CASE REPORT

Oral focal mucinosis: a rare case with literature review

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SUMMARY

Oral focal mucinosis (OFM) is an unusual disease that affects diverse localised areas of the mouth, where the connective tissue undergoes focal degeneration. It was described for the first time by Tomich in 1974. It presents as an asymptomatic pedunculated or sessile growth, commonly on the gingiva, with most cases being in women. Its pathogenesis is linked to overproduction of hyaluronic acid by fibroblasts during collagen production, ensuing in focal myxoid degeneration. It has no characteristic features and diagnosis depends on histological scrutiny. We report a rare case of oral focal mucinosis affecting a 54-year-old man who presented with a sessile gingival growth mimicking a common tumour-like lesion.

BACKGROUND

Oral focal mucinosis (OFM) is an uncommon soft tissue lesion of unidentified aetiology. Clinically, it is most commonly found on the gingiva and presents as a painless, sessile or pedunculated mass that is the same colour as the surrounding mucosa.¹ It is a disease of unknown aetiology, perhaps resulting from overproduction of hyaluronic acid by fibroblasts.² It is considered to be the oral counterpart of cutaneous focal mucinosis, which is characterised by a focal myxoid degeneration of connective tissue.^{3–4} OFM occurs largely in adults during the fourth and fifth decades of life, and more commonly on keratinised mucosa; it has also been reported, though rarely, in children and adolescents. Its diagnosis mainly relies on histological analysis and the treatment involves complete surgical excision. Its recurrence is yet to be reported.⁵

It is a rare lesion and so far only 63 cases have been reported in the English literature. The present case report adds one such rare case of OFM manifested clinically as a gingival growth and details the routine histopathological and immunohistochemical (IHC) staining that was significant in the final concluding diagnosis.

CASE PRESENTATION

A 54-year-old man presented to the department of oral medicine and diagnosis with a gingival overgrowth in the lower right back tooth region. He first noticed the growth 3 months earlier; it started out small and gradually attained its size at presentation. It was interfering with chewing and was associated with mild pain. The patient's medical, dental, family and personal histories were non-contributory. The patient was moderately built, cooperative and no gross facial asymmetry was

observed. On intraoral examination, a well-defined, pinkish, pedunculated, roughly ovoid shaped growth measuring approximately 2×1.5 cm was present on the buccal gingiva and vestibular mucosa of region 46. The tooth was completely covered by the lesion on the buccal aspect and occlusal indentations of the opposing tooth were noted on the superior surface of the lesion (figure 1). On palpation, all inspector findings were confirmed: it was firm in consistency and non-tender, and extended superiorly to the occlusal surface and inferiorly to the buccal vestibular region. On the basis of history and clinical examination, a provisional diagnosis of fibrous epulis of gingiva was given.

INVESTIGATIONS

The lesion was excised surgically under local anaesthesia using a scalpel, and sent for histopathological evaluation (figure 2). The bleeding from the surgical area was normal, and no complications were noted during the procedure. H&E-stained microscopic slides of the case revealed a stratified squamous hyper-parakeratinised epithelium and the underlying connective tissue stroma was composed of loose fibromyxoid stroma with stellate-shaped fibroblasts (figure 3). Deeper stroma showed the myxomatous stroma with plump fibroblasts and no inflammatory cells (figure 4). These histopathological features were suggestive of OFM. Further, as an adjuvant, IHC analysis was carried out for vimentin and S-100, and showed a positive reaction for vimentin (figure 5) and was negative for S-100, ruling out other myxomatous lesions of neural origin.

OUTCOME AND FOLLOW-UP

The surgical wound healed satisfactorily without any complications and with no recurrence. The patient

Figure 1 Preoperative intraoral view of the lesion.

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Figure 2 Surgically excised specimen.

has been routinely followed-up over the past 1 year with no signs of discomfort and no recurrence.

DISCUSSION

OFM is a rare disease where the connective tissue undergoes a focal myxoid degeneration.⁶ In 1966, Johnson and Helwig described an asymptomatic dome-shaped skin nodule, frequently on the face, trunk, or extremities, which they termed cutaneous facial mucinosis.⁷ The oral counterpart of the cutaneous myxoid cyst and cutaneous focal mucinosis was described by Tomich.⁸

Clinically, OFM can start in the form of small nodules up to 2 cm in diameter, rounded and asymptomatic, sessile, of firm consistency, pinkish in colour, similar to adjoining normal mucosa and, rarely, presenting secondary ulceration. Skin onsets do not appear to be associated with oral lesions. Largely, cases of OFM are described as lesions on keratinised mucosa, such as found on the gums or palate, with 80% of lesions.²⁻⁹ The aetiology remains unknown, but a possibility is an increase in the production of hyaluronic acid by fibroblasts during collagen production.⁹ The review of the available literature shows that it is most commonly diagnosed as fibroma or epulis. Radiologically, the OFM can be seen as a radiolucent homogeneous and well-defined lesion. Thus, the histopathological diagnosis becomes decisive in these conditions. Recently, immunohistochemistry has been found to provide an added advantage to the routine histopathological diagnosis.^{4-8,10} The histopathological features of OFM demonstrate a submucosal, well-localised but

Figure 4 H&E-stained photomicrograph showing the myxomatous stroma with plump fibroblasts and no inflammatory cells (×400).

non-encapsulated nidus of very loose, myxomatous or 'mucinous' connective tissue. Other superficial lesions may produce atrophy and loss of rete ridges of the overlying squamous epithelium. Fibroblasts are seen least within the mucinous area, often signifying delicate, fibrillar processes. The mucinous zone is much less vascular than the adjoining connective tissues, and inflammatory cells are not connected with the lesion except as a perivascular infiltrate of lymphocytic T-cells at the periphery.²⁻⁵

The standard therapy for OFM is surgical treatment, after which the condition has generally not been seen to recur.³ Since the first report by Tomich in 1974, approximately 57 cases of OFM have been recorded in the English literature, of which only one case by Narayana and Casey noted recurrence in a 25-year-old woman.¹⁰

The present case report provides a thorough knowledge of this odd lesion. It must be emphasised that in many focal gingival lesions, clinically preoperative diagnosis is not possible due to their scarcity. However, OFM should be considered in the differential diagnosis of soft tissue lesions in adults. A review of many cases revealed that there has previously never been a clinical diagnosis of 'oral focal mucinosis' and that the histological characteristics are the basis for the diagnosis, indicating the need for biopsy to be performed well and the importance of pathological examination to pinpoint the diagnosis.

Figure 3 H&E-stained photomicrograph showing the surface epithelium with underlying myxoid stroma (×100).

Figure 5 Immunohistochemically-stained fibroblasts showing positivity for vimentin in the stroma (×100).

Learning points

- ▶ Most focal gingival lesions are difficult to diagnose accurately without histological evaluation of the tissue.
- ▶ It must be emphasised that in many cases, histopathological and immunohistochemical evaluation serve as the basis for diagnosis.
- ▶ Oral focal mucinosis is an uncommon condition and should be included in the differential diagnosis of tumour-like lesions even though the treatment and clinical behaviour may be similar to other reactive lesions.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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