Case Report: Management of Oral Mucous Membrane Plasmacytosis with Gingivectomy

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DOI:10.61139/ijdor.1248921

Abstract

Mucous membrane plasmacytosis (MMP) is a rare benign condition. Neoplastic lesions should be considered in the differential diagnosis for this condition, which is clinically involved in different anatomically locations and presents clinically different appearances. Cellular, polyclonal plasma cell infiltration of mucosal tissue is in this condition observed. This case report discusses diagnosis and treatment in a patient with oral involvement of MMP. In the case diagnosed with MMP, no recurrence was observed in the short-term follow-up. However, long-term follow-up of such cases is recommended. Detailed information about the diagnosis and treatment of this clinical condition is still needed.

Case Reports (HRU Int J Dent Oral Res 2023; 3(2):104-107

Key words: Mucous membrane plasmacytosis, biopsy, gingivectomy**.**

Introduction

First described by Zoon in 1952 and was located at the glans penis which he described as ‘’chronic benign balanoposthitis with plasmacytes’’ (1). Mucous membrane plasmacytosis (MMP) is a rare benign condition consists of an intense plasma cell infiltration of the mucous membrane. It is seen that this clinical picture in gingiva was also named as atypical gingivostomatitis, idiopathic gingivostomatitis and allergic gingivostomatitis in previous years (2). It was suggested that it was common with hypersensitivity reactions and defined also as an idiopathic, inflammatory clinical entity (3). This condition was thought to be and allergic reaction to unknown ingredient mouthwash, toothpaste and chewing gum other causes like decreased vitamin intake and local infections was suggested (4). Upper respiratory-digestive system and oral mucosal regions were seen as anatomical sites affected (5). Fogarty et al. presented a patient with disease progressed from gingiva to upper airway then treated with lower dose of radiation to achieve symptomatic improvement and minimal toxicity (6). In general, clinical examinations reveals a well-circumscribed, soft, slightly raised, edematous mass. If it occurs in the lip and gingival area, they complain of symptoms such as pain, swollen lips and gingiva. When it occurs in other parts of the air-digestive system, such as the larynx, pharynx, patients
moan about of oral pain, dysphasia, dysphonia and difficulty breathing (7).

There is no agreed upon or consistently successful treatment option for this clinical picture. Corticosteroids, surgical removal, antibiotics, low-dose radiation, cryotherapy are some of suggested therapies. Polyclonal plasma cell infiltration of mucosal tissue is represented in MMP which is a rare variant of mucositis (8). MMP is a rare, idiopathic and inflammatory mucositis that frequently presents as an erythematous lobulated mucosa. MMP is known for ulcerated plaques that are known to bleed (9). This report represents a case of MMP in the right maxillary canine area.

Case Reports

A 53 year-old female patient was referred to the Department of Periodontology, because of swelling in the right maxillary canine buccal area (Figure 1). The patient's medical history revealed that there was no significant systemic disease or allergy. According to the information given by the patient, the lesion has been present in the mouth at least for 9 months. In addition, clinic examination showed caries in the right lateral incisor. No other orofacial symptoms were observed.

Figure 1: Clinical appearance of the lesion.

Clinical periodontal examination revealed, pink, inflamed mass below the free gingival margin/interdental papilla. There was a notable ulceration on the center of the enlargement.

After the periodontal phase I treatment a further review for two weeks showed no improvement. The patient was not cooperative to the periodontal treatment. Desired oral hygiene levels could not be obtained in phase 1 treatment. However, it was decided to remove the lesion. Written informed consent was obtained from patient and ethics committee approval was not needed.

Infiltrative local anesthesia (Ultraver D-S) applied to area. External bevel incision is made and lesion is removed with a Kirkland type gingivectomy knife. Specimen is carefully examined (Figure 2) and sent to pathology department for advanced examination. Gingival contour is corrected with gingivoplasty procedure and a periodontal dressing was applied to the area. The patient was seen again in the 2nd week after the surgical procedure (Figure 3). However, the patient did not continue the following control sessions afterwards.

Dimensions of the biopsy specimen were 1.2x0.6x0.4 cm. The biopsy material was cream brown in color and its hardness was medium hard. Sections were observed in homogeneous appearance.

Figure 2: The appearance of the specimen.

Figure 3: Clinical appearance in the second week after the procedure.

Biopsy sections showed hyperplasia, irregular acanthosis with small formations of plasma cells under the epithelium. Plasma cells were companied but in few by lymphocyte, histiocytes and neutrophil leukocytes. Haematoxylin and eosin staining (H&E) revealed plasma cells with an eccentric nucleus with oval coarse chromatin, with abundant dark blue cytoplasm and perinuclear hoof, marked with arrows are observed (Figure 4 A). Immunochemical analysis of inflammatory cells under the epithelium showed that LCA, CD45, CD 79a, CD38, MUM1 staining were positive. Kappa, Lambda and CD138, staining was also positive (Figure 4 B-D). According to Ki67 staining proliferative index was %4. Pancytokeratine showed a positive staining in the surface epithelium. In the immunochemical analysis performed, it was noted that the infiltration in the plasma cells under the epithelium which were polyclonal.

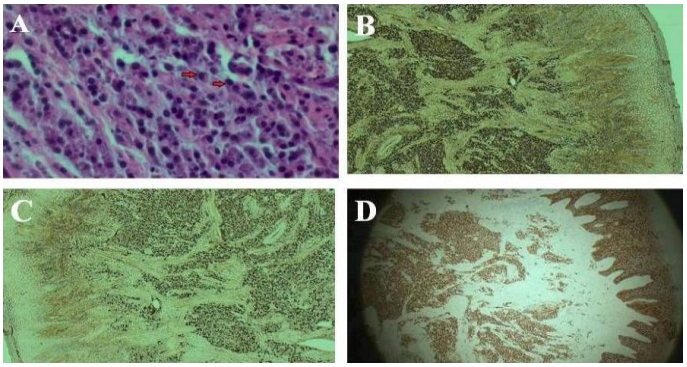

Figure 4: Pathological evaluation
A: H&E, x63 magnification;
B: Kappa, x5 magnification,
C: Lambda, x5 magnification,
D: CD138, x5 magnification.

Discussion

In the nomenclature cases have been reported with variety of titles such as idiopathic plasmacytosis (10,11), plasma cell orificial mucositis (12), oral papillary plasmacytosis (13) and mucous membrane plasmacytosis (5). The clinical differential diagnosis of MMP consist of Wegener granulomatosis, pemphigus, erythroplasia, erosive lichen planus, sarcoidosis, fungal infections, squamous cell carcinoma, mucous membrane pemphigoid, and allergic gingivostomatitis (14).

The diagnosis of MMP can be difficult considering histological resemblance with neoplastic lesions. Treatment of MMP is challenging because of lack of consensus on its management. There is one reported case that shows accompanying squamous cell carcinoma with unidentified etiopathogenetic relation (15).

Unlike plasma cell gingivitis, in which the link with allergic reactions is well established, the association between MMP and allergy is less well recognized (16). Previous reports have described the use of corticosteroids, antibiotics and surgical debulking procedures, with inconsistent results (6).

Najarian et al. achieved long lasting remission with cryotherapy. They used liquid nitrogen with the open-spray technique. Freeze time was 30 to 60 seconds for lower lip localized MMP. In this way they achieved 6 months remission and no functional side effects.[9] Mahler et al. used 2% fusidic acid combination with tetracaine 4 times a day. After regular use continuous improvement was achieved. There were no side effects to be specified and non-invasive treatment was accomplished (17).

MMP is rare type of mucositis, there are no long-term studies with large numbers, and regular clinical review is recommended. This report adds to the literature another case of the rare plasma cell disorder MMP of gingiva. It also demonstrates a successfully excisional treatment of the MMP.

Conclusion

Plasma cell malignancies occur from an anormal clonal proliferation of plasma cells. Despite the fact MMP is identified as idiopathic, other formations of plasma cell conditions may involve neoplastic diseases such as solitary plasmacytoma of bone and multiple myeloma. Therefore, a high index of suspicion is required to identify MMP and biopsy can be taken into consideration for ulcerated gingival enlargements.

One-sentence summary describing the key finding(s): Diagnosis and treatment stages of an individual with mucous membrane plasmacytosis are discussed.

Author contribution statement: All authors have made substantial contribution to the case. AGT and HH treated the patient. AND and NBA made the pathological evaluations. All authors drafted and approved the final version of the manuscript. AGT performed the measurement and involved in the follow-up.

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