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Case report

# A case of bullous pemphigoid with initial onset in oral mucosa

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ABSTRACT

Bullous pemphigoid is an autoimmune bullous disease occurring primarily in the skin, and initial onset in the oral mucosa is quite rare. A case of bullous pemphigoid with initial onset in the gingiva is reported. The patient was a 42-year-old female who presented with a primary complaint of recurrent gingival bullae that had appeared 9 months earlier. Enzyme-linked immunosorbent assay (ELISA) analysis of the patient's serum showed a positive reaction for anti-bullous pemphigoid 180 kDa antibodies (anti-BP180-NC16a antibodies). Immunohistochemical staining of a biopsy sample also revealed deposition of IgG and complement C3 in the basement membrane. With the use of an azulene gargle and topical application of a steroid ointment, the condition remained stable, but six weeks later, bullae also appeared around the navel, on the back, and on one lower leg. Use of a topical steroid ointment prescribed by a dermatologist was ineffective, so that, after one month, the patient was treated with oral administration of minocycline, and the symptoms in the mouth and skin improved. In this case, bullous pemphigoid with initial gingival onset was diagnosed at an early stage by ELISA analysis, and a satisfactory outcome was obtained without systemic use of steroid, which has commonly been used in the past.



## 1. Introduction

Pemphigoid is an autoimmune bullous disease characterized pathologically by the formation of subepithelial bullae and the production of autoantibodies against the basement membrane. Pemphigoid conditions typically include mucous membrane pemphigoid (MMP) and bullous pemphigoid (BP), with the former occurring mainly in the ocular and oral mucosa, and the latter occurring mainly in the skin, but only rarely in the oral mucosa [1]. A rare case of bullous pemphigoid with initial onset in the gingiva is reported.

## 2. Case report

The patient was a 42-year-old Japanese female with a bullae on the upper and lower gingiva. She presented to a local dentist because of recurrent, bilateral, maxillomandibular bullae that would erupt on the molar region. The patient was treated with a gargle and antibacterial medication, but the condition did not improve, so she was referred to the Department of Oral and Maxillofacial Surgery of Mie University Hospital. The skin over the whole body and the ocular mucosa were normal. Panoral redness of the gingival membrane was visible, and both bullae and erosions were found bilaterally on the buccal gingiva of the maxillary molars (Fig. 1). The submandibular lymph nodes were also tender and swollen bilaterally.

General hematology test values were normal, and the patient tested negative for both anti-desmoglein 1 and anti-desmoglein 3 antibodies. On enzyme-linked immunosorbent assay (ELISA) analysis (measured by SRL, Inc., Tokyo, Japan), anti-bullous pemphigoid 180 kDa antibodies (anti-BP180-NC16a antibodies) had a high index value of 45 (reference value <9), but the patient tested negative for anti-bullous pemphigoid 230 kDa antibodies (anti-BP230 antibodies).

Histopathological findings: a biopsy was performed on the right maxillary molar buccal gingival mucosa including bullae. Dissociation directly beneath the basement membrane of the squamous epithelium was apparent, and this was accompanied by chronic inflammatory cell infiltration of the lamina propria mucosa, but

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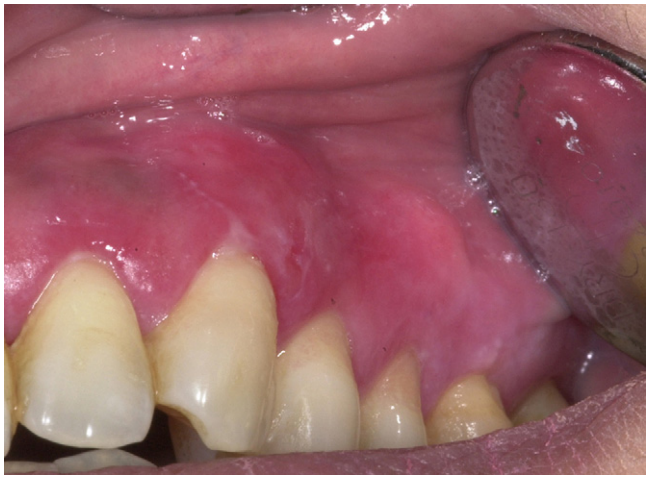

**Fig. 1.** Photo of the mouth at the time of initial diagnosis. Bullae and erosions can be seen in the buccal gingiva of the maxillary molars.

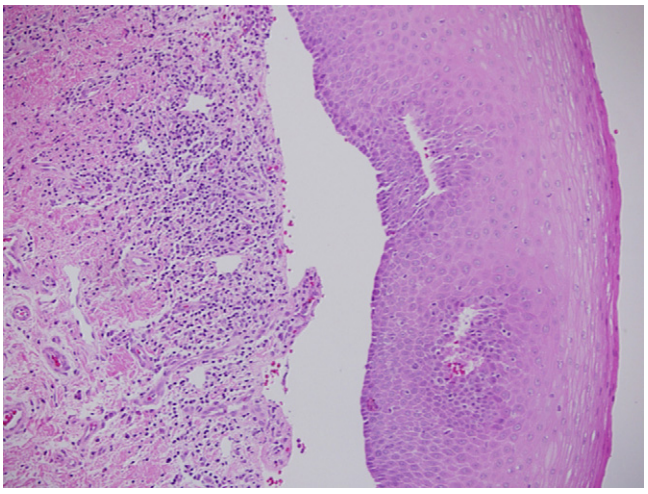

**Fig. 2.** Histopathological image of a biopsy specimen. There is dissociation directly beneath the basement membrane, accompanied by infiltration of chronic inflammatory cells at the periphery (haematoxylin and eosin stain; original magnification, 200×).

no Tzanck cells were seen (Fig. 2). Immunohistochemical staining using direct immunofluorescence revealed deposition of IgG and complement C3 in the basement membrane. IgM was slightly positive in some areas, but IgA was negative (Fig. 3). The diagnosis of bullous pemphigoid was made based on the above findings.

At the time of initial presentation, the Department of Dermatology of this hospital was also consulted to verify the absence of dermal manifestations. Because the oral symptoms were relatively mild, the patient was treated with an azulene gargle and topical application of triamcinolone acetonide ointment. The oral symptoms did not worsen, but after six weeks, bullae and ulceration appeared around the navel, on the back (Fig. 4a), and on the lower left leg (Fig. 4b). While no histological or immunofluorescence tests were performed on the skin lesions, skin eruption due to BP was diagnosed by the Department of Dermatology, and the patient was treated with topical application of a beclomethasone dipropionate ointment, but no clear improvement was seen. At about the same time, the patient reported pain on swallowing, so the Department of Otorhinolaryngology-Head and Neck Surgery was also consulted, but no abnormalities of the pharyngeal or laryngeal mucosa were found. Starting one month after the skin eruption appeared, the Department of Dermatology began treating the patient with oral administration of 100 mg/day of minocycline and 120 mg/day of fexofenadine. As of this writing, the bullous eruptions in the mouth and on the skin have decreased, and the symptoms have improved.

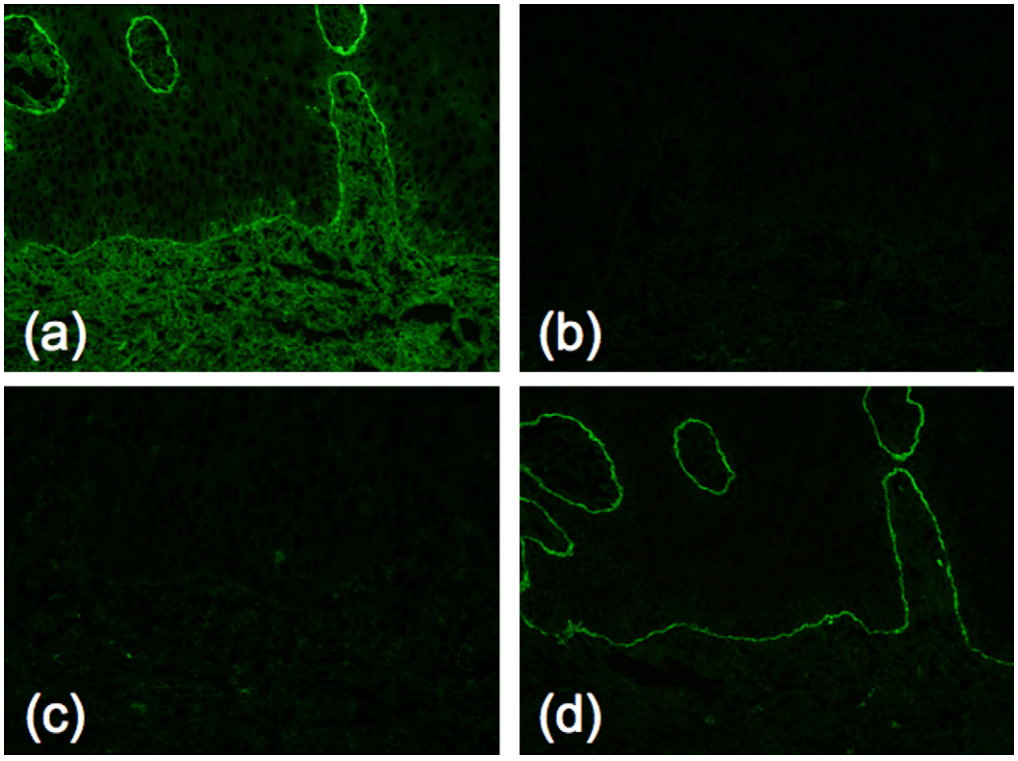


**Fig. 3.** Immunohistochemical staining by direct immunofluorescence. The basement membrane is positive for IgG and C3 (a: IgG, b: IgA, c: IgM, and d: C3, 400×).