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Angina bullosa hemorrhagica: A case study of two patients with unusual presentations

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Abstract
Angina Bullosa Hemorrhagica (ABH) is a rare but benign oral mucosal disorder characterized by spontaneous, blood-filled blisters that rupture within hours, leaving behind shallow ulcers that heal without scarring. This case study presents two patients a 52 and 25-year-old male both experiencing recurrent episodes of ABH with no systemic disease or predisposing factors. The 52-year-old patient exhibited a large hemorrhagic blister on the soft palate, whereas the 25-year-old patient presented with a smaller lesion at the junction of the hard and soft palate. Both patients underwent clinical and laboratory evaluations, including complete blood count and coagulation profiles, which were within normal limits. The diagnosis of ABH was confirmed based on clinical findings and exclusion of autoimmune and hematologic disorders. This report highlights the diagnostic challenges and the importance of differentiating ABH from other vesiculobullous and hemorrhagic conditions such as pemphigus, thrombocytopenia, and vascular abnormalities. The cases reaffirm the self-limiting nature of ABH and emphasize the need for increased clinical awareness to prevent unnecessary investigations and patient anxiety.

Keywords: Angina bullosa hemorrhagica, hematologic disorders, soft palate, pemphigus

Introduction

Angina Bullosa Hemorrhagica (ABH) is a rare but benign oral mucosal disorder characterized by the sudden onset of blood-filled blisters in the absence of systemic disease, trauma, or identifiable hematologic abnormalities. First described by Badham in 1967, ABH remains an underrecognized condition that is often misdiagnosed due to its clinical resemblance to other vesiculobullous and hemorrhagic oral conditions (1). The lesions appear spontaneously, rupture within a short duration (usually a few hours), and heal without scarring or significant discomfort. While it predominantly affects middle-aged and elderly individuals, younger adults may also present with ABH, as seen in the cases discussed in this report (2).

The exact pathophysiology of ABH remains unclear. It has been proposed that ABH results from mechanical or minor traumatic injury to the oral mucosa, leading to sudden subepithelial hemorrhage and blister formation (3). Factors such as friction from food, dental procedures, or irritation from hot or spicy foods have been suggested as triggers, although many cases occur without any identifiable cause (4). Additionally, some studies suggest that corticosteroid inhalers and anticoagulant therapy might predispose individuals to ABH, although neither patient in this report had a history of medication use (5).

Clinically, ABH is most frequently seen on the soft palate, followed by the buccal mucosa, lips, and tongue. The lesions are typically tense, blood-filled blisters that appear suddenly, often without pain. Once ruptured, they leave behind shallow ulcerations that resolve within a few days (62). Unlike autoimmune blistering diseases such as pemphigus vulgaris and mucous membrane pemphigoid, ABH does not exhibit persistent ulceration or systemic involvement, making histopathological examination and immunofluorescence studies unnecessary in most cases (7).

Although ABH is self-limiting and does not require specific treatment, its sudden and recurrent presentation may cause distress to patients. Therefore, it is essential for clinicians to recognize ABH and differentiate it from more serious conditions, including hematological disorders (thrombocytopenia, leukemia), vascular abnormalities (angioedema, purpura), and infectious diseases (herpetic gingivostomatitis) (8). This report discusses two cases of ABH in a young and an older adult, illustrating the variability in presentation and the importance of recognizing this condition in different age groups.

Case Presentation

Case 1: A 25-Year-Old Male with Recurrent Oral Blood Blisters

A 25-year-old male presented with a history of multiple spontaneous blood blisters in the oral cavity over the past six months. The blisters typically appeared on the soft palate and inner cheek, ruptured within a few hours, and healed without scarring. The patient denied pain but reported mild discomfort for a short period after the rupture. There was no history of trauma, dental procedures, or irritant food intake preceding the episodes.

The patient had no history of systemic illnesses such as diabetes, hypertension, or bleeding disorders. He was not on any medications, including corticosteroids or anticoagulants. No family history of similar lesions or autoimmune disorders was reported.

On intraoral examination, a ruptured lesion with an erythematous base was noted on the soft palate. The surrounding mucosa appeared normal with no signs of secondary infection. Systemic examination was unremarkable.

Laboratory tests, including complete blood count, coagulation profile, and autoimmune panel, were all within normal limits. Based on the clinical findings and the episodic nature of the lesions, a diagnosis of ABH was made. The patient was advised to maintain good oral hygiene and avoid hard or spicy foods that could irritate the mucosa. Over a six-month follow-up period, the patient reported occasional recurrences but no complications.

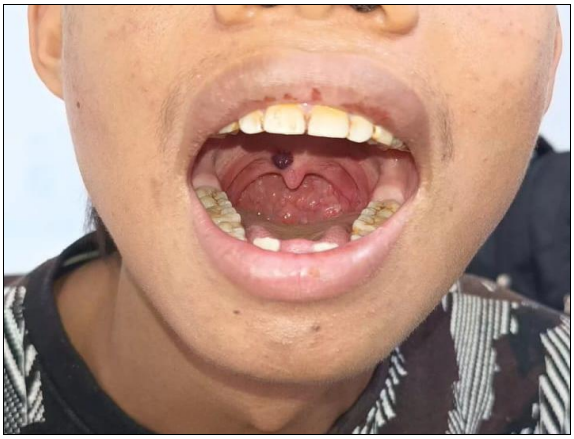

Fig 1: Angina Bullosa Hemorrhagica in a 25-Year-Old Male: Isolated Hemorrhagic Lesion on the Soft Palate

Case 2: A 52-Year-Old Male with a History of Multiple Oral Blood Blisters

A 52-year-old male presented with recurrent, spontaneous blood blisters in the oral cavity, particularly on the soft palate and lateral tongue. He had been experiencing these episodes for the past year, with the lesions rupturing within a few hours and healing without residual damage. He described a sensation of fullness in the affected area prior to blister formation, followed by rapid rupture and mild discomfort.

The patient had no known systemic illnesses, and his medical history was unremarkable. He denied any history of medication use, allergies, or recent dental procedures. No contributing factors, such as sharp food intake or local trauma, were identified.

On examination, a fresh, intact blood blister measuring about 0.8 cm was noted on the left buccal mucosa, along with a recently ruptured lesion on the soft palate. The mucosa appeared healthy otherwise, with no evidence of ulceration or inflammation beyond the affected sites. No systemic abnormalities were found.

Comprehensive laboratory investigations, including coagulation studies, blood glucose levels, and autoimmune markers, were all normal. The patient was diagnosed with ABH and reassured about the benign nature of the condition. He was advised to avoid excessive hot food intake and given symptomatic treatment with topical antiseptics. Follow-up visits over the next six months showed sporadic recurrences but no significant complications.

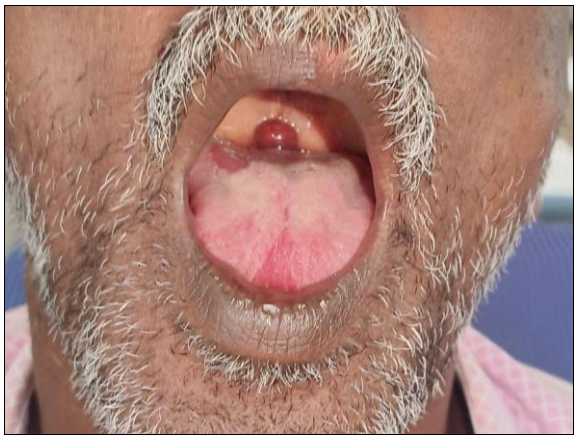

Fig 2: Angina Bullosa Hemorrhagica in 52-Year-Old Male: Palatal Blood Blister Presentation

Discussion

Angina Bullosa Hemorrhagica remains an underrecognized clinical entity, often misdiagnosed due to its sudden onset and resemblance to various hemorrhagic and vesiculobullous disorders (9). It primarily affects middle-aged and elderly individuals, but as demonstrated in this case report, younger adults may also develop the condition. Despite being benign and self-limiting, the sudden appearance of oral blood blisters can cause significant distress, prompting patients to seek medical attention. Proper clinical recognition is essential to prevent unnecessary investigations and interventions (11).

The exact etiology of ABH remains unclear, but several theories have been proposed. The condition is thought to result from increased fragility of the subepithelial capillaries, leading to spontaneous hemorrhage and blister formation (11). Some studies suggest that ABH may be related to minor trauma, hot food consumption, or mechanical irritation (12). However, in many cases, including the two presented here, no clear predisposing factor can be identified, raising the possibility of idiopathic vascular fragility. Other proposed contributing factors include mechanical trauma from hard or rough food, ill-fitting dentures, or accidental cheek biting, which can trigger ABH in susceptible individuals (13). Corticosteroid use, particularly inhaled steroids, has also been implicated in some cases due to their effect on mucosal thinning and increased capillary fragility (14). Some reports have suggested an association with diabetes, hypertension, or clotting abnormalities, although no definitive link has been established (15).

The rapid onset and self-limiting nature of ABH distinguish it from other systemic or autoimmune diseases. The lesions resolve within hours to days without scarring, and patients typically do not experience systemic symptoms. However, because of its hemorrhagic nature, ABH must be carefully differentiated from more serious conditions. Autoimmune vesiculobullous diseases such as pemphigus vulgaris and mucous membrane pemphigoid present with persistent, painful erosions and require immunofluorescence testing for confirmation (16). Hematologic disorders such as thrombocytopenia and leukemia can also cause hemorrhagic oral lesions, but these conditions are typically associated with persistent bleeding, petechiae, and abnormal blood counts (17). Vascular and allergic conditions, including angioedema and hereditary hemorrhagic telangiectasia (HHT), may mimic ABH but present with additional systemic signs such as facial swelling or recurrent epistaxis.

Several previous studies have reported similar presentations of ABH, reinforcing its characteristic clinical course. Messadi DV, Mirowski described that recurrent oral blood blisters mimicking a hematologic disorder, but with no systemic disease, and lesions that resolved spontaneously (18). Narang and Kanwar reported two cases of ABH in otherwise healthy adults, both presenting with isolated blood blisters on the palate and buccal mucosa (19). López-Jornet *et al*. (2012) documented five cases, some with a history of minor trauma before lesion onset, while others had no identifiable triggers, mirroring the case of the 25-year-old patient presented here.

The management of ABH is primarily supportive. Since the lesions resolve spontaneously, no specific treatment is required. However, symptomatic care may include patient education and reassurance about the benign nature of the

condition, avoidance of hot, spicy, or hard foods that may exacerbate mucosal irritation, and maintaining good oral hygiene to prevent secondary infection. In cases with mild discomfort, topical anesthetics such as lidocaine gel can provide symptomatic relief. The prognosis of ABH is excellent, with no long-term complications or malignant potential. While recurrent episodes may occur, they do not indicate a progressive or systemic disorder. Patients should be reassessed if lesions persist beyond a few days, involve extraoral sites, or are accompanied by systemic symptoms, which would warrant further investigations.

In conclusion, ABH remains an often-overlooked oral mucosal disorder, with its sudden presentation leading to unnecessary concern and investigations. The cases discussed, involving a young adult and a middle-aged male, highlight the variability in presentation and reinforce the need for clinicians to recognize ABH as a benign, self-limiting condition. Although the condition is more common in individuals over 50, its occurrence in younger patients suggests that age alone should not be considered a diagnostic criterion. The absence of systemic involvement, spontaneous resolution, and normal laboratory parameters are key to distinguishing ABH from more serious conditions. Clinicians should be aware of ABH to prevent misdiagnosis and avoid unnecessary medical interventions. Future studies are needed to further explore its underlying pathophysiology and any potential predisposing factors contributing to its occurrence in younger individuals.

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