Angina Bullosa Haemorrhagica -A case report

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Abstract:

Introduction: Angina bullosa haemorrhagica (ABH) is characterized by the recurrent appearance of blood vesicles/blisters on the oral mucosa, most commonly affecting the soft palate which usually rupture spontaneously and mostly are asymptomatic.

Case report: We report here a case of a 39year old male who reported to the department with a complain of sudden red color blister eruption of the palate for which he was quiet concerned.

Conclusion: Angina bullosa haemorrhagica (ABH) is a benign, self-limiting, and under-reported disease that should be kept in differential diagnoses of blood-filled lesions in the oral cavity, as it can prevent misdiagnosis and unnecessary treatment

Keywords: Angina, Erosion, Bleeding, Vesicle, Haemmorrhagica

Introduction:

Angina bullosa hemorrhagica (ABH) was first reported and narrated by Badham in 1967, as an alteration that causes recurrent hemorrhagic blisters, which rupture easily located exclusively in the oropharyngeal or oral mucosa in sites particularly exposed to trauma[1] whose color ranges from dark red to purple and may result in mild discomfort.[2] However, this condition can occur in any location of the oral cavity. They are most commonly seen as asymptomatic vesicles and are observed when their content is spilled over the oral cavity.[3] The etiology of ABH remains poorly understood, and lesions are often underdiagnosed.[4] ABH is also associated with several medical conditions, such as hypertension, diabetes mellitus, drug-induced thrombocytopenia, chronic use of steroid inhalers, and masticatory trauma[5]

Case report:

A 39-year-old male patient with no relevant past medical and dental history presented to the department with a chief complaint of sudden blister formation two in number over his hard palate and soft palate lasting for one hour. On taking a detailed case history the patient revealed that before the formation of the blood filled vesicle, the patient was reportedly eating pizza , whose corner hard crust scratched the roof of his soft palate region. Once it scratched the patient immediately gargled with warm saline gargle and soon after that he noticed a painless blisters formation one over his hard and other over is soft palate. The blood filled vesicles were asymptomatic and one of them ruptured with an hour to result in a red bleeding spot on soft palate. The patient immediately after that reported to the department for his check up. The patient had no chronic bleeding disorders and was not on any regular medications, including blood thinners. The patient had no family history of any chronic diseases.

On intraoral examination a small solitary blood filled vesicle was seen over the hard palate measuring about 3x3mm in diameter, slightly raised, purplish red in color and on palapation was soft, smooth and non tender. Another ruptured vesicle was seen on the soft palate associated with a bleeding spot

Facial profile
Intraoral examination

Based on the history and clinical examination a provisional diagnosis of Angina bullosa hemorrhagica was given.

Discussion:

ABH, which is also known as stomatopompholyx hemorrhagica, benign hemorrhagic bullous stomatitis, and oral hemophlyctenosis, was first described in 1967 by Badham as blood-filled blisters in the oral, oropharyngeal, and/or esophageal mucosa.[1] Although a rare condition, it usually goes underdiagnosed due to the clinicians' lack of knowledge and its self-limited nature and spontaneously favorable evolution. [4]

ABH can occur in the structures of the oropharyngeal cavity, including the soft palate, hard palate, cheeks, floor of the mouth, anterior pillar of the tonsillar fossa, epiglottis, arytenoids, and the esophagus, but it most commonly manifests in the soft palate.[6]

ABH tends to occur in adults between the fifth and the seventh decades of life, with female predominance of 1.4:1.[4]

The etiology and pathogenesis of ABH remain unclear and several hypotheses have been proposed .[4] The various etiologic factors include-

Longterm use of inhaled corticosteroids.[7] Chronic use of these drugs may impair collagen formation and cause atrophy of the epithelial tissue.[8] Disorders of the collagen and elastic fibers of the oral mucosa result in less anchorage of blood vessels, which can cause hemorrhagic lesions after trauma,[9] predisposing patients to these conditions. Trauma was the causative agent in the present case reported here.

A possible association of ABH with hypertension, diabetes mellitus, hyperglycemia, and a family history of diabetes have been reported in literature. [10]

A previous history of trauma has been the main finding reported in most cases (> 80%), and it seems to be the most relevant etiological agent.[4] Trauma associated with hard, hot, or spicy food intake was the most frequently reported factor,[4] similar seen in the present case. The trauma leads to a loss of cohesion between the epithelium and connective tissue.[4] A possible fragility of the vasculature and/or elastin and/or collagen in

some patients could favor subepithelial hemorrhages.[4,11,12]

Clinical examination remains the mainstay of the diagnosis of ABH. A comprehensive review of the patient's systemic condition, especially in patients with suspected hemostasis abnormalities (tests such as platelet counts and coagulation tests).[4] Biopsy and direct immunofluorescence (DIF) have also been suggested or underlying systemic disease should be done. Besides, the presence of a triggering event (chewing), isolated or recurrent lesions on the soft palate with favorable evolution within a few days without leaving scars also favors the diagnosis of ABH.[13,14] Rarely, a biopsy is performed on an intact blister because they usually rupture easily within a few hours. In these situations, extravasation of red blood cells will be observed below the lining epithelium.[4] Therefore, clinicopathological correlation is essential to ensure a correct diagnosis. The presence of hemostatic disorders, antithrombotic treatment, or positive DIF may exclude the diagnosis of ABH.4,[13,14]

For confirmation of the diagnosis of ABH a diagnostic criteria was given by Ordioni et al.[4] mentioned below

Main criteria

(I) Clinically noticeable hemorrhagic bulla or erosion with a history of bleeding of the oral mucosa
(II) Exclusively oral or oropharyngeal localization

Additional criteria

(III) Palatal localization
(IV) Triggering event or promoting factor (food intake)
(V) Recurrent lesions
(VI) Favorable evolution without leaving a scar in a few days
(VII) Painless lesion, tingling, or burning sensation
(VIII) Normal platelet count and coagulation profile
(IX) Negative direct immunofluorescence

It has been suggested that a combination of at least six of the nine proposed criteria for an accurate diagnosis, with criteria I and II as required for the diagnosis of ABH.[4]

Reassurance of the patient is necessary in symptomatic patients.[4] Analgesic drugs and local care (chlorhexidine 0.12-0.2%) can be provided. Large intact lesions, especially on the soft palate, should be incised and drained to avoid a possible obstruction of the upper aerodigestive tract.[4,15] Interestingly, some authors have suggested combining ascorbic acid and citroflavonoids as a strategy to prevent recurrences.[16]

Conclusion:

ABH of the oral cavity is considered a rare medical condition. Abrupt blister development in the oral cavity might be frightening at first glance. However, a good approach that involves detailed history-taking in addition to a proper physical examination can lead to the correct diagnosis of ABH. The etiology behind ABH remains unclear. It can share some clinical and histological characteristics with

more serious diseases, making diagnosis difficult. A careful clinical examination is essential to rule out autoimmune or hematological disorders. Also, diagnostic criteria for this condition have recently been proposed based mainly on clinical examination, allowing an accurate diagnosis without the need for all complementary exams, which are sometimes unnecessary, invasive and/or costly

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