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DOI:
10.4103/ijaai.ijaai_24_18

Angina bullosa hemorrhagica: A rare case report in known asthmatic on inhaled corticosteroids

Sonal Suresh Arsude¹, Bhagyashree Babanrao Supekar², Ankita Sunilrao Jire³

Abstract:

Angina bullosa hemorrhagica (ABH) is characterized by development of acute onset of painless solitary or multiple blood-filled vesicles or bullae in oral mucosa which are not attributed to blood dyscrasias, vesiculobullous disorder, and systemic diseases. These lesions rupture spontaneously to form ragged, often painless, superficial erosions that heal spontaneously within 1 week without scarring. Trauma appears to be the most common identifiable precipitating factor. We report a case of ABH to emphasize the importance of recognition of this relatively rare benign condition and to differentiate it from other dermatological and systemic causes of oral blisters.

Keyword:

Angina bullosa hemorrhagica, blood blisters, buccal mucosa, inhaled corticosteroids, trauma

Introduction

Angina bullosa hemorrhagica (ABH) is a benign phenomenon, usually occurring on soft palate of middle-aged individuals, that is, characterized by the sudden appearance of a blood blister on the oral mucosa in the absence of an identifiable cause or systemic disorder. This condition was first described in 1933 as traumatic oral hemophlyctenosis. The term ABH was first used by Badham in 1967.^[1] The lesions of ABH may be indistinguishable from blood blisters due to thrombocytopenia; however, blood investigations and the absence of areas of ecchymosis, epistaxis, or gingival bleeding are helpful signs to rule it out.^[2]

Case Report

A 32-year-old female patient presented with breathlessness since 3 years along with chest tightness and seasonal variation. She had also complaints a frequent cough, cold,

and running nose. History of wheezing was present. On respiratory examination, the patient had bilateral rhonchi. On pulmonary function test, she had mild obstruction with good bronchodilator reversibility. Chest X-ray was normal. Serum immunoglobulin E levels were raised. Rest blood investigations were normal. On the basis of clinical examination and investigations, a diagnosis of bronchial asthma was made. She was started on inhaled corticosteroids (Budesonide Tran capsules via lupihaler). After starting of 6 months of treatment, the patient experienced recurrent episodes of sudden appearance of oral blood-filled blisters which were associated with mild burning sensation. Lesions ruptured spontaneously and healed without scarring within a week. She was referred to dermatologist in view of oral blisters. She did not give a history of trauma before lesions. She was not a known case of systemic hypertension and diabetes. On intraoral examination, a blood-filled blister was present on the right buccal mucosa, which was painless, raised, round, dark red in color, and measured around 1 cm in

¹Department of Respiratory Medicine, NKP Salve Institute of Medical Sciences and RC, Departments of ²Dermato-Venereo- Leprology and ³Pharmacology, Government Medical College and Hospital, Nagpur, Maharashtra, India

Address for correspondence:

Dr. Bhagyashree Babanrao Supekar, C/O Supekar Niwas, Behind Shinde Hospital, D. P. Road, Adarsh Nagar, Beed - 431 122, Maharashtra, India. E-mail: bhagyashree.supekar.23@gmail.com

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How to cite this article: Arsude SS, Supekar BB, Jire AS. Angina bullosa hemorrhagica: A rare case report in known asthmatic on inhaled corticosteroids. Indian J Allergy Asthma Immunol 2019;33:56-8.

diameter [Figure 1]. Blister ruptured spontaneously and healed without scarring within a week [Figure 2]. Other cutaneous examination was normal. All hematological parameters including platelet count, bleeding time, clotting time, prothrombin time, white blood cell count, and blood sugar random were within normal limits. The patient did not give consent for biopsy. On the basis of the history and clinical presentation, a diagnosis of ABH was made. No treatment was given for oral blister as the condition is self-limiting.

Discussion

ABH is an uncommon and benign subepithelial disorder appearing as blood blisters on the oral and oropharyngeal mucosa with no relation with any dermatological, hemostatic, or systemic condition. Badham in 1967 defined these lesions with this term.^[1] This entity has received multiple names, such as benign hemorrhagic bullous stomatitis, localized oral purpura, and stomatopompholyx hemorrhagica. However, despite all the attempts in changing its name, ABH continues as the most commonly used term in the literature.

Conventionally, ABH is an idiopathic condition. ABH usually affect the soft palate, but these lesions can also occur in the anterior pillar of the fauces, epiglottis, arytenoids, pharyngeal wall, and esophagus.^[3] Both middle-aged and elderly individuals are more affected by ABH which ruptures spontaneously and heals without scarring.

Causes that have been mentioned in the literature are related to the minor trauma of hot and spicy foods,^[1,2,4] dental procedures,^[5] steroid inhalers,^[6,7] systemic diseases such as diabetes mellitus (DM),^[3,4,8-11] hypertension,^[4,9-11] and chronic renal failure^[11,12] and may be a contributing factor in developing ABH.

Trauma is the most common cause in the development of ABH. Mastication increases blood flow rate in soft

palate via parasympathetic reflex vasodilation. Hence, the soft palate is easily injured during mastication of hard or crispy food and is prone to ABH.^[5]

Grinspan *et al.* postulated the contribution of an alteration in glucose metabolism in their patients with ABH because they found DM, a positive family history of DM, or a sign of abnormal glucose metabolism in 44% of their patients.^[8] There are reports of association of ABH with hypertension, and chronic renal failure may be associated with the development of ABH although pathogenesis not yet cleared.^[9-12]

Our case of ABH was a known asthmatic on inhaled corticosteroids. There have been several reports of ABH related to steroid inhalers.^[6,7] High and Main reviewed the occurrence of ABH in 106 inhaler users and pointed out the association between the development of ABH and the use of steroid-based inhaler preparations for periods longer than 5 years.^[6] It is known that up to 80% of any inhaled preparation remains within the oral cavity and that clearance of such is slowest from the palate.^[7] Inhaled corticosteroids may cause skin thinning, purpura and may also lead to formation of ABH in asthmatics using high-dose preparations.^[13]

The diagnosis of ABH essentially is clinical; however biopsy and immunofluorescence studies may be useful to exclude other blistering diseases. The microscopic examination of ABH reveals a subepithelial bulla filled with blood and an underlying mild and nonspecific mononuclear inflammatory cell infiltrate which is limited to the region of the lamina propria. Direct immunofluorescence staining for IgA, IgG, IgM, and fibrin is negative and can demonstrate equivocal staining along the basement membrane zone for complement 3.^[14] Differential diagnosis must include thrombocytopenia pemphigus, bullous pemphigoid, bullous lichen planus, dermatitis herpetiformis, epidermolysis bullosa, and oral amyloidosis.



Figure 1: Single nontender hemorrhagic bullae over right buccal mucosa



Figure 2: Healed ruptured bulla without scarring

The lesions of ABH can be distinguished from blood blisters due to thrombocytopenia with the help of blood investigations and the absence of areas of ecchymosis, epistaxis, or gingival bleeding. Vesiculobullous disorders causing oral blisters can be differentiated from ABH by its clinical presentation, histopathological examination, and immunofluorescence studies. No treatment is required for ABH because the blood blisters spontaneously rupture and heal.

We report this case to create awareness among chest physician as ABH is self limiting condition, asymptomatic which can occur in patients on treatment with inhaled steroids and heals spontaneously without treatment.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Badham NJ. Blood blisters and the oesophageal cast. *J Laryngol Otol* 1967;81:791-803.
2. Rai S, Kaur M, Goel S. Angina bullosa hemorrhagica: Report of two cases. *Indian J Dermatol* 2012;57:503.
3. Giuliani M, Favia GF, Lajolo C, Miani CM. Angina bullosa haemorrhagica: Presentation of eight new cases and a review of the literature. *Oral Dis* 2002;8:54-8.
4. Horie N, Kawano R, Inaba J. Angina bullosa hemorrhagica of the soft palate: A clinical study of 16 cases. *J Oral Sci* 2008;50:33-6.
5. Garlick JA, Calderon S. Oral blood blisters in angina bullosa haemorrhagica secondary to trauma of eating and dental injection. *Br Dent J* 1988;165:286-7.
6. High AS, Main DM. Angina bullosa haemorrhagica: A complication of long-term steroid inhaler use. *Br Dent J* 1988;165:357.
7. Higgins EM, du Vivier AW. Angina bullosa haemorrhagica – A possible relation to steroid inhalers. *Clin Exp Dermatol* 1991;16:244-6.
8. Grinspan D, Abulafia J, Lanfranchi H. Angina bullosa hemorrhagica. *Int J Dermatol* 1999;38:525-8.
9. Rosa A, Geraldo Pappen F, Neutzing Gomes AP. Angina bullosa hemorrhagica: A rare condition? *RSBO* 2012;9:190-2.
10. Beguerie JR, Gonzalez S. Angina bullosa hemorrhagica: Report of 11 cases. *Dermatol Reports* 2014;6:5282.
11. Yamamoto K, Fujimoto M, Inoue M, Maeda M, Yamakawa N, Kirita T, *et al.* Angina bullosa hemorrhagica of the soft palate: Report of 11 cases and literature review. *J Oral Maxillofac Surg* 2006;64:1433-6.
12. Shashikumar B, Reddy RR, Harish M. Oral hemorrhagic blister: An enigma. *Indian J Dermatol* 2013;58:407.
13. Capewell S, Reynolds S, Shuttleworth D, Edwards C, Finlay AY. Purpura and dermal thinning associated with high dose inhaled corticosteroids. *BMJ* 1990;300:1548-51.
14. Stephenson P, Lamey PJ, Scully C, Prime SS. Angina bullosa haemorrhagica: Clinical and laboratory features in 30 patients. *Oral Surg Oral Med Oral Pathol* 1987;63:560-5.

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