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CASE REPORT

Ligneous alveolar gingivitis in the absence of plasminogen deficiency

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A case of localized, longstanding, asymptomatic ligneous gingivitis affecting the crest of the edentulous lower left posterior alveolar ridge (ligneous alveolar gingivitis) of a middle-aged Caucasian woman is presented. This patient did not have any associated ophthalmic lesions (ligneous conjunctivitis) and did not have a plasminogen deficiency.
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Keywords: amyloid; conjunctivitis; fibrin; gingival accumulation; ligneous alveolar gingivitis; plasminogen

Case report

A 46-year-old female was referred to the Oral Medicine Department, Glasgow Dental Hospital, for assessment of a red area on the lower left alveolar ridge. The patient stated that the lesion had been present for several years and had been completely painless.

Her past medical history was clear with no known allergies and she was taking diclofenac sodium for an ankle injury. The patient was a non-smoker and drank alcohol socially.

On examination she was partially dentate with a granular, ulcerated, erythematous area on the alveolar ridge in the lower left molar region (Fig. 1), no other intra-oral lesions were detected. On further questioning the patient reported that 36 and 37 had been extracted several years previously. Incisional biopsy of the lesion was performed under local anaesthesia and submitted for histopathological examination.

Histological examination of the tissue revealed a lightly keratinized epithelium, which was atrophic in parts and acanthotic in others. There was also an area of non-specific chronic ulceration. A scattered chronic inflammatory cell infiltrate was present. Within the upper lamina propria were areas of hyalinization (Fig. 2), the configuration of which was suggestive of blood vessel walls. The material within the lamina propria did not stain with congo red or Van Giesen histochemical stains indicating that the material was neither amyloid nor collagen. MSB Trichrome stained some of the material red/pink, indicative of fibrin (Fig. 3); however, much of the material did not stain red/pink, which may indicate the presence of other protein components. Immunohistochemistry showed positive staining for both CD31 (Fig. 4) and CD34 (not shown) highlighting the presence of endothelial cells within the hyalinized areas, while staining for factor VIII was strongly positive (Fig. 5), consistent with a vascular origin.

On reviewing the patient 3 weeks after the biopsy the area of the surgery had healed uneventfully but the lesion had persisted. The patient was questioned regarding any visual disturbances and ophthalmic examination revealed no pathology. On further follow up 3 months later the lesion remained unchanged. Haematological investigations were performed to exclude a plasminogen deficiency as this has been reported in other cases of ligneous gingivitis (1) as well as ligneous conjunctivitis (2). Unlike the other reports, our patient was not found to be plasminogen-deficient, with a value of 90 U/dl (normal range: 73–140).

Comments

Ligneous gingivitis is a rare disorder characterized by generalized gingival enlargement, caused by amyloid-like material deposits in the subepithelial connective tissue (3, 4). The condition has been associated with ligneous conjunctivitis, which is a rare autosomal recessive form of chronic membranous conjunctivitis sometimes seen with associated lesions in the larynx, nose, cervix and gingivae (5, 6).

The aetiology and pathogenesis of these oral lesions remains unclear, but an uncontrolled immune-mediated inflammatory process possibly triggered by minor trauma, together with abnormal epithelial cell

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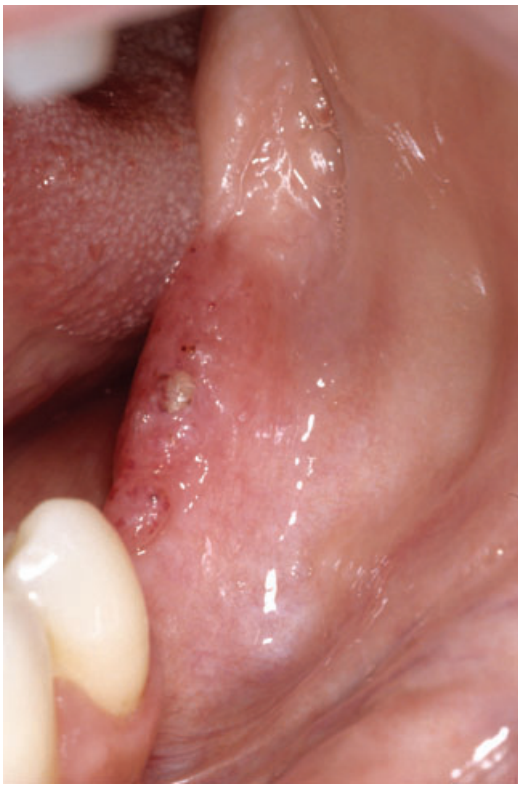

Figure 1 Clinical picture of the ligneous lesion in the lower left molar region.

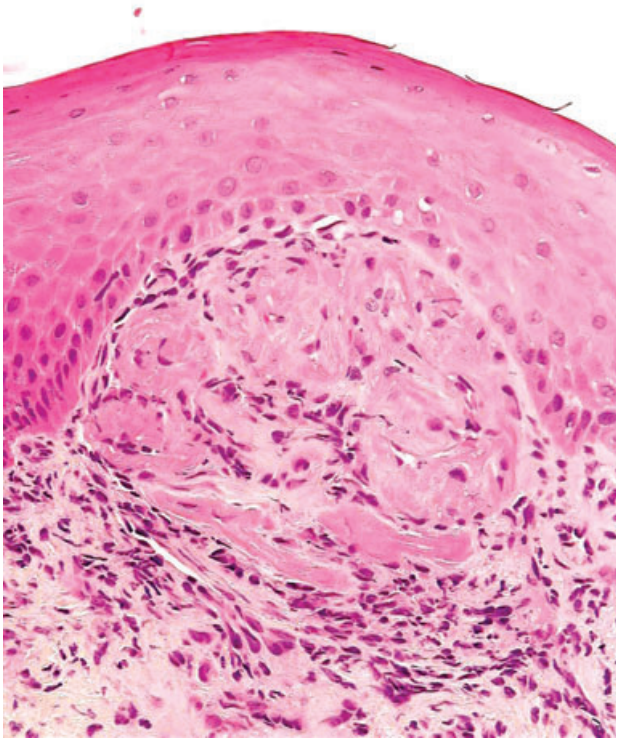

Figure 2 Haematoxylin and eosin-stained histological section of the ligneous lesion (×200).

destruction, fibrin leakage and defective fibrin breakdown may be the cause (7). Scully et al. (1) suggested drugs as another possible aetiological factor.

The case presented in this report is unusual in a number of aspects. In previously reported cases, the lesions affected mucosa associated with teeth, most often in more than one quadrant (3). However, this lesion was localized and found on alveolar mucosa that was, at the time of presentation, not associated with teeth; thus the name ligneous alveolar gingivitis is suggested.

Previous investigations have shown that both ligneous gingivitis and conjunctivitis may be associated with a plasminogen deficiency resulting in fibrin deposition in the connective tissues (1). In the case presented the general histopathological features, together with the negative congo red and Van Giesen stains, and the positive MSB Trichrome, factor VIII and CD31/CD34 immunostaining, are consistent with those previously described in the lesions of ligneous conjunctivitis and ligneous gingivitis (3, 4). However, whilst Scully et al. state that the amyloid-like material was mostly fibrin, other components have been demonstrated in conjunctival lesions, including immunoglobulin and albumin (1). The fact that in our case only some of this material stained positively for fibrin may indicate other components are also present, but these have not been identified. Additionally, unlike the other cases reported previously, this patient was not found to have a plasminogen deficiency.

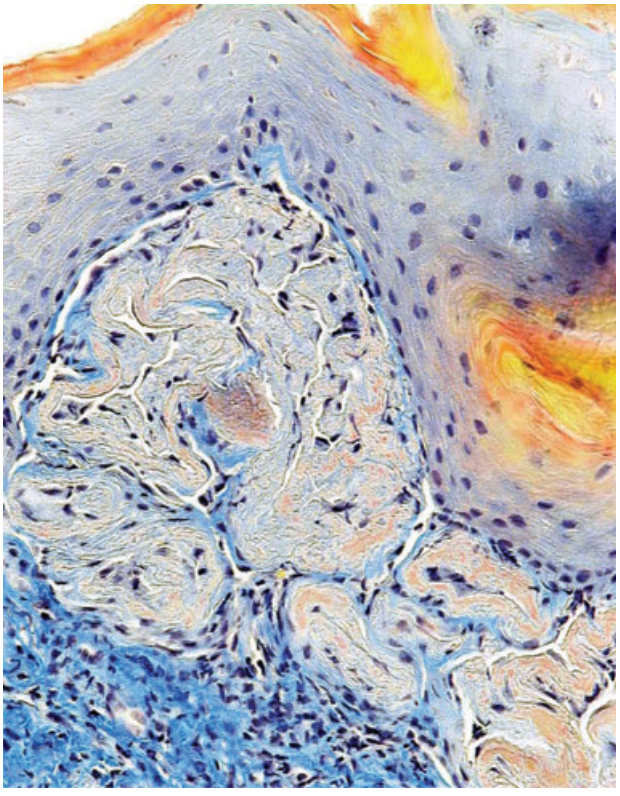

Figure 3 MSB Trichrome-stained histological section of the ligneous lesion (×200).

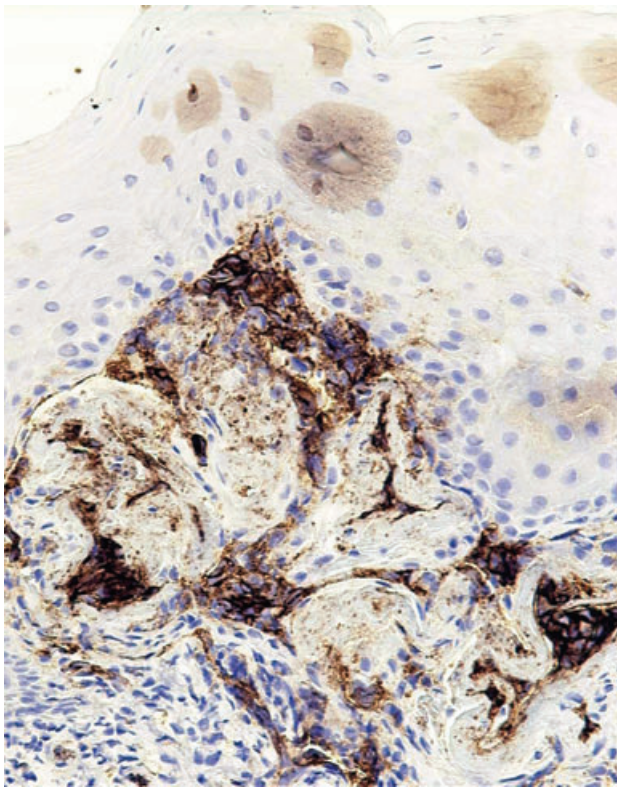

Figure 4 Immunohistochemistry staining showing positivity for CD31 (×200).

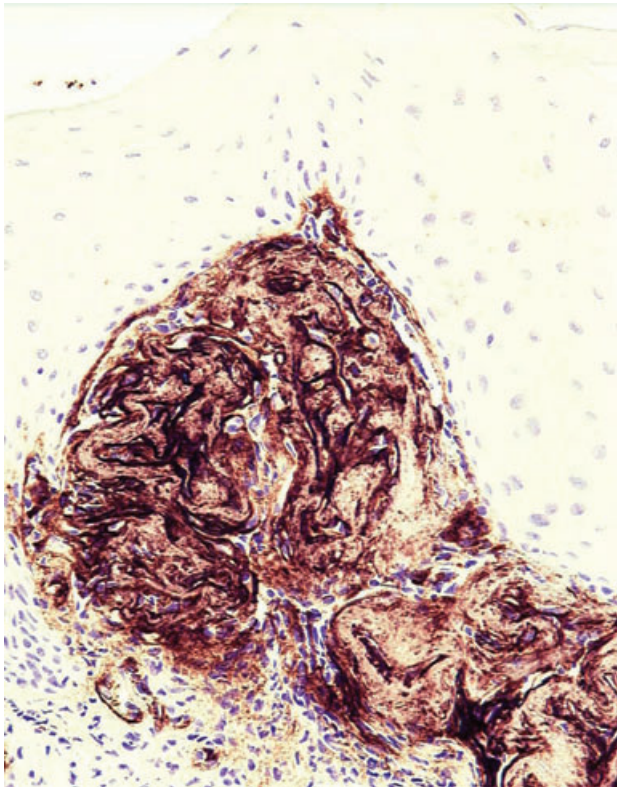

Figure 5 Immunohistochemistry staining showing strong positivity for factor VIII (×200).

Conclusion

This case report highlights the fact that ligneous gingivitis is not always associated with plasminogen deficiency. However, it is unclear whether either functional defects in the plasminogen cascade or deficiencies in other regulatory elements in the pathways have contributed to the pathogenesis of this lesion. It may also present as a persistently localized rather than a generalized lesion (3), and does not necessarily have to be associated with teeth as was reported in previous studies (7).

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Acknowledgement

It is with deep regrets that the Editor of this Journal has learned that Dr D. G. MacDonald, Glasgow Dental Hospital and School, has passed away.