

Bilateral oral focal mucinosis on the palate of a 2-year-old child: a case report

JOHNATHAN WOO & WA SHAM CHEUNG

Department of Dentistry, British Columbia Children's Hospital, Vancouver, British Columbia, Canada

International Journal of Paediatric Dentistry 2015; 25: 70–72

Background. Oral focal mucinosis (OFM) is an uncommon benign oral lesion. The aetiology of the lesion is unknown. Histologically, it appears as a well-circumscribed myxomatous mass surrounded by denser, collagenous connective tissue. Most cases of OFM were found in adults. It is very unusual for young children to have OFM.

Case Report. A case of OFM in a 2-year-old child is reported. The patient was presented with

non-painful bilateral enlargements on the palate. The overlying mucosa was smooth and not ulcerated and appeared in the same colour as the adjacent tissue. The histology of the lesion showed myxomatous mass indicative of OFM. Treatment consisted of surgically removing the lesions under general anaesthetic.

Conclusion. Paediatric dentists should consider OFM in their differential diagnosis of soft tissue oral lesions in children.

Introduction

Oral focal mucinosis (OFM) is a rare lesion of unknown aetiology that was first described by Tomich in 1974¹. What is thought to be the oral counterpart to cutaneous focal mucinosis¹ presents as an innocuous soft tissue swelling in the oral cavity that may be pedunculated or sessile². Most cases are located on the gingiva or hard palate^{1–5} but has also been reported to occur on the tongue^{1,4}. It arises predominantly in adults during the fourth and fifth decades of life⁵ and has been reported infrequently in children and adolescents³. OFM shows a distinct female/male ratio of 5 : 3⁴.

The present report describes a case of bilateral OFM on the palate of a 2-year-old child. The clinical and histological features, the current pathological knowledge and management of OFM are discussed.

Case report

A 2-year-old female was referred to the Department of Dentistry of the British Columbia Children's Hospital, with a 3-month history of prominent bilateral palatal rugae/enlargements in the hard palate. Medical history was significant for global developmental delay. There was no history of pain, trauma, infection nor were airway or feeding difficulties reported. There were no genetic or other significant medical conditions revealed in the background histories of the parents. Aggressive non-nutritive sucking habits were also ruled out. The parents were concerned about the impact the swellings would have on their child's feeding habits.

Clinically, the patient appeared to be healthy and in no distress. Intraoral examination revealed bilateral, pink, sessile masses involving the anterior and posterior palate in a symmetrical fashion (Fig. 1). The masses did not extend across the midline and were firm to touch. The right lesion was oval and 20 mm at its longest, whereas the left lesion was round and 15 mm in diameter. The overlying mucosa was smooth, non-ulcerated and appeared in the same colour as the adjacent tissue. The patient also had ankyloglossia. Hard tissue

Correspondence to:

Dr Wa Sham Cheung, BC Children's Hospital
Rm K0 -162, 4480 Oak Street, Vancouver, BC, Canada
V6H 3V4. E-mail: swcheung@shaw.ca

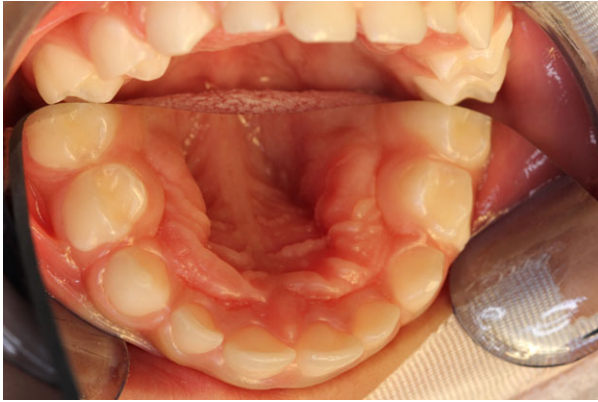


Fig. 1. Intraoral view of the palate.

examination revealed a non-carious and spaced primary dentition. No pathological mobility of the teeth was noted. A complete blood count, urine organic acid profile and plasma amino acid profile were requested. The results were all within normal ranges.

Differential diagnoses at the time of the clinical examination included palatal exostosis, fibrous hyperplasia, peripheral ossifying fibroma, pleomorphic adenoma, lymphoma and Langerhans cell histiocytosis.

A general anaesthetic appointment was arranged to accomplish the following procedures: complete dental radiographic examination, excisional biopsy of the palatal lesions and lingual frenectomy to correct the ankyloglossia.

The dental radiographic examination was unremarkable, and no radiographic changes in the area of the soft tissue palatal lesions were noted. The palatal region was infiltrated with 1.8 mL of 2% lidocaine with 1 : 100,000 epinephrine, and an excision of the bilateral palatal lesions was performed. Electrocautery was used to achieve haemostasis. Right and left specimens were then placed in formalin and sent for pathological examination. The lingual frenum was released, and an interrupted 4-0 absorbable suture was placed. The histology of the specimen showed squamous mucosa overlying submucosa with a subepithelial band of myxoid tissue of low cellularity (Fig. 2). The nuclei appeared spindled and bland (Fig 3). These histopathological features were suggestive of OFM. The child was followed up 1 month post operatively. The palate has since completely mucosalized without complications.

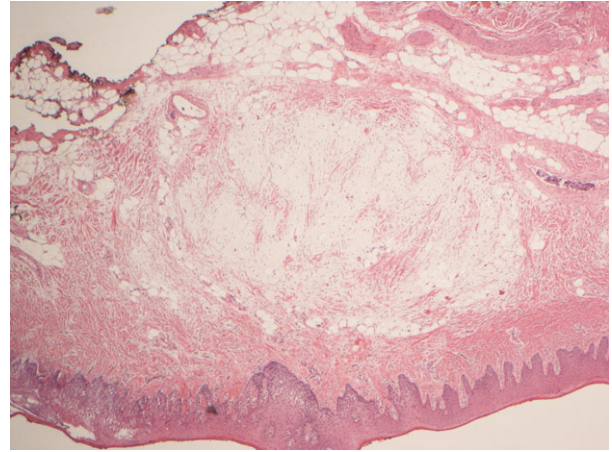


Fig. 2. Photomicrograph (Low power view) showing non-encapsulated area of loose, myxomatous connective tissue surrounded by denser, collagenous connective tissue.

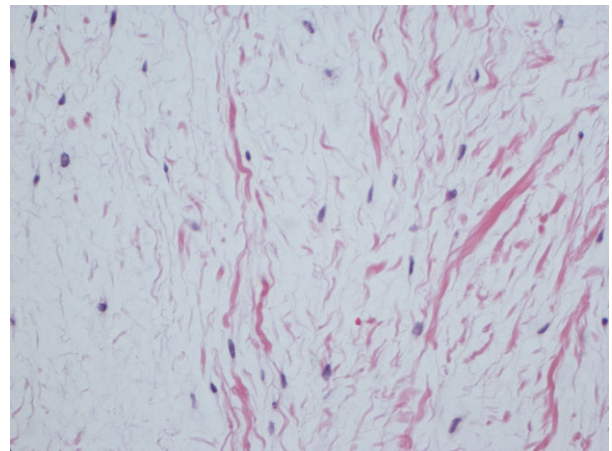


Fig. 3. Photomicrograph (High power view) showing myxoid tissue of low cellularity.

Discussion

This article presents a case of bilateral OFM on the palate of a very young child. The aetiology of OFM is unknown but is thought to be the result of an overproduction of hyaluronic acid by fibroblasts at the expense of collagen production¹⁻³. The reason for this change is not clear, but trauma may be a contributing factor³.

This current case is only the second case of OFM occurring in a child under 5 years of age and the first case of bilateral lesions on the palate. The previous case under 5 years of age was a 4-year-old child with OFM occurring on the hard palate³. There have been 5 reported cases of OFM in patients under

18 years old⁵. The present case is the sixth one in this age group.

The clinical and histological findings of this case are in correlation with those of the previous reports of OFM. Due to the uncommon occurrence and lack of distinctive clinical features, histological features are used as the basis for diagnosis. Current recommended management of OFM is surgical excision^{1–5}.

The lesions in the present case were quite large and bilateral. The other differential diagnoses should also be considered including juvenile myofibroma, pyogenic granuloma, peripheral odontogenic fibroma and peripheral giant cell granuloma. As the lesions occupied a large portion of the palate, failure to diagnose and manage them promptly may potentially lead to dysphagia and/or speech impairment. Pre-operative diagnosis of OFM is difficult due to its rare occurrence and lack of distinctive clinical features. Diagnosis is hence based solely on histopathological features. Despite its rarity, it should be considered one of the differential diagnoses for oral soft tissue lesions in the paediatric population.

Why is this clinical report important to paediatric dentists

- Although OFM is rare in children, paediatric dentists should be sufficiently knowledgeable to consider it in their differential diagnosis of oral soft tissue lesions that may present similarly in children.

Conflict of Interest

The authors declare no conflict of interest.

Acknowledgements

The authors wish to thank Dr M. Reichman, an staff oral and maxillofacial surgeon, BC Children's Hospital, Vancouver, BC, Canada, for his expertise in the surgical management of this case; Dr A. Oviedo, staff pathologist at BC Children's Hospital, Vancouver, BC, Canada, for her histopathological assistance; Dr K. Campbell, chief of dentistry, BC Children's Hospital, Vancouver, BC, Canada, for her editorial assistance of the manuscript and Dr W. Zhao, paediatric dentist, for her photographic assistance.

References

- 1 Tomich CE. Oral focal mucinosis. A clinicopathologic and histochemical study of eight cases. *Oral Surg Oral Med oral Pathol* 1974; **38**: 714–724.
- 2 Bharti V, Singh J. Oral focal mucinosis of palatal mucosa: a rare case report. *Contemp Clin Dent* 2012; **3**(Suppl. 2): S214–S218.
- 3 Gnepp DR, Vogler C, Sotelo-Avila C, Kielmovitch IH. Focal mucinosis of the upper aerodigestive tract in children. *Hum Pathol* 1990; **21**: 856–858.
- 4 Soares de Lima AA, Naval Machado MA, Martins WD *et al.* Oral focal mucinosis. *Quintessence Int* 2008; **39**: 611–615.
- 5 Lee JG, Allen G, Moore L, Gue S. Oral focal mucinosis in an adolescent: a case report. *Aust Dent J* 2012; **57**: 90–92.