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A rare presentation of nodule-proliferative plasma cell mucositis: Clinical insight and discussion

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Introduction

Plasma cell mucositis (PCM) is a rare proliferative disorder involving plasma cells in the upper aero-digestive tract. Although its cause remains unknown, it is recognized as a benign condition that primarily affects adults. Over time, the terminology for this condition has evolved, with alternative names including plasma cell orificial mucositis, idiopathic plasmacytosis, mucous membrane plasmacytosis of the upperaero-digestive tract, and oral papillary plasmacytosis; therefore, the term "plasma cell mucositis" has been suggested for standardization [1].

Oral plasma cell mucositis (OPCM) is a typically idiopathic hypersensitivity reaction, although specific antigens such as components of chewing gum, toothpaste, khat, or certain foods, may be potentially contributing factors. Its clinical presentation varies, but is commonly characterized by pronounced erythema of the oral mucosa, accompanied by surface changes (cobblestone-like, nodular, papillomatous, granular, or velvety) and Symptoms including dysphagia, oral pain, sore throat, and pharyngitis [2,3].

The principal histological feature of OPCM is a dense polyclonal plasmocytic infiltrate in the superficial lamina propria. Immunohistochemical demonstration of polyclonality with no kappa or lambda light chain restriction allows the differentiation of OPCM from myeloma, lymphoma, and extra-medullary plasmacytoma [4].

Patients with OPCM often have a prior history of autoimmune or immune-mediated diseases, such as Sjögren's syndrome, autoimmune hepatitis, diabetes mellitus etc. However, these associations are inconsistent, and no single condition has been definitively linked [5].

The therapeutic management of PCM is primarily symptomatic. Steroid therapy is often effective in achieving disease stabilization or even complete regression but has limiting side effects. Severe cases may require immunosuppressive drugs, systemic chemotherapy, and low-dose radiotherapy have been reported [6].

Case report

A 17-year-old unmarried male presented to the Oral and Maxillofacial OPD in Isra university Hyderabad Sindh in December 2023, with a4 week history ofpersistent "rough mucosa" on inner left cheek.

The patient reported a solitary lesion with mild to moderate pain and a burning sensation, particularly when consuming spicy foods. It developed abruptly, initially as small nodule, then rapidly extended to surrounding mucosa rapidly. Additionally, no systemic symptoms, skin/genital lesions or family history of similar lesions were reported. The patient's medical, surgical, and social history was unremarkable.

Clinical examination revealed non-tender submandibular lymphadenopathy on the ipsilateral side. Intraorally, a nodulo-proliferative lesion was present on the left buccal mucosa, extending from commissure to the posterior buccal mucosaof molar region.

Morphologically lesion appeared heterogeneous with areas of erosion, ulceration, and proliferation, with irregular margins and pronounced erythema. It was raised, soft, non-tender with ill-defined borders on palpation with the rest of the oral mucosa appearing normal (Figure 1).

No similar lesions were found elsewhere on the body.

The differential diagnosis, considering the clinical presentation, included ulcerative lichen planus, mucous membrane pemphigus, plasma cell mucositis, and systemic lupus erythematosus.

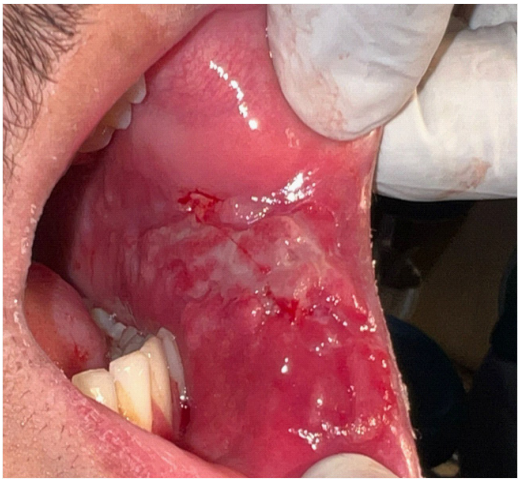

Figure 1: Oral mucos appearing normal.

An incisional biopsy was performed with the specimens sent for histopathology (preserved in 10% formalin) and direct Immunofluorescence (DIF) (preserved in Zeus medium). Meanwhile,the patient was prescribed a superpotent topical steroid (clobetasol 0.05%) four times daily for one week.

The histopathology showed ulcerated mucosa with inflammation, granulation tissue, vascular proliferation, plasma cell rich inflammatory infiltrate with admixed eosinophils consistent with immune mediated mucositis. PASDAB stains were unremarkable and DIF for IgG, IgA, IgM, C3, and C1q antibodies were negative.

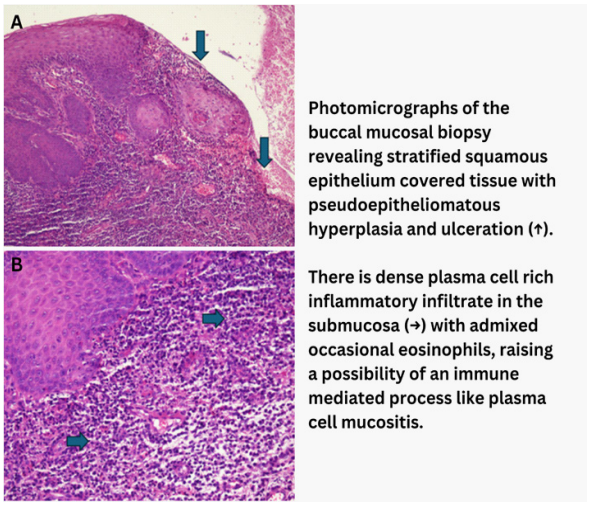

Figure 2: Clinical image.

Immunofluorescence studies on a separate mucosal biopsy (not shown here) were negative (H&E, 10X & 20X).

At one week follow-up, the lesion showed near-complete resolution with the use of topical clobetasol 0.05%. He was advised to identify potential antigenic antigens triggers to prevent recurrence.

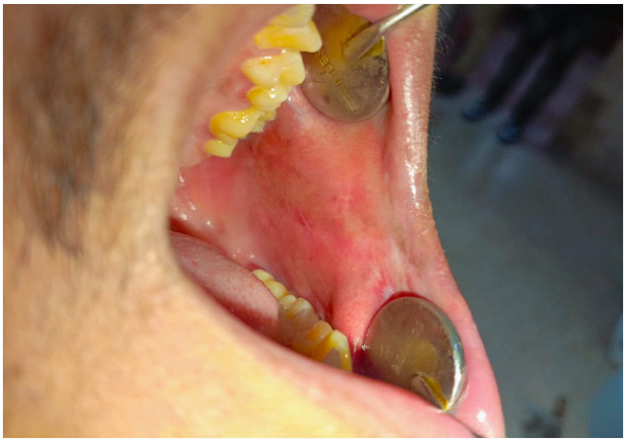

Figure 3: Clinical image.

The patient presented a few weeks later with a similar but less severe lesion and nearly identical symptoms. He noted that the lesion appeared about two hours after breakfast which included sandwich with white sauce, which upon further reflection, was also consumed during the previous episode. However, the specific trigger within white sauce remains unknown.

He was treated with intralesional triamcinolone, injected at the lesion base and advised to avoid white sauce. He is still in contact and has reported no further eruptions after avoidance.

Discussion

Plasma Cell Mucositis (PCM) is a rare inflammatory disorder affecting mucous membranes, characterized histologically by dense plasma cell infiltration. Oral PCM (OPCM) is considered the oral counterpart of plasma cell balanitis (Zoon's balanitis), an inflammatory condition of the glans penis first described by Zoon in 1952, which exhibits a band-like infiltrate of plasma cells in the upper dermis. Clinically and histopathologically identical manifestations of have been observed on various orificial surfaces, including the vulva, buccal mucosa, palate, lips, tongue, epiglottis, and larynx. Collectively, these conditions are referred as PCM [7].

The demographic data on PCM varies across studies. A 2021 study reported a mean age of 60.3 years (range: 11-82 years), while earlier studies indicated a younger age of 45 years with (4:1) female predominance. While the younger age aligns with our case, the gender predilection deviates. However, Solomon's 2008 study found a slight male predominance, which supports our case demographic. PCM most commonly affects gingiva (30%), also involving alveolar mucosa. However, our case primarily involved the buccal mucosa, which suggests potential variations [8].

Three potential etiological hypotheses for PCM have been proposed [1]. Allergic hypersensitivity developing after exposure to a sensitizing antigen, identified in 27.21% of cases in. Some local irritants include chewing gum and herbal toothpaste [9]. As identified by Gargiulo et al., an inflammatory process with trauma or parafunctional habits triggering the development of sub-epithelial plasma cell infiltrates [2]. Additionally, the presence of plasma cell-related inflammatory lesions in contact and friction-prone areas outside the oral cavity—such as the genital region, axillae, and skin—suggesting that repetitive microtrauma may play a significant role in the etiology of plasma cell infiltration [10]. Furthermore, exposure to fungal (Candida albicans in particular), viral (Herpes virus), or bacterial (dental plaque) infections may act as potential triggers [11].

Chronic inflammation causing immune dysregulation may be a likely contributor but no single antigen, chemical mediator, or specific cell type has been identified as the sole trigger [3].

Clinical presentation varies widely with pain being most common, particularly in areas with tissue loss, with occasional bleeding. Only 6.33% of patients are reported asymptomatic. If the disease extends to the pharynx or larynx, symptoms like pharyngeal globus, cough, and hoarseness may occur. Histological analysis reveals a dense plasma cell infiltrate predominantly within the lamina propria but occasionally extending into the epithelium separated by fibrous connective tissue septa. In some cases, polymorphic infiltrate contains varying proportions of lymphocytes, neutrophils, and eosinophils. It is important to note that a plasma cell infiltrate in an oral biopsy may indicate an underlying systemic disease. Therefore, it is essential to rule out other conditions that present with plasma cells in their oral histopathology [12]. Additionally, microscopy frequently shows epithelial hyperplasia (papillary, pseudoepitheliomatous or psoriasiform hyperplasia). Patients may exhibit epithelial changes like atrophy, edema, spongiosis, acanthosis, and micro-abscess formation [13]. Russel bodies may also be present.

Immunohistochemical analysis, though performed in limited number of cases, typically reveals a normal kappa-to-lambda ratio, with a predominance of kappa light chains in some patients. Immunohistochemistry should always be performed in PCM cases to confirm the polyclonality of immunoglobulins, thereby confirming polyclonality and distinguishing PCM from neoplastic processes confirming it as a benign reactive process [14].

The diagnosis of PCM relies on thorough Assessment of patient's medical history, physical examination, serological tests, and histopathological analysis. The lack of an international agreement on the best treatment reflects the differences in symptoms and clinical presentation. Identifying and eliminating local irritants has been successful in up to 52% of cases. However, this alone is not always sufficient [15].

Corticosteroids remain the primary treatment option commonly including Triamcinolone Acetonide, Prednisolone, Prednisone, and Betamethasone, though their immediate and long-term effectiveness remains uncertain. In our case, the patient showed near-complete resolution after using topical clobetasol 0.05% supporting evidence of short-term steroid effectiveness [16].

Success of topical steroid therapy depends on epidermal thickness and the extent of plasma cell infiltration, sometimes leading to a poor response. Intra- or peri-lesional injection enhance efficacy in refractory cases as was observed in our case with intralesional triamcinolone administration upon recurrence [17]. The effectiveness of immunosuppressants and immunomodulators remains relatively low, with success rates of 23.81% in monotherapy and 19.05% in combination therapy with 57.14% of cases showing no improvement. Among these drugs, Tacrolimus has shown the best results, acting by inhibiting the transcription of cytokine genes such as IL-3, IL-4, IL-5, and TNF. Additionally, non-surgical periodontal therapy,in combination with other treatments, helps in achieving clinical remission of gingival lesions [14].

Conclusion

This case underscores the importance of considering PCM in the differential diagnosis of oral mucosal lesions, particularly those with a noduloproliferative appearance and a plasma cell-rich infiltrate. Histopathology and immunohistochemistry are essential for distinguishing PCM from other plasma cell-related conditions, ensuring accurate diagnosis and management. While corticosteroids remain the mainstay of treatment. Identifying and eliminating potential triggers, as demonstrated in this case, may offer a more sustainable approach to disease control. Further researches are needed to establish standardized diagnostic criteria and treatment protocols. This case contributes to the growing evidence suggesting that PCM may have a broader clinical spectrum than previously recognized, necessitating ongoing research, enhancing clinical awareness.

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