

An Oral Blood-filled Blister: A Quiz

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A 32-year-old woman was admitted to the dermatology department due to recurrent blisters in the oral cavity. She had experienced recurrent blisters for the last 2 years, which ruptured spontaneously within a few hours. She denied any pain or discomfort. The patient had allergic rhinitis and hypothyroidism, managed with intranasal mometasone and levothyroxine. On physical examination, an erosive lesion was present on the lateral border of the tongue, measuring approximately 2 × 2 cm. The patient reported that the lesion had appeared after a blister had burst 2 weeks prior to admission. Photographs provided by the patient presented a tense haemorrhagic blister on the lateral border of the tongue (Fig. 1). She was also undergoing orthodontic treatment and had dental braces. Her platelet count was

231 × 10³/μl. Prothrombin time and partial thromboplastin time were within normal ranges. A biopsy and histopathological examination showed acanthosis without features of dysplasia. Local inflammatory infiltration was found in the underlying connective tissue. Direct immunofluorescence (DIF) for IgG, IgA, IgM, C1q, and C3c from the perilesional mucosa were negative. IgG antibodies against desmoglein 1, desmoglein 3 and BP180 on enzyme-linked immunoassay (ELISA) were absent. Moreover, stratified epithelium-specific antinuclear antibodies (SES-ANA) characteristic for chronic ulcerative stomatitis (CUS) were also negative.

What is your diagnosis? See next page for answer.



Fig. 1. (A, B) Tense blood-filled blister on the lateral border of the tongue. (C) An erosive lesion appeared after rupture of the blister.

ANSWERS TO QUIZ

An Oral Blood-filled Blister: A Commentary

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Diagnosis: Angina bullosa haemorrhagica

Based on the clinical picture and the aforementioned examinations the diagnosis of angina bullosa haemorrhagica (ABH) was established. The term ABH was introduced by Badham (1) in 1967 to describe uncommon, recurrent blisters filled with blood in the oral cavity. The real prevalence is unknown. This pathology is more common among older adults, with a peak incidence in the 5th decade (2). ABH usually occurs on the palate, or could present as diffuse lesions of the oral cavity, located on the tongue (especially on its lateral aspect, just like in the present case), buccal or labial mucosa (3, 4). Clinical manifestation is non-specific, although it may cause mild pain, burning or itching sensation (5). However, due to lesion size, it may lead to dyspnoea or upper airway obstruction (6). The blisters have a sudden onset and rupture spontaneously. The superficial ulcers that are formed heal within 7–14 days without scarring (7). This disorder has a tendency to recur, but the frequency of recurrence varies. The blisters may appear after local trauma, with the most common trigger being mastication of hard, crunchy and hot food (5, 8). It could also be associated with a constitutional predisposition to a weakened junction between the epithelium and connective tissue (9). Dental procedures and endoscopic diagnostic treatment might predispose to tissue damage. Some authors have suggested that long-term use of inhaled steroids may cause epithelial atrophy and modify the distribution of elastic fibres (10, 11). Grinspan et al. (12) described an association with diabetes and hyperglycaemia; whereas other authors reported hypertension as a potential contributing factor (11).

In 2019, diagnostic clinical criteria, based on previously reported cases, were proposed (5). Nonetheless, considering potential differential diagnoses, more investigations are needed to exclude other pathologies. The most common conditions are mucocutaneous immune disorders, such as pemphigus vulgaris, pemphigoid, erythema multiforme and bullous lichen planus (7). Histological examination might reveal non-specific ulceration with inflammatory cell infiltration. Although the results are negative in ABH, direct immunofluorescence for IgG, IgA, IgM and C3 are also recommended to exclude autoimmune blistering dermatoses, as performed in the current patient (3).

Another group of diseases presenting with similar lesions are haematological abnormalities e.g. thrombocytopenia, von Willebrand disease or leukaemia. Blood tests and a coagulation panel should be sufficient to rule out systemic pathologies (2). Despite the benign nature of ABH, patients might find it worrying due to its size and location (5). The

treatment is symptomatic. Any traumatic factors and irritating agents, such as metal crowns, hard and hot food, should be eliminated or avoided, as the current patient was instructed. Patients using inhaled steroids could be recommended to rinse their mouth after each use. However, in the current case, therapy with intranasal mometasone was discontinued and replaced with clemastine. To prevent secondary infections, antiseptic chlorhexidine gluconate (0.12–0.25%) mouthwash might be needed (2). Likewise, recommendations were also applied to the current patient; however, the periodic reoccurrence of lesions in the current case could be associated with ongoing orthodontic treatment.

In conclusion, we present here a rare case of recurrent subepithelial blood-filled blisters. The diagnosis of ABH is mainly clinical. Nevertheless, due to broad differential diagnosis, more diagnostic procedures are required in order to exclude other vesiculobullous diseases of the oral cavity. The patients might benefit from avoidance and potential discontinuation of various contributing factors that are known to exacerbate the disease. However, long-term follow-up might be necessary.

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