

## CASE REPORT

# Plasma cell granuloma of lip

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Received : 14-01-11  
Review completed : 07-10-11  
Accepted : 27-01-12

### ABSTRACT

Plasma cells are medium-sized round-to-oval cells with eccentrically placed nuclei, usually found in the red pulp of the spleen, tonsils, medulla of the lymph nodes, nasal mucosa, upper airway, lamina propria of the gastrointestinal tract, and sites of inflammation. Plasma cell granuloma is a rare reactive tumor-like proliferation composed chiefly of plasmacytic infiltrate. Here, we present a case of plasma cell granuloma of lip in a female patient.

**Key words:** Plasma cells, plasma cell granuloma, lip

Plasma cells are medium-sized round-to-oval cells with eccentrically placed nuclei. The cells range in size from 10 to 20  $\mu$ m and the nuclear: Cytoplasmic ratio is 1:2. The nuclear chromatin is often arranged in a cartwheel-like or clock-face pattern.<sup>[1]</sup> Plasma cells are usually found in the red pulp of the spleen, tonsils, medulla of the lymph nodes, nasal mucosa, upper airway, lamina propria of the gastrointestinal tract, and sites of inflammation. Their main function is to produce immunoglobulins or antibodies.<sup>[2]</sup>

Plasma cell granuloma is a rare, reactive, tumor-like proliferation composed chiefly of plasmacytic infiltrate.<sup>[3]</sup> It was first described by Bahadori and Liebow in 1973.<sup>[4]</sup> Plasma cell granuloma has been known by different terms, e.g., inflammatory pseudotumor, inflammatory myofibroblastic tumor, inflammatory myofibrohistiocytic proliferation, and xanthomatous pseudotumor.<sup>[2]</sup>

The lung is the most common site of involvement, although it may occur in any organ.<sup>[5]</sup> Plasma cell granuloma of the oral cavity is seen primarily in the periodontal tissues.<sup>[6]</sup> Other than the gingiva it may also be seen on the tongue,<sup>[7]</sup> lip,<sup>[8]</sup> buccal mucosa,<sup>[9]</sup> palate<sup>[10]</sup> and submandibular gland.<sup>[11]</sup>

The etiology of this lesion is still unknown. Hypotheses have suggested that the pathogenesis has infectious, autoimmune, and vascular origins.<sup>[12]</sup>

Arber *et al.* recently detected Epstein-Barr virus (EBV) association in 7 of 18 cases of inflammatory pseudotumor arising in the lymph node, spleen, and liver, and suggested that EBV might play a role in the pathogenesis of plasma cell granuloma.<sup>[13]</sup> Ballesteros *et al.*, however, failed to detect EBV genomes in the oral cavity of these patients by *in situ* hybridization.<sup>[14]</sup>

### CASE REPORT

A 55-year-old female patient reported to our private dental clinic with a complaint of ulceration in both upper and lower lips for the past 6 months. The lesion on the lip crossed the midline and involved the vermilion and mucosa of the lip [Figure 1]. Medical history was not significant except for history of tobacco chewing for the past 15 years; she said that she used to pouch the tobacco in the lower vestibular sulcus for nearly half an hour before swallowing it.

We made a provisional diagnosis of allergic stomatitis. A complete hemogram was done, which showed all blood counts to be within normal limits. Urine examination was normal. Investigations oriented to viral infection were not conducted due to absence of prodromal symptoms. Due to inflamed appearance of the lesions, an incisional type of biopsy was planned and carried out after the routine preliminary investigations had been done.

The hematoxylin and eosin-stained soft tissue section showed the lesional tissue covered by ulcerated stratified squamous epithelium. The fibrovascular connective tissue with minimal stroma exhibited a dense, diffuse, mixed inflammatory cell infiltrate, composed predominantly of

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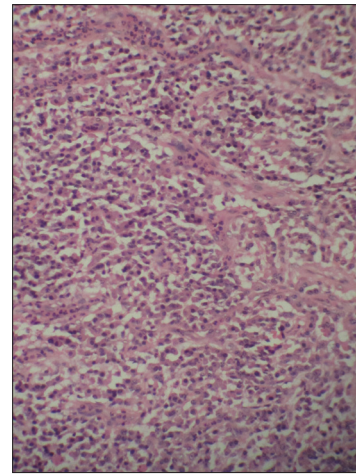


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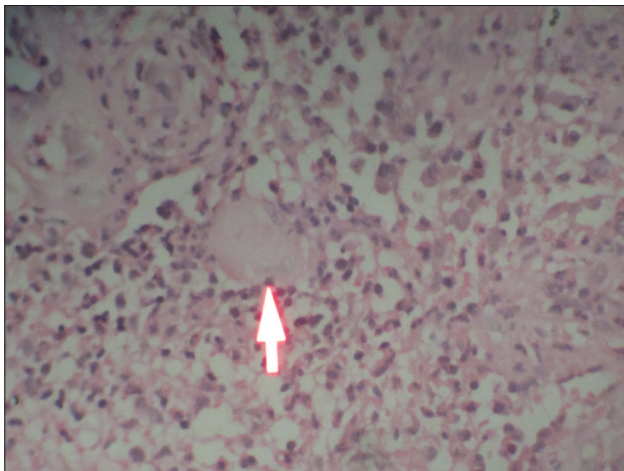
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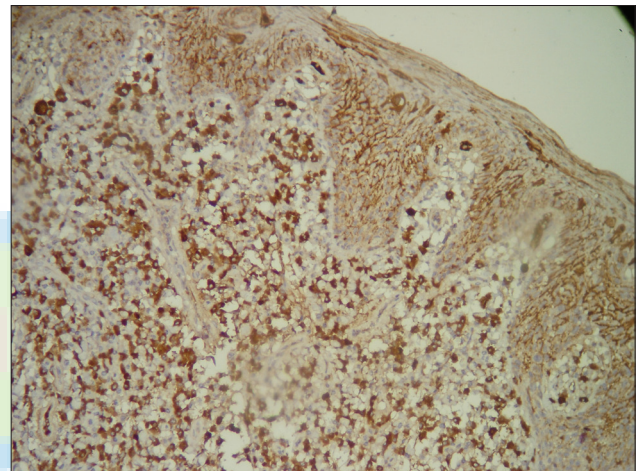
**Figure 1:** Image showing the lesion on the lip



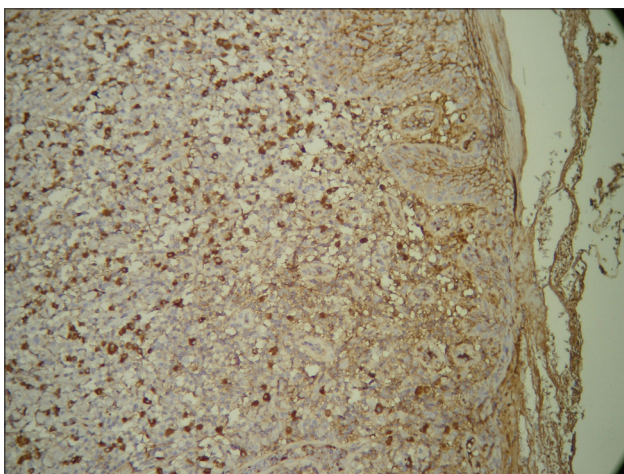
**Figure 2:** Hematoxylin and eosin stained section showing sheets of plasma cells



**Figure 3:** Fig3 Hematoxylin and eosin stained section showing russell bodies



**Figure 4:** Immunoreactivity for kappa marker



**Figure 5:** Immunoreactivity for lambda marker

plasma cells but also showing neutrophils, lymphocytes, and macrophages [Figure 2]. Occasional multinucleated giant cells were present. Numerous small round homogeneous eosinophilic masses suggestive of Russell bodies were

observed [Figure 3]. Inflammatory cell exocytosis and intra- and inter-cellular edema were also observed. The lesion was diagnosed as plasma cell granuloma and the patient was advised kappa and lambda light-chain gene rearrangements investigation to rule out a malignancy such as plasmacytoma.

On immunohistochemical evaluation, both kappa [Figure 4] and lambda [Figure 5] markers shows positive reaction and there was no light-chain restriction, which confirmed the previous diagnosis. The patient was treated with complete excision of the lesion. She has, however, so far failed to turn up for follow-up.

## DISCUSSION AND REVIEW OF LITERATURE

Plasma cell granuloma is also called inflammatory pseudotumor and inflammatory myofibroblastic tumor due to its heavy content of lymphoid and plasmacytic elements. Since these terms are interchangeably used in the literature, accurate data regarding the incidence and



anatomic distribution of plasma cell granuloma is difficult to obtain.<sup>[15]</sup>

The presence of polyclonal plasma cells, lymphocytes, and histiocytes suggests an infectious or autoimmune origin.<sup>[16]</sup> Plasma cell granuloma is thought to result from inflammation following minor trauma or surgery or to be associated with malignancy but this was not so in the present case and our patient was free of any history of trauma or malignancy.<sup>[17-19]</sup>

In the present case, the lesion was present on the lip. Though plasma cell granulomas are usually solitary, our patient had multiple ulcerated lesions. This lesion has no sex predilection and may occur at any age.

Kim *et al.* reported gingival plasma cell granuloma in patients with cyclosporine-induced gingival overgrowth and suggested that interleukin-6 (IL-6) and phospholipase C-γ1 may induce heavy plasma cell infiltration in cyclosporine-induced gingival overgrowth.<sup>[20]</sup> The patient we present in this case report was not receiving any medication.

A kappa:lambda ratio 1:6 is suggestive of a monoclonal proliferation.<sup>[21]</sup> It is generally accepted that lesions consisting of monoclonal plasma cells are neoplastic, whereas lesions with polyclonal plasma cells are inflammatory.<sup>[22]</sup>

It is always important to differentiate plasma cell granuloma from extramedullary plasmacytoma and multiple myeloma, considering the poor prognosis of the latter.<sup>[23]</sup>

Recurrence has been suggested to be more common if the lesions are extrapulmonary, when recurrence rates are reported to be up to 25%–40%.<sup>[24]</sup> Since our patient failed to turn up for follow-up we are unable to report if there has been any the recurrence.

Although plasma cell granuloma in the oral cavity is rare, it is important to recognize this entity as a benign inflammatory lesion.

## CONCLUSION

In conclusion, plasma cell granuloma might be misinterpreted as a malignant neoplasm due to its aggressive clinical appearance. Biopsy is usually necessary to reach a definitive diagnosis. The correct recognition of the lesion is important to avoid needless extensive and radical surgical procedures.

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**How to cite this article:** Sabarinath B, Sivapathasundharam B, Vasanthakumar V. Plasma cell granuloma of lip. *Indian J Dent Res* 2012;23:101-3.

**Source of Support:** Nil, **Conflict of Interest:** None declared.