Angina Bullosa Hemorrhagica with a Possible Relation to Dental Treatment, Diabetes Mellitus, Steroid Inhaler and Local Trauma: Report of 3 Cases

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ABSTRACT
Angina bullosa hemorrhagica is a rare condition characterized by one or more blood filled blisters or bullae predominantly in the soft palate region caused either by local mucosal trauma, dental treatment, underlying systemic conditions or use of steroid inhalers. We report three cases of angina bullosa hemorrhagica with different etiological factors.

Keywords: Angina bullosa hemorrhagica, Dental treatment, Diabetes mellitus, Steroid inhaler, Local trauma.

INTRODUCTION

In 1967, Badham advocated the term angina bullosa hemorrhagica (ABH) for an oral mucosal blood blister that occurs with no evidence of blood dyscrasias or vesiculobullous disorder. Clinically, ABH is a solitary and often painful lesion, primarily affecting soft palate of middle-aged adults. Multiple lesions are exceptional and repeated occurrences have been reported in few cases.1,2

ABH heals spontaneously, but for advanced lesions with suspected airway obstruction active treatment is recommended. Although, the diagnosis is nonproblematic in most cases and it is important to critically distinguish ABH from more serious conditions, including various hemorrhagic disorders and bullous diseases. As to etiopathogenesis of ABH, local mucosal trauma, underlying systemic diseases and the use of long-term steroid inhalers have been suggested.2

In the present case reports, we were able to identify the causative agents to be local mucosal trauma secondary to oral prophylaxis in case 1, underlying systemic illness in the form of diabetes mellitus and bronchial asthma for which the patient was using steroid inhaler in case 2 and local trauma secondary to coarse food in case 3.

CASE REPORTS

Case 1
A 40-year-old male patient reported the OPD with complaint of a blood filled blister in lower front teeth region. Medical history and general physical examination was noncontributory. Careful intraoral soft tissue examination revealed a solitary, black colored, oval blood filled bulla measuring 5 mm × 15 mm in the mandibular labial vestibule extending from 31 to 41 region (Fig. 1). The bulla ruptured after two days leaving shallow ulcer, which healed within a week without leaving a scar. H & E stained sections revealed nonspecific inflammatory features (Fig. 2).

There was no recurrence, when the patient was followed for the next six months.

Fig. 1: Solitary oval blood filled bulla in the mandibular labial vestibule
Case 2

A 54-year-old male patient reported with a complaint of oral ulcer since two days.

The patient gave a history of recurrent bulla every three months, which persisted for 2 minutes and ruptured discharging blood. The bulla ruptured leaving an irregular shallow ulcer and healed within two weeks without scarring. Also the size of bulla increased with every recurrence and there is tingling and tightness in the same region before the appearance of new bulla.

Patient was controlled diabetic and asthmatic and is using ipratropium bromide as a steroid inhaler for past three years.

Intraoral soft tissue examination revealed a large, diffuse shallow ulceration on the soft palate extending anteroposteriorly from 1 cm posterior to hard palate up to the uvula, and mediolaterally in the midline approximately 2 cm with multiple hemorrhagic points, irregular margins and surrounding erythema. Pharyngeal mucosa appeared inflamed (Fig. 3). A diagnosis of angina bullosa hemorrhagica secondary to diabetes mellitus and the use of steroid inhaler was considered.

Incisional biopsy of the lesion was done to rule out mucous membrane pemphigoid, bullous lichen planus and amyloidosis. H & E stained sections revealed nonspecific inflammatory features.

The patient was prescribed with topical antiseptic mouthwash and referred to the general physician, where the steroid inhaler was replaced with systemic medications and the lesion completely healed within a week.

Case 3

A 34-years-old male patient reported to the outpatient department with complaint of two blood filled blisters below the tongue, which appeared one hour before, after the intake of coarse food (fried papad). Patient gave a history of similar blisters eight months back after the ingestion of similar food. Careful intraoral soft tissue examination revealed two solitary, bluish black colored, oval blood filled blisters measuring 5 mm × 5 mm each located on the left side of the frenulum in the floor of the mouth (Fig. 4). The blisters then ruptured leaving small shallow ulcers, which healed without leaving a scar within five days. Patient was advised to avoid the similar coarse food in near future.

Clinical history and histopathology confirmed the diagnosis of traumatic angina bullosa hemorrhagica.

DISCUSSION

Angina (painful), bullosa (a blister), hemorrhagica (blood filled) (ABH) describes a condition where an, often painful, tense, blood-filled blister or blisters develop in the mouth. 3 Angina bullosa hemorrhagica (ABH) is the term used to describe acute, benign, and generally subepithelial oral mucosal blisters filled with blood, that are not attributable to any hemostatic defect4.

This condition was first described in 1933 as traumatic oral hemophlyctenosis.4 Badham first described ABH in 1967 as blood filled blisters in the oral, pharyngeal and oesophageal mucosa.5

The onset is sudden and may follow trauma caused by eating, hot drinks, dental procedures or shouting. The use of steroid inhalers with salbutamol and ipratropium bromide (IB) in asthmatic
patients is a possible aetiological factor. In the largest published series of 30 patients, no precipitating factor was found in 47%. Lesions predominantly occur on the soft palate. Other sites inside the mouth may be involved. There is usually a solitary lesion. Multiple blisters may be developed. The blisters usually rupture spontaneously and the sites heal uneventfully. Blisters usually reach 2 to 3 cm in diameter and burst spontaneously leaving ragged ulcers that heal without scarring. Clinically, the lesions may recur.

Pathological studies have yielded nonspecific findings and the underlying etiopathology remains obscured. Various differential diagnoses pertaining to angina bullosa hemorrhagica (ABH) have been reported in the dental literature. These differential diagnoses include mucous membrane pemphigoid, bullous pemphigoid, bullous lichen planus, epidermolysis bullosa, dermatitis herpetiformis, linear IgA disease, and oral amyloidosis.

ABH is a self-limiting condition occurring exclusively in the oral mucosa characterized clinically by tense hemorrhagic blisters that heal without any sequelae and histologically by a pauci-inflammatory subepithelial hemorrhagic bulla with occasional lymphocytic infiltrate.

No treatment is required for angina bullosa hemorrhagica (ABH) because the blood blisters spontaneously rupture and heal. Any large, intact blood blister should be incised to prevent further enlargement that could cause airway obstruction. The treatment for ABH has been described as symptomatic, using a mouthwash and analgesics. Avoidance of coarse food, medical management of underlying systemic diseases, precaution to prevent soft tissue injury while performing any operative dental procedures have been advocated.

Angina bullosa hemorrhagica seems to be more common than reported in the literature and knowledge of the condition is important for right diagnosis and treatment approach when necessary.

REFERENCES