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Case Report

Angina Bullosa Hemorrhagica: Unique Case Report of Blood Blisters

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Abstract

Angina bullosa hemorrhagica (ABH) is a rare condition with clinical features 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. Asthma patients are often prescribed inhaled glucocorticoids (GCs) in the long term; these GCs negatively affect collagen formation. Since the majority of the patients with ABH are long-term takers of GCs, it is clear that GCs play a role in the onset of ABH. This study examines a case with ABH who used nonsteroidal anti-inflammatory drugs (NSAIDs) and inhaled GC. ABH does not require treatment.

Keywords: *Angina Bullosa Hemorrhagica; Hemorrhagic Bulla; Inhaled Glucocorticoids; Oral Hemorrhagic Blister*

Introduction

'Angina bullosa hemorrhagica' (ABH) refers to a rare condition with clinical features by one or more blood-filled blisters or bullae predominantly in the soft palate region that cannot be attributed to defects in hemostasis or to a systemic disorder. Superficial erosion occurs after a lesion spontaneously bursts. The lesion is gone within a day and the healing does not leave scars. The first mention of this condition came from Argentina in 1933 [1] where it was called traumatic oral haemophlyctenosis. ABH, the term used today, was coined in 1967 by Badham [2].

Case Reports

A 44-year-old male outpatient reported to Subharti Dental College due to his tongue developing a blood-filled blister. The patient had asthma for the past 12 years, for which he was using inhaled GCs, and he was also undergoing the treatment of low platelet count for 2 years. A lesion had previously formed in the same area around four times, each time bursting spontaneously and leaving no trace within seven days. Overall, clinical pictures were suggestive of Angina bullosa hemorrhagica (Figure 1-4). A blood-filled bulla was found on the soft palate upon inspection (Figure 1). Round blood-filled blisters on the left lateral border of tongue (Figure 2), Blood-filled vesicle on the dorsum of the tongue (Figure 3), Blood-filled vesicle on the posterior aspect of left buccal mucosa (Figure 4).

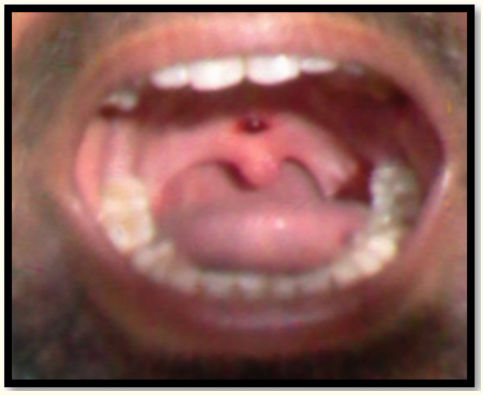

Figure 1: *Hemorrhagic bulla on the soft palate.*

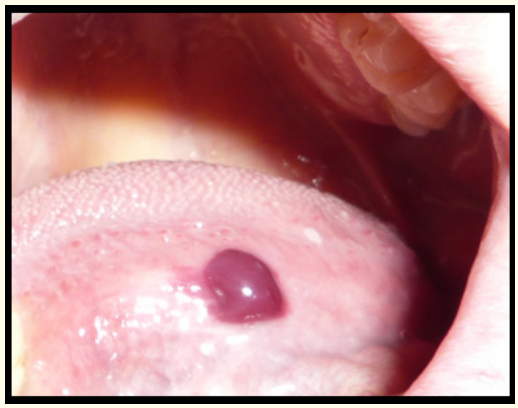

Figure 2: *Round blood filled blisters on the left lateral border of tongue.*

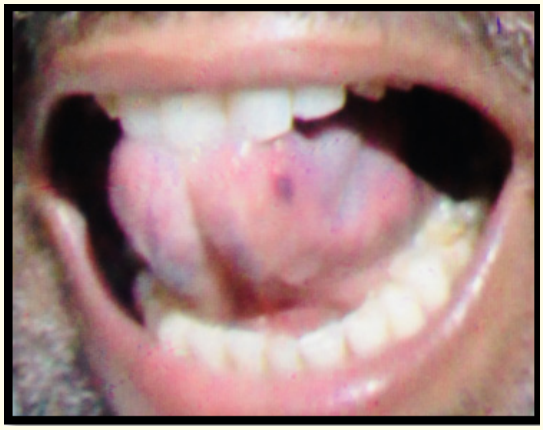

Figure 3: *Blood-filled vesicle on the dorsum of the tongue.*

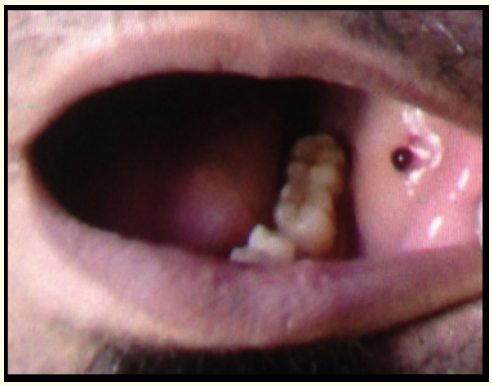

Figure 4: *Blood-filled vesicle on the posterior aspect of left buccal mucosa.*

Differential diagnosis

Differential diagnosis was made to exclude other mucosal or cutaneous diseases such as erythema multiforme, bullous lichen planus, pemphigus, pemphigoid, epidermolysis bullosa, blood dyscrasias, angiomatoid lesions of Rendu-Osler-Weber disease and other systemic and dermatological disorders.

Lab reports indicated normal results for activated partial thromboplastin and prothrombin time; a complete blood count was done in the lab investigations. A lesional biopsy was not completed for the lesion as it was not malignant, and the patient had requested to avoid any procedure that was invasive. The final diagnosis was ABH, based on the results of the lab procedures and the medical history. The patient was comforted that his condition was non-malignant and a follow-up visit was scheduled. Benzydamine-hydrochloride mouthwash was prescribed twice daily. The patient returned after 15 days and the lesion was healed.

Discussion

Kirtschig and Happle [3] considered the term ABH 'angina' to be misleading and coined a more appropriate term: 'Stomatopompholyx haemorrhagica.' However, some large blisters may cause a sensation of choking and Gibson justified the term 'angina.' Other designations used include localized oral purpura [4], benign hemorrhagic bullous stomatitis and recurrent or traumatic oral hemophlyctenosis.

The condition's major pathogenetic mechanism is the epithelial-connective tissue connection's mechanical instability, which may make non-keratinized mucous membranes more vulnerable to damage [5]. ABH is a result of various factors, and recurring use of inhaled GCs caused the condition in this case. Inhaling of steroids with salbutamol and ipratropium bromide in asthmatic patients is an aetiological factor.

Applying GCs on skin in the long-term has been associated with atrophy; the usage can lead to bruises, ruptures, and more transparent skin [6]. Lipocortin protein which reduces phospholipase A2's activities is synthesized when skin atrophies as a result of GCs used on the skin. The suppression of phospholipase A2, and thus a decrease in acid arachidonic acid's release, causes impairment in both mitotic activities and protein synthesis and leads to inhibition of inflammatory processes [7].

Conclusion

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. To identiy typical clinical characteristics and the effects of NSAIDs and inhaled GCs in the development of ABH, more cases need to be investigated.

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Nil.

Conflicts of Interest

The authors report no conflicts of interest related to this research.

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