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E–Case Report

Angina Bullosa Hemorrhagica: Report of Two Cases

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Abstract

Angina bullosa hemorrhagic (ABH) describes the acute and sometimes painful onset of oral blood-filled vesicles and bullae, not attributable to blood dyscrasia, vesiculobullous disorders, systemic diseases or other known causes. The haemorrhagic bullae spontaneously burst after a short time resulting in ragged, often painless, superficial erosions that heal spontaneously within 1 week without scarring. Trauma appears to be the most common identifiable precipitating factor, but the essential tissue defect is yet unidentified. This paper presents two cases of ABH with the aim to create awareness regarding occurrence of this lesion, thus avoiding any misdiagnosis.

Key Words: *Angina bullosa hemorrhagica, blood blisters, soft palate, trauma*

What was known?
Angina bullosa hemorrhagic is an example of traumatic lesion characterized by oral blood-filled vesicles and bullae, not attributable to blood dyscrasia, vesiculobullous disorders, systemic diseases or other known causes. The haemorrhagic bullae spontaneously burst after a short time resulting in ragged, often painless, superfi cial erosions that heal spontaneously within 1 week without scarring.

Introduction

Traumatic lesions of the oral mucosa occur frequently in clinical practice. Most of them represent acute or chronic injuries of soft tissues arising from incorrect hygienic procedures. Only sometimes do they become artefactual problems, burns and posttraumatic mucosal lesions. However, their origin, location and clinical signs may considerably differ. They can appear atypically, and sometimes may present bizarre characteristics.[1,2] Angina bullosa hemorrhagica (ABH) is an example of oral mucosal traumatic lesion caused by various mechanical stimuli, especially by ingestion of hard and crispy food.[2,3] It 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. Badham, first used the currently accepted term ABH in 1967.[2,3] The lesions of ABH may be indistinguishable from blood blisters related to thrombocytopenia; however, blood tests and the absence of areas of ecchymosis, epistaxis, or gingival bleeding are helpful signs to rule it out.[2-5]

The awareness of ABH in the field of dermatology and dentistry is very much necessary to avoid misdiagnosis, since this condition spontaneously rupture and heal without any treatment. This case report creates awareness regarding occurrence of the lesion especially on soft palate.

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Case Reports

Case 1

A 40-year-old man patient presented to the Oral Medicine and Radiology Department with chief complaint of decayed teeth. He was a non smoker and non alcoholic. His dental and family history was unremarkable. On intraoral examination, a blood blister was present on the soft palate, which had almost converted into an ulcer [Figure 1]. The patient was unaware of the same lesion and therefore gave no significant history regarding the same. He was also unable to recall any similar previous episodes in the past. Patient was then subjected for routine blood examinations, which yielded normal results. On the basis of clinical appearance and the history, a provisional diagnosis of ABH was made. Incisional biopsy of the lesion was done to rule out pemphigus, bullous pemphigoid, bullous lichen planus etc. Hematoxylin and Eosin stained sections revealed hemorrhagic areas with mild inflammatory cell infiltrate. No treatment was carried out and the lesion spontaneously healed without scarring within 3 days.

Case 2

Another 45-year-old man patient presented with the chief complaint of blister on the palate, since day one. He developed the same soon after taking meal, the previous night. On examination, a blood filled blister was present on the soft palate, which was painless, raised, round, dark red in color and measured around 1 cm in diameter [Figure 2]. Routine blood examination, which included platelet count, bleeding time, clotting time, prothrombin time, WBC

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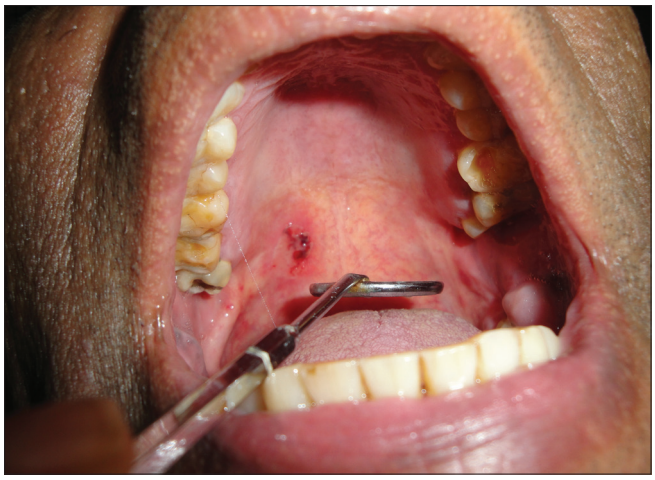

Figure 1: Blood blister on the soft palate in Case 1

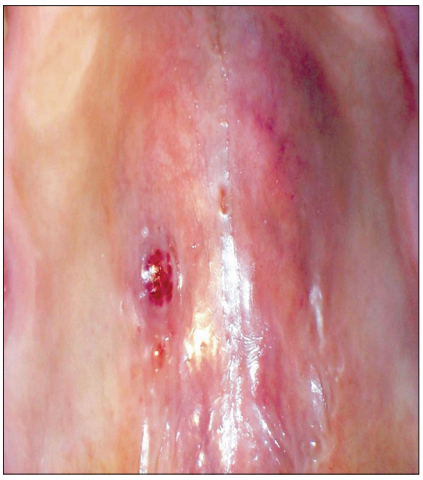

Figure 2: Painless, round blood filled blister on palate in case 2

count and blood sugar random were within normal limits. This lesion was also diagnosed as ABH. H and E stained sections revealed non-specific inflammatory features. The lesion persisted for few hours and then spontaneously ruptured and eventually healed in next 2 days.

Discussion

Traditionally, ABH is an idiopathic condition. Causes that have been mentioned in the literature are related to the minor trauma of hot foods, restorative dentistry, periodontal therapy, dental injections of anesthetics, steroid inhalers and chlorhexidine gluconate mouthrinse.[6-8] Diabetes mellitus may be a contributing factor in developing ABH. Some authors suggest mild trauma as the causative agent in ABH to break the epithelial-connective-tissue junction, causing bleeding of superficial capillaries and resulting in the formation of a subepithelial hemorrhagic bullae.[2,3,8,9]

The blisters last only few minutes and then spontaneously rupture, leaving a shallow ulcer that heals without scarring, discomfort or pain, as seen in both the present cases. The lesions reach an average size of 1-3 cm in diameter. The soft palate is the most commonly affected site. Occasional lesions have been reported in the buccal mucosa and tongue. Similar lesions in other mucous membranes or the skin have not been reported. Considering the fragility of soft palate mucosa, it is easy to speculate that submucosal hemorrhage may be elicited even by subclinical trauma. It is also noteworthy that mastication significantly increases the blood flow rate in the soft palate via parasympathetic reflux vasodilatation. Collectively, the soft palate is easily injured during mastication of hard and crispy food and is therefore prone to ABH.[2,3,9,10]

The diagnosis of ABH essentially is clinical; however, the cases in which a biopsy has being taken, the microscopic examination reveals a subepithelial bulla filled with blood and an underlying mild and nonspecific mononuclear inflammatory cell infiltrate, that generally is limited to the region of the lamina propria. Occasionally, neutrophils may be seen.[2,11,12] Direct immunofluorescence staining for IgA, IgG, IgM and fibrin is negative and can demonstrate equivocal staining along the basement membrane zone for Complement Component 3 Biopsy and immunofluorescence studies may be useful to exclude other blistering diseases.[11,12]

Differential diagnosis must include pemphigus, bullous pemphigoid, bullous lichen planus, dermatitis herpetiformis, epidermolysis bullosa, oral amyloidosis and thrombocytopenia. No treatment is required for ABH because the blood blisters spontaneously rupture and heal. Use of benzydamine hydrochloride provides symptomatic relief.[2,3,10]

What is new?
Most of the general practitioners are unaware of this lesion leading to misdiagnosis and patient is subjected to unnecessary treatment since this condition spontaneously rupture and heal without any treatment. This case report creates awareness regarding occurrence of the lesion especially on soft palate.

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