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Cicatricial pemphigoid presenting as desquamative gingivitis



KEYWORDS

Cicatricial pemphigoid;
Desquamative gingivitis;
Direct immunofluorescence;
Mucous membrane pemphigoid

Cicatricial pemphigoid, also known as mucous membrane pemphigoid, is a group of chronic, blistering, mucocutaneous autoimmune diseases in which tissue-bound autoantibodies are directed against one or more components of the basement membrane.¹ Here, we presented a case of cicatricial pemphigoid presenting as desquamative gingivitis in a 42-year-old female patient.

This 42-year-old female patient came to oral mucosal disease clinic of our hospital for treatment of pain and gingival bleeding when tooth brushing. Oral examination revealed erosive lesions on the buccal gingivae of teeth 24, 25, 26, 34, 35, 36, 44 and 45 (Fig. 1A). The erosive lesions were sensitive to hot and spicy food and present for 4 months. The patient was referred to oral surgeon for biopsy under the clinical impression of desquamative gingivitis. A small piece of the perilesional buccal gingival tissue near tooth 24 was removed and sent for histopathological examination. Microscopically, it showed a split between the stratified squamous epithelium and the underlying connective tissue with a moderate chronic inflammatory cell infiltrate in the superficial region (Fig. 1B and C). A small subepithelial vesicle with the edematous stratified squamous epithelium at the roof and the inflamed connective tissue at the base was also found near the margin of the

tissue specimen (Fig. 1D). Therefore, the final histopathological diagnosis was cicatricial pemphigoid.

The cicatricial pemphigoid occurs more frequently in older female patients with a female-to-male ratio of 2:1. Oral lesions are seen more commonly at the gingivae, presenting as desquamative gingivitis, but lesions may also found at other mucosal sites including conjunctival, nasal, esophageal, laryngeal, and vaginal mucosae. Desquamative gingivitis is more commonly seen in patients with erosive oral lichen planus and less frequently found in patients with pemphigus vulgaris. Although the cicatricial pemphigoid can be easily diagnosed without the help of immunohistochemical staining to identify the tumor or cell origin,^{2–5} direct immunofluorescence studies of the perilesional mucosa showing a linear deposition of immunoglobulins (IgG, IgA or IgM) or complement 3 (C3) at the basement membrane zone of the epithelium are very helpful for the final diagnosis of cicatricial pemphigoid. Moreover, this specific immunofluorescence pattern can be found in nearly 90% of patients with cicatricial pemphigoid. In addition, direct immunofluorescence finding for pemphigus vulgaris lesion is a lattice-like deposition of immunoglobulins (IgG or IgM) or C3 in the intercellular areas among epithelial cells and that for erosive oral lichen planus lesion is a deposit of fibrinogen in a shaggy pattern at the basement membrane zone in the absence of immunoglobulin and complement. Thus, direct immunofluorescence is often used to differentiate cicatricial pemphigoid from pemphigus vulgaris and erosive oral lichen planus. The mild oral lesions of cicatricial pemphigoid can be treated by topical application of corticosteroid ointment 2 to 3 times per day. For more severe cicatricial pemphigoid oral lesions, systemic administration of a relatively large dose of prednisolone (30 mg–50 mg per day) for a week usually can control the disease. The treatment outcome can be maintained by alternate-day oral administration of a low dose of prednisone (5 mg per day).

<https://doi.org/10.1016/j.jds.2019.05.004>

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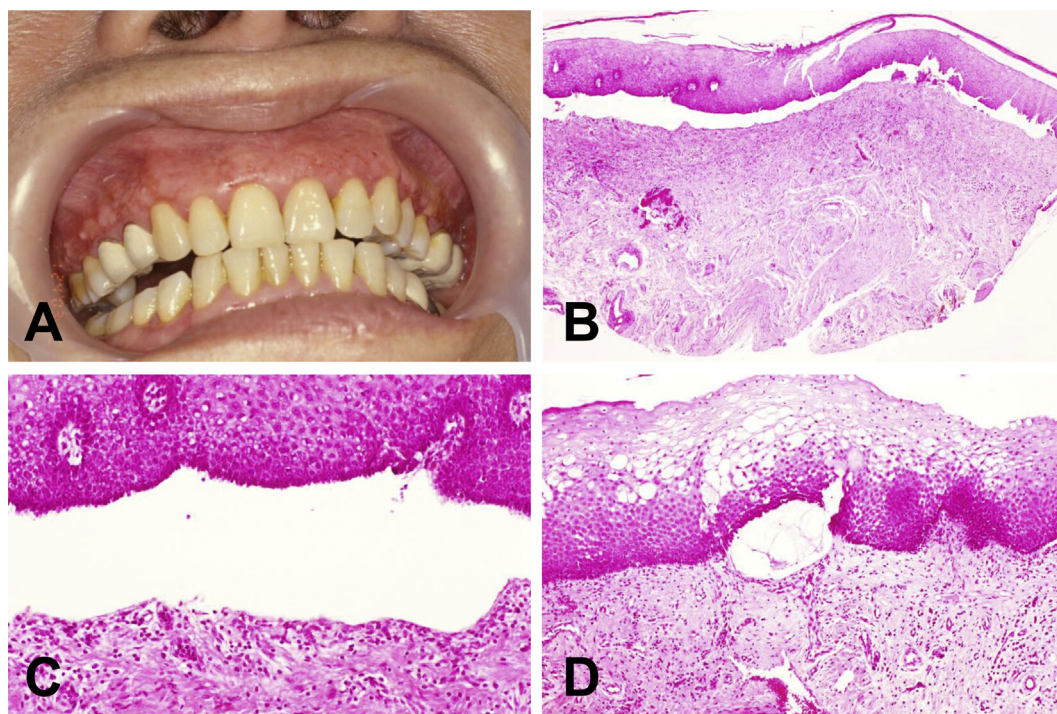


Figure 1 Clinical and histological photographs of our case of cicatricial pemphigoid. (A) The clinical photograph showed desquamative gingivitis with erosive lesions on the buccal gingivae of teeth 24, 25, 26, 34, 35, 36, 44 and 45. (B and C) Low-power (B, original magnification, 4 \times) and high-power (C, original magnification, 20 \times) microphotographs demonstrated a split between the stratified squamous epithelium and the underlying connective tissue with a moderate chronic inflammatory cell infiltrate in the superficial region (hematoxylin and eosin stain). (D) Medium-power microphotograph showed a small subepithelial vesicle with the edematous stratified squamous epithelium at the roof and the inflamed connective tissue at the base near the margin of the tissue specimen (hematoxylin and eosin stain, original magnification, 10 \times).

Conflicts of interest

The authors declare no conflicts of interest relevant to this article.

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Hou-Chiang Chou
Department of Dentistry, Far Eastern Memorial Hospital,
New Taipei City, Taiwan

Yu-Hsueh Wu
Department of Dentistry, Far Eastern Memorial Hospital,
New Taipei City, Taiwan

Graduate Institute of Clinical Dentistry, School of
Dentistry, National Taiwan University, Taipei, Taiwan

Chun-Pin Chiang
Department of Dentistry, Far Eastern Memorial Hospital,
New Taipei City, Taiwan

Graduate Institute of Clinical Dentistry, School of
Dentistry, National Taiwan University
Department of Dentistry, National Taiwan University
Hospital, College of Medicine, National Taiwan University,
Taipei, Taiwan

Graduate Institute of Oral Biology, School of Dentistry,
National Taiwan University, Taipei, Taiwan

Ju-Hsuan Yang*
Department of Dentistry, Far Eastern Memorial Hospital,
New Taipei City, Taiwan
Graduate Institute of Clinical Dentistry, School of
Dentistry, National Taiwan University, Taipei, Taiwan

*Corresponding author. Department of Dentistry, Far
Eastern Memorial Hospital, No. 21, Section 2, Nanya South
Road, Banciao District, New Taipei City 220, Taiwan.
E-mail address: larkysurf@yahoo.com.tw (J.-H. Yang)

Received 1 May 2019
Available online 23 December 2019