CASE REPORT

Angina Bullosa Haemorrhagica: A Case Report on Recurrent Oral Blood-Filled Blister

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Abstract

Angina Bullosa Haemorrhagica is a disorder where blood filled bullae occurs inoral mucosa in absence of hematological conditions, systemic diseases and mucocutaneous conditions. They burst within 2-3 days releasing blood which heals without scarring. They develop rapidly with reported airway obstruction and respiratory distress.A 57-year-old man complained of recurrent blood-filled swelling onbuccal mucosa, not associated with pain. Selective grinding of sharp cusps along with Vitamins, Iron supplements and mouthwash Chlorhexidine was prescribed which resolved the problem. The knowledge of this entity is crucial in ensuringit doesn't get misdiagnosed and lead the patient to undergo numerous investigations.

Keywords: Blister; case report; mouth mucosa; oral hemorrhage.

Introduction

Angina bullosa Haemorrhagica is a painless benign lesion; where a blood-filled bulla occurs in the sub epithelial region of the oral, oropharyngeal and esophageal mucosa. It was first described by Bradham in 1967 AD and usually occurs in the soft palate followed by lateral border of tongue and buccal mucosa.[1] They resolve within 1 or 2 days leaving an erosive lesion that heal without scarring. They have a high rate of recurrence of 30-62%.[1,2] It is seen in the absence of underlying systemic conditions, hematological or mucocutaneous conditions.[3] Our patient had a similar lesion in the right buccal mucosa after chewing on hard food.

Case Report

A 57-year-old male patient came to the Department with the chief complaint of swelling in the lower right back region of cheek which occurred while eating hard food a day back and was painless. He gave a history of frequent similar lesions (4-5 episodes) appearing in the cavity in the last five years; always after eating hard food. The lesions were not painful, did not increase in size and subsided on their own after a few days. He had a similar lesion in the soft palate a few years back that enlarged rapidly which he burst using a geometry compass fearing airway obstruction. These lesions released blood from the mouth in a scanty amount which stopped on its own.
There was no familial history of similar lesions and no history of adverse habits of the patient.

A single blood-filled bullae 1 cm in diameter was present on the buccal mucosa opposite to 47 and 48 at the level of occlusion. (Figure 1)

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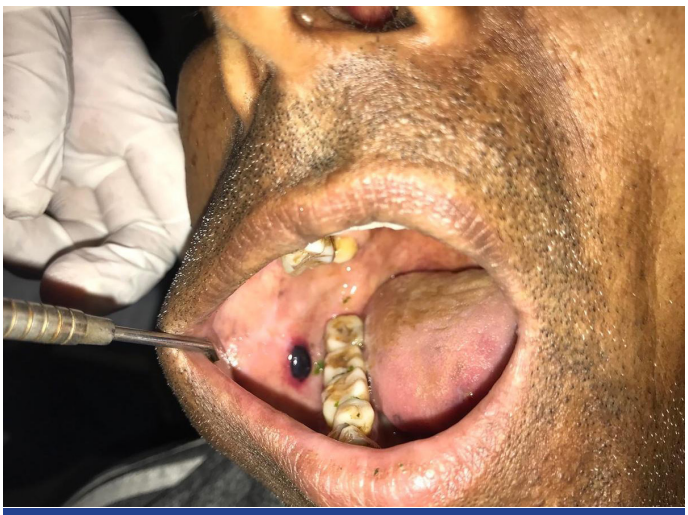

Figure 1. Blood filled Blister in the right buccal mucosa.

It was surrounded by an erythematous halo and had a well-defined border. The bullae appeared bluish red with pale surrounding mucosa. It was soft, non-tender, non-mobile with a smooth surface texture. Nikolsky's sign was negative.

Sharp cusps were present with respect to 47 and 48.

Patient was advised serological tests to rule out viral infections and blood tests to rule out hematological conditions. The serological tests were insignificant. Patient tested negative for Human Immunodeficiency virus, hepatitis, syphilis, tuberculosis. The only positive findings were increased Transaminase level and decreased serum hemoglobin level. Complete blood count and bleeding profile were within normal range.

The patient was prescribed Chlorhexidine 0.2% mouthwash for 2 weeks. Ascorbic acid and iron supplements in response to low hemoglobin levels were prescribed and a soft diet was also advised. Selective grinding of the buccal cusps with respect to 47 and 48 was done reduce trauma and future recurrences.

On one week follow up, the patient presented with healed

lesion.(Figure 2)

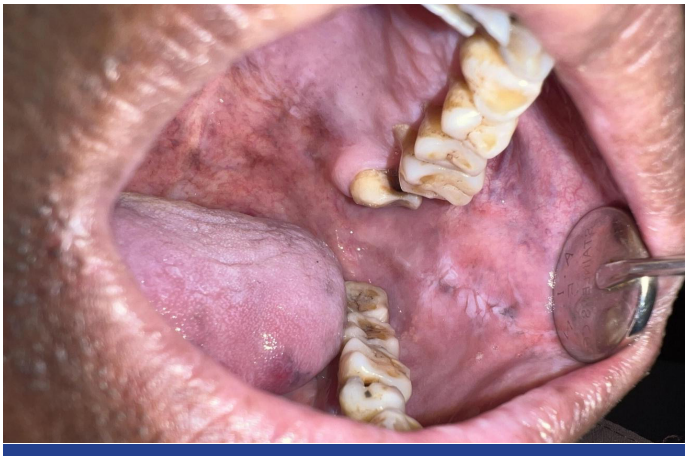

Figure 2. One year follow-up.

Follow ups after one month, six month and one year after treatmentshowed no history of recurrences.

Discussion

Angina bullosa Haemorrhagica is a benign lesion where a blood-filled bulla occurs in the subepithelial region of the oral, oropharyngeal and esophageal mucosa. They completely resolve within 1 to 2 days but they have a high rate of recurrence usually in the same region. Systemic conditions, hematological or mucocutaneous conditions are absent in these patients.[3] They are seen in the 50-70 age population and have 1.1:1 - 1.4:1 predilection for males.[1,2] Ours was 57-year-old male patient. There is not much available on the incidence and prevalence rate in the present literature pool. Though two cases of suffocating lesions leading to dyspnea and respiratory distress have been reported.[1] Our patient also described a similar incident in which he burst a lesion in the soft palate using a geometric compass.

The etiology is not confirmed and is said to be multifactorial. Trauma during mastication is said to be the most common contributing factor and lesions are seen frequently after meals as was in our case. The lesion appeared frequently at the level of the occlusion.[1] Hard food, dental intubation is also said to cause ABH.[3] Grinspan found that 44.4% of 54 cases they analyzed had Diabetes, Hyperglycemia history but no family history.[4] Horie et al also found cases with Hypertension while High and Main suggested use of steroids in the long term led to the lesions. In all cases there was no history of coagulopathies and Nikolsky's sign was negative. Similar findings were noted in the present case.

ABH is a diagnosis based on clinical findings. Biopsy is usually not feasible due to short lived nature of the lesion. Plus, the friable mucosa may be further traumatized during biopsy procedure. Biopsy of the lesion usually shows ulcers which were nonspecific and lamina propria filled with inflammatory cells of chronic nature.[2] The differential diagnosis includes vesiculobullous lesions; Hematological disorders like thrombocytopenia, von Willebrand disease, Leukemia; Mucocutaneous lesions like mucous membrane pemphigus, pemphigus vulgaris, linear IgA disease and epidermolysis bullosa; traumatic hematoma which is symptomatic at the time of injury.

In 2019, Ordioni gave diagnostic criteria for angina bullosa Haemorrhagica.[1] The criterias includes (I) Clinically notable hemorrhagic bullae or erosion with a history of bleeding in the oral mucosa, (II) Exclusively oral or oropharyngeal localization, (III) Palatal localization, (IV) Triggering event or promoting factor (food intake), (V) Recurrent lesions, (VI) Favorable evolution without a scar within a few days, (VII) Painless lesion, or a tingling or burning sensation, (VIII) Normal platelet counts and coagulation test results, (IX) Negative DIF results. ABH can be diagnosed if 6 out of the 9 criteria are fulfilled, among them the 1 and 2 criteria must be present. Out of 9 criteria 6 were present in our case with 1, 2, 4, 5, 6, 7, 8 criteria being present, diagnosing it as an ABH case.

Treatment is usually aimed at symptomatic relief and includes analgesics for pain, topical antiseptics Chlorhexidine 0.12-0.2%, Grinspan suggested ascorbic acid and citroflavinoids to prevent recurrence. Some cases involving the soft palate or the oropharyngeal mucosa require incision and drainage to prevent airway obstruction as well. Antibiotic prophylaxis

and antiseptic rinses are recommended to prevent secondary infections.[5] Sources of trauma should be removed which might be sharp cusps as was also in our case.[1] The patient was advised soft diet and micronutrients were prescribed. In response to low serum hemoglobin levels, iron supplements were also prescribed. The patient was also counselled.

Angina bullosa Haemorrhagica bullosa is a rare subepithelial lesion that resolves on its own. Overall prognosis remains good for ABH. Due to its rarity, it has been frequently misdiagnosed. Few cases with airway obstruction and respiratory distress have been reported which necessitates that emergency responders should know about the disease as well.

Declarations

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

Acknowledgement: Not applicable

Conflict of Interest: The authors declare that they have no competing interests

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