Unusual presentation of more common disease/injury

Glycogenic acanthosis presenting as leukoplakia on the tongue

L Montebugnoli,¹ L Felicetti,¹ F Cervellati,¹ M P Foschini²¹ Department of Oral Science, University of Bologna, Bologna, Italy² Section of Anatomic and Histopathology, Department of Haematology and Oncology 'L. and A. Seragnoli', University of Bologna, Bellaria Hospital, Bologna, Italy**Correspondence to** Lucio Montebugnoli, lucio.montebugnoli@unibo.it**Summary**

This report describes a rare case of glycogenic acanthosis (GA) located on the tongue of a 72-year-old man. The patient presented with a white plaque that had been on the right side of the tongue for 5 years. Histologically, the lesion was characterised by thickened squamous epithelium with abundant intracellular glycogen deposits. No epithelial dysplasia was noted. This is the first report of GA involving the tongue. This benign condition should be added to the large number of lesions responsible for leukoplakia in this anatomical location.

BACKGROUND

White lesions of the oral mucosa must be carefully investigated and oral leukoplakia properly diagnosed, since it is considered a risk factor for oral cancer.¹

Oral leukoplakia is a focal white plaque varying in size and shape without any specific clinical or histological characteristic. Hence the diagnosis of leukoplakia is established by a process of exclusion, ruling out all other causes of white lesions such as candidiasis, white sponge nevus, leukoedema, frictional keratosis, etc.²

Glycogenic acanthosis (GA) is a benign lesion characterised by small white raised plaques commonly detected in the lower third of the oesophagus.³ On histology GA is composed of thickened squamous epithelium, with glycogen-rich keratinocytes.⁴

This report describes a case of GA presenting as a white plaque of the tongue requiring differentiation from oral leukoplakia.

CASE PRESENTATION

A 72-year-old man was referred to our department for evaluation of a white lesion on the right side of the tongue. The lesion had been present for 5 years and appeared as a 4 cm area of plaque-like thickening of the mucosa much whiter than the surrounding tissue (figure 1). The patient enjoyed otherwise good health. His personal and family histories were unremarkable. In particular, there was no history of smoking or local trauma and no other oral lesions were detected at clinical examination.

Figure 1 Lesion appearing as a 4 cm area of plaque-like thickening of the mucosa on the right side of the tongue.

Figure 2 A. Histologically the lesion presented as a slightly raised plaque composed of epithelium with basal cell hyperplasia and clear keratinocytes filled with glycogen (magnification 20 $\times$), as demonstrated with periodic acid–Schiff (PAS) stain (B, magnification 100 $\times$). Immunohistochemistry with anti-human papilloma virus (HPV) antibody gave negative results (inset positive control consisted of vulvar condyloma) (C, magnification 100 $\times$).

A first biopsy, performed at presentation, failed to disclose any dysplastic or malignant features. The lesion had been slightly enlarging during the last 5 years so a further biopsy was performed to exclude the development of dysplasia and to investigate the possibility of any other disease responsible for the white plaque.

The biopsy material was fixed in buffered formalin and paraffin embedded. Serial sections were stained with H&E and periodic acid–Schiff (PAS) stain with and without diastase digestion to highlight the intracytoplasmic glycogen. In addition immunohistochemical staining was performed to disclose possible infection by human papilloma virus (HPV, monoclonal, clone K1HB, diluted 1:100; Thermo Scientific, Waltham, MA, USA). The first biopsy was retrieved and the histological findings compared. Both showed the same histological picture and therefore will be described together.

The histological sections showed thickened squamous epithelium with basal hyperplasia and acanthosis (figure 2A). The cells of the prickle layer had clear cytoplasm filled with glycogen as demonstrated on PAS staining (figure 2B). There was no evidence of epithelial dysplasia according to the criteria defined by the WHO blue book.⁵ No inflammatory infiltration or fungal structures were present. Immunohistochemistry failed to reveal evidence of HPV infection (figure 2C).

DISCUSSION

The histological features observed in the tongue lesion of our patient are the same as those observed in oesophageal GA. GA of the oesophagus was first described by Rywlin and Ortega in 1970.⁶ GA is commonly encountered during endoscopic procedures in older patients^{3 7} and is characterised by multifocal slightly elevated whitish plaques located in the lower oesophageal third. Histologically, the lesion is composed of hyperplastic squamous epithelium with intracytoplasmic glycogen.⁶ GA has no malignant potential as reflected in our case in which the lesion had been present for 5 years with no features of malignant transformation. Lesions similar to GA may be associated with impaired glucose metabolism as in diabetes or glycogenosis, but our patient had neither of these conditions.⁸

Despite their common occurrence in the oesophagus, glycogenic plaques have seldom been detected in other locations. A recent case report described GA in the larynx as an occasional finding during autopsy.⁹ Features similar to those of GA have been reported in patients with Cowden syndrome,¹⁰ a rare genetic disorder characterised by multiple hamartomas affecting various organs,¹¹ but our patient did have any other mucosal or cutaneous lesions indicative of Cowden syndrome. In addition, most cases with this condition reported so far had multiple and small lesions, and the paucity of the plaques in this case together with the large diameter is even more unusual.

In conclusion, our patient presented the typical histological features of oesophageal GA that led us to establish a diagnosis of GA of the tongue. This diagnosis should be added to the many lesions presenting as whitish plaque and included in the differential diagnosis of oral leukoplakia.

Competing interests None.

Patient consent Obtained.

REFERENCES

1. **Lodi G**, Porter S. Management of potentially malignant disorders: evidence and critique. *J Oral Pathol Med* 2008;**37**:63–9.
2. **Warnakulasuriya S**, Johnson NW, van der Waal I. Nomenclature and classification of potentially malignant disorders of the oral mucosa. *J Oral Pathol Med* 2007;**36**:575–80.
3. **Stern Z**, Sharon P, Ligumsky M, *et al*. Glycogenic acanthosis of the esophagus. A benign but confusing endoscopic lesion. *Am J Gastroenterol* 1980;**74**:261–3.
4. **Fenoglio-Preyser CM**, Noffsinger A, Stemmermann GN, *et al*. The non-neoplastic esophagus. *Gastrointestinal pathology, an atlas and text*. 3rd edn. In Kluwer W, ed. Chapter 2. Philadelphia, Pennsylvania, USA: Lippincott, William & Wilkins, 2008:79.
5. **El Naggar AK**, Reichart PA. Proliferative verrucous leukoplakia and precancerous conditions. In: Barnes L, Eveson JW, Reichart P, Sidranski D, eds. *Pathology and genetics head and neck tumours*. Lyon: World Health Classification of Tumours, 2005:180–1.
6. **Rywlin AM**, Ortega R. Glycogenic acanthosis of the esophagus. *Arch Pathol* 1970;**90**:439–43.
7. **Glick SN**, Teplick SK, Goldstein J, *et al*. Glycogenic acanthosis of the esophagus. *AJR Am J Roentgenol* 1982;**139**:683–8.
8. **Ghahremani GC**, Ruschovich AM. Glycogenic acanthosis of the oesophagus: radiographic and pathologic features. *Gastrointest Radiol* 1984;**9**:93–8.
9. **Fyfe BS**, Garcia FU. Laryngeal glycogenic acanthosis presenting as leukoplakia. *Arch Otolaryngol Head Neck Surg* 1998;**124**:1029–30.
10. **Nishizawa A**, Satoh T, Watanabe R, *et al*. Cowden syndrome: a novel mutation and overlooked glycogenic acanthosis in gingiva. *Br J Dermatol* 2009;**160**:1116–18.
11. **Eng G**. PTEN: one gene, many syndromes. *Hum Mutat* 2003;**22**:183–98.

This pdf has been created automatically from the final edited text and images.

Copyright 2010 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit

<http://group.bmj.com/group/rights-licensing/permissions>.

BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Please cite this article as follows (you will need to access the article online to obtain the date of publication).

Montebugnoli L, Felicetti L, Cervellati F, Foschini MP. Glycogenic acanthosis presenting as leukoplakia on the tongue. *BMJ Case Reports* 2010; 10.1136/bcr.01.2010.2634, date of publication

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow