The Journal of Laryngology & Otology (2008), ***122***, *1112–1114.*

doi:10.1017/S0022215107000631

Clinical Record

An unusual presentation of oropharyngeal mucosal plasmacytosis related to toothpaste

D C Tong, M R Leaper*, A N Colquhoun†, A M Rich

Abstract

Objective: We present the case of a 59-year-old Chinese patient with an unusual presentation of mucosal plasmacytosis involving the oropharynx, related to the use of toothpaste.

Method: Case presentation and review of English medical literature involving mucosal plasmacytosis.

Results: Mucosal plasmacytosis is an uncommon disease process and has been associated with hypersensitivity reactions. Most cases involve the gingival mucosa, although there have been reports of cases involving other oral mucosal sites and the upper aerodigestive tract. Our case provides an example of oropharyngeal plasmacytosis related to toothpaste. A resolution of signs and symptoms followed withdrawal of the suspected allergens.

Conclusion: Mucosal plasmacytosis is a benign inflammatory process that may appear to be more sinister on clinical examination. Skin patch testing is a useful adjunct in confirming the diagnosis.

Key words: Oral Cavity; Mucosa, Hypersensitivity; Toothpaste

Introduction

Mucosal plasmacytosis is an uncommon, non-neoplastic condition characterised by an intense plasma cell proliferation in the affected tissue.

The aetiology and pathogenesis of this condition are often unknown. We present an unusual case of oropharyngeal mucosal plasmacytosis related to the use of toothpaste.

Case report

A healthy, 59-year-old Chinese man presented to the ENT out-patient clinic with a two-month history of persistent and progressive hoarse voice, which had failed to improve after two courses of oral antibiotics from his general medical practitioner.

In addition to his hoarseness, the patient had a dry, non-productive cough and subjective difficulty in swallowing solid food. There was no associated pain or other symptoms. The patient was a lifelong non-smoker and non-drinker.

Examination revealed a diffusely oedematous supraglottic area involving the epiglottis and both aryepiglottic and median glossoepiglottic folds. There was no surface ulceration (Figure 1).

Haematological investigations were normal, including white cell count and erythrocyte sedimentation rate.

An initial clinical diagnosis of laryngopharyngeal reflux was made and treated with a proton pump inhibitor.

No changes were noted at a two-month review appointment, and oral fluconazole was prescribed to cover the possibility of laryngeal candidiasis.

A further review four weeks later showed no improvement. The patient therefore underwent an elective examination and biopsy under general anaesthesia. The biopsy showed florid inflammatory changes, including a dense, mixed inflammatory infiltrate involving the full thickness of the epithelium and a dense, lymphoplasmocytic infiltrate within the subepithelial connective tissue.

The lymphoid cells were a mix of CD20-positive B-cells and CD3- and CD4-positive T-cells, suggesting a benign, reactive tissue process.

Lymphoma was excluded following further investigation with flow cytometry.

A revised diagnosis of mucosal plasmacytosis was made based on the histopathological findings.

Independently, the patient had been referred to the oral and maxillofacial surgery unit by his dentist for evaluation of raised, erythematous lesions in the left maxillary buccal sulcus and alveolar mucosa (Figure 2). The lesions did not involve the attached gingiva. A biopsy was performed, which showed sheets of chronic inflammatory cells, predominately plasma cells, in the fibrous stroma of the specimen (Figure 3). The plasma cells were uniform and no significant mitotic activity was observed. The surface was partly covered by oedematous epithelium with focal areas of ulceration. Immunochemistry studies showed that the plasma cells were polyclonal for kappa and lambda light chains. A diagnosis of oropharyngeal mucous membrane plasmacytosis was made.

A possible association with exposure to cinnamon or mint found in toothpaste was explored. The patient was advised to stop using toothpaste when brushing his teeth and to augment his oral hygiene with chlorhexidine mouthwash.

A subsequent review showed a dramatic improvement, with complete resolution of the oropharyngeal symptoms (Figure 4).

From the Department of Oral Diagnostic and Surgical Sciences, School of Dentistry, University of Otago, Dunedin, the *Department of Otolaryngology, Dunedin Hospital, and the †Department of Oral and Maxillofacial Surgery, Waikato Hospital, Hamilton, New Zealand.
Accepted for publication: 11 June 2007. First published online 25 September 2007.

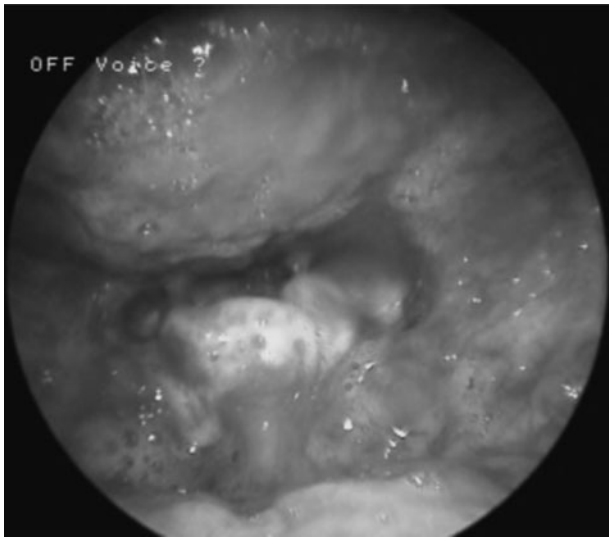

FIG. 1
Endoscopic view of diffuse swelling involving the epiglottis and aryepiglottic and glossoepiglottic folds.

Skin patch testing was requested in order to confirm and investigate other potential causative agents; this included exposure to antimicrobials, perfumes and flavourings, especially those commonly found in toothpaste. The results showed significant reactions to cinnamaldehyde, cinnamyl alcohol and a fragrance mix containing cinnamaldehyde.

Twelve months later, the patient had continued to avoid brushing with toothpaste and remained symptom-free.

Discussion

Twenty to 30 years ago, there were a number of reports of patients presenting with a sudden onset of painful, erythematous gingivae, which when biopsied were shown to have a dense, polyclonal, plasmacytic infiltrate in the connective tissue.[1,2] This condition was known by a variety of terms, particularly plasma cell gingivitis,[3,4] and, at least in some instances, resolved after the withdrawal of allergens.[4,5] Hypersensitivity to components of chewing gum was implicated. Once the putative allergens were removed from this product, the number of cases of plasma cell gingivitis reduced, although sporadic cases were still reported.[6,7]

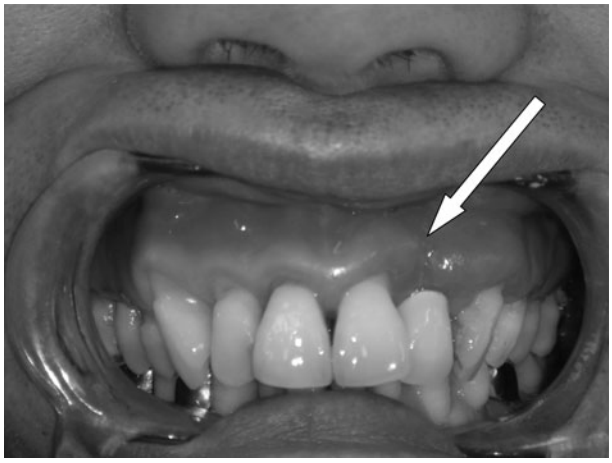

FIG. 2
Clinical photograph of intraoral involvement, showing swollen alveolar mucosa. The arrow indicates the alveolar gingival tissue biopsy site.

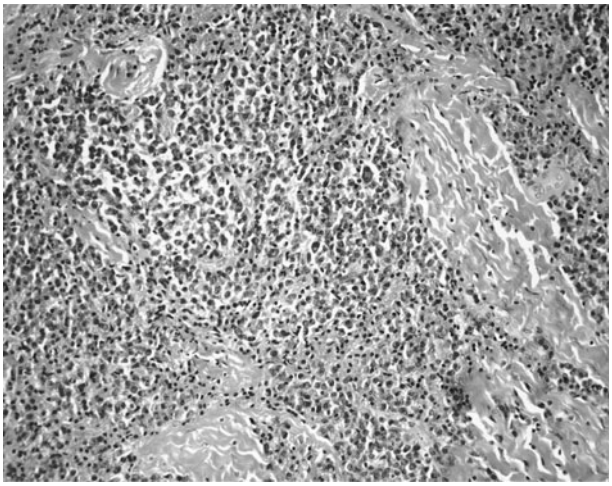

FIG. 3
Photomicrograph of biopsy, showing sheets of plasma cells throughout the submucosa (H&E; ×20).

Plasma cell lesions involving other oral mucosal and upper aerodigestive tract sites have been less frequently reported. These more extensive lesions have been reported in the pharynx, palate, buccal mucosa, tongue, larynx and trachea, and may or may not be accompanied by gingival lesions.[8,9] This condition has been termed plasma-cell orificial mucositis, plasma cell mucositis or mucous membrane plasmacytosis.[8,10,11] It affects middle-aged people, with a male predominance. The usual description is of a red, slightly elevated, 'cobblestone' lesion, without ulceration, which may be associated with discomfort in the region, dysphonia, dysphagia and difficulty breathing.[9,10]

The presenting symptoms and the clinical appearance in the current case fit this description. The histology in the present case also fits the previous reports of oedematous epithelium overlying connective tissue with an intense infiltrate of mature plasma cells.

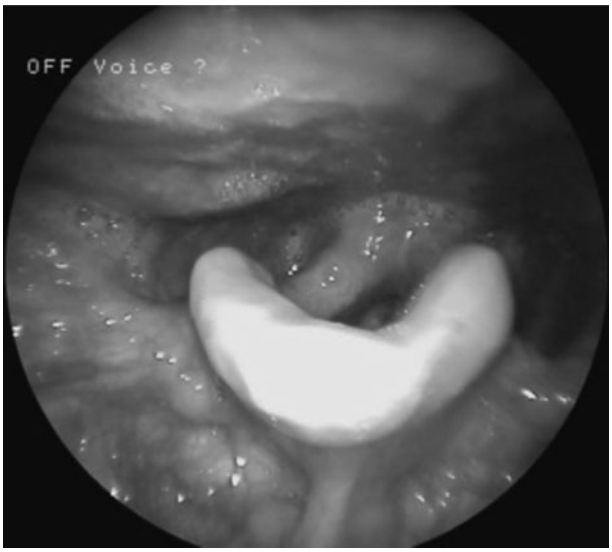

FIG. 4
Endoscopic view of supraglottic region, showing resolution of signs following withdrawal of suspected allergens.

https://doi.org/10.1017/S0022215107000631 Published online by Cambridge University Press

In such cases, a neoplastic plasmacytic proliferation should be ruled out by demonstrating the polyclonal nature of the infiltrate, or via gene rearrangement studies.

- **Mucosal plasmacytosis is an uncommon, non-neoplastic condition characterised by an intense plasma cell proliferation in the affected tissue**
- **This paper describes the case of a 59-year-old male patient with an unusual presentation of mucosal plasmacytosis involving the oropharynx, related to the use of toothpaste**
- **The likelihood of an allergic aetiology in this case was strengthened by the strong reaction to skin testing of cinnamon products**

Mucous membrane plasmacytosis is not reported to progress to malignancy.[9] However, since the condition is rare, there are no long-term studies with large numbers, and regular clinical review is recommended.

Unlike plasma cell gingivitis, in which the link with allergic reactions is well established, the association between mucous membrane plasmacytosis and allergy is less well recognised. The present case is unusual in that resolution of the mucous membrane plasmacytosis followed withdrawal of the suspected allergens, without further medication. The likelihood of an allergic aetiology in this case was strengthened by the strong reaction to skin testing of cinnamon products. Previous reports have described the use of corticosteroids, antibiotics and surgical debulking procedures, with inconsistent results.[9]

Conclusion

This report adds to the literature another case of the rare plasma cell disorder mucous membrane plasmacytosis. It also demonstrates an association with cinnamon products, acting as putative allergens.

References

1 Owings JR. An atypical gingivostomatitis: a report of four cases. *J Periodontal* 1969;**40**:538–42
2 Perry HO, Deffner NF, Sheridan PJ. Atypical gingivostomatitis. Nineteen cases. *Arch Dermatol* 1973;**107**:872–8
3 Lubow RM, Cooley RL, Hartman KS, McDaniel RK. Plasma cell gingivitis. Report of a case. *J Periodontol* 1984;**55**:235–24
4 MacLeod RI, Ellis JR. Plasma cell gingivitis related to the use of herbal toothpaste. *Br Dent J* 1989;**166**:375–6
5 Kerr DA, McClatchey KD, Regezi JA. Allergic gingivostomatitis (due to chewing gum). *J Periodontol* 1971;**42**:709–12
6 Silverman S, Lozada F. An epilogue to plasma cell gingivostomatitis (allergic gingivostomatitis). *Oral Surg Oral Med Oral Pathol* 1977;**43**:211–17
7 Sollecito TP, Greenberg MS. Plasma cell gingivitis. Report of two cases. *Oral Surg Oral Med Oral Pathol* 1992;**73**:690–3
8 White JW, Olsen KD, Banks PM. Plasma cell orificial mucositis. Report of a case and review of literature. *Arch Dermatol* 1986;**122**:1321–4
9 Dermatology Online Journal. http://dermatology.cdlib.org/95/case_reports/plasmacytosis/bharti.html [20 September 2006]
10 Ferreiro JA, Egorshin EV, Olsen KD, Banks PM, Weiland LH. Mucous membrane plasmacytosis of the upper aerodigestive tract. A clinicopathologic study. *Am J Surg Pathol* 1994;**18**:1048–53
11 Kaur C, Thami GP, Sarkar R, Kanwar AJ. Plasma cell mucositis. *J Eur Acad Dermatol Venereal* 2001;**15**:566–7

Address for correspondence:
Mr D C Tong,
Consultant Oral and Maxillofacial Surgeon,
Dept of Oral Diagnostic and Surgical Sciences,
PO Box 647,
Dunedin, New Zealand.

Fax: 64 3 479 3937
E-mail: darryl.tong@stonebow.otago.ac.nz

Mr D C Tong takes responsibility for the integrity of the content of the paper.
Competing interests: None declared