*Case Report*
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# Angina Bullosa Hemorrhagica: A Review and Report of Two Cases

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## Abstract

Angina Bullosa Hemorrhagic (ABH) is described as an acute and sometimes painful onset of oral blood-filled vesicles and bullae, that are not attributable to blood dyscrasia, vesiculobullous disorders, systemic diseases or other known causes. After a short time, these hemorrhagica bullae spontaneously burst, resulting in ragged, superficial erosions that are often painless. They heal spontaneously within a week without scarring. ABH is underrecognized, and clinicians should be familiar with it because of its potential association with underlying conditions, such as diabetes mellitus, hypertension, or inhaled steroids. Here we present two cases of ABH in an attempt to create awareness regarding this lesion.

**Keywords:** Angina Bullosa Hemorrhagica, Blood Blisters, Soft Palate, Trauma

## 1. Introduction

General dentists often come across cases of traumatic lesions involving the oral mucosa. These lesions may appear atypically, with strange presentations[1,2]. Angina Bullosa Hemorrhagica (ABH) is one of many trauma-induced lesions of the oral cavity caused by friction due to routine activities, such as eating crunchy hard food[2,3]. This benign condition usually occurs on the soft palate of middle-aged individuals. It typically presents as a blood-filled blister on the oral mucosa that suddenly appears without probable cause or underlying systemic condition. This was termed traumatic oral hemophlyctenosis and was first described in 1933. Badham et al. gave the presently used term Angina Bullosa Hemorrhagica in 1967[2,3]. The differential diagnosis of lesions of ABH is the blood blisters related to thrombocytopenia. The two are differentiated based on blood investigations and the absence of gingival bleeding, epistaxis, or ecchymosis[2-5].

Both dermatologists and dentists should be aware of ABH to help prevent incorrect diagnosis since this condition does not require any treatment. This article helps create awareness about the lesions, especially those occurring on the soft palate.

## 2. Case Reports

### 2.1 Case 1

A 68-year-old male patient presented with the chief complaint of recurrent blusters arising anywhere in the oral cavity, especially the mucosa. The blisters were filled with blood and burst leaving raw painful areas which healed in a few days and recurred in a different location intra-orally. The patient was diabetic, asthmatic and hypertensive. For the medical conditions, he was on drugs and using oral inhalers for asthma regularly. The patient had been experiencing this around for the past 3 years and had been to several dentists, ENT specialists and Physicians.

On examination, a blood-filled blister was present on the lateral border of the tongue, which was painless, raised, round, dark red in colour and measured around 1 cm in diameter (Figure 1A). The patient also reported a similar lesion on the soft palate which had occurred a few weeks ago (Figure 1B).

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### 2.2 Case 2

A 54-year-old female complained of the sudden appearance of blood-filled blisters in different locations in the oral cavity predominantly the palate. The patient reported that these lesions appear as small blood-filled, painless macules, progressively increasing in size and bursting spontaneously. After bursting it has a burning sensation and heals in a few days without scarring. These lesions reoccur in a few days to months at a different location. No similar skin lesion were reported anywhere else on the body (Figure 2). The patient has a medical history of diabetes mellitus type II, Hypertension and is on inhalers for asthma.

### 2.3 Laboratory Investigations

For both patients, blood investigations of the coagulation panel, platelets, and blood counts were within normal limits. There was no history of any skin dyscrasias and Diagnosis of ABH was arrived at based on clinical presentation after ruling out other diseases based on blood investigations work-up.

## 3. Discussion

Angina Bullosa Hemorrhagica (ABH) term was coined in 1967 by Badham. He described ABH as oral blood-filled vesicles or bullae that could not be attributed to a systemic disease, vesiculobullous disorders, blood dyscrasia or other known causes[1]. This condition manifests as a blood-filled blister in the oral cavity. The most commonly involved site is the soft palate in oral cavity. Older aged individuals are usually affected.

The term traumatic oral haemophlyctenosis was earlier used by Balina in 1933 (Argentina) to describe the same lesion. He suggested these to be trauma-induced in origin, particularly in older patients who had age-related vascular changes. 14 cases were recorded in 1969; in 1976, the clinical and histological features were specified and described in detail[2]. Based on these features the condition was renamed Recurrent Oral Hemophlyctenosis (ROH). The term ABH is misleading because most bullae arise in the oral cavity and are not consistent with lesions usually called 'angina' as noted by Kirtschig and Happle. They suggested the term: stomatopompholyx Hemorrhagica. It is believed that this condition was first described by Balina who proposed the use of ROH[3].

ABH is listed as an idiopathic condition. It has a sudden onset and can be caused by even minor mucosal insults. It can also occur after dental work, eating, drinking hot beverages, or yelling. Interestingly mastication greatly raises the rate of blood flow via the parasympathetic reflex causing vasodilatation in the soft palate, and hard or crispy food hurts the palate, which may cause blisters of ABH[5]. One potential aetiological component for asthmatic individuals is the usage of steroid inhalers. Out of 30 cases in the biggest documented series, 47 per cent had no known triggering cause[6]. In a case of post-operative ABH brought on by intubation and extubation, Hosain *et al.,* mentioned a postoperative case of ABH.



**(A)** **(B)**

**Figure 1.** Intra-oral photographs depict. **(A)** blood blood-filled vesicle on the lateral border of the tongue; **(B)** multiple small red petechiae and flat lesions on soft palate.



**Figure 2.** Intra-oral photograph depicts a red (blood-filled) flat lesion on the soft palate measuring 10x10mm.

The patient had a solitary blister at the posterior of the palate, not compromising the airway. The soft palate is where lesions are most common. The colour of the intact bulla is red to purple. Typically, lesions are 2-3 cm in dimension and rupture on their own, producing ulcers with ragged borders that do not cause scarring on healing. Reoccurrence of lesions has been noticed[8].

The majority of ABH diagnoses are made clinically, although histology may be used to rule out additional disease processes. The epithelium is parakeratotic with subepithelial split from the underlying connective tissue and this is one of the histopathological characteristics of ABH. Vesicles containing fibrin and erythrocytes are visible in the superficial region. When present, the inflammatory cell infiltration is mainly made up of lymphocytes. The eosinophils and neutrophils that are found in other vesiculobullous disorders are not present. There is no indication of the presence of IgG, IgM, IgA or C3 antibodies within the basement membrane zone or the epithelium, according to immunofluorescence studies[5,7].

ABH lesions are frequently mistaken for those seen in various other systemic and dermatological conditions. ABH is typically diagnosed despite a usual history of rapid blistering even if there are no systemic, dermatological, or haematological symptoms and the ulcers heal normally. Intraoral blood-filled lesions can be seen in individuals with bleeding disorders (thrombocytopenia and von Willebrand's disease); however, a haemostatic function test can differentiate between the two[6,7]. It can be distinguished from benign mucous membrane pemphigoid by the lack of desquamative gingivitis and mucosal involvement in the nose or conjunctiva. A pruritic rash typically allows for the differentiation of dermatitis herpetiformis from linear IgA illness. Planus bullae in oral bullous lichen are frequently connected to a striated pattern. Erythema multiforme can be distinguished by its target-like skin lesion[8]. Along with the appearance of bullous lesions on the skin and macroglossia, other clinical symptoms of amyloidosis include hemorrhagic bullae, which are typically chronic.

Ordioni *et al.*[9] listed the nine diagnostic criteria for ABH:

i. A clinically visible hemorrhagic blister or erosion in the oral cavity with a history of bleeding.
ii. Localization in the oral or oropharyngeal region only
iii. Localization on the palate
iv. An initiating event or promoting factor like the intake of food
v. Lesions tend to recur
vi. Favorable prognosis without scarring within a few days
vii. Lesions are painless without any burning or tingling sensation
viii. Blood investigations of coagulation parameters and platelet count are in normal ranges
ix. Direct immunofluorescence test results should be negative

In our cases, the two patients satisfied eight of the nine diagnostic criteria listed above (primary criteria A and B included). Since it can repair itself most of the time, active treatment is not necessary. Prophylactic antibiotic coverage and antiseptic rinses, like chlorhexidine digluconate at a dosage of 0.25% to 0.12%, are advised if the ABH has damaged the soft palate and the blister has burst in order to help avoid secondary infections[9]. Surgery should be used to repair intact big palatal or pharyngeal blister that is creating a choking sensation. ABH has a favourable prognosis.

## 4. Conclusion

Due to the rarity of this ailment, many people are unaware of it, which leads to needless testing and incorrect diagnoses. After eliminating other possible causes of blood-filled bullous lesions of the oral cavity, ABH can be accurately identified based on its classic clinical appearance. The medical community has become more

aware of ABH thanks to this case report. To handle this illness, the patient only has to be reassured and counselled.

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