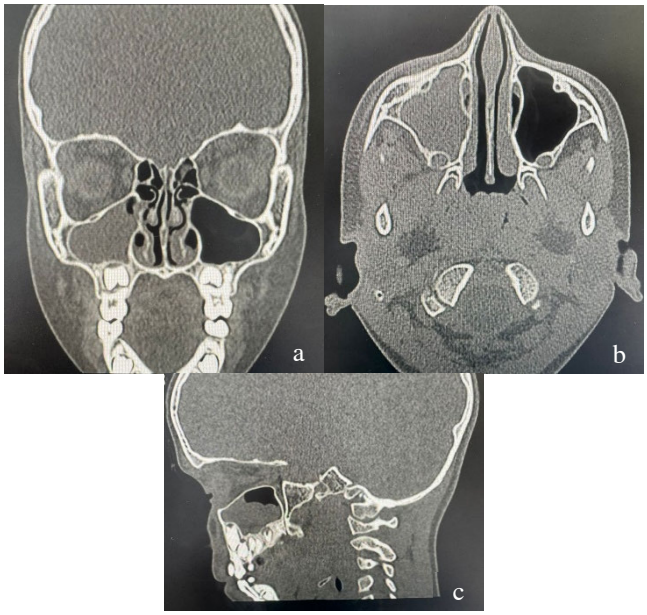

Figure 3(a,b,c): (a) Coronal view. (b) Axial view. (c) Sagittal view of computed tomography scan shows mucosal thickening of the right maxillary sinus, with the mucosa extending superiorly and nearly filling the antrum.

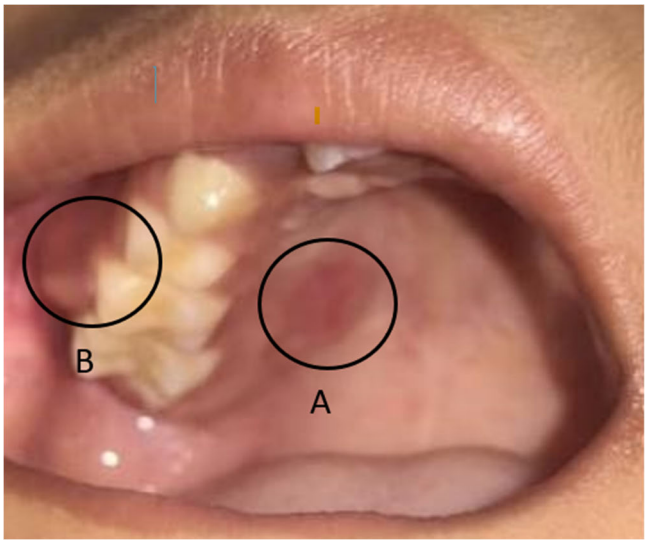

Figure 4: (A) Final clinical image showing complete healing a few days after the biopsy procedure. (B) Development of a periapical infection.

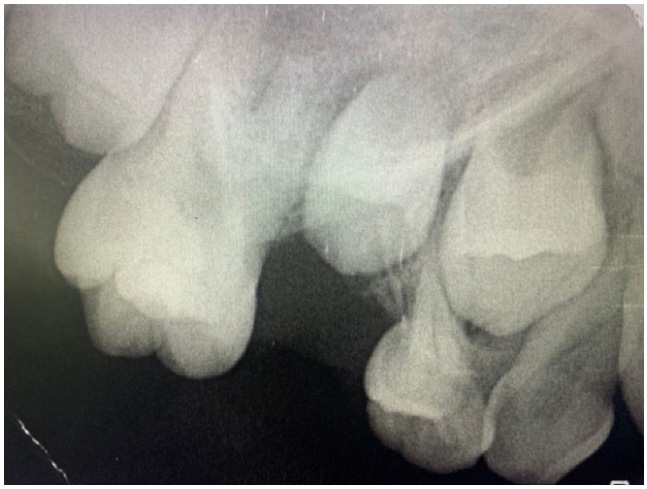

Figure 5: Development of a periapical infection related to the upper right second primary molar tooth #55.

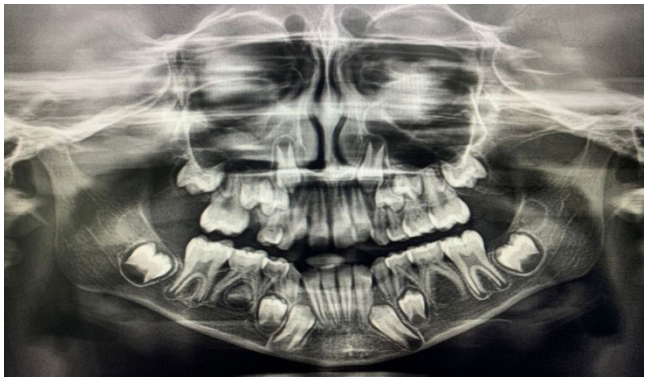

Figure 6: Panoramic radiograph (OPG).

the patient required only supportive care focused on pain management, which included analgesics administered every six hours for two days. Following the biopsy, the lesion showed progressive healing and achieved complete resolution within a few days (Figure 4a).

DISCUSSION

Just over 200 cases of necrotizing sialometaplasia (NS) have been reported in the English-language literature, making it a rare disorder [7]. NS accounts for less than 1% of all oral biopsies. First described in 1973 by Abrams and colleagues, this condition can occur anywhere salivary glands are present. However, most cases are intraoral, typically involving the minor salivary glands of the hard or soft palate. Major salivary glands may also be affected, though less commonly.

As demonstrated in the present case, non-ulcerated NS poses a diagnostic challenge. It is essential to rule out salivary gland tumours, particularly malignant ones, when a palatal swelling or nodular lesion is observed. Pain and colour changes (especially bluish or purplish hues) have been suggested as independent predictors of malignancy. Since pain is a common symptom, early-stage NS, before the development of central ulceration, may clinically resemble

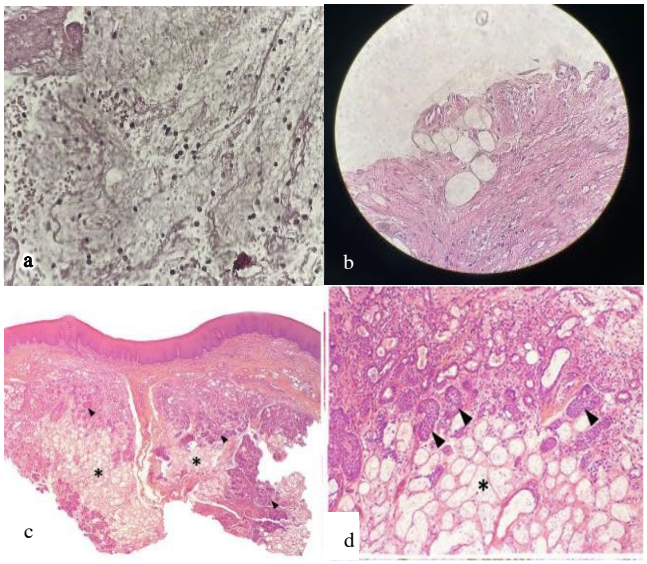

Figure 7(a,b,c,d): (c) Photomicrographs of the biopsy showing large areas of acinar necrosis (*) associated with foci of squamous metaplasia (arrowhead) beneath a normal surface epithelium. (d) Ductal squamous metaplasia and extensive acinar necrosis, with preservation of the lobular glandular architecture.

malignant tumours. However, salivary gland malignancies typically lack the rapid progression seen in NS [1].

Overall, NS is a benign, self-limiting condition that usually presents as an ulcerated lesion. Nonetheless, it can occasionally appear in a non-ulcerated form. This case highlights the importance of including early-stage, non-ulcerated NS in the differential diagnosis of minor salivary gland tumours. Non-ulcerated Necrotizing Sialometaplasia May Mimic a Salivary Gland Tumour.

Males appear to be affected by necrotizing sialometaplasia (NS) at approximately twice the rate of females [8]. NS often begins as a non-ulcerated area of oedema, which may be accompanied by pain. After 2–3 weeks, the necrotic tissue typically sloughs off, leading to the formation of a crater-like ulcer [9]. In the absence of treatment, complete healing generally occurs within 4 to 90 days [10]. The pathophysiology of NS has been described in five histological stages: infarction, sequestration, ulceration, reparative stage and healed stage.

In our case, the incisional biopsy was performed after a CBCT scan. No bone involvement of the hard palate was observed, which is consistent with previous findings. Although rare, bone involvement does not exclude a diagnosis of NS [8].

The aetiology of NS can vary. In paediatric cases, necrosis may not always be evident. For instance, Ylikontiola *et al.* described a 2-year-old girl diagnosed with sialometaplasia of the soft palate following adenoidectomy, where clinical and histopathological evaluation revealed no frank necrosis [11]. Imbery and Edwards have suggested that when the infarct remains localized, ulceration and

sequestration may not occur [12]. Ylikontiola *et al.* [11] hypothesized that infarction of the salivary gland tissue in their case was likely triggered by the adenoidectomy.

Additional risk factors for NS include dental injections, alcohol abuse, smoking, cocaine use, upper respiratory infections, radiation therapy and the presence of nearby tumours, as well as a history of surgery, trauma or injury. Bulimia has also been proposed as a possible contributing factor [8,13–15]. Although tumour-related NS is uncommon, Brannon *et al.* reported 8 cases of NS associated with adjacent tumours or cystic lesions out of a sample of 184 patients, with 4 of these involving the palate [3].

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