CASE REPORT

Angina Bullosa Hemorrhagic – A Rare Mucosal Condition of Oral Cavity

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ABSTRACT

Introduction: Angina bullosa hemorrhagic is a benign, rare condition of oral cavity, characterized by a sudden onset blood filled blisters in mucosal of oral cavity which rupture and heals spontaneously without scarring, considered to present in 0.05% of cases of oral ulcer. It resolves spontaneously, but in 30% cases it has tendency to reoccur.

Case report: A 48-year-old female presented with sudden onset, self-resolving oral ulcer clinically diagnosed as Angina Bullosa Hemorrhagic following Ordioni et al. criteria for diagnosis.

Conclusion: Angina Bullosa Hemorrhagic is a rare, benign disease of oral cavity that rupture and heals spontaneously without scarring, often triggered by trauma or associated with systemic factors such as hypertension, diabetes or prolonged use of corticosteroid. Diagnosis is mainly clinical. Differentiation form vesicobullous, hematological disorder is essential to avoid misdiagnosis and unnecessary interventions.

INTRODUCTION

Angina bullosa hemorrhagic is a benign and rare condition of oral cavity characterized by a sudden onset blood filled blisters in mucosal of oral cavity which rupture and heals spontaneously without scarring and could not be attributed to any hematological, dermatological and systemic disease.[2] It is considered to be present in 0.05% of cases[1] Middle aged to elderly people are more effected[2] with slightly higher prevalence in female 55.3%. May occur as a solitary or multiple lesions with palate is the most common site of the lesion but can involve faucial pillars, epiglottis, arytenoid, pharyngeal wall and esophagus.[3,9] Although disease is self-limiting, but the risk of recurrence is 30 %.[4] There is no specific cause for the disease, but it is considered to be associated with trauma with hard hot and spicy food, also there is possible association with diabetes, hyperglycemia, hypertension and long-term use of steroid and stress.[5] Diagnosis of the disease is mostly clinical and without any need for specific management. It is mostly underdiagnosed due to asymptomatic and self-limiting behavior, but clinician should be aware of the disease to reduce investigation, avoid misdiagnosis with some autoimmune or hematological disorder and to avoid wrong treatment.

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CASE REPORT

A 48 years female presented with an ulcer inside oral cavity for 4 days which is sudden onset as a blister inside oral cavity which ruptured spontaneously with bleeding. There is no history of any trauma to oral cavity, no history of fever or any other lesions in the body. She is a known hypertensive on medication, not a known diabatic. She used to chew betel nut. Furthermore, she also gave similar history of ulcer which appeared 3 in last year which rupture with bleeding and

heels spontaneously. On examination there is a red to pinkish appearing ulcer of 3×3 cm, with well demarcated margin with whitish slough over the ulcer (Figure 1). It is soft on palpation; no induration and it did not bleed on touch. There is no cervical lymphadenopathy. Her complete blood count, coagulation profile, serology, liver function test all are within normal limit.

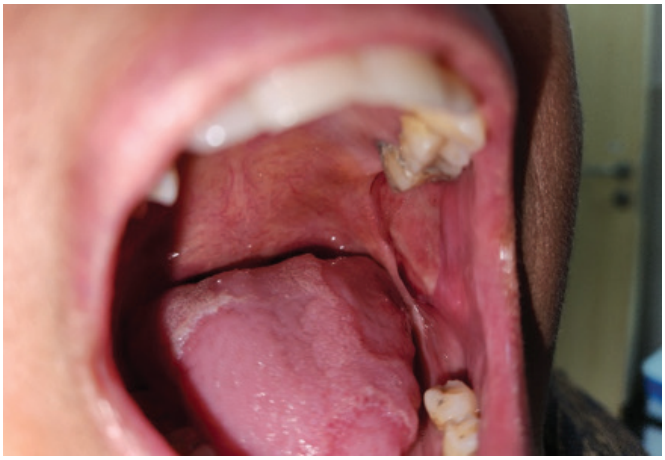

Figure 1: on the day of presentation(day1) after the blister ruptured with bleeding.

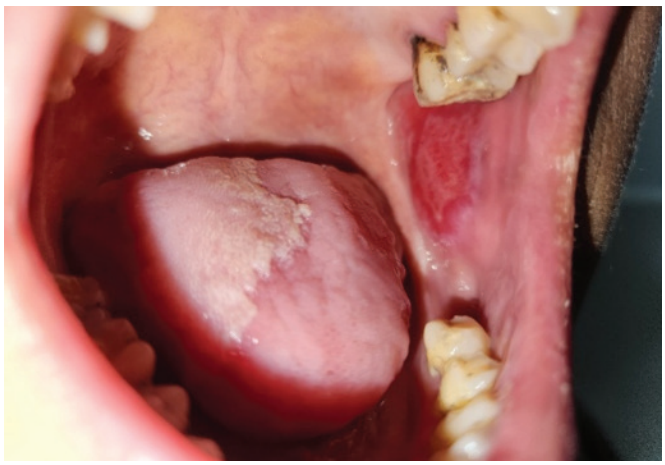

Figure 2: on day 5, ulcer started healing spontaneously

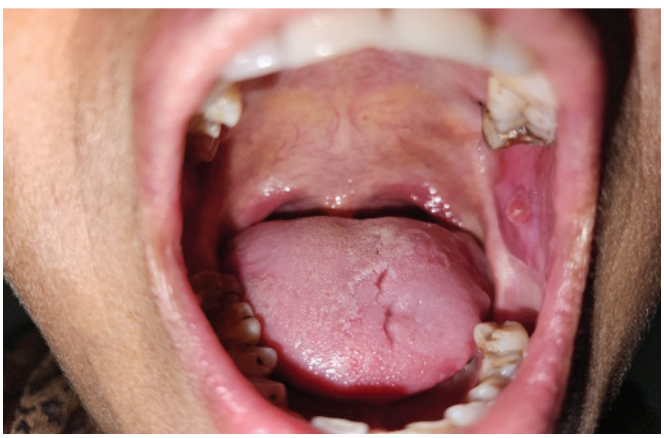

Figure 3: on day 14, ulcer mucosalised fully.

Wedge biopsy from the ulcer shows vascular tissue with fibroblast reactive endothelial cell and mixed inflammatory infiltrate predominantly of lymphocyte, plasma cell and occasional neutrophil suggestive of granulation tissue. Patient was treated symptomatically with benzadamine mouth wash, tab vit c, topical ointment. Patient was followed in day 5 (Figure 2) and day 14 (Figure 3), and it was seen that the ulcer was progressively healed without any scarring.

DISCUSSION

Angina bullosa hemorrhagic term is use for a acute, benign subepithelial mucosal blister filled with blood. It was first described in 1933 as traumatic oral haemophlyctenosis, later in 1967 Badham et al first used the currently accepted term angina bullosa hemorrhagic. The disease is mainly seen in middle-aged or elderly people. The median age at presentation is 54 years, with 60% of the patients in the range of 45–70 years. In a 14-year multi-institutional retrospective study from Brazil where 23 ABH cases from 2006 to 2020 were reviewed, the prevalence of ABH was found to be 0.18%.[11]

ABH patient has a genetic predisposition of loose adhesion between the epithelium and corium of the mucosa or a weak anchorage of the mucosal vessels which results in subepithelial hemorrhage. The traumatic Angina bullosa hemorrhagic commonly occurs in soft palate where the covering squamous epithelium of the nonkeratinized type is thin and friable. The break in the epithelial-connective–tissue junction causes bleeding of the superficial capillaries, resulting in the formation of the subepithelial hemorrhagic bullae. Angina bullosa hemorrhagic which is associated with prolonged use of corticosteroid is considered to be due to collagen synthesis modifications lead to mucosal atrophy and a decrease in submucosal elastic Fibers, especially in the elderly population and inhalers may also induce capillary breakdown. Similarly, vascular fragility is considered to be the pathology in patients with diabetes mellitus.[10]

Diagnosis of angina bullosa hemorrhagic is mainly clinical. Ordioni et al. proposed criteria for diagnosis of the disease.[6]

Main criteria

- Clinically noticeable hemorrhagic bulla or erosion with a history of bleeding of the oral mucosa
- Exclusively oral or oropharyngeal localization

Additional criteria

- Palatal localization
- Triggering event or promoting factor (food intake)
- Recurrent lesions
- Favorable evolution without leaving a scar in a few days
- Painless lesion, tingling, or burning sensation

- Normal platelet count and coagulation profile
- Negative direct immunofluorescence

For a positive diagnosis of ABH using these criteria, the case should meet a minimum of 6 out of 9 defined criteria, with criteria I and II as required. In this case our patient fulfilled 6 criteria among the 9 including the first 2 major criteria.

Histopathological findings from biopsy tissue before its rupture show a subepithelial blister with blood content and an atrophic squamous epithelium seen surrounding the lesion. Infiltration of chronic inflammatory cell is also seen. An acute subepithelial inflammatory infiltrate with a perivascular disposition is also seen in certain case. The biopsy of the ulcer after the rupture of the blister shows nonspecific ulcer with chronic inflammatory infiltrate, mainly lymphocytic.[8]

Angina bullosa hemorrhagic should be differentiated from other vesicobullos disorder of oral cavity including hematological, immunological and cystic pathology. Hematological pathologies, such as thrombocytopenia, Von Willebrand Disease, may present lesions similar to ABH. The mucocutaneous immunological diseases are the most important differential diagnosis of ABH and should include pemphigus vulgaris, mucous membrane pemphigoid, lineal IgA disease, epidermolysis bullosa acquisita and bullous amyloidosis. The differential diagnosis with oral cystic pathologies includes superficial mucocele.[8] So, in a case of oral blistering disease a thorough history and clinical evaluation is of utmost importance. For single benign ulcer histopathological examination is always not needed, only to be performed in cases of multiple, recurrent ulcer with cutaneous involvement.

The disease is self-limiting and as such doesn't warrant any active intervention. Management is mainly conservative which includes Benzydamine hydrochloride or chlorhexidine gluconate mouthwash to reduce the damage to topical steroids and prevent infection. Ascorbic acid can be given to the patient which is considered to prevent the relapse.[7]

CONCLUSION

Angina Bullosa Hemorrhagic is a rare, benign blister forming disease of oral cavity that rupture and heals spontaneously without scarring. It mainly affects middle-aged populations often triggered by trauma or associated with systemic factors such as hypertension, diabetes or prolonged use of corticosteroid. Diagnosis is mainly clinical, histopathological examination is reserved for recurrent or complex cases. Differentiation form vesicobullous, hematological disorder is essential to avoid misdiagnosis and unnecessary interventions. ABH is self-limiting, only requires conservative management in the form of mouth wash and vitamin C supplementation. Awareness among clinicians ensures accurate diagnosis and appropriate care for this underdiagnose condition.

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