

ANGINA BULLOSA HEMORRHAGICA; A CASE REPORT OF AN UNCOMMON CONDITION

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ABSTRACT

Introduction Angina bullosa hemorrhagica (ABH) is a rare benign disorder characterized by one or multiple blood-filled bullae in the oral cavity and oropharynx with a sudden onset. Lesions are mostly isolated characterized by rapid onset, development and healing process. Trauma, inhalation corticosteroids and diabetes are common factors in ABH development. **Case report:** In this article we are presenting 3 ABH cases for three different patients. All of them were presented to the dental clinic complaining of the sudden appearance of recurrent blood-filled lesions. They reported that the lesions shortly rupture with a self-limited bleeding after 1 to 2 days from their appearance. Complete remission is expected within 7 to 10 days. ABH should be differentiated from other vesiculobullous disorders and hematologic disease.

Conclusion: The typical clinical presentation in addition to routine hematologic investigations is usually sufficient to diagnose ABH. Patient's education and reassurance are mandatory. No treatment is required for such cases

KEYWORDS: angina bullosa hemorrhagica, ABH, blood-filled blister, case report

INTRODUCTION

Angina bullosa hemorrhagica (ABH) is a rare benign disorder characterized by one or multiple blood-filled bullae in the oral cavity and oropharynx with a sudden onset⁽¹⁾. ABH occurs almost exclusively in the oral cavity⁽²⁾. Knowledge of its clinical presentation is crucial in order to facilitate proper diagnosis, treatment, and patient reassurance & counseling⁽¹⁾. Middle-aged and elderly are more

commonly affected by these lesions than younger individuals, mostly in the nonkeratinized mucosa, especially the soft palate. In a short period of time these blisters break and release their bloody content, leaving an asymptomatic ulcerative surface that heals within few days⁽³⁾.

Local recurrences are frequent and usually occur at the same site as previous lesions⁽²⁾. Although ABH blisters are not associated with any blood

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disorder, systemic diseases or vesiculobullous disorders⁽³⁾. ABH may be clinically similar to small hemangiomas or other blood filled lesions during aspiration. Also they may be similar ecchymosis.

The pathogenesis of ABH is unknown, but trauma seems to be a major factor in its onset⁽²⁾. In addition, inhaled corticosteroids and diabetes mellitus may predispose to ABH⁽³⁾. In this article, three ABH cases will be discussed. Informed consent was assigned from each patient to share their clinical data and photographs.

CASE REPORT

Case 1

A 45 years old male patient presented at the Oral Medicine Department, Faculty of Dentistry, Cairo University complaining of a hemorrhagic blister that suddenly appeared in the left buccal mucosa the following day after having his last dinner, resulting in discomfort. The patient documented that the blister has ruptured after few hours releasing small amount of blood in his oral cavity. He also reported the appearance of similar blisters, in the palate few times before that. A general physical examination and laboratory investigations determined that the patient was in good health, without history of blood disorders, anticoagulant use, liver disease, skin manifestations or any other systemic disease. An intraoral examination revealed the presence of fine tissue tags on the buccal mucosa and the site of the ruptured blister was painless (Figure 1a). There was mild inflammation around the site of the ruptured blister. According to the patient's history, the size of the blister was the same since the beginning. Two weeks after that, this patient came with another small blood-filled blister in the soft palate with the same history as the previous one (Figure 1b). This blister didn't blanch during diascopic examination. The complete blood investigations were normal.

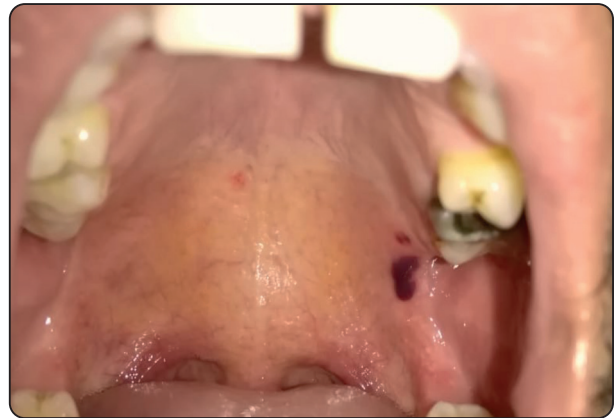


Fig. (1A) representing angina bullosa hemorrhagica in the soft palate



Fig. (1B) representing fine tissue tags that appeared after ABH rupture in the right buccal mucosa

Case 2

A 55 years old female presented at the oral Medicine Department Faculty of Dentistry, Cairo University complaining of sudden appearance of asymptomatic hemorrhagic blister on the buccal mucosa one day earlier. No history was given for blood disorders, anticoagulant drug use, liver disease, skin manifestations or any vesiculobullous disorders. During intraoral examination we observed a blood-filled lesion that didn't blanch during diascopic examination (Figure 2).



Fig. (2) representing angina bullosa hemorrhagica in the buccal mucosa



Fig. (3B) representing angina bullosa hemorrhagica in the postrolateral surface of the tongue after healing

Case 3

A 47 years old female presented to the Oral Medicine department, Faculty of Dentistry, October 6th University complaining of asymptomatic blood-filled blister related to the left postero-lateral border of the tongue after accidental traumatic bite. Medical and family history were insignificant and there were no other extraoral lesions.

Clinical examination revealed a bluish blood-filled blister related to the left postero-lateral border of the tongue. A diascopic test was made and there was no blanching. The lesion lasted for 24 hours only then ruptured (Figure 3a &b).



Fig. (3A) representing angina bullosa hemorrhagica in the postrolateral surface of the tongue

DISCUSSION

ABH was first described in 1967 as one or multiple subepithelial blood-filled blisters that cannot be attributed to any systemic disease, bleeding disorders or vesiculobullous diseases⁽¹⁾.

The exact incidence and prevalence of ABH are not known. However, Grinspan et al.¹ observed over 10,000 patients between 1990 and 1996 in their dental hospital and found 54 patients presenting with ABH. Between 1985 and 2016, a total of 225 cases were reported in literature. Males and females are equally affected between the fifth and eighth decades⁽⁴⁾. ABH is characterized by rapidly developing blue blisters reaching 2–3 cm in diameter within minutes, mostly on the soft palate, although localization on the hard palate, tongue, and buccal mucosa is also described^(5,6). The lesions are painless but burning sensation may precede the lesion development. Blisters rupture spontaneously within days⁽⁷⁾. When the lesion ruptures, the bloody content flows into the oral cavity. Bleeding is usually self-limited, ceasing after a few minutes⁽⁸⁾. After the blister ruptures, a reddish erosion and scraped white epithelial lining representing the roof of the ruptured blister is evident⁽⁹⁾. Lesions heal without scar formation⁽⁸⁾. Recurrence rate is about 30 percent and the blisters may appear at different locations⁽¹⁰⁾.

The exact mechanism of ABH is unclear; however, in most cases, it seems to exist after eating hard food. During chewing, vasodilation of the oral mucosa due to the parasympathetic reflex combined with the friable oral mucosa and the trauma of chewing hard food may make oral mucosa prone to bleeding⁽⁴⁾. ABH is also reported after sneezing routine dental procedures, thermic injury and intubation^(4,5). Also the use of inhalation steroids for long time. Other systemic contributing factors might be diabetes mellitus and hypertension^(7,8,10). The chronic use of inhaled steroids can result in reduction of collagen synthesis and maturation. This will result in oral mucous epithelium atrophy and reduction in tissue elasticity. In addition, the lack of support to the small blood vessels in the areas surrounding AHB could explain the development of ABH even in response to minor trauma in chronic steroid inhalation⁽⁶⁾.

The isolated nature of the blister, typical clinical presentation, and rapid healing are often sufficient to diagnose ABH. However, diagnosis is often delayed by the unfamiliarity of the ABH and fear of other more serious diseases. During diagnosis, laboratory testing, including complete blood count, and bleeding & coagulation assessment tests are indicated to exclude any possible underlying hemostatic disorders. Biopsy or analysis of blister fluid is not indicated, because it may cause a secondary infection⁽⁷⁾.

The differential diagnosis of ABH includes hematological diseases and blistering disorders⁽⁴⁾. ABH can be distinguished from hematological causes by typical clinical presentation i.e.: isolated nature of the swelling and rapid development, rupture and healing process in addition to laboratory tests to exclude presence of bleeding disorders. Other bleeding manifestations in other sites such as epistaxis and hematuria may be concomitant symptoms, guiding the clinician in the differential diagnosis. Blistering disorders often present as a

more generalized condition, mostly associated with other skin manifestations⁽⁸⁾.

Histopathological analysis reveals the presence of nonspecific ulcers containing a chronic inflammatory infiltrate primarily lymphocytes⁽¹¹⁾.

No specific treatment is required for AHB. The overall benign character of the condition and its favorable prognosis should be highlighted to the patient⁽⁸⁾. It is recommended to use anti-inflammatory or antibacterial mouthwashes (0.25% chlorhexidine gluconate) 2 to 3 times a day for 3 days to prevent secondary infection at the site of the lesion⁽³⁾. When lesions rupture, bleeding is self-limited. Complete remission is expected within 10 days⁽⁸⁾. Only in the case of a potentially threatened airway, the incision of the bulla may be necessary. In rare occasions, an intubation or even a tracheotomy might be needed to secure the airway in case of a deeper protruding swelling^(7,12).

CONCLUSION

ABH is a rare, benign disorder with acute, post-traumatic blood-filled blister development, in the oral cavity. The typical clinical presentation that is characterized by isolated nature of the blister, rapid development, rupture and healing are often sufficient to diagnose ABH. Diagnosis should start with a detailed history-taking of the lesion and concomitant dermatologic, bleeding and systemic manifestations, thorough physical examination and laboratory tests focusing on hematological causes to exclude other disorders. No treatment is required. Follow up is advised to exclude underlying disorders.

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