

Angina Bullosa Haemorrhagica on the Ventral Surface of the Tongue- a rare case report

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ABSTRACT

Angina Bullosa Hemorrhagica is a condition affecting the oral mucous membrane characterised by the presence of oral subepithelial blood filled blisters that ruptures and heals spontaneously by itself without any scarring.

Key words : Blood filled Blisters, Trauma,ventral surface of tongue.

Patient named shadasivam of age 68 / M came to our department of oral medicine and radiology, Vinayaka missions sankarachariyar Dental college with a chief complaint of missing teeth .He was referred to the Department of Prosthodontics.After 1 week the Patient reported back to the Department of oral Medicine and Radiology with a complaint of Blood tinged saliva. On eliciting his Personal Habits, he was a non smoker and non alcoholic. His family history was unremarkable.Medical History reveals that he is a Known Diabetic and Hypertensive and under Medication . He gives no history of any drug allergy. Intraoral examination revealed the presence of two intact bullae near the left side of the ventral surface of the tongue.On the right side ventral surface of the tongue, patient presents with ruptured bullae.On eliciting the history of present illness, patient gives history of trauma near the ventral surface of the tongue while doing a border molding procedure for the fabrication of mandibular complete denture. Correlating the history of trauma ,clinical Features and the chief complaint , a provisional diagnosis of Angina Bullosa Hemorrhagica was made.

INTRAORAL EXAMINATION



FOLLOW UP AFTER 7 DAYS



DISCUSSION

Angina bullosa hemorrhagic (ABH) describes the acute and sometimes painful onset of oral blood-filled vesicles and bullae, not attributable to blood dyscrasia, vesiculobullous disorders, systemic diseases or other known causes. The haemorrhagic bullae spontaneously burst after a short time resulting in ragged, often painless, superficial erosions that heal spontaneously within 1 week without scarring. Trauma appears to be the most common identifiable precipitating factor.

This condition was first described in 1933 by Balina of Argentina¹ as traumatic oral haemophlyctenosis. He also postulated a trauma-induced origin, especially in patients with senile capillary changes and Badham² in 1967 first used the currently accepted term ABH. This entity was then named recurrent oral hemophlyctenosis (ROH). As Kirtschig and Happle pointed out, the term ABH is misleading because most bullae arise in the oral cavity and are not consistent with lesions usually called 'angina'; they proposed a more appropriate name for the disease: stomatopompholyx haemorrhagica. The authors believe that Balina was the first to describe this condition and suggested the use of the name ROH¹

The awareness of ABH in the field of dermatology and dentistry is very much necessary to avoid misdiagnosis, since this condition spontaneously ruptures and heals without any treatment. This case report creates awareness regarding occurrence of the lesion especially on ventral surface of the tongue.

Routine blood examination, which included platelet count, bleeding time, clotting time, prothrombin time, WBC count and blood sugar random were within normal limits.

The lesion persisted for few hours and then spontaneously ruptured and eventually healed in next 2 days.

The described cases of angina bullosa hemorrhagica (ABH) had spontaneous onset or were related to minor trauma of ingestion of hot drinks, hard, rough, and crispy food, trauma from sharp edges of adjacent attrited teeth, Smokeless Tobacco, coughing, sneezing, shouting, restorative dentistry such as Metal crowns and Prosthesis or periodontal therapy².

Garlick JA reported a case of Angina Bullosa Hemorrhagica (ABH) following secondary to trauma of eating and dental injection³

Hosain and colleagues reported a case of postoperative ABH caused by intubation and extubation, describing a patient with a single blister at the junction of the soft and hard palate that did not compromise the patient's airway.⁴

High AS et al reported a case of angina bullosa haemorrhagica as a complication of long term steroid inhaler use.⁵

Pahl C et al reported a case of angina bullosa haemorrhagica occurring in the oropharynx in the posterior Pharyngeal wall obstructing the airway, requiring tracheal intubation⁶

Diabetes mellitus may be a contributing factor in developing ABH. Some authors suggest mild trauma as the causative agent in ABH to break the epithelial-connective-tissue junction, causing bleeding of superficial capillaries and resulting in the formation of a subepithelial hemorrhagic bullae.

The blister of angina bullosa hemorrhagica (ABH) appears tense, dark red to purple in color, and blood-filled surrounded by an ecchymotic halo. It has an average size of 1-3 cm in diameter.

The soft palate is the most commonly affected site in angina bullosa hemorrhagica. Occasional lesions have been reported in the buccal mucosa, alveolar ridge, tongue, hard palate, and, rarely, the gingiva. If located on the tongue, the Lateral borders of the tongue which are frequent sites of trauma is most commonly affected. The vermilion border of the lips are almost always spared. Angina bullosa hemorrhagica also may involve the pharynx and the esophagus. Approximately one third of the patients exhibit lesions in more than one location.

Grinspan et al reported that 44% of his patients in a series of 24 cases published in 1999 had from type II diabetes, hyperglycemia, or family history of diabetes. No conclusive evidence of a cause-and-effect relationship exists between the presence of angina bullosa hemorrhagica and glucose metabolism.⁷

Stephenson published a large series of 30 patients, not finding a clear precipitating factor in 47% of the cases. There have been many precipitating factors described: trauma by a sharp cusp or edge of an adjacent tooth or metal crown, masticatory trauma, hot drinks, use of steroids, as well as dental or injection of local anesthesia prior to extraction of teeth or vital crown preparation or root canal treatment.⁸

One case of Angina Bullosa Hemorrhagica was reported in a 50 year old chronic renal failure Patient since two years who was on Hemodialysis for 8 months with Hypertension.⁹

Differential diagnosis must include pemphigus, bullous pemphigoid, bullous lichen planus, dermatitis herpetiformis, Erythema Multiforme and thrombocytopenia.

In Pemphigus the Bullae are not Blood filled whereas in ABH the Bullae are Blood filled.

Bullous pemphigoid is differentiated from ABH in that the Bullae of Bullous pemphigoid is tense or thick walled and not easily rupturable whereas in Angina Bullosa Hemorrhagica, the Bullae are easily rupturable. Characteristic Ocular signs such as Entropion (inwardly placed eyelashes), Trichiasis (inwardly placed eye lashes injuring the cornea) and Symblepharon

(fibrous adhesions between the palpebral conjunctiva and the sclera of the eyeball restricting the movements of the eyeball) can be seen in Cicatricial Pemphigoid (Cicatricial – refers to scarring). No such ocular signs occur in Angina Bullosa Hemorrhagica (ABH).

In Erythema Multiforme there is characteristic Presence of Haemorrhagic crusting on the Vermilion Border of Lips whereas Vermilion Border of lip is not involved in ABH. Thrombocytopenia is a blood dyscrasia where there is reduction in the number of platelets that results in bleeding from all the orifices of the body (Epistaxis -Bleeding from Nose, Blood in urine and stools, Bleeding from Gingiva). Superficial bleeding into the skin that appears as a rash of pinpoint-sized reddish-purple spots (petechiae), usually on the lower legs. No treatment is required for ABH because the blood blisters spontaneously rupture and heal. RAS gel (20% Benzocaine Local anesthetic gel) provided symptomatic relief. Patient was recalled after 7 days. Healing appears satisfactory.

CONCLUSION

Most of the general practitioners are unaware of this lesion leading to misdiagnosis and patient is subjected to unnecessary treatment since this condition spontaneously ruptures and heals without any treatment. This case report creates awareness regarding occurrence of the lesion especially on the ventral surface of the tongue. Patient affected with ABH has blood tinged saliva that can be mistaken as carcinoma by them.

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