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Case Report

Angina bullosa hemorrhagica: A rare and benign oral mucosal disorder– A case report

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ABSTRACT

The oral cavity plays a crucial role in overall health, with certain oral lesions potentially indicating underlying conditions. Angina bullosa hemorrhagica (ABH) is a rare, benign oral mucosal disorder characterized by spontaneous blood-filled bullae unrelated to systemic diseases or bleeding disorders. This case report aims to describe a clinical presentation and management of a patient with recurrent ABH. A 17-year-old patient presented with a blood-filled lesion on the maxillary left buccal mucosa, persisting for one week, causing discomfort while chewing, with a history of a similar episode nine months back. No systemic or hematologic abnormalities were detected. Clinical examination revealed a solitary, tense, blood-filled bulla. Histopathology confirmed the diagnosis of ABH, ruling out other vascular or inflammatory lesions. There are no established guidelines for managing this condition. In the present case, an excisional biopsy was performed to distinguish it from other blood-containing bullae and alleviate discomfort. Follow-up was carried out at 15 days, 6 months, and 1 year. In conclusion, ABH is a rare but significant condition requiring accurate diagnosis to avoid diagnostic dilemmas and to prevent unnecessary treatments. Awareness of its clinical features helps clinicians differentiate it from other oral vesiculobullous disorders, ensuring appropriate patient management.

Keywords: Angina bullosa hemorrhagica, Blood-filled bullae, Excisional biopsy, Oral mucosal disorder, Vesiculobullous lesions

INTRODUCTION

"Angina bullosa hemorrhagica" (ABH) is the term given by Badham in 1967 to describe a rare, idiopathic condition in the oral mucosa characterized by a blood-filled blister or bulla, which cannot be attributed to a bleeding disorder or is not associated with a systemic disease.[1] ABH has been documented under various diagnostic terms, such as "*recurrent or traumatic oral hemophlyctenosis*" and "*benign hemorrhagic bullous stomatitis*."[2] It is primarily observed in middle-aged and elderly individuals, with a slightly greater female predilection of about 55.3%.[3] The prevalence and incidence of ABH have not been precisely determined; however, it is estimated to occur in approximately 0.05% of the population.[4,5] The exact etiopathogenesis of ABH remains unclear and is therefore regarded as multifactorial.[6]

In susceptible individuals, trauma to the oral mucosa is believed to trigger the characteristic bullous lesions. Clinically, this condition is distinguished by the abrupt appearance of one or more red-blue, blood-filled subepithelial bullae, primarily affecting the soft palate, lateral borders of the tongue, and buccal mucosa.[6] The progression of the bullae varies but is typically brief, they rapidly enlarge and spontaneously rupture within 24–48 h, leaving behind a superficial ulcer

that heals within one to 2 weeks without scarring.[3,7-9]

Frequent recurrence of ABH lesions is common, occurring either in the same location or in different areas.[4,10] ABH is not associated with hematologic disorders or other vesiculobullous diseases.[11] Although the condition is benign and typically resolves on its own, it often leads to patient anxiety and presents a diagnostic challenge for healthcare professionals. Hence, recognition of this condition is crucial for clinicians through clinical findings and microscopic features to establish an accurate differential diagnosis, distinguishing it from other oral bullous disorders and ensuring proper management while avoiding unnecessary interventions. Given its rare occurrence and diagnostic overlap with hematologic and immunobullous disorders, ABH warrants academic attention as a distinct clinical entity within oral mucosal pathology. Thus, the purpose of this paper is to present a case of ABH, discuss its clinical features, and review relevant literature to provide a comprehensive understanding of this uncommon oral disorder. This case report has been written in accordance with the CARE guidelines to ensure accurate and transparent reporting.

CASE REPORT

A 17-year-old patient presented to the Department of Pediatric Dentistry with a blood-filled, red swelling on the maxillary left buccal mucosa, persisting for the past 1 week. The patient reported previous episodes with a similar lesion in the same area 2 months ago, which ruptured after 2 days, resulting in an ulceration that healed within a week without scarring. However, the lesion reappeared in the same region 1 week ago. The patient had no relevant medical history, no family history, no toxic habits, and no known allergies. No notable extraoral abnormalities were detected.

On intraoral examination, a solitary, tense, blood-filled bullae was observed on the left buccal mucosa, located adjacent to teeth #24 and #25 [Figure 1]. The lesion measured approximately 1.2 cm × 1.0 cm in size and presented as a bright red, smooth-surfaced bullae. It was soft in consistency and non-tender on palpation.

The differential diagnosis included hemangioma, pyogenic granuloma, and bleeding disorders such as thrombocytopenia.

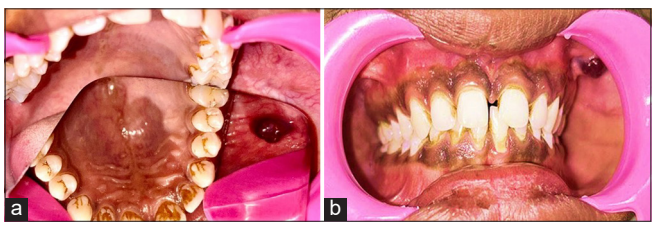

Figure 1: (a-b) Blood-filled vesiculo-bullous lesion on the left buccal mucosa.

To exclude hematological disorders, comprehensive blood investigations and coagulation profiles were conducted, all of which yielded results within normal physiological limits.

In accordance with the diagnostic criteria proposed by Ordioni *et al*. (2019) for ABH, which require the presence of at least six out of nine specified features, including mandatory fulfillment of criteria I and II, our patient exhibited several key findings supportive of this diagnosis.[10] Clinically, the patient presented with a characteristic hemorrhagic bulla or erosion accompanied by a history of oral mucosal bleeding. The lesions were exclusively localized to the oral/oropharyngeal region, recurrent in nature, and healed spontaneously within a few days without scarring. Additional findings included the painless nature of the lesions, a normal platelet count, and a coagulation profile. These seven fulfilled criteria, as outlined in Table 1, are consistent with the diagnostic framework of ABH and support the diagnosis in this case.

Table 1: Clinical criteria for the diagnosis of angina bullosa hemorrhagica as proposed by Ordioni *et al*.,[10] demonstrating fulfillment of seven out of nine criteria in the present case.

I	Clinically notable hemorrhagic bulla or erosion with a history of bleeding of the oral mucosa
II	Exclusively oral or oropharyngeal localization
III	Palate localization
IV	Triggering event or food-promoting factor (food intake)
V	Recurrent lesions
VI	Favorable evolution without a scar within a few days
VII	Painless lesion, tingling, or burning sensation
VIII	Normal platelet count and coagulation profile
IX	Negative direct immunofluorescence

Treatment

There are no established guidelines for the management of ABH.[12] Initially, the management approach focused on patient education, emphasizing the nature of the condition, possible causes, and self-care measures to prevent exacerbation. Supportive management was implemented to relieve symptoms and facilitate healing. This included anti-inflammatory therapy with 0.1% triamcinolone acetonide applied 3–4 times daily for 5 days, topical application of 0.3% hyaluronic acid gel 2–3 times a day for 5 days, and the use of 0.15% benzydamine mouthwash (15 mL rinsed for 30 s) 2–3 times daily for 5 days to aid mucosal repair. In addition, antiseptic agents such as chlorhexidine gluconate were used to help prevent secondary infections and dietary modifications such as avoiding spicy, acidic, and rough foods that may irritate the lesion. The patient was reviewed after 5 days of medication, but the lesion exhibited no clinical signs of resolution. At this point in time, the lesion had persisted for over 12 days in total. Due to the patient's discomfort

and to rule out other blood-containing bullae, an excisional biopsy was performed as a therapeutic measure.

Local anesthesia (e.g., 2% lignocaine with epinephrine 1:100,000) was administered through infiltration. A silk suture was placed around the lesion to provide traction and elevation, facilitating precise excision [Figure 2a]. A No. 15 scalpel blade was used to create an incision through the full thickness of the mucosa, and the lesion was excised with clear margins [Figure 2b]. The excised specimen was immediately preserved in 10% formalin, appropriately labeled, and submitted for histopathological examination [Figure 2c]. Hemostasis was achieved using a pressure pack, followed by wound closure with a non-resorbable suture using a three-simple interrupted technique [Figure 2d].

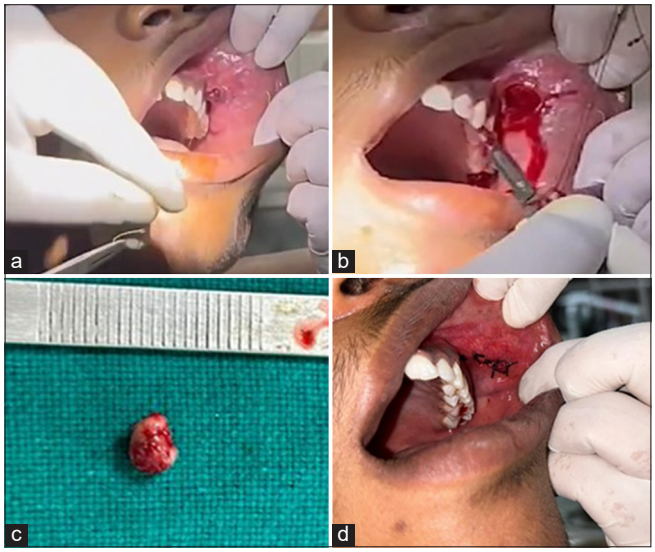

Figure 2: (a) Encircle the lesion with a silk suture to provide traction and elevate it from its base, facilitating precise excision. (b) Aseptic excision was performed under sterile conditions to minimize the risk of infection and ensure optimal surgical outcomes. (c) Removed soft-tissue specimen was sent for histopathological examination. (d) Three interrupted sutures were placed to ensure proper wound approximation and promote optimal healing.

Histopathological examination

Microscopic examination of the excised specimen revealed a subepithelial blister filled with erythrocytes and minimal inflammatory infiltrate. The absence of endothelial proliferation or vascular malformation excluded hemangioma, while the lack of granulation tissue ruled out pyogenic granuloma. Integrating the patient's clinical history, examination findings, and histopathological features, the lesion was diagnosed as "*Angina bullosa hemorrhagica.*"

Follow-up and outcomes

The patient was recalled for follow-up after 15 days and 1 year after surgical excision and exhibited no signs of recurrence or associated complications [Figure 3a and b].

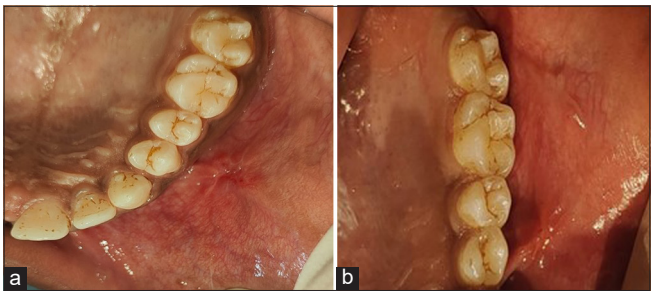

Figure 3: Follow-up photograph at (a) 15 days after surgical excision, (b) One year after surgical excision.

DISCUSSION

ABH is a term used to describe benign sub-epithelial oral mucosal blisters filled with blood not attributable to a systemic disorder or hemostatic dysfunction.[3] Despite its recognition as a distinct clinical entity, its true prevalence remains uncertain. Only a limited number of case reports have been published in the literature. Grinspan *et al.* and Rosa *et al.* suggested that while epidemiological data are limited, it is estimated to affect approximately 0.05% of the population.[4,5] However, due to its transient nature and frequent misdiagnosis, the actual prevalence may be under-reported. The disease predominantly affects individuals in their sixth to eighth decades of life, with a slight predominance among females.[7,10,12]

The etiopathogenesis of ABH remains unclear and is believed to be multifactorial. While the precise triggers are yet to be fully understood, several studies propose that minor trauma to the oral mucosa plays a key role in the development of lesions in susceptible individuals.[6] Common predisposing factors include dental procedures, hot food or beverages, chronic irritation, mechanical trauma from sharp food particles, and even stress-related factors. Some reports have also linked ABH to the prolonged use of inhaled corticosteroids, suggesting that mucosal fragility may contribute to lesion formation. Alberdi-Navarro *et al*., Dias *et al*., and Ordioni *et al.* observed that the lesions typically resolve within 1 week without any residual scarring, although recurrences are frequent.[3,7,10] Recurrent episodes may involve the same anatomical site or different locations within the oral cavity, often causing discomfort but not leading to long-term complications.

According to Joshi *et al.*[13] there are no established guidelines for its management. Various authors have advocated a conservative approach in the management of ABH, as the condition is typically self-limiting. Conservative treatment primarily includes symptomatic management with analgesics such as ibuprofen or paracetamol for pain relief, along with topical corticosteroids like triamcinolone acetonide to reduce inflammation. Antiseptic mouth rinses, such as chlorhexidine gluconate, are used to prevent secondary infection, while mucosal protective agents like hyaluronic acid gel promote

healing.[14,15] Dietary modifications, including avoidance of spicy, acidic, and rough foods, along with adequate hydration, further aid in symptom control.[15] However, in certain cases where diagnostic uncertainty or significant discomfort persists, invasive interventions have been performed. Martini *et al*.[16] conducted an incisional biopsy to obtain a histopathological diagnosis, while Joshi *et al*. performed incision and drainage in cases presenting with larger hemorrhagic bullae causing functional impairment and discomfort.[13]

In our case, a conservative approach was initially implemented; however, as there was no relief even after 12 days, an excisional biopsy was subsequently performed. The decision to proceed with an excisional biopsy was based on several clinical considerations, including the need to rule out malignancy, given the lesion's recurrence at the same site. Unlike previous episodes, it failed to regress spontaneously and showed no response to conservative management. Moreover, the lesion demonstrated progressive enlargement, ultimately extending onto the occlusal table and resulting in difficulty with mastication.

An incisional biopsy was not performed because the lesion measured <3 cm in size. According to Neville *et al*.,[17] Marx and Stern,[18] and Greenberg *et al*.,[19] an excisional biopsy is the preferred approach for lesions under 3 cm, as it allows for both definitive diagnosis and therapeutic excision in a single procedure. There was no evidence of recurrence or complications observed even after 1-year of follow-up.

CONCLUSION

ABH is a distinct benign yet clinically significant oral disorder that requires accurate diagnosis to prevent unnecessary interventions. Diagnosis is primarily clinical, based on characteristic presentation and exclusion of other vesiculobullous or hematological conditions. Its self-limiting nature supports an initial conservative management approach and patient reassurance. Clinicians must increase awareness of ABH and utilize standardized diagnostic criteria to improve identification and care. Invasive procedures should be reserved for cases with diagnostic uncertainty or persistent lesions. Clear communication with patients about the benign course of ABH is essential. Future research should focus on epidemiological studies to determine the true prevalence, explore the underlying pathogenesis, and develop evidence-based management guidelines. Long-term studies on recurrence patterns and treatment outcomes will further improve clinical practice.

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