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Angina Bullosa Hemorrhagica: A Rare Case Report

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ABSTRACT

Angina bullosa hemorrhagica(ABH) is the term used todescribe benign subepithelial oral mucosal blisters filledwith blood that are not attributable to a systemicdisorder or haemostatic defect. The lesions are characterizedby their sudden onset. The pathogenesis isunknown, although it may be a multifactorialphenomenon. The soft palate is most commonly affected, but ABH may also appear on the buccal mucosa, lips and tongue. They appear as apainless, dark red and blood-filled blisterin the mouth that rapidly expand and rupturespontaneously in 24-48 hours. We present the case of a 30 year old male with Angina bullosa haemorrhagica.The clinical features of this case are discussed.

Key words: Blood-filled blister,ABH,Soft palate**.**

INTRODUCTION

Angina bullosa hemorrhagica (ABH) is a relatively uncommon pathology of unclear etiology with acute onset of blisters in the oral and oropharyngeal mucosa that cannot be related to Vesiculo bullous conditions, hemostatic defects, autoimmune pathologies or any other systemic disease. [1] The pathophysiology remains obscure with multifactorial phenomenon. Though ABH traditionally was considered rare previously however studies by Slezak and Yamamoto et al considered it to be relatively common. [2] ABH is generally a clinical diagnosis.[3]

Case report

A 30 year old male patient reported to Tagore Dental College with the chief complaint of blister on the palate for the past two days. The patient was apparently normal before 2 days when he first noticed the blister at night when the patient was consuming hot food during dinner. The patient gave a history of rapid onset of the blister which broke within few minutes after its appearance and there was associated bleeding from the blister. It was not associated with pain, fever, difficulty in speech or swallowing. However mild burning sensation and slight dryness of mouth was felt for the past one day. There was no similar history in the past. There was no medical history related to blood dyscrasias, anti coagulant therapy, liver disorder. There was no relevant personal history. Family history was negative.

General examination was non contributory and there was no associated skin, eye or any mucosal lesions in other regions of the body. On extra oral examination there were no significant findings. On intra oral examinationareas of mild erosion were noticed in the soft palate, along with petechiae at the region of the ruptured blister (Figure 1). The adjacent area revealed a solitary well defined dark red colored blister measuring approximately 1 cm in diameter on the soft palate.

The surface of the blister was smooth and there were no secondary changes. There were no visible pulsations.

On palpation the inspectory findings were confirmed and the blister was non tender.Routine blood examination which included bleeding time, clotting time, platelet count, prothrombin time, WBC count and random blood glucose levels were within normal limits.

Based on history and clinical examination the lesion was clinically diagnosed as ABH.There was no treatment planned since the lesion ruptured and the patient was asymptomatic. However the patient was observed and advised for follow up and review.

DISCUSSION

Angina bullosa hemorrhagica is defined as an acute, benign and usually subepithelial oral mucosal blisters filled with blood which is not attributed to any hemostatic defect or systemic disorder. The history of ABH dates back to 1933, when Balina of Agentina described it as traumatic oral hemophlyctenosis. It was later named as recurrent or al hemophlyctenosis. In 1967, Badham was the pioneer to first use the currently accepted term ABH. However Kirtschig and Happle considered it to be inappropriate since generally the bullae arise in the oral cavity and are not consistent with lesions usually called angina. So they recognized it as stomatopompholyx hemorrhagica.

Though the etiology of ABH is considered idiopathic, yet it is usually hypothesized that minor traumatic insults to the mucosa, long term use of steroid inhalers may be a common predisposing factor. In 47% of cases no precipitating factor was identified. However mostly the common reason being traumatic insult may be associated with mechanical, thermal injuries wherein intake of solid crispy food items, hot drinks, hot foods and occasionally even shouting or sneezing ; sometimes dental procedures or endoscopic trauma may be predisposing factors. [4]

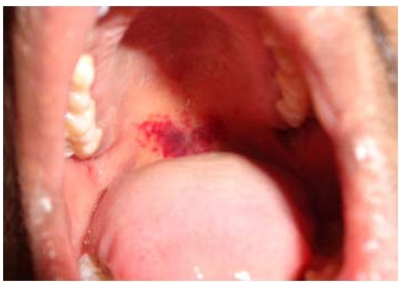

Fig. 1: Shows mild erosion along with petechiae on the soft palate

The fundamental reason underlying these predisposing factors is related to the weakened junction between epithelial and connective tissue especially in non keratinized mucosa resulting in sub epithelial oral blisters filled with blood. Predominantly the soft palate is affected by virtue of its fragility and the fact that masticatory forces increases the flow of blood in soft blood which is related to the parasympathetic reflex vasodilatation.[5] The sites commonly affected apart from soft palate include lips, lateral and ventral aspect of tongue, floor of mouth and buccal mucosa. Occasionally other areas such as anterior pillar of fauces, epiglottis, arytenoids, pharyngeal arch and esophagus were affected[6] and very rarely gingiva and hard palate may be affected.[7]

As regards to the age distribution middle aged and elderly population is commonly affected with a median age group of 54 years and rarely children below 10 years are affected.[8] Though generally there is no gender predilection yet few studies by Slezak has reining revealed that 55.3% of females are usually affected.[9]

The manifestation of ABH usually has a sudden onset which is preceded by burning sensation and xerostomia. In common circumstances the blisters of ABH are usually not associated with pain; they tend to break down spontaneously within few hours from the onset and they liberate red blood contents a in erosive areas which heal within a week to 10 days.[1]

The differential diagnosis for ABH includes benign mucous membrane pemphigoid, linear IgA disease, dermatitis herpetiformis, oral bullous lichen planus, erythema multiforme, amyloidosis, epidermolysis bullosa. The above

diagnostic conditions can be differentiated in that conjunctival mucosal involvement occurs in benign mucous membrane pemphigoid; pruritic rash appears in linear IgA disease, dermatitis herpetiformis; striated pattern is characteristic of oral bullous lichen planus; target lesions are seen in erythema multiforme; persistent hemorrhagic bulla along with macroglossia and petechiae are present in amyloidosis; bullous skin lesions are present in epidermolysis bullosa.[10, 11]

ABH generally is a clinical diagnosis arrived by excluding the above similar diagnostic conditions. The histopathological features include parakeratotic epithelium with a subepithelial separation from the underlying lamina propria. Superficially located vesicles filled with erythrocytes and fibrins are seen. Lymphocytes are the common inflammatory cell infiltrate; the other inflammatory cells such as neutrophils and eosinophils are absent. Immunofluorescence does not demonstrate IgG, IgM, IgA or C3 antibodies within the epithelium or basement membrane zone.[7]

As regards to management since the oral blisters are generally painless and rupture spontaneously within a few hours from their onset liberating red blood contents and resulting in eroded areas which heal within a week to 10 days without scarring, there is generally no intervention for smaller lesions . However occasionally larger lesions present in the palate may cause a feeling of suffocation wherein surgical drainage may be indicated.[12]

To conclude it is of paramount importance that general dentists be aware regarding the occurrence of ABH. It is also essential to avoid misdiagnosis and unnecessary intervention except in exceptionally large lesions which may cause upper airway obstruction.[3, 4]

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