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Case Report

Oral Mucous Membrane Pemphigoid with Contact Dermatitis - A Rare Clinical Entity.

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Abstract

Mucous membrane pemphigoid (MMP) is a rare inflammatory, autoimmune, subepithelial vesiculobullous lesion. Oral mucosa is affected in almost 90% of cases. Its development is chronic with a possible involvement of ocular, laryngeal and genital mucosa. Spontaneous remission is rare. Currently, improving oral hygiene, topical corticosteroid treatment is used to control the oral lesions of MMP. In the present case report, a 53 year old female patient with a known history of hypothyroidism reported to the department of Periodontology with a complaint of burning sensation and tenderness in the gums on intake of spicy food for the past one week .The patient had also noticed the formation of blisters on the gums which would break off on their own. Hence provisional diagnosis of MMP was considered based on the clinical and histopathological examination which was later confirmed with immunofluorescent interpretations. The treatment objective is to suppress extensive blister formation, to promote healing, and to prevent scarring. The above objectives were met in our case by thorough periodontal maintenance and steroid therapy. She also presented with skin lesions due to contact dermatitis caused by occupational exposure to tobacco which makes this case report unique.

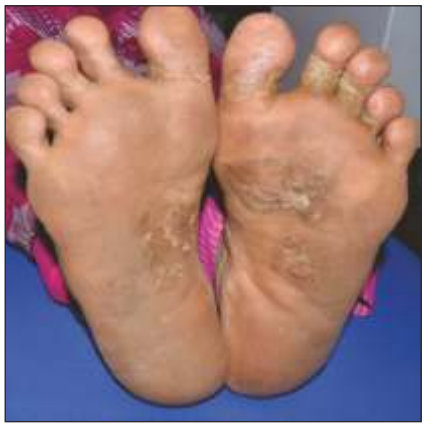
Introduction

There are various autoimmune disorders of oral cavity. One among them is mucous membrane pemphigoid (MMP) which can present with variety of clinical manifestations including both oral and skin lesions. The most common manifestation seen is desquamative gingivitis which can be suggestive of a systemic disease. The following case, reports a patient showing MMP as well as skin lesions due to contact dermatitis caused by occupational exposure to tobacco.

Case Report

A 53 year old female patient who is under thyroid therapy (thyronorm 75µg) reported to the department with the chief complaint of burning sensation in gums on consumption of hot and spicy food since one week. The patient was well built and nourished with no other systemic condition. The patient also reported with a history of working in a beedi rolling factory (tobacco maker). She had noticed the presence of ulcerations in her palate over the period of six months that regresses on its own and recurs after few days and she also presented with the peeling of the skin of the under surface of the foot, thumb and scalp since 4- 5 months for which she was on alternative medication(fig 1,2,3). On intra oral examination, patient had a fair oral hygiene with generalized gingival recession and an erosive lesion was noticed on the posterior hard palate corresponding to the palatal mucosa in the region of right upper molars, measuring around 0.5 x 0.5 cm in dimensions, with erythematous borders and slough (fig 4). The lesion was non tender on palpation and did not bleed on touch. The marginal gingiva in relation to the maxillary and mandibular anteriors appeared erythematous and desquamated and the Nikolsky's sign was negative. Hence a provisional diagnosis of chronic generalized periodontitis and erosive lichen planus of right side posterior hard palate

with desquamative gingivitis of maxillary and mandibular labial mucosa was given. Exfoliative cytology was done and a thorough scaling and root planing along with palliative treatment was given. Patient was referred to dermatology for the diagnosis of skin lesions and was recalled after a week. After a week the symptoms persisted and patient continued to present with same clinical findings. Hence incisional biopsy was taken from the gingiva of the right upper molar for histopathological examination. The histopathological section showed the presence of stratified squamous orthokeratinised epithelium with hyperkeratosis (fig 5). The epithelium appeared to be separated from the underlying connective tissue suggestive of a split at the interface with an intact basement membrane. Direct Immunofluorescence was further advised for the confirmation of vesiculobullous lesions which showed a linear pattern of Ig G, C3 and fibrinogen in the basement membrane (fig 6). Correlating with the clinical, histopathological and Direct immunofluorescence examination, a diagnosis of Mucous Membrane Pemphigoid was confirmed. The dermatologist confirmed skin lesion was irritant contact dermatitis.The patient was put on topical steroids, 0.1% Triamcinolone acetonide (Tenovate®) thrice daily for the gingival lesion and was recalled after a week. Triben B® cream and cetrizine for dermal lesion was given. The mucosal and skin lesion reduced significantly within a month of follow up(fig 7,8,9).Then patient was referred to department of prosthodontics for prosthetic rehabilitation and the patient is kept on a regular follow up.



Figures 1 : Lesion on feet

Figure 2 : Lesion on scalp

Figure 3 : Lesion on thumb

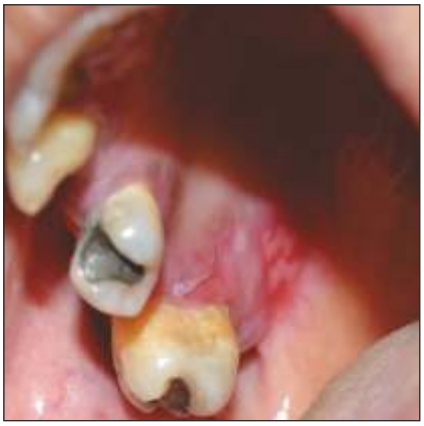


Figure 4 : Palatal Lesion

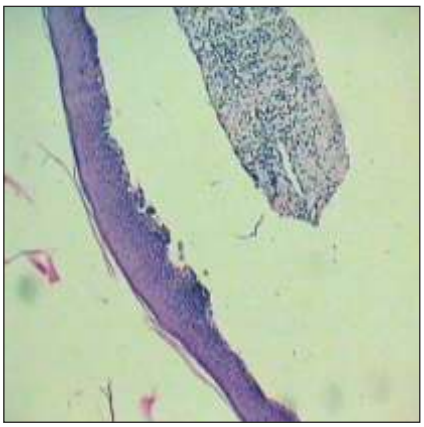


Figure 5 : Histopathology

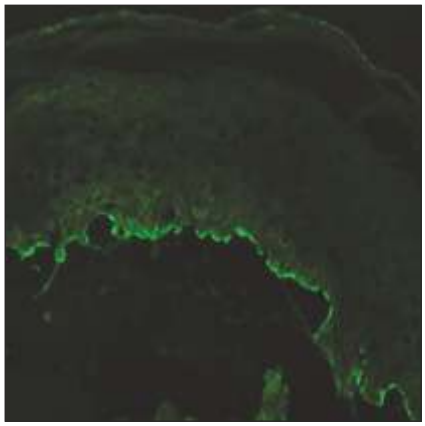


Figure 6 : Immunofluorescence

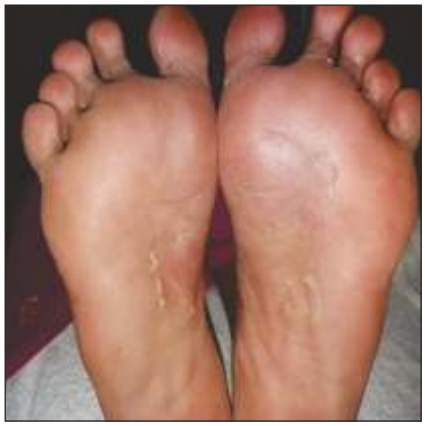


Figure 7 : Healing after 7 days.

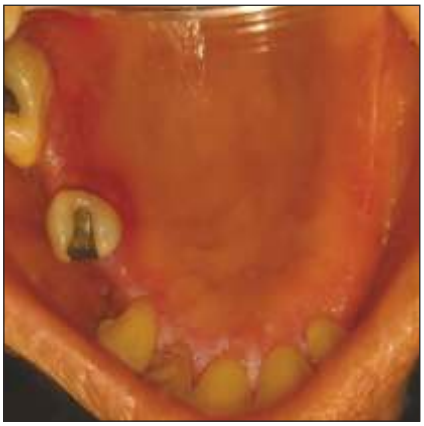


Figure 8 : Healing after 7 days

Figure 9 : Healing after 7 days

Discussion

Mucous membrane pemphigoid presents with mucosal involvement at multiple sites with occasional skin involvement[1]. Oral mucosal involvement is most common, present in about 85% of the patients followed by ocular conjunctiva (65%), nasal mucosa (20-40%), skin (25-30%), anogenital area and pharynx (20%), larynx and oesophagus (5-15%)[1]. The most common oral manifestation is desquamative gingivitis[2] which can also be seen in erosive lichen planus[3] which lead us to the provisional diagnosis in the present case. A compliment mediated antibody induced process results in the detachment of epithelium from the underlying connective tissue within the basement membrane zone which can either be due to direct cytotoxic effects or by the action of lysosomal proteolytic enzymes[1].

Patients usually present with burning sensation, with gingival erythema and loss of stippling. Presence of a vesicle, bullae or ulceration can also be noticed[2,6]. Patients can exhibit a positive "Nikolsky's sign"with epithelial sloughing and exposure of bleeding surfaces[2] but it was negative in the present case. Oral lesions rarely heal by scarring[1]. The case reported here had only oral mucosal manifestations. Ocular involvement and the rest of the clinical features were absent.

Definitive diagnosis of Mucous Membrane Pemphigoid is done on the basis of histopathological and immunofluorescence examinations. Histologically, it is characterized by the presence of subepidermal vesicles and bullae with no evidence of acantholysis[4,2]. The basement membrane appears to be detached from the underlying connective tissue with a sub- epithelial cleft. Direct Immuno fluorescence (DIF) features shows the

presence of tissue bond basement membrane zone antibodies with a liner deposition of Ig G, Ig A, and C3 at the basement membrane[4, 2]. The immunologic tolerance to structural proteins is lost in the basement membrane zone that leads to the development of auto antibodies[1].The histopathological and immunofluorescence reports of the present case also had the similar presentation.

Management depends upon the location, severity and rate of progression of the disease. The first international consensus on MMP had categorized the patients into "low risk" and "high risk" depending on the sites involved[5, 6]. Low risk patients are those having only oral mucosal and skin involvement, similar to the present case. Such patients are treated with topical corticosteroids like 0.1% triamcinolone acetonide, 0.05% fluocinolone acetonide, or 0.05% clobetasol propionate for 3-4 times a day for 9-24 weeks[2, 7]. Similarly, the present case has also been treated with 0.1% Tenovate ® (0.05% clobesatol propionate) three times a day for two weeks and is kept on a regular follow up.

The skin lesion presented here was different from the skin lesions associated with mucus membrane pemphigoid as they commonly occur in head and neck region. Occupational exposures to exogenous agents are the most common cause of irritant dermatitis. Similarly our patient presents with a history of working in a beedi rolling factory. Resolution of the lesion was observed once the exposure to the occupational hazard was reduced. In this case the contact dermatitis was mainly caused due to tobacco, hence the term tobacco dermatitis.[8]

The earliest reports of tobacco dermatitis are anecdotal only. Blaisdell (1924) described a middle aged male cigar roller with a work-related hand dermatitis and Davis (1924) a 56-year-old cigar salesman with an episodic hand dermatitis, which spread to his face and trunk[8].

Only 2 studies analyzing the relationship between mucosal pemphigoid and smoking have been published, neither of which found any significant association[9,10] .Hence a diagnosis of irritant dermatitis depends on careful clinical examination, patient history and assessment of exposure to irritants including occupational and non-occupational exposures.

Conclusion

MMP can present with a variety of clinical manifestations including skin lesions. Till date there has not been any documented cases showing an association of tobacco and its affect on autoimmune disorders like vesiculobullous lesions. Proper history taking and a thorough histopathological examination plays a key role in the diagnosis. Further research and studies have to be conducted and more emphasis should be given in order to find a possible causal association between mucous membrane pemphigoid and tobacco dermatitis.

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