Angina Bullosa Hemorrhagica - A Case Report and Brief Review

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ABSTRACT
Angina bullosa hemorrhagica is a rare recurrent oropharyngeal disorder clinically characterized by subepithelial blood filled bullae in the oral and oropharyngeal mucosa with no association with other systemic disorders which often creates a diagnostic dilemma among clinicians because of its clinical presentation. This article is a case report and a brief review on etiology, clinical features, lab investigation, diagnostic criteria, differential diagnosis and management of Angina bullosa haemorrhagica.

Keywords: Angina bullosa hemorrhagica, blood filled blister, ordioni’s criteria, bullous disorders.

INTRODUCTION
Angina bullosa hemorrhagica (ABH) is a rare oral disorder characterized by blood-filled bullous lesions in the oral cavity and the oropharynx in the absence of an underlying systemic, hematological or mucocutaneous condition (1). This condition was first described in 1933 by Argentina as traumatic oral haemophlyctenosis and Badham in 1967 was first to name this condition as ABH (2). Aetiology is unknown but most commonly associated with masticatory trauma(3), followed by long term usage of inhalational steroids, hot beverages, increased progesterone and some authors have described certain familial predisposition in the development of ABH (1,4–7). Patients often complaints of burning or tingling sensation and discomfort on taking food patient develops anxiety due the chocking sensation caused by bullae (8). Common sites in oral cavity are soft and hard palate(9), tongue, buccal mucosa, labial mucosa, floor of the mouth and rarely gingiva(4,8). Usually appears as a single lesion measuring approximately 3mm to 3.5cm. The bullae ruptures and heals without scarring (5,8,10). Lab investigations such as bleeding time, cloting time, platelet count, activated partial thromboplastin time, prothrombin time are usually with in normal limits. Biopsy is done in some of the studies but usually showed negative direct immunofluorescence(1). The lesion is usually self-limiting and does not required any treatment unless pharynx and larynx are involved (11).

CASE REPORT:
A 60-year-old male patient was referred from department of ENT of chettinad medical college to the Department of Oral Medicine Radiology with a chief complains of blood-filled blister in the roof of the mouth. History of presenting illness revealed that Patient had similar recurrent episode around 3 – 4 times a year for past 10 years which are usually seen after consuming hard food involving Various sites in oral cavity which gradually increase in size and rupture leaving a raw area. On probing the medical history patient revealed that he was diabetic for past
20 years and was under homeopathic medication. On Intraoral examination a single dark reddish purple dumbbell shaped blood-filled bullae with well-defined margins was seen at the junction of hard & soft palate. Measuring 1.5 X 1 cm in size extending anteriorly 3 cm from palatal rugae posteriorly on to the soft palate, mediolaterally 2cm from the mucogingival junction of 28 and 18 (figure 1). On palpation the bulla was soft with smooth surface, fluctuant yielding to pressure with no pulsation. Based on the history and clinical presentation a Provisional diagnosis of angina bullosa hemorrhagica and differential diagnosis of hematoma and mucous membrane pemphigoid was given. Patient was subjected to blood investigations and reviewed the following day on examination increase in size of bullae was noted measuring approximately 3cm in diameter roughly oval in shape (figure 2). Advised blood investigation report remained within normal limits (Table:1). Considering the history, clinical presentation and lab investigation reports and also 7 out of 9 criteria for angina bullosa hemorrhagica proposed by Ordioni et al being positive. Hematoma and mucous membrane pemphigoid were excluded as haematoma usually resolves in 2 weeks and does not rupture. Whereas bullae in mucous membrane pemphigoid appears to be filled with clear fluid rather than blood in ABH. Hence final diagnosed of Angina bullosa hemorrhagica (ABH) was given. On consequent follow up after 2 days bullae ruptured leaving eroded surface (figure 3) Patient was advised to use chlorhexidine 0.12% mouth wash and report after 7 days for review. After 7 days the healing was good and palatal mucosa appeared normal (figure 4).
Figure 3: Erosion following rupture of bullae.

Figure 4: Review after 7 days showing healed mucosa without scaring.

Table 1: Blood report of the patient – values in normal range.

<table>
<thead>
<tr>
<th>Lab investigations</th>
<th>Lab results</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bleeding time</td>
<td>2 mins</td>
</tr>
<tr>
<td>Clotting time</td>
<td>4 mins</td>
</tr>
<tr>
<td>Prothrombin time</td>
<td>12.1 sec</td>
</tr>
<tr>
<td>Activated partial thromboplastin time</td>
<td>32.1 sec</td>
</tr>
<tr>
<td>International normalised ratio (INR)</td>
<td>1.12</td>
</tr>
<tr>
<td>Platelet count</td>
<td>2.59 Lac/C.mm</td>
</tr>
<tr>
<td>Random glucose</td>
<td>391mg/dl</td>
</tr>
<tr>
<td>Total protein</td>
<td>7.6g/dl</td>
</tr>
<tr>
<td>Albumin</td>
<td>4.3 g/dl</td>
</tr>
<tr>
<td>Globulin</td>
<td>3.3 g/dl</td>
</tr>
<tr>
<td>A/G ratio</td>
<td>1.3:1</td>
</tr>
</tbody>
</table>
Bilirubin total | 0.73mg/dl  
---|---  
Bilirubin direct | 0.096 mg/dl  
Aspartate aminotransferase (AST) | 50 U/L  
Alanine aminotransferase (ALT) | 97 U/L  
Alkaline phosphatase | 121 U/L  
Gamma GT | 70 U/L  

**Discussion:**

ABH is a benign disorder of unknown etiology. It is characterized by sudden onset of blood filled blister in oral and oropharyngeal mucosa(6). Mechanism of development of ABH has been suggested that a loss of cohesion between the epithelium and the chorion can cause the rupture of the subepithelial capillaries after trauma and condition the emergence of a blood containing blister(1,12,13). Most frequently affecting 3rd -5th decade of life(8,10) with no sex predilection. High prevalence of ABH among diabetics was described by Greenspan et al in 1999 is worth mentioning as 44% of ABH patients showed alternated serological levels of glucose or familial history of diabetes mellitus(1,12). It is possible that considering that both entities share the same age range, and diabetes has a high incidence among adults, it could be a coincidental presentation not a direct pathological association, moreover several cases of patient with chronic kidney failure also described in literature. In 2020 systemic review conducted by Ordioni et al found that 15% of the patient had positive history of diabetes. various other etiological factors suggested in literature are dental procedure, shouting, coughing, sneezing and menstruation(8). Parents usually give history acute formation of bullae which appears abruptly within seconds. Clinical features presenting as single blood filled bullae measuring 2-3 cm, dark red to purple in colour which ruptures and heals spontaneously without scaring with 30% risk of recurrence (10). Treatment is not required as the lesion is self-limiting. When oropharynx is involved breathing difficulty and choking requires immediate management such as Incision of bullae, intubation and even tracheostomy (10). Prognosis is good(14).

**Conclusion:**

Angina bullosa hemorrhagica is a benign blistering disorder often creating anxiety among patients. Oral lesions do not require any treatment, but oropharyngeal involvement should be treated with care to prevent life-threatening complications such as bullae obstructing respiratory passage. Hence clinician should be well aware of this naive lesion, prompt treatment when necessary and effective patient counselling.

**Reference**


