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Case Report

Dermatology Section

Giant Angina Bullosa Haemorrhagica Associated with Severe Anaemia: A Case Report

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ABSTRACT

Angina Bullosa Haemorrhagica (ABH) is a rare oral disorder characterised by blood-filled bullous lesions in the oral cavity and oropharynx. In 1933, Balina of Argentina first characterised this condition as traumatic oral haemophlyctenosis. Subsequently, Badham introduced the term ABH to describe the disorder. The condition is also referred to by alternative names, including localised oral purpura and stomatopompholyx haemorrhagica. The lesions appear acutely and are primarily distributed over the tongue and oral mucosa without any systemic involvement. They are frequently isolated and rupture quickly, leaving an ulcerated area. The clinical appearance of this lesion can be alarming for patients and poses a diagnostic challenge for clinicians, as it may closely resemble more serious conditions such as haematological disorders and other vesiculobullous diseases. Here, we present a 72-year-old female diagnosed with ABH, notable for the unusually large size of the lesion and its rare association with severe anaemia.

Keywords: Blister on tongue, Haemorrhagic blister, Severe anaemia, Spontaneous resolution, Sudden onset

CASE REPORT

A 72-year-old female presented with a complaint of an asymptomatic, sudden onset of a blood-filled blister over the tongue for a duration of one week, associated with bleeding from the lesion during friction. While consuming hard food, she suddenly experienced a tingling sensation on her tongue, which was soon followed by a feeling of heaviness and the development of a blood-filled blister on the right lateral side of her tongue, which was initially of pea-nut size, gradually increasing in size and attained the present size in 48 hours, and after which it was static.

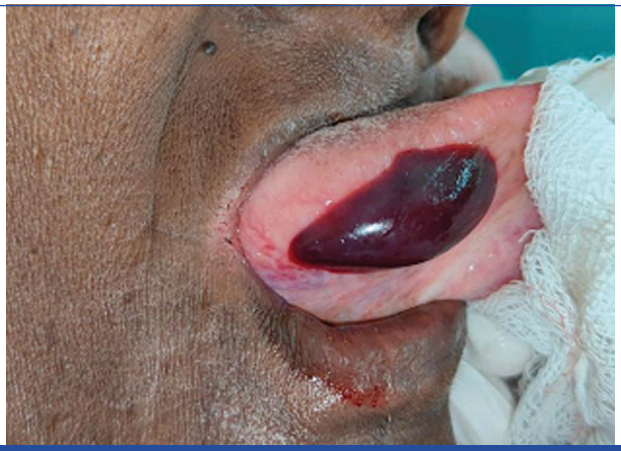
She gave no history of wearing dentures or having a similar lesion in the past. The patient was not a known case of diabetes, hypertension, or bleeding disorder. On general examination, the patient appeared moderately built and nourished, with marked pallor observed. There was no evidence of cyanosis, clubbing, or any generalised or regional lymphadenopathy.

On dermatological examination, there was a single well-defined oval soft haemorrhagic bulla of size 4.5 cm in length and 2 cm breadth present over the right inferolateral aspect of the tongue [Table/Fig-1]. There was no pulsation, and the surrounding mucosa appeared normal. On palpation, the lesion was non-tender and fluctuant.

Her complete haemogram demonstrated severe anaemia, with a haemoglobin concentration of 4 g/dL. The total leukocyte count was 6,700/mm^3, the erythrocyte count was 4.3 million/mm^3, and the platelet count was 260,000/mm^3. Peripheral blood smear revealed microcytic hypochromic anaemia. Since the patient declined a biopsy on the tongue, histopathological examination was not performed. Her renal function, liver function, lipid profile, and coagulation profile, such as prothrombin time, partial thromboplastin time, and bleeding time, were within normal limits.

Based on the history and clinical examination, a clinical diagnosis of giant ABH was considered.

Absence of cutaneous involvement, painless nature of blister, and complete remission without treatment in three days excluded autoimmune blistering disorders. In addition, the absence of an allergic history to drugs ruled out fixed drug eruption.



[Table/Fig-1]: Showing a single oval soft haemorrhagic bulla on the right inferolateral aspect of the tongue.

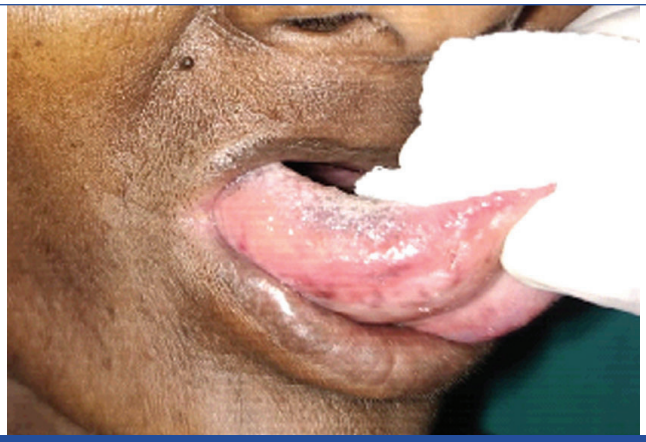
Blood disorders such as thrombocytopenia, von Willebrand disease, leukaemia, haemophilia, vasculitis, and Rendu-Osler-Weber disease were excluded based on the absence of characteristic genetic and clinical features such as petechiae, ecchymoses, epistaxis, gingival bleeding, and abnormal haematological parameters, which were not present in this case.

This case also met six out of the nine clinical diagnostic criteria for ABH proposed by Ordioni U et al., [1].

After excluding all the possible causes of blister in the oral cavity, the patient was clinically diagnosed with ABH.

On day 3 of admission, the bullous lesion spontaneously ruptured and healed completely. [Table/Fig-2] shows the completely healed lesion on day 5.

The general physician recommended a blood transfusion due to the severity of the anaemia; however, the patient declined this intervention. As an alternative, four doses of intravenous iron sucrose 200 mg in 100 mL of normal saline infusion over 20 minutes on alternate days were administered, and the patient was advised to repeat haemoglobin testing after two weeks. The patient did not return for further follow-up.



[Table/Fig-2]: Showing a completely healed lesion (On day 5 of admission).

DISCUSSION

The ABH is a self-resolving condition marked by the sudden appearance of a haemorrhagic bullous lesion. They are mostly distributed over the soft palate, buccal mucosa, lateral side of the tongue, and the lips [2-4]. Gingiva may very rarely be affected [5]. Lesions are often asymptomatic; however, ulceration and pain can develop because of bulla rupture [3,6]. Infrequently, the lesions can involve the pharynx, oesophagus, oral cavity floor, and epiglottis [6-8].

ABH's aetiology is unknown. Mucosal vascular anomalies and a breakdown in the cohesiveness between the epithelium and mucosal dermis are thought to contribute to its pathogenesis. The documented triggers include hot, spicy foods, dental trauma, intubation, the use of local anaesthetics, endoscopy, and air travel [4,6,8,9]. Topical steroids are known to induce epithelial atrophy and changes in the elastic fibres of the lamina propria, which can increase the risk of developing subepithelial bullous lesions after trauma [10,11].

Clinically, ABH most commonly affects individuals between 60 and 70 years of age, with a slightly higher incidence in females. ABH is rare in children under 10 years of age. The condition presents as single or recurrent episodes. Most cases involve solitary lesions, though multiple lesions have also been documented. The bullae are typically small, only a few millimetres in size, but larger bullae can occasionally occur. They usually appear suddenly following minor trauma and are red to purple in colour, often surrounded by an ecchymotic halo. The blisters may have a collapsed roof or rupture spontaneously, resulting in an irregular ulcer that generally heals within a week without leaving a scar [12].

When a biopsy is taken from a blood-filled blister, the detachment most commonly occurs at the subepithelial level, although intramucosal and intradermal separations have also been documented in the literature. Parakeratosis may be observed in the adjacent tissue with mild to moderate infiltrate of non-specific mononuclear inflammatory cells limited to the lamina propria. Direct immunofluorescence typically shows no staining for immunoglobulins G, A, or C3, which aids in distinguishing this condition from autoimmune bullous diseases [13].

The differential diagnoses considered for ABH are mucous membrane pemphigoid, epidermal lysis bullosa acquisita, linear IgA dermatosis, erythema multiforme, oral amyloidosis, pemphigus, dermatitis herpetiformis, and bullous lichen planus. Certain haematological conditions, such as thrombocytopenia, von Willebrand disease, leukaemia, and certain vasculitides, can present clinically in the oral cavity similar to ABH [14,15].

The diagnosis is mainly clinical. Biopsy is not mandatory. In 2019, Ordioni U et al., published proposed diagnostic criteria for ABH based on a systematic review of previously reported cases [1]. According to their recommendations, a diagnosis of ABH can be made if a case meets at least six out of the nine specified criteria, which includes: (I) clinically notable haemorrhagic bulla or erosion with a history of bleeding in the oral mucosa; (II) exclusively oral or oropharyngeal localisation; (III) palatal localisation; (IV) triggering event or promoting factor (food intake); (V) recurrent lesions; (VI) favourable evolution without a scar within a few days; (VII) painless lesion, or a tingling or burning sensation; (VIII) normal platelet count and coagulation test results; and (IX) negative Direct Immunofluoroscence (DIF) results.

Currently, there is no specific treatment available for ABH. It has been reported that topical antiseptics, such as chlorhexidine, at doses ranging from 0.12% to 0.2%, can be used to prevent secondary infection at the site of erosion [15]. Furthermore, it is advised to avoid any triggers like irritants or traumatic elements that could lead to recurrence of the lesion [1,7]. The development of ABH has also been associated with systemic conditions like hypertension and diabetes mellitus [3,15]. To date, there have been no previous reports in the literature of ABH occurring in association with severe anaemia. The present case was unique because of the larger size of the lesion and its rare association with severe anaemia and a normal coagulation profile.

CONCLUSION(S)

A 72-year-old woman who presented with a painless haemorrhagic blister on the tongue without any co-morbidities was clinically diagnosed with ABH. The lesion resolved spontaneously without leaving any scar. ABH should be taken into consideration whenever a patient presents with an acute onset of an asymptomatic bullous lesion in the oral cavity that heals spontaneously. Despite the fact that both the patient and the clinician often initially view this as an alarming presentation, ABH has a benign course that recovers on its own. It is imperative for treating clinicians to distinguish this benign condition from other blistering disorders in the oral cavity.

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