Case Report

Recurrent episodes of angina bullosa hemorrhagica of the tongue

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Abstract

Angina bullosa hemorrhagica is a relatively rare lesion of uncertain etiology affecting the oral cavity and the oropharynx characterized by blood-filled blisters or bullae of varying size. The lesions are sudden in onset, usually asymptomatic and bullae often rupture leaving ulcers which heal without scarring. No specific treatment is usually advised since it is a self-limiting lesion. Rarely, larger lesions in the oropharynx may cause air obstruction and require immediate attention. The clinical presentation of this lesion may cause apprehension to the patient and diagnostic challenge to the clinician since they clinically mimic other serious disorders such as blood disorders and vesiculobullous lesions. An awareness among clinicians is needed to prevent misdiagnosis and unnecessary diagnostic procedures of this distinct entity.

Keywords: Angina bullosa hemorrhagica, benign condition, blood blister, diabetes mellitus, hemorrhagic bulla, hypertension, idiopathic, inhaled glucocorticoids, oral lesions, tongue

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INTRODUCTION

Angina bullosa hemorrhagica (ABH) is a rare mucosal lesion of uncertain etiology characterized by short-lived blood-filled blisters. In 1933, this condition was described as traumatic oral hemophlyctenosis by Balina of Argentina. Badham was the first person to give the name ABH in 1967.[1] This condition is also known by other names like localized oral purpura and stomatopompholyx hemorrhagica.[2] Although the pathogenesis is still unclear, ABH seems to be a multifactorial with trauma being the major triggering factor.

CASE REPORT

A 48-year-old male reported with an asymptomatic blood-filled blister over the tongue. On examination, a solitary hemorrhagic bulla of size 1.5–2 cm in diameter was present over the left dorsal surface of the tongue [Figure 1]. No visible pulsation was seen and adjacent mucosa appeared to be normal. On palpation, it was nontender, turgid and fluctuant in consistency. On eliciting the history, the patient revealed that there was the sudden onset of the lesion while having meal and he had similar smaller lesions on the tip of the ventral surface of

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the tongue [Figure 2] and on the right lateral border of the tongue previously [Figure 3] which ruptured spontaneously and healed within a day or two for which he provided the clinical photographs. The patient was a known case of controlled diabetes mellitus and hypertension. Routine blood tests and coagulation tests were done to rule out blood disorders and results were normal. No dermatologic finding was evident on clinical examination ruling out the dermatologic disorders. Based on the characteristic history, clinical presentation and lack of laboratory findings, a diagnosis of ABH was made. No biopsy was performed since it is not mandatory and the patient did not want any invasive procedure. He was instructed to maintain proper oral hygiene and was prescribed antiseptic mouthwash and topical analgesics for pain relief if required and recalled after 2 days. He was advised about the symptoms if similar lesions appeared on the oropharynx. The bulla ruptured spontaneously in the next morning leaving an ulcer which healed uneventfully [Figure 4].

DISCUSSION

The etiology of ABH is not known. Several causative factors have been suggested like trauma due to sharp cusps, hard/course food, trauma during treatment procedures such as dental procedures, endoscopy, local anesthesia, genetic predisposition with increased mucosal vascular fragility, shouting/sneezing, long-term use of corticosteroid inhalers as in asthmatics, systemic diseases such as diabetes-mellitus, hypertension, chronic renal failure, rheumatoid arthritis, gastrointestinal disturbances, hyperuricemia and localized amyloidosis.[3-7] An imbalance in glucose metabolism as in the familial occurrence of diabetes mellitus has been linked with the development of ABH though the mechanism is not clear.[3] The use of corticosteroids for more than 5 years is said to cause changes in the collagen, a decrease in elastic fibers and epithelium atrophy resulting in weakened epithelial-connective tissue junction or a weak anchorage of mucosal vessels.[8,9]

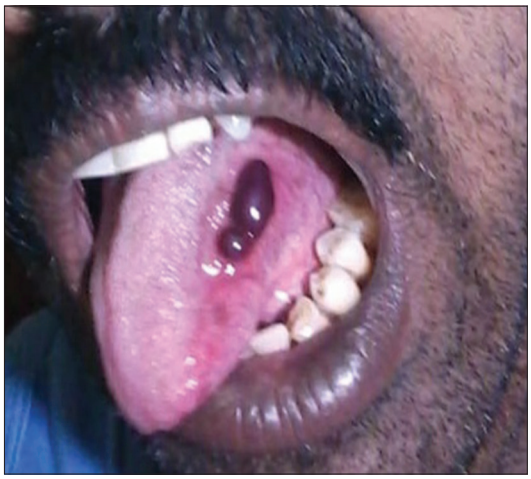

Figure 1: Clinical photograph showing a solitary blood-filled blister over the left dorsal surface of the tongue

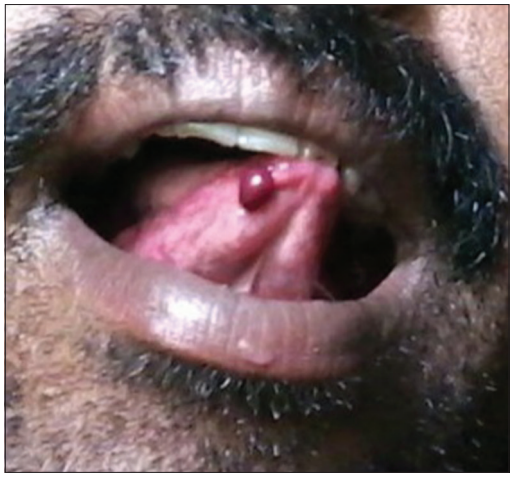

Figure 2: Clinical Photograph showing a blood-filled blister on the tip of the ventral surface of the tongue

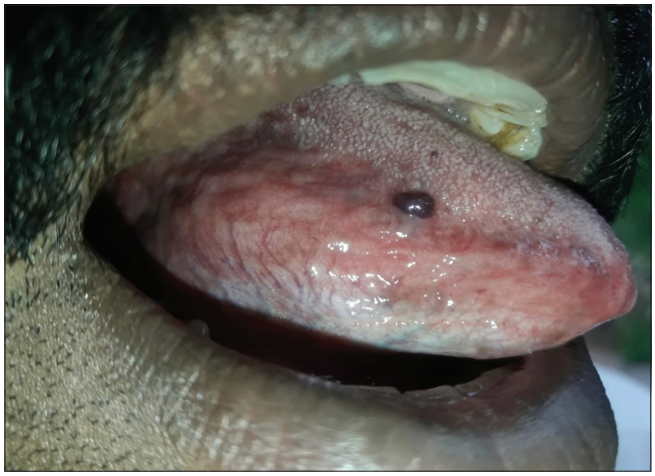

Figure 3: Clinical photograph showing blood-filled blister on the right lateral border of the tongue

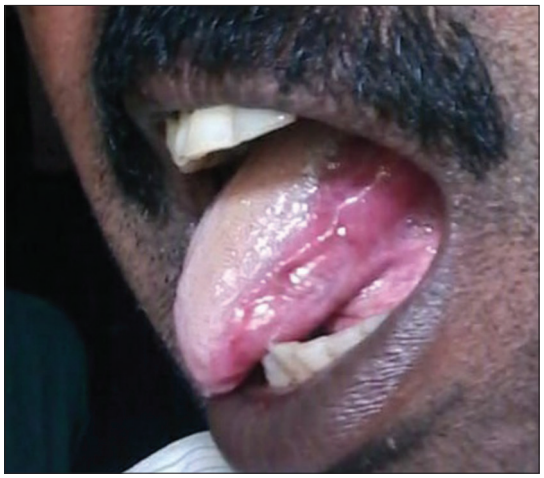

Figure 4: Clinical photograph showing healed lesion on left dorsal surface of the tongue

Persons with increased fragility of the mucosal epithelium may develop a subepithelial hemorrhagic bulla following minor trauma and capillary bleeding.[10] This was true in the present case as described by the patient that he developed bullae immediately after eating hard or rough food. He described that he could feel the blood gushing into the bullae shortly after trauma to form a small blister initially and later within a day or two the blister increased in size with further fill and finally ruptured spontaneously leaving a large ulcer in a day or two. The patient used to take analgesic acetaminophen tablet if necessary at times.

Clinically, ABH is often seen in middle aged and elderly in 60–70 years with no sex predilection.[3,4] Some authors report female preponderance with episodes during premenstruation.[3,6] ABH is not common in children <10 years.[3] There may be a single episode or recurrent episodes as seen in the present case. Majority of cases show solitary lesions, but multiple lesions have also been reported.[5] The bullae are usually small measuring only a few millimeters but sometimes larger bullae may be seen.[7,11] Bullae are sudden in onset usually after minor trauma and red to purple with an ecchymotic halo. They may show a collapsed roof or rupture spontaneously leaving an irregular ulcer which heals in a week with no scarring. ABH lesions do not involve masticatory mucosa.[5,10] The soft palate is the most common site of occurrence followed by the buccal mucosa and the tongue.[5,7] Tongue involvement is less frequent compared to the palate and buccal mucosa. Few authors have reported tongue involvement.[4-6] The gingiva and lips are less commonly affected.[3] It is believed that during mastication there is increased blood flow to the soft palate via parasympathetic reflex vasodilatation which might cause ABH more frequently in the soft palate.[9] Less frequently, the lesions have also been reported in the oropharynx and esophagus.[12] The lesions are usually asymptomatic however, they may be associated with pain and burning sensations of the mucosa due to ulcerations. Few patients complain of slight discomfort, xerostomia, tingling and tightness, difficulty in swallowing, choking (angina means choking), or gagging reflex if the lesions are large and seen in faucillar pillars, arytenoids, oropharynx, posterior pharynx, epiglottis, or esophagus.[12] Airway obstruction has also been reported very rarely with palatal or pharyngeal blisters. The patient should be made aware of the complications and advised to report immediately if symptoms persist as bullae have to be ruptured under supervision.[12,13] The free blood in the larynx requires intubation of the trachea by either fibreoptic endoscopy or direct laryngoscopy, or surgical tracheostomy.[13]

The diagnosis of ABH is mainly by clinical examination and history.[7] Biopsy is not mandatory and is performed only to rule out other clinical mimics when in doubt. The histopathologic examination of the intact bullae may show subepithelial blisters filled with red blood corpuscles with few inflammatory cells in the subjacent connective tissue. A decrease in elastic fibers and capillary hypertrophy in PAS stain was noted by Grinspan *et al*.[3] If the bulla has ruptured, the histopathology reveals an ulcerated epithelium and connective tissue showing chronic inflammatory infiltrate mainly comprising lymphocytes and few neutrophils. Some have suggested direct immunofluorescence staining for IgA, IgG, IgM and fibrin which is often negative.[5] Immunoglobulin G and compliment 3 was demonstrated at the basement membrane by some authors.[3]

The differential diagnosis of ABH includes vesiculobullous disorders, blood disorders and fixed drug eruptions. Vesiculobullous lesions like erythema multiforme, pemphigus vulgaris, mucous membrane and cicatricial pemphigoid, bullous lichen planus, acquired epidermolysis bullosa, linear IgA and dermatitis herpetiformis may be ruled out by the presence of blistering lesions on the skin, histopathologic examination and immunofluorescence studies which are not seen in ABH.[5] The absence of allergic drug history and eosinophils in histopathology rules out drug-induced eruptions.[5,7] Tissue deposition of amyloid in histopathology can be seen in amyloidosis which is absent in ABH.[1,3] Blood disorders like thrombocytopenia, von Willebrand's disease, leukemia, hemophilia and vasculitis, Rendu– Osler–Weber disease can be ruled out based on the presence of other genetic and clinical features symptoms such as petechie, ecchymosis, epistaxis or gingival bleeding and abnormal blood parameters which are not evident in ABH.[4] In Rendu– Osler–Weber disease, the angiomatoid lesions mimic bullae, but they are not episodic. In ABH, the absence of clotting abnormalities helps to establish the diagnosis.[3] It is also important to do a systemic examination since ABH is associated with other systemic diseases most common being diabetes mellitus and hypertension which was present in this case.

No specific treatment is advised for ABH.[7] Some authors have prescribed mouthwash containing 0.25% or 0.12% chlorhexidine digluconate thrice a day for a week to prevent pain and secondary infection.[5] Benzydamine hydrochloride mouthwash or spray has also been used.[12] Ascorbic acid/citroflavonoid (200 mg twice daily) has also been prescribed.[3] Avoidance of trauma, coarse food, or avoiding unnecessary treatment procedures in susceptible patients, grinding of sharp teeth and monitoring the

patients with systemic illness is essential. Oropharyngeal lesions should be monitored carefully since they may require surgical intervention.[13]

CONCLUSION

The diagnosis of ABH is challenging. The clinical presentation may cause apprehension to both the clinician and the patient. Reassuring the patient of its benign nature is essential. Typical history, absence of skin lesions and negative hematological investigations will aid the clinicians in the appropriate diagnosis and ruling out other serious disorders. Awareness and information about ABH among the clinicians and the patients are thus needed.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initial s will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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