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Case Report

Angina bullosa hemorrhagica, an uncommon oral disorder: Case report

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ABSTRACT

Angina bullosa hemorrhagica is one of the rare oral lesion which appears clinically as blood blisters within the oral cavity. This lesion may occur in response to any mild trauma to the oral tissue. It is an acute oral disease mainly seen at the lining mucosa. It is characterized by blood-filled ballooning of the oral mucosa in the form of vesicles and bullae. These lesions are single and rupture easily leaving an ulcerated area.
In this article the authors are describing the case of Angina Bullosa Hemorrhagica attended in Department of Periodontology at Hazaribag College of Dental Sciences & Hospital, Hazaribag.

1. Introduction

Angina bullosa hemorrhagica (ABH) is an uncommon and a benign sub epithelial disorder appears as haematic blister on oral and oropharyngeal mucosa, although its real prevalence is still unknown. This condition was first described in 1933 as traumatic oral hemophlyctenosis later by Badham, in 1967 coined the term Angina bullosa hemorrhagica for this lesion. The lesions of ABH may be indistinguishable from blood blisters related to thrombocytopenia; however, blood tests and the absence of areas of ecchymosis, epistaxis, or gingival bleeding are helpful signs to rule it out. Irrespective of gender this lesion distinctively affecting adults, with the highest incidence over the 5^{th} decade of life.[1]

The present case report describes a case of patient diagnosed with ABH and discusses the characteristics and clinical evolution of lesions, possible complications, and adequate treatment approaches, considering masticatory trauma as main etiologic agent.

1.1. Description of case

A 42-year old female patient reported at the Department of Periodontology and Oral Implantologyy, Hazaribag College of Dental Sciences and Hospital, Hazaribag, Jharkhand, India is described below. With the chief complain of sudden appearence of a blood blister on her lateral aspect of tongue while having dinner. On examination, a blood filled blister was seen on the lateral border of tongue, which was initially painless, raised round and dark red – violet in colour.

Medical and dental history was taken of the patient. Patient is under medication for type II Diabetes Mellitus (DM) since 7 years and did not give history of taking any drug or systemic agent that could affect blood coagulation (acetylsalicylic acid or blood-thinners) or any other underlying condition that could cause further systemic abnormality.

Patient was then subjected for routine blood examination, which yielded normal result.

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Complete information related to the nature of the disease was explained to the patient. Non surgical treatment plan was initiated and patient was kept under observation.

After 3 days the blister spontaneously got ruptured by itself. The patient experienced minor pain, followed by ulceration of the area, for which palliative treatment approach was done and the lesion got healed within 7 days.

Table 1: Data for the patient

S. No.	Description of patient	Data
1	Sex	Female
2	Age (years)	42
3	Smoking habit	No
4	Medical history	Patient is under medication for diabetes since 7 years.
5	Site affected	Side of tongue, Buccal musoca
6	Cause	Dietary habit
7	Drug treatment	None
8	Laboratory tests	Bleeding time : 2.35 Clotting time : 5.32
9	Secondary infection	None
10	Treatment	Non-surgical approach

Fig. 1: Intraoral lesion seen on buccal mucosa, Single, well defined

2. Discussion

ABH is characterized by the sudden appearance of a blister with blood on the oral mucosa, with no identifiable cause or related systemic disorder. While its etiology is still uncertain, it has been described as a multifactorial phenomenon, in which functional or dental trauma seem to be the most important risk factor.[2]

Traditionally, ABH is an idiopathic condition. ABH usually affect the soft palate, but these lesions can also occur in the anterior pillar of the fauces, epiglottis,

Fig. 2: Single, well defined, nontender, bulla at lateral border of tongue

Fig. 3: Lesionspontaneously ruptured and eventually healed

arytenoids, pharyngeal wall, and esophagus.Both middle-aged and elderly individuals are more affected by ABH which ruptures spontaneously and heals without scarring. Grinspan et al.postulated the contribution of an alteration in glucose metabolism in their patients with ABH because they found DM, a positive family history of DM, or a sign of abnormal glucose metabolism in 44% of their patients. There are reports of association of ABH with hypertension, and chronic renal failure although pathogenesis is not yet cleared.[3]

In clinical practice the isolated nature of the swelling, typical clinical presentation, and rapid healing process of the blister are often sufficient to diagnose ABH. During the workup, laboratory testing, including differential blood count, and coagulation assessment tests are indicated to exclude any possible underlying hemostatic defects. A tissue biopsy or analysis of blister fluid is not advised,

because it may cause a secondary infection at the blister site.[4]

Reports have suggested a possible association between ABH and systemic disorders such as diabetes[5] and hypertension,[6] although further studies aimed at confirming such preliminary findings must be performed to confirm it.

Table 2: Association between AnginaBullosa Hemorrhagica and Diabetes Mellitus and Hypertension[7]

Ref.	n	Diabetes	Hypertension
Grinspan et al[8]	54	24(44)%	00 (0%)
Giuliani et al[9]	08	01 (12.5%)	00 (0%)
Yamamoto et al[10]	11	04 (36.4%)	03 (27.3%)
Horie et al[11]	01	01 (6.25%)	06 (37.5%)
Deblauwe and Van der Waal[12]	09	01 (11.1%)	00 (0%)
Serra et al[13]	04	00 (0%)	02 (50%)
Martini et a1[14]	04	00 (0%)	02 (50%)
Rosa et al[15]	47	4 (8.5%)	17 (32.2%)

‘

Even though the diagnosis of ABH is purely clinical, the patient's systemic condition must be considered. Singh et al point out that a history of continuous trauma of the teeth to the mucosa may lead to a presumptive diagnosis of ABH;[2] however, the differential diagnosis excludes cutaneous,

mucous or blood pathologies, such as erythema multiforme, lichen planus, pemphigus, pemphigoid, dermatitis herpetiformis, epidermolysis bullosa, oral amyloidosis and thrombocytopenia and Willebrand disease. As it is observed that blood blister in ABH spontaneously gets ruptured and heals by itself, and so no treatment is required. However benzydamine hydrochloride can be prescribed to provide symptomatic relief.[16,17]

The first step in ABH management is patient counseling. The overall benign character of the condition and its favorable prognosis should be highlighted.

Clinical criteria for the diagnosis ofABH[18]

(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)

The above mentioned table includes 9 criteria to diagnose ABH out of which 7 criteria were observed in our patient , are as follows:-

1. Clinically notable hemorrhagic bulla or erosion with a history of bleeding of the oral mucosa.
2. Exclusively oral or oropharyngeal localization.
3. Triggering event or food promoting factor (food intake).
4. Recurrent lesions.
5. Favourable evolution without a scar within few days.
6. Painless lesion, tingling or burning sensation.
7. Normal platelet count and coagulation profile.

Table 3: Clinical differential diagnosis of Angina bullosa hemorrhagica with mucocutaneous diseases of an immunological basis

Disease	Type of lesion	Content of the blister	Location	Cutaneous involvement	Involvement of other mucosal membranes
Angina Bullosa hemorrhagica	Subepithelial blister	Hematic	Lining Mucosa (soft palate)	No	Oropharynx and esophageal
Mucous membrane pemphigoid	Subepithelial blisters and vesicles	Serous and serohematic	Masticatory mucosa and Lining mucosa (gingiva)	Yes	Ocular, genital, oropharynx, nasal and esophageal
Pemphigus vulgaris	Intraepithelial blisters and vesicles	Serous	Masticatory mucosa and Lining mucosa (areas of friction)	Yes	Nasal, ocular, esophageal, genital, pharyngeal
Linear IgA disease	Subepithelial blisters and vesicles	Serous and serohematic	Masticatory mucosa and Lining mucosa	Yes	Ocular, nasal, genital
Bullous amyloidosis	Subepithelial blister	Serous, serohematic or hematic	Masticatory mucosa and Lining mucosa	Yes	Ocular, anal, vaginal, esophageal (depending on the subtype)

Table 4: Clinical criteria for the diagnosis of ABH[18] (For a positive diagnosis of ABH using thesecriteria, the case should meet a minimum of 6 out of 9 defined criteria, withcriteria I and II as required)

I	Clinically notable hemorrhagic bulla or erosion with a history of bleeding of the oral mucosa
II	Exclusively oral or oropharyngeal localization
III	Palate localization
IV	Triggering event or food promoting factor (food intake)
V	Recurrent lesions
VI	Favourable evolution without a scar within few days
VII	Painless lesion, tingling or burning sensation
VIII	Normal platelet count and coagulation profile
IX	Negative direct immunofluorescence

3. Conclusion

The knowledge related to ABH in dental practice is very much necessary in order to avoid misdiagnosis, since this condition itself gets rupture and heal within 10 days of duration. It is important for the dentist to acknowledge this condition as to differentiate it from other oral vesicular processes with a poorer prognosis such as Pemphigus Vulgaris, Mucous Membrane Pemphigoid or certain hematological diseases.

4. Conflict of Interest

The authors declare that there are no conflicts of interest in this paper.

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None.

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