A rare case of plasma cell mucositis in a young patient

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Abstract:
Plasma cell mucositis (PCM) is a rare benign proliferative disorder affecting various mucosal surfaces usually seen in elderly patients. In the present case, a 13-year-old young boy reported with the complaint of swollen upper lip and enlarged gums in relation to upper front teeth. The diagnosis of plasma cell mucositis is complex and frequently requires histopathological examination and immunohistochemistry. Management included conventional gingivectomy and intralesional steroid. Regular follow-up of PCM is essential based on the possibility of recurrence, the potential involvement of other mucosal surfaces and unknown potential for malignant transformation.

Key words:
Gingivectomy, kappa and lambda, multiple myeloma, plasma cell mucositis

INTRODUCTION

Plasma cell mucositis (PCM) is a benign inflammatory condition characterized by dense plasma cell infiltrate in the mucosa close to the orifices. PCM was first reported by Zoon[1] as involving glans penis. It can affect vulva, lips, buccal mucosa, palate, gingiva, tongue, epiglottis, larynx, and also other parts of the body. The other nomenclatures used for PCM are plasma cell orificial mucositis, idiopathic plasmacytosis, and oral papillary plasmacytosis.[2] According to Smith et al.,[3] PCM has been proposed as a name to facilitate the documentation of such cases. PCM is an extremely rare condition, with less than 50 cases reported.[4] A review by Solomon et al.,[2] on PCM reported that the average age of affected patients as 56.6 years with a slight male predominance of 1.2:1 and the youngest patient as 27-year-old. Here, we report a case of plasma cell infiltration of gums and labial mucosa in a 13-year-old boy.

CASE REPORT

A 13-year-old, 40 kg male patient reported to the department of periodontics, Mar Baselios Dental College, Kothamangalam, Kerala with the chief complaint of swollen and bleeding gums in upper anterior region. The patient first noticed a mild swelling of the upper lip around six months back with the gradual involvement of the gums, which showed bleeding since the last one month. No contributory medical or dental history could be elicited.

On extraoral examination, the upper lip was swollen and soft in consistency [Figure 1].
ml (Kenacort) were given once a week for three weeks on the lip mucosa and mucogingival junction of the affected area. Excisional biopsy of the gingival lesion was done by means of an external bevel gingivectomy. Histopathological examination of excised tissue showed a parakeratinized stratified squamous ulcerated surface epithelium associated with fibrovascular connective tissue. The connective tissue exhibited dense chronic inflammatory cell infiltrate comprised predominantly of plasma cells [Figure 3a and b]. Immunohistochemistry (IHC) evaluation showed biclonal expression of kappa and lambda with marginal overexpression of kappa [Figure 4]. The histopathological and IHC findings confirmed the diagnosis of PCM. The surgical site healed uneventfully and a near normal appearance of the lip and gingiva [Figure 5] was seen after six weeks. A one-year post-surgical follow-up showed no evidence of recurrence.

**DISCUSSION**

PCM is a rare variation of mucositis comprising of a polyclonal plasma cell infiltration of the mucosa. The coexistence of autoimmune or immunologically mediated diseases is not rare with PCM. However, these diseases are not present...
PCM is generally considered to be a benign condition, which shows favorable prognosis. The identification of monoclonal or polyclonal expansion can help in the differential diagnosis of various plasma cell disorders. The confirmation of PCM requires IHC showing polyclonal expression with kappa and lambda chain restrictions other than clinical and histopathological investigations. Kappa and Lambda are immunoglobulin free light chains which are considered as a marker for plasma cell activation. PCM shows a predominance of kappa chain expression than lambda. Monoclonal proliferations of plasma cells are seen in neoplasms such as multiple myeloma and extramedullary plasmacytoma. The presentation of PCM complicated by evidence of monoclonal proliferation of plasma cells has been reported. However, the potential for full malignant transformation is unknown.

Management of PCM is mainly targeted at symptomatic relief. PCM has been treated with topical, intralesional, and systemic steroids, antibiotics, antifungals, and cryotherapy. Corticosteroid is considered as the most frequent treatment modality, but this has unreliable results and is of questionable benefit. Majority of these results in disease stabilization but complete regression is rarely noticed. Even though a rare entity, the clinical and histological features of PCM may resemble many common benign and neoplastic conditions of oral cavity and hence it is a diagnosis of exclusion requiring extensive investigation and multidisciplinary evaluation.

CONCLUSION

PCM is rarely reported in dental literature, and it is important to recognize PCM in dental community. The diagnosis is based on clinical pathologic correlation, and therefore, close communication between specialists in several disciplines are required for proper diagnosis of such rare disorder. Documentation and reporting of such cases with regular follow-up will bring greater awareness of the condition and can be helpful in identifying the condition at a younger age to prevent future complications.

Declaration of patient consent

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

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Conflicts of interest

There are no conflicts of interest.

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