

Angina Bullosa Hemorrhagica: A Case Report

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Abstract

Angina bullosa hemorrhagica (ABH) is a rare and benign process which presents acutely with blood- containing bullae within the oral or oropharyngeal mucosa. The disease process is self-limiting as the bullae spontaneously evolve into erosions, though recurrence commonly occurs. In the following case, we describe a patient who presented with a complaint of mouth swelling with associated drainage of blood following consumption of pizza. Examination revealed a bullous lesion on the hard and soft palate with sanguineous drainage and the patient was subsequently diagnosed with ABH. Management involved recommendation of Chlorhexidine rinses, and icing which is consistent with the symptomatic treatment approach for this condition.

Categories: Oral Medicine, Dermatology, Otolaryngology

Keywords: oral erosions, bullous lesion, oral mucosal lesions, angina bullosa hemorrhagica (abh), hemorrhagic bullae.

INTRODUCTION

Angina bullosa hemorrhagica (ABH) is a benign process leading to the acute formation of blisters containing blood within the oral or oropharyngeal mucosa [1-3]. The bullae are subepithelial in nature with a red-blue color [3]. Subsequently, the lesions progress to erosions secondary to spontaneous rupture of blisters. The healing process occurs within a few days up to two weeks and there is no residual scarring, but recurrence is common [1-3]. The pathophysiology and etiology of ABH are not yet fully elucidated. The disease process is thought to stem from multiple factors with trauma, such as from spicy food, commonly being considered a trigger. There may be possible weaker associations between ABH and prolonged inhaled corticosteroid use, connective tissue disorders, hypertension, hyperglycemia, chronic renal failure, and personal or family history of diabetes mellitus [1-3]. The approach to diagnosis is widely variable though it tends to be a clinical diagnosis. There does not appear to be a consensus regarding the necessity of laboratory

studies such as platelet count, coagulation tests, and biopsy. However, the following nine diagnostic criteria have been suggested: (I) hemorrhagic bulla or erosion with a history of oral mucosal bleeding, (II) exclusive oral or oropharyngeal involvement, (III) palate localization, (IV) triggering event or promoting factor such as food consumption, (V) recurrence of lesions, (VI) favorable evolution within a few days without scarring, (VII) painless or presence of burning or tingling sensation, (VIII) normal platelet count and coagulation studies, and (IX) negative direct immunofluorescence. It has been suggested that six of the nine criteria, including criteria I and II, be present for diagnosis [4]. ABH can be managed symptomatically along with providing reassurance. This may involve implementation of topical antiseptics, analgesics, and avoidance of triggers [2,3].

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ABH is a relatively rare condition primarily occurring in individuals older than 30 years. There is no clear distribution conveying that prevalence is higher in males or females. Although it is a rare disorder, it is important to consider it as a differential diagnosis for patients presenting with oral or oropharyngeal bullae containing blood. Additionally, the prognosis for this condition is generally positive and it follows a benign course, but it is important to consider the possibility of complications such as airway obstruction [3].

Increasing health care provider awareness of this condition may translate to mitigating misdiagnosis and the likely present underdiagnosis of ABH [2]. There is a broad list of conditions to consider on the differential along with ABH, including mucocutaneous bullous disorders and hemostasis disorders [1-4].

CASE PRESENTATION

A 64-year-old male with a past medical history significant for diabetes mellitus type II presented to the emergency department (ED) with a chief complaint of mouth swelling that began after eating pizza prior to arrival. He noted that after eating a few bites of pizza, that there was a feeling of fullness and swelling in the back of his throat that worsened until his airway was obstructed. The onset of symptoms was under five minutes in length, with the airway obstruction lasting only seconds before there was a "popping sensation" and approx. 5-10 cc of blood was spit out by the patient. He denied changes in phonation. The patient denied a history of similar symptoms.

On exam, vitals revealed blood pressure of 177/112 mmHg, heart rate of 86 beats per minute, respiratory rate of 18 cycles per minute, and oxygen saturation of 96% on room air. A large hemorrhagic bulla, measuring approximately 2.5 x 2.5 cm, was visualized at the hard and soft palate and was draining minimal blood spontaneously at the inferior aspect near the uvula. The mass was soft and non tender.

Computed tomography (CT) scan of the head was obtained out of concern for possible mass or malignancy. However, findings only showed slightly prominent and heterogeneous uvula, suggesting edema. Upon some further research and case review, there had been reports of similar lesions secondary to food which are often self limiting. The patient was observed for several hours without clinical deterioration. The patient was informed of the findings and Chlorhexidine rinses and icing was recommended. The patient was discharged for follow-up with outpatient otolaryngology.

DISCUSSION

Despite ABH being a very benign condition, with most cases self-resolving once the blister membrane ruptures, recognition of ABH as a differential is important when considering treatment options within the emergency department in order to avoid unnecessary treatments. For example, in this patient the unnecessary exposure to radiation via CT Head could have been avoided if the differential for ABH was able to be considered earlier on, as well as the outpatient follow-up with otolaryngology.



Figure 1. Photo of a large hemorrhagic bulla at the hard and soft palate [8]

Knowledge of the treatment of ABH is also important due to the avoidance of using other treatments indicated for other disorders with similar presentations. For example, some case reports report utilization of steroids within the ED, which is a treatment for other blistering disorders, however, does not aid in the healing process of ABH.

With the presentation of a blister in the emergency department that is rapidly expanding after minimal trauma, differentials can include bullous pemphigoid, bullous lichen planus, mucous membrane pemphigoid, and pemphigus vulgaris. The presentation that distinguishes ABH from bullous pemphigoid is, most notably, that the blisters of ABH are filled with blood, not the clear yellow fluid seen in conditions such as bullous pemphigoid. Mucous membrane pemphigoid is also lighter in appearance when compared to the surrounding oral mucosa, whereas ABH blisters are darker in appearance. Bullous lichen planus also, notably, has the presence of purple planar patches, and mucous membrane pemphigoid lacks the raised blisters of ABH.

Due to the rapid expansion of the blood-filled blister of ABH in the oral mucosa, airway obstruction can occur as seen in this case. In this case report, the patient was able to

regain his airway immediately after rupture of the blister sac. While an airway obstruction is a serious matter and can cause severe panic in patients even after resolution, reassurance that the condition is not likely to recur is appropriate. In a review of 54 cases, 30% of patients were reported to have recurrence of ABH [5]. It is not specified whether recurrence was in the same location as the initial eruption, however, due to the extremely fragile state leading to self-resolution of any potential airway obstruction, assuring patients about the benign nature of this condition is appropriate.

Continued bleeding is a potential complication of ABH as well, particularly in patients who are on anticoagulation. Use of tranexamic acid-soaked gauze was described successfully. Other more readily available alternatives would also include application of gauze-soaked epinephrine solution to the area to induce vasoconstriction of the superficial blood vessels [6,7]. Topical 0.12% Chlorhexidine solution is recommended in order to minimize risk of infection, with usage before and after meals to decrease exposure of the wound to food particles.

The exact mechanism of angina bullosa hemorrhagica is unknown, however commonalities between case reports include trauma, often after exposure of the oral mucosa to hot, crunchy foods. Other cases demonstrate ABH after dental procedures and intubation. The occurrence of ABH after consumption of food is theorized to be related to the vasodilation of superficial arteries in response to parasympathetic stimulation, which when combined with frail oral mucosa, can lead to blood vessel damage leading to the rapid blood blister formation. Predisposing factors include use of inhaled steroids, hypertension, and diabetes mellitus, which is thought to predispose to oral mucosa atrophy [8].

CONCLUSIONS

Angina bullosa hemorrhagica is a benign condition which presents acutely with blood-containing bullae within the oral or oropharyngeal mucosa. Due to its low incidence and relatively low awareness of the disorder, ABH is often misdiagnosed and under-diagnosed. A meticulous history, complete physical evaluation with possible laboratory studies, and thorough differential diagnosis formation is vital for correct diagnosis of this condition. It is important to further health care provider awareness and knowledge of this condition in order to minimize misdiagnosis and contribution to further under-diagnosis.

ADDITIONAL INFORMATION

Disclosures

Human subjects

Consent was obtained or waived by all participants in this study.

Conflicts of interest

In compliance with the ICMJE uniform disclosure form, all authors declare the following:

Payment/services info

All authors have declared that no financial support was received from any organization for the submitted work.

Financial relationships

All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work.

Other relationships

All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

REFERENCES

1. Supekar B, Sawatkar G, Wankhade V: Angina bullosa hemorrhagica. *Indian Dermatol Online J.* 2019, 10:89.
2. Silva-Cunha J, Cavalcante I, Barros C, et al.: Angina bullosa haemorrhagica: A 14-year multi-institutional retrospective study from Brazil and literature review. *Med Oral Patol Oral Cir Bucal.* 2022, e35-e41.
3. Alberdi-Navarro J, García-García A, Cardona-Tortajada F, et al.: Angina bullosa hemorrhagica, an uncommon oral disorder. Report of 4 cases. *J Clin Exp Dent.* 2020, e509-e513.
4. Ordioni U, Hadj Saïd M, Thierry G, et al.: Angina bullosa haemorrhagica: A systematic review and proposal for diagnostic criteria. *Int J Oral Maxillofac Surg.* 2019, 48:28-39.
5. Grinspan D, Abulafia J, Lanfranchi H: Angina bullosa hemorrhagica. *Int J Dermatol.* 2001, 38:525-528.
6. Beguerie J, Gonzalez S: Angina bullosa hemorrhagica: Report of 11 cases. *Dermatol Reports.* 2014, 6:5-7.

7. Rodrigues S, Chagury A, Lorenzetti F: Angina bullosa agica in an emergency service of otorhinolaryngology. *Int Arch Otorhinolaryngol.* 2014, 18:
8. Peters J, van Kempen P, Robijn S: Angina bullosa hemorrhagica: Post-traumatic swelling in the oral cavity— A case report. *J Adv Oral Res.* 2020, 11:97-100.

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