Ref: Ro J Stomatol. 2021;67(4)
DOI: 10.37897/RJS.2021.4.5

Case report of a rare bullous variant of oral lichen planus

Ioanina Parlatescu[1], Serban Tovaru[1], Carmen Nicolae[1], Dragos Epistatu[2], Cristina Pirvu[3], Valentin Varlas[4,5], Roxana Elena Bohîlțea[4,5], Paula Perlea[6]

[1] Department of Oral Pathology, Faculty of Dental Medicine, "Carol Davila" University of Medicine and Pharmacy, Bucharest, Romania
[2] Department of Radiology, Faculty of Dental Medicine, "Carol Davila" University of Medicine and Pharmacy, Bucharest, Romania
[3] Department of Research Methodology-Ergonomics, Faculty of Dental Medicine, "Carol Davila" University of Medicine and Pharmacy, Bucharest, Romania
[4] Department of Obstetrics and Gynecology, "Carol Davila" University of Medicine and Pharmacy, Bucharest, Romania
[5] Department of Obstetrics and Gynecology, Filantropia Clinical Hospital, Bucharest, Romania
[6] Department of Endodontics, Faculty of Dental Medicine, "Carol Davila" University of Medicine and Pharmacy, Bucharest, Romania

ABSTRACT

The aim of this report is to describe the lichen planus on the oral mucosa in the bullous variant of the disease. It is often misdiagnosed with other mucosa disorders (allergies, bullous dermatosis). A 37-year-old-female patient presented for oral mucosa painful lesions of 3 months duration. A microscopic examination of the lesional areas was consistent with the diagnosis of oral lichen planus and direct immunofluorescence confirmed it. This case showed that although a rare condition the variant of bullous lichen planus can be encountered in daily clinical practice.

Keywords: lichen planus, oral mucosa, bullous variant, oral lesions

BACKGROUND

Oral lichen planus (OLP) is a chronic autoimmune disease with an incidence of 0.98% in the general population [1]. The disease affects middle-aged patients and slightly more women [2,3]. Unlike skin lesions, oral lesions have a chronic evolution with symptomatic periods during patients' lifetimes. The diagnosis for oral lesions is based on the clinical criteria or by both clinical criteria and histological examination [4].

The OLP etiology is unclear yet. Stress, systemic medication, dental materials, chronic liver diseases, diabetes, other autoimmune diseases are predisposing factors. Cell mediated immunity with CD4+, CD8+ T cells, natural killer cells, mast cells trigger and induce apoptosis in oral keratinocytes [5].

There are six clinical variants of OLP: reticular, papular, plaque, atrophic, erosive and bullous [5]. The reticular form is more common and presents with white stria. The atrophic variant presents red areas with or without white stria. The erosive variant shows erosions, atrophy and white stria. The plaque and papular types have only white lesions with different aspects. The bullous variant shows ulcers resulting from the rupture of bulla surrounded by other types of lesions [4]. In OLP the lesions are located on buccal mucosa, dorsal tongue and gingiva [4]. The lesions(white stria, plaques, erosions, atrophy, ulcers and bullae) are usually multiple, polymorphic and most of the cases have bilateral involvement. Symptoms vary from mild discomfort to burning sensation or pain and are correlated with the lesions type and severity.

Corresponding author:
Dragos Epistatu
E-mail: dragos.epistatu@umfcd.ro

Article History:
Received: 15 December 2021
Accepted: 22 December 2021

The OLP histological features are characterized by hyperparakeratosis, hyperorthokeratosis, atrophy, acanthosis, Civatte bodies, basal cell hydropic vacuolation, saw-tooth rete ridges, a band-like lymphocytic infiltrate in the lamina propria [5]. It also rules out the presence of oral dysplasia and has distinctive characteristics from oral lichenoid lesions.

Direct immunofluorescence is an adjuvant investigation which strengthens the diagnosis of oral lichen planus. Deposits of fibrinogen in the basement membrane region and no immunoglobulins and complement are the characteristic patterns in OLP [5].

CASE PRESENTATION

A 37-years-old Caucasian female patient presented with the chief complaint of painful lesions on the oral mucosa. She reported that the lesions developed in the last 4 months and had noticed some bullae which turned to painful ulcers. Since the onset of the lesions, the patient was recommended a topical ointment with A and E vitamins with no improvement of the symptoms or the lesions. The patient was a non-smoker and worked as a medical nurse with a high level of psychological stress. No cutaneous or genital lesions were reported by the patient.

The patient's personal medical history revealed depression, gastroesophageal reflux disease and Hashimoto's thyroiditis with hypothyroidism. All the associated diseases were under treatment.

During the oral clinical examination, we observed keratosis-white lesions, erosions and ulcers affecting different areas of the oral cavity, with positive Nikolsky sign (Figure 1). On the left buccal mucosa the white lesions associated a small ulcer. Desquamative gingivitis was present on the vestibular upper and lower gingiva and affected mainly the interdental papillae (atrophy, keratosis and ulcers).

FIGURE 1. Intraoral photographs showing multiple keratotic lesions on bilateral buccal mucosa, desquamative gingivitis of the upper and lower gingiva, ulcerative lesions on the ventral tongue

The routine hematological investigations were in the normal range. An additional evaluation of the suspicion of oral candidiasis revealed no fungal superimposed infection.

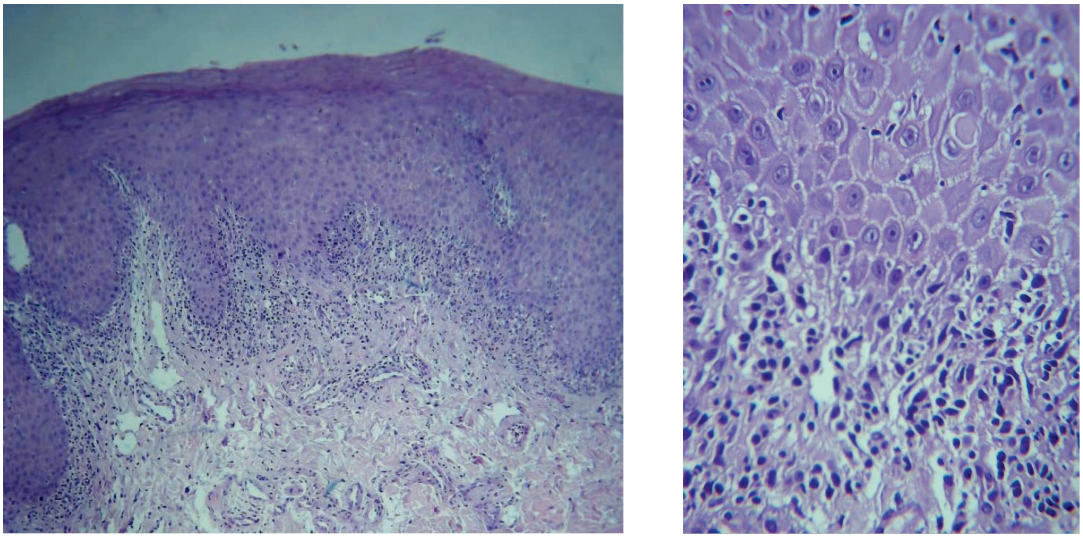
We considered the differential diagnosis of autoimmune bullous dermatosis (pemphigus or pemphigoid), lichenoid reaction and lichen planus. A gingival biopsy of the lesional area was performed and we asked for both anatomopathological examination and direct immunofluorescence evaluation.

The histopathological examination revealed epithelium with hyperorthokeratosis, hypergranulosis, acantosis, and elongated epithelial ridges, and in the connective tissue a chronic band-like inflammatory infiltrate predominantly lymphocytic suggestive for oral lichen planus (Figure 2).

The direct immunofluorescence evaluation was negative for immunoglobulin A, immunoglobulin G and complement. Rare deposits of immunoglobulin M(IgM) were observed under the basal membrane. The fibrinogen was intensely present with the granular pattern in the basal membrane area (Figure 3).

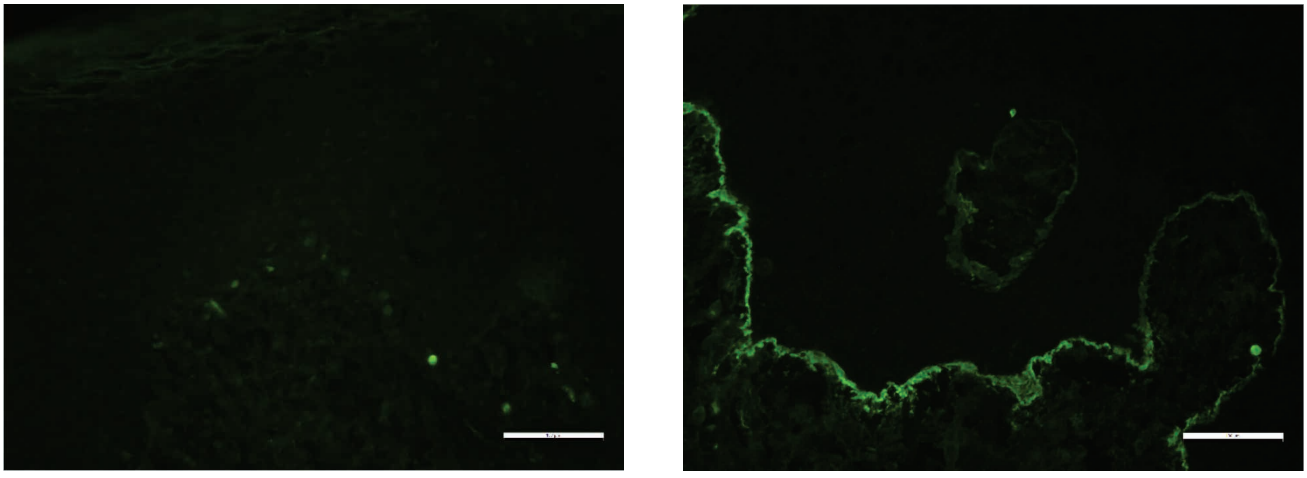
DISCUSSION

An appropriate diagnosis of oral lichen planus can raise difficulties if clinical features are not very typical. The usual clinical aspect of the Wickham striae was not encountered in the present case. But the erosions and ulcers causing pain and discomfort determined the patient to seek diagnosis. Desquamative gingivitis is a clinical descriptive term



A B

FIGURE 2 A,B. Histopathological findings. Hematoxylin-eosin staining. A: Epithelial hyperkeratosis, acanthosis and hypergranulosis and in the lamina propria a band-like inflammatory infiltrate predominantly lymphocytic(original magnification x100) B: Detail of the basal membrane area with a Civatte body and dense lymphocytic infiltrate(original magnification x400).



IgM Fibrinogen

FIGURE 3. Direct immunofluorescence showed rare deposits of IgM under the basal membrane and intense and granular fibrinogen in the basal membrane

for gingival lesions in autoimmune diseases such as OLP or bullous dermatosis. It is not a diagnosis but when present, it suggests that the histopathological examination should be accompanied by direct immunofluorescence evaluation. Nikolsky sign positive and the patient's observations of previous oral bullae were useful hints for this investigation.

The reported case associated another general autoimmune disease – Hashimoto's thyroiditis.

A recent cross-sectional study on OLP and thyroid diseases determined that Hashimoto's thyroiditis prevalence is significantly higher in OLP patients when compared to the general population [6]. Some authors consider that both diseases have as a common pathogenesis the predominance of T cells inflammation [7].

Direct immunofluorescence, although not specific for OLP, has the scope to determine the location of the autoimmune process in the tissue and to differentiate between different types of bullous diseases [1]. There are cases in which the gingival involvement entitled desquamative gingivitis may be the only clinical manifestation. Thus direct immunofluorescence is the key investigation to clear the differential diagnosis. In the reported case, the direct immunofluorescence reinforced the diagnosis.

The OLP treatment intends to reduce the symptoms, heal the lesions and avoid dysplastic epithelial changes. The reported patient was recommended a topical cream formula with a moderate corticosteroid and close follow-up.

CONCLUSIONS

The present case report emphasizes that diagnosing a bullous oral lichen planus case can be a challenge for oral healthcare professionals. A detailed anamnesis and a thorough clinical examination are the starting point for the accurate diagnosis.

Note

The authors have equal contributions for making this article.

REFERENCES

1. Li C, Tang X, Zheng X, et al. Global Prevalence and Incidence Estimates of Oral Lichen Planus: A Systematic Review and Meta-analysis. *JAMA Dermatol.* 2020;156(2):172-181.
2. Boñar-Alvarez P, Pérez Sayáns M, Garcia-Garcia A, et al. Correlation between clinical and pathological features of oral lichen planus: A retrospective observational study. *Medicine* (Baltimore). 2019; 98(8):e14614.
3. Carrozzo M, Porter S, Mercadante V, Fedele S. Oral lichen planus: A disease or a spectrum of tissue reactions? Types, causes, diagnostic algorhythms, prognosis, management strategies. *Periodontol* 2000. 2019;80(1):105-125.
4. Ismail SB, Kumar SK, Zain RB. Oral lichen planus and lichenoid reactions: etiopathogenesis, diagnosis, management and malignant transformation. *J Oral Sci.* 2007;49(2):89-106.
5. Cheng YS, Gould A, Kurago Z, Fantasia J, Muller S. Diagnosis of oral lichen planus: a position paper of the American Academy of Oral and Maxillofacial Pathology. *Oral Surg Oral Med Oral Pathol Oral Radiol.* 2016;122(3):332-354.
6. Tang Y, Shi L, Jiang B, Zhou Z, Shen X. A Cross-Sectional Study of Oral Lichen Planus Associated With Thyroid Diseases in East China. *Front Endocrinol* (Lausanne). 2020;10:928.
7. Wu P, Luo S, Zhou T, et al. Possible Mechanisms Involved in the Cooccurrence of Oral Lichen Planus and Hashimoto's Thyroiditis. *Mediators Inflamm*. 2020;2020:6309238.