

Angina bullous haemorrhagica: Case report and review of literature

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Abstract

Angina Bullosa Haemorrhagica (ABH) is a benign lesion of the oral cavity categorized by sudden onset of single or multiple blood filled lesions that burst leaving an ulcer that usually heals in 7-10 days without leaving a scar. We report a case of ABH diagnosed on the basis of history, clinical examination and blood, renal and liver function tests. The case findings and its management is discussed. Literature of the cases presented in the last 5 years was searched from Google Scholar and Pubmed. The review was summarized in a tabulated form.

Keywords: Angina Bullosa Haemorrhagica, Oral lesions, benign condition, idiopathic, case report, Pakistan.

Introduction

Angina Bullosa Haemorrhagica (ABH) is a term first introduced by Badham in 1967 to describe a benign condition of acute onset identified by presence of mostly solitary and occasionally multiple blood filled blisters in the oral cavity or oropharynx.¹ It is also known as "Recurrent oral haemophlyctenosis" or "Haemorrhagic bullous stomatitis".²

ABH has an idiopathic etiology. The lesion is not attributed to blood dyscrasias, vesicullo- bullous conditions, systemic diseases or any other known cause.³ Some association has been seen with trauma, dental procedure, hereditary predisposition, diabetes, ingestion of hard, hot and crispy food or inhaled long term steroids.^{2,3} The condition has been seen more commonly in middle-aged and elderly patients irrespective of gender.²

The blood-filled vesicles rapidly expand and rupture to release contents of blood, leaving an ulcerative surface of oral mucosa that heals in 7 to 10 days, usually asymptotically but sometimes preceded by mild burning sensation and xerostomia.⁴ The diagnosis is primarily based on clinical assessment. Medical and dental history as well as assessment of haematological and coagulation disorders and absence of ecchymosis,

epistaxis or gingival bleeding are helpful in distinguishing it from dental trauma and blood dyscrasias which include anaemia, thrombocytopenia, leukaemia and haemophilia.^{3,5} As it is a benign condition, management is usually directed at relieving the discomfort by prescribing NSAIDs and mouthwash containing chlorhexidine digluconate. Antibiotics are occasionally prescribed to prevent secondary infections.³ We report a case of ABH with idiopathic cause after obtaining informed consent from the patient along with the summary of such lesions reported in literature in last 5 years (Table-1).

Case Report

A 52 year old female presented to the emergency department of Fatima Memorial Hospital, Lahore in January 2017, complaining of a sudden appearance of bluish-black nodule on the junction of the hard and soft palate that rapidly increased in size causing feeling of suffocation (Figure-1a). The lesion ruptured spontaneously resulting in release of blood contents. After clinical examination of the lesion, the patient was instructed to apply pressure with gauze drenched in Tranexamic acid (500mg) at the bleeding site. Bleeding ceased in 15 minutes. Oral examination showed a 3cm wide lesion of clotted blood on the junction of the hard and soft palate (Figure-1b). There was no previous history of any such lesion in oral cavity or any other part of the patient's body.

Medical history relating to hypertension, diabetes mellitus, blood dyscrasias, use of asthma inhaler, liver or kidney disease, allergies or epistaxis was absent. The patient had a history of hypercholesterolaemia and was on anti hypercholesterolaemics for the last 2 years. Complete blood count, Coagulation profile (Prothrombin time=13.2sec, Activated partial thromboplastin time=28sec, International normalized ratio=1.01) and Liver function tests (Alkaline phosphate= 230U/L, S.G.O.T= 31U/L, S.G.P.T= 29U/L and Serum total Bilirubin= 0.4mg/dL) were found to be normal and the patient was generally in good health.

Mucous membrane pemphigoid, epidermolysis bullosa, linear IgA, dermatitis herpetiformis were ruled out on the basis of solitary occurrence, healing

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Table-1: Review of ABH cases in the last five years (2012-2017).

Authors	No. of cases	Site	Suspected associating factor/systemic condition	Treatment advised
Park ¹⁰	1	Right lateral border of tongue	Hyperglycaemia	Topical steroid mouthwash
Abhinav ⁹	2	Lip and oral cavity	Drug induced thrombocytopenia (lesions reappeared within a few hours of Rifampicin and within a day with pyrazinamide)	Not specified
Begueire ²	11	Right buccal mucosa, a few petechiae on the forearms and legs but no frank bleeding from any site 8 cases: Lesions on palate. 1: gum 1: tongue 1: jugal mucosa	Drug-induced thrombocytopenia (patient had ingested aceclofenac one night prior to manifestation) 3 patients: hypertension 1: colon cancer 1: prostate cancer 1: diabetes and myocardial infarction 1: alcoholic while 4: no associating factor.	Not specified. Patient avoided aceclofenac. All cases were safely monitored with regular follow-ups
Shashikumar ⁴	2	Lateral aspect of tongue.	No associated disease or drug intake	Not specified in both cases
Singh ⁷	1	Junction of buccal mucosa and alveolar ridge Right buccal mucosa	Chronic renal failure since 2 years. High blood pressure, elevated serum urea and creatinine Sharp cusps were present in relation to right upper and lower molars	Grinding of sharp cusps. Ointment mucopain (benzocain 20%) and tantum oral rinse (benzylamine hydrochloride)
Shoor ⁶	1	Right side of posterior palate	No associating factor established	0.2% chlorhexidine gluconate and topical anaesthetic.
Martins ³	6	4 cases: soft palate. 2 cases: tongue	1 patient : smoker	Antibiotic therapy in 4 cases
Rosa ⁸	47	36: palate. 11: not specified	17 cases: recognized lesion following trauma out of which 2 were Diabetic. 17 cases: hypertensive and 16 on hypertensive drugs	Not specified
Rai S ⁵	2	Palate	None	No treatment
Present case	1	Palate	No associating factor	Chlorhexidine mouthwash and acetaminophen

without any scarring, no association with allergens and negative family history. Blood dyscrasias were excluded from the differential diagnosis after observing normal test results. Additionally, absence of ecchymosis, bleeding from gingiva or epistaxis helped in ruling out thrombocytopenia. Therefore, on clinical presentation the lesion was diagnosed as ABH.

Patient was reassured of the benign nature of the lesion. Pain and accompanying burning sensation was managed by prescribing chlorhexidine-based mouthwash and acetaminophen as an analgesic. Patient was evaluated every week for 4 weeks and written informed consent was taken to report her case. The lesion healed by the end of 4 weeks without leaving any scar in the palatal region (Figure-1c).

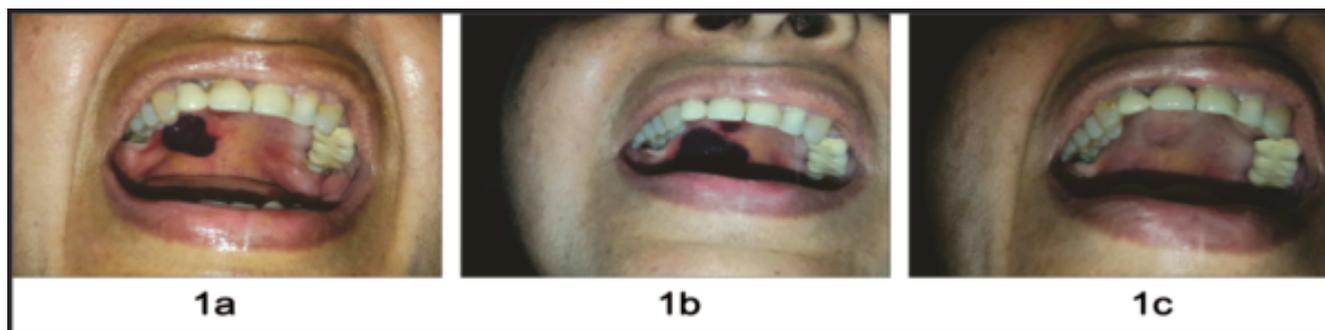


Figure-1: Clinical picture of lesion at different stages. a) Bluish-black nodule on the junction of hard and soft palate. b) Lesion of clotted blood after cessation of bleeding. c) Healed lesion without scarring after 4 weeks.

Discussion

Angina Bullosa Haemorrhagica (ABH) is considered a rare condition. However, overview of English language literature of the case reports in the last 05 years revealed that the condition is not as uncommon as it was considered previously (Table-1). Pubmed and Google Scholar were searched for similar case reports related to ABH from 2012-2017. To our knowledge, seventy-three cases have been reported globally in the last 5 years but this is the first case report of ABH from Pakistan.

Among the reported cases, palate is the most common site of occurrence followed by buccal mucosa and tongue. Less frequently, the lesions have also been reported on oropharynx and oesophagus.^{2,6} The condition usually affects middle aged or elderly patients, irrespective of gender.² In the present case, the lesion occurred at the junction of soft and hard palate of the middle aged female patient.

The ABH lesion has a sudden onset either during or immediately after meals, growing in size up to 1-3cm in diameter, lasting only a few minutes before rupturing spontaneously and releasing contents of blood. The shallow ulcer left behind heals in 7-10 days mostly asymptotically or occasionally accompanied with burning sensation and xerostomia.^{3,5} In the case discussed, there was sudden onset of a single blood filled blister during meal that grew in size and ruptured spontaneously leaving a 3cm wide ulcer which healed in 4 weeks accompanied with discomfort and burning sensation. The lesion took longer to heal than average time taken by cases reported in the literature. The reason for this could be the large size of the lesion.

Generally the condition has been rendered idiopathic but some reports associate it to local trauma from hot or spicy foods, dental procedures, endoscopy, local anaesthesia, and chronic steroid inhalation.⁷⁻⁹ Systemically, it has been associated with hypertension, asthma, diabetes, rheumatoid arthritis, gastrointestinal disturbances and hyperuricaemia.³⁻⁵ From the literature reviewed, it was found that ingestion of hot or spicy food, hypertension and diabetes mellitus are most commonly associated with ABH. No association with hypercholesterolaemia has been reported in the cases reviewed. In the present case, our patient had hypercholesterolaemia for the last 2 years and had a family history of the condition. However, the lesion was not associated with local trauma, dental procedure,

steroid inhalation, hypertension, asthma, diabetes, rheumatoid arthritis, gastrointestinal disturbances and hyperuricaemia.

The specific clinical history allows confirmation of diagnosis without the need for a biopsy. Diagnosis starts with obtaining detailed medical history along with thorough clinical examination to rule out other more serious diseases such as blood dyscrasias, mucous membrane pemphigoid, epidermolysis bullosa, linear IgA and dermatitis herpetiformis. Leukaemia or vasculitis may also have an appearance of a haemorrhagic blister.^{2,5,8} A complete blood count, coagulation tests and liver function tests should be carried out to exclude blood disorders. Histopathological analysis is not required; however, researchers that have carried out histopathological analysis of such lesions have revealed the presence of nonspecific ulceration with normal epithelium over nonulcerated mucosa. Most common features include acanthosis and chronic inflammatory infiltrate consisting primarily of lymphocytes in the lamina propria with a subepithelial split.² Direct immunofluorescence staining for IgA, IgG, IgM and fibrin is negative and can demonstrate indistinct staining along the basement membrane zone for Complement Component 3.⁵

On rare occasions lesions occurring on oropharynx can cause feeling of suffocation or result in asphyxiation. This requires tracheal intubation or surgical tracheostomy.⁶ Management of lesion begins with reassuring the patient of the benign nature of the condition. In the cases reviewed, symptomatic relief was provided by NSAIDs, topical steroids and chlorhexidine or benzydamine hydrochloride-based mouthwashes. In some cases antibiotic therapy has been given in order to prevent secondary infections.^{2,3}

Conclusion

ABH was considered a rare condition, however the number of cases reported in last 5 years suggest that it is an idiopathic disorder that is occurring more frequently. The lesion develops as blister spontaneously but has a good prognosis. Palliative management with topical steroid, chlorhexidine gluconate-based mouthwashes or NSAIDs is recommended with a follow-up examination of at least 4 weeks.

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Conflict of Interest: None to declare.

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