

Letter to Editor

Solitary Tongue Nodule in a Case of Pemphigus Vulgaris: A Diagnostic Enigma

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Dear Editor,

A 50-year-old male patient presented with a 5-year history of recurrent bullae and erosions on the skin and oral cavity. Despite prior treatment with oral steroids and immunomodulators, his disease remained refractory. A biopsy and direct immunofluorescence performed at our institute confirmed a diagnosis of pemphigus vulgaris (PV). At this point in time, the Autoimmune Bullous Skin Disorder Intensity Score was 42.5. Hence, the patient was transitioned to rituximab infusions along with a low dose of oral steroids. Approximately four weeks after the infusion, he reported a smooth, reddish, broad-based, firm nodule, occasionally bleeding about 2 × 2 cm in diameter on the dorsal aspect of his tongue. Furthermore, there was a thick whitish scrapable coating on the tongue, partially obscuring the nodule [Figure 1a]. At this juncture, the activity of pemphigus was well controlled. Differential diagnoses included hemangioma, pyogenic granuloma (PG), fibroma and Kaposi sarcoma. An excisional biopsy of the nodule revealed numerous endothelium-lined congested vascular spaces with partly ulcerated lining epithelium suggestive of PG. A single row of keratinocytes reminiscent of tomb stone appearance of PV was seen overlying the vascular proliferation. Dense fibrotic stroma and chronic inflammation are also present [Figure 1b]. A diagnosis of PG with coated tongue was made. Six months later, the patient did not experience a recurrence of the excised nodule [Figure 1c].

The case emphasises the rarity of PG in PV patients, with limited documented cases in English language. Previous reports, such as those by Burgan *et al.* and Fahmy *et al.*, describe similar occurrences in younger patients, typically presenting with multiple nodules.^[1,2]

Therefore, our case represents only the second reported instance in an adult patient.^[3] Unlike previous cases, which presented with multiple PG lesions, our case exhibited a different clinical pattern.

Cawson categorised oral PG histopathologically into two types: Lobular capillary hemangioma (LCH) and non-LCH. In our case, it exhibited characteristics more akin to the LCH type.^[4] The pathogenesis of PG in PV patients is hypothesised to be linked to localised micro-injuries in the oral epithelium due to inflammatory responses, loss of keratinocyte adhesion and tissue protein alterations. In addition, poor oral hygiene leading to bacterial colonisation and immunosuppressive drug use may contribute to overzealous proliferation of vascular connective tissue.^[5]

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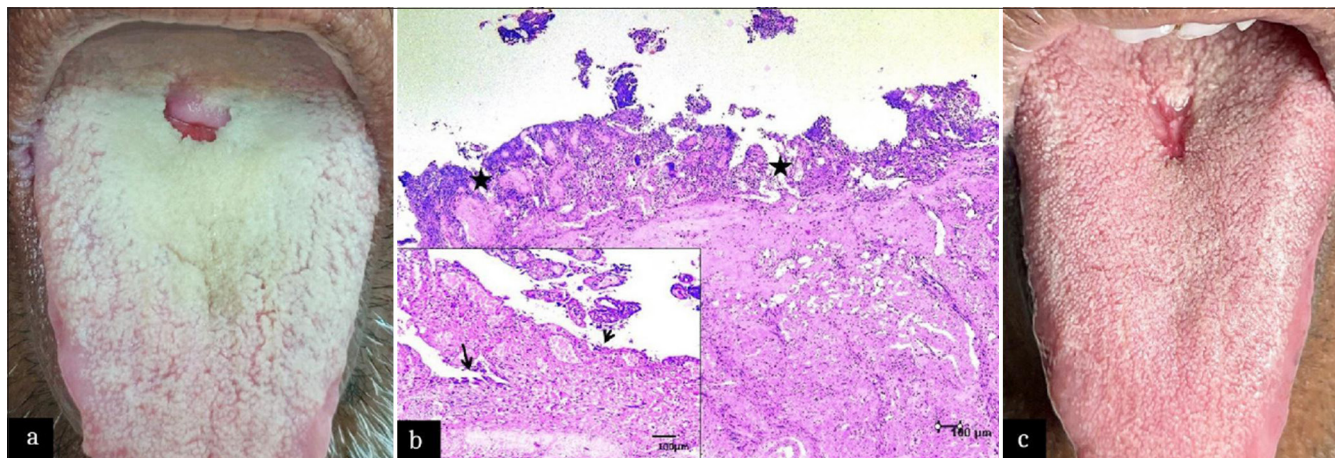


Figure 1: (a) Solitary tongue nodule 2×2 cm on the dorsal aspect of the tongue along with thick whitish coating all over the tongue. (b) Section shows numerous endothelia lined congested capillary sized blood vessels (black stars) with underlying fibrotic tissue (Haematoxylin and Eosin, $\times 50$). Inset shows single row of tombstone-like keratinocytes overlying the vascular proliferation (black arrows) Haematoxylin and Eosin, $\times 100$. (c) Six months after excisional biopsy resolution of tongue nodule.

Complete surgical excision is the treatment of choice for such lesions. To reduce chances of bleeding and in non-responding cases, various lasers such as neodymium-doped yttrium aluminium garnet, flash lamp-pumped pulsed dye laser or cryotherapy can be used.^[6]

Reviewing the few cases that have been published earlier shows a pattern for predicting oral PGs in patients with oral pemphigus. First, PGs often develop in long-standing oral pemphigus. In our patient, PG appeared 5 years after pemphigus diagnosis, and in other reports, it was 1–3 years. Poorly controlled pemphigus and concurrent skin diseases as in our patient may increase the risk of developing PG.

The PG's atypical central tongue location in this patient, lack of antecedent trauma and the presence of tombstone-like basal keratinocytes overlying lobular capillary proliferation on histopathology strongly support its origin in PV-affected mucosa rather than as a trauma-induced lesion.

To sum up, PG should be considered in the differential diagnosis for a patient with oral pemphigus presenting with a rapidly growing, ulcerative and haemorrhagic oral nodule.

Ethical approval: Institutional Review Board approval is not required.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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