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ANGINA BULLOSA HEMORRHAGICA - A RARE CASE REPORT

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ABSTRACT

Angina bullosa hemorrhagica is a benign subepithelial bullous condition of oral cavity characterized by an acute onset blood blister in the oral cavity which ruptures spontaneously within 24 hours and heals in 7 days. It is unrelated to any other dermatological or blistering disease. It has a benign course. Etiology is multifactorial. Can be local trauma or inhaled steroids. Here we report a case, 36-year-old female presenting with this rare disorder. Very less cases are reported in literature due to its rarity and benign course which resolves spontaneously. It can be diagnosed clinically and treated conservatively. It is crucial to differentiate it from other serious conditions. Prognosis is good.

INTRODUCTION

Angina bullosa hemorrhagica is a singular benign subepithelial bullous disorder of the oral cavity. It was first described in 1933 by Balina as traumatic oral haemophlyctenosis. After that it was exemplified by many others.[1] In 1967, Badham first framed the term Angina bullosa hemorrhagica to describe blood blisters, mostly in middle aged females, in the oral cavity, predominantly in soft palate, which ruptures spontaneously within 24 hours and heals in 7 days.[2] The diagnosis of angina bullosa hemorrhagica is challenging as it is not commonly seen and rarely reported in literature. Here we intend to highlight the clinical features and pathogenesis of this rare condition.

CASE REPORT

A 36-year-old Female presented to the ENT OPD with chief complaint of acute onset, painless, solitary reddish fluctuant shining mass of 1.5 cm in diameter in the junction between hard and soft palate for 1 day (Fig. 1).

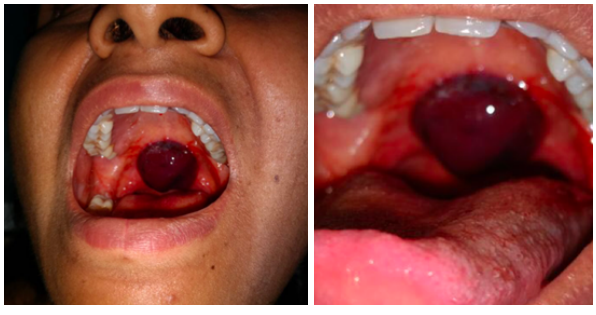

Fig. 1. Blister before rupture.

It was not progressing in size. It developed after taking hot tea the previous evening. There was no history of similar episodes in the past. She was not a diabetic or hypertensive. On digital examination, it ruptured spontaneously (Fig. 2) leaving behind remnants of cyst wall which got peeled off eventually leaving an ulcerated area with superficial erosion of mucosa with an erythematous halo (Fig. 3,4).

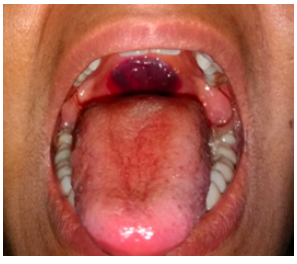

Fig. 2. Blister after rupture with remnants of cyst wall

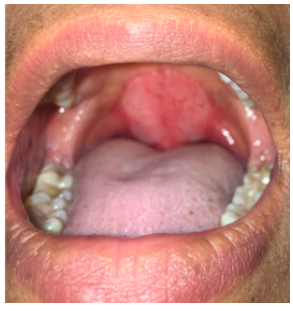

Fig. 3. Blister after peeling off of cyst wall. Shows erythematous halo with superficial erosion of mucosa.

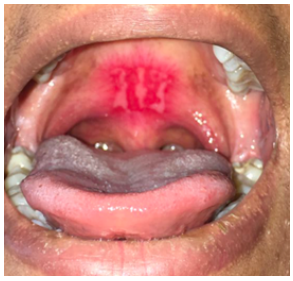

Fig. 4. 7 days after rupture of the lesion.

Routine blood investigations including bleeding time, clotting time, platelets, etc. were found to be normal. Dermatology opinion was obtained. They suspected subepithelial Rhinosporidiosis of the palate. Scrapping from the lesion was taken. It was negative for spherules with endospores.

Diagnosis is by the acute onset, short course, presence of aggravating factor, absence of evidence of other disorders and normal blood investigations. Treatment is symptomatic. Lesions were treated successfully with topical steroids. For local discomfort and pain, anti- inflammatory drugs were given. To avoid possible recurrences, ascorbic acid was administered to the patient. Antibiotics were given to avoid superinfection.

DISCUSSION

Angina Bullosa Hemorrhagica presents with painful hemorrhagic bulla in the oral cavity mostly in the junction between hard and soft palate, bursts spontaneously relieving pain in 24 hours, heals without scar in 1 week. It is seen in the absence of any other disease (1). Etiology is multifactorial. It can be Local trauma or topical glucocorticoids.

Local trauma due to excessive forces associated with mastication or mild thermal injury can cause loss of adherence between

epithelium and chorion and lead to rupture of subepithelial capillaries. Local trauma can also be due to intake of hard or crunchy food, hot food or drink, chicken or fish bone, citrus fruit or dental procedures (2). In our case, trauma to soft palate due to intake of hot food seems to be the triggering factor.

Topical glucocorticoids can cause atrophic changes like bruising, rupture or increased transparency. It is due to synthesis of lipocortin which suppresses phospholipase A2 by direct interaction. Phospholipase A2 is necessary for biosynthesis of mediators of inflammation and it is one of first link in arachidonic cascade. This leads to decreased release of arachidonic acid which is a central regulator of inflammatory response. This inhibits the inflammatory process, mitotic activity and protein synthesis. Coagulation cascade is interrupted as arachidonic acid is a precursor for thromboxane A2 which is a vasoconstrictor and also induces platelet aggregation. It also affects collagen synthesis. This leads to atrophy of the epithelium of mucosa. Collagen content of the mucosa becomes reduced which leads to decrease in tissue elasticity in this region. This results in decreased support of blood vessels present in this region which results in rupture of blood vessel in response to minor trauma. NSAID can play an additive role because of its property to inhibit cyclooxygenase and antiplatelet. (1) In our patient there is no history of using any inhaled steroid.

Angina Bullosa Hemorrhagica usually occurs in the junction of hard and soft palate. It can also occur in lips, side of tongue, floor of mouth, base of tongue or cheek. Large lesions in the base of tongue can sometimes cause respiratory obstruction. These needs to be drained immediately to prevent this. It has a sudden onset, during or after meals with prior burning sensation. The blood blisters are painless and rupture spontaneously and empty their blood content. They heal within 10 days. In our patient, it ruptured on manual examination and the eroded area healed in 10 days.

Histopathological examination will show nonspecific ulcers with inflammatory infiltrate with lymphocytes. Previous history of trauma, dental procedures should be meticulously evaluated to rule out other differential diagnosis. Coagulation disorders should be assessed.

Differential diagnosis can be benign mucous membrane pemphigoid which will present with nasal or conjunctival mucosal involvement, linear IgA disease and dermatitis herpetiformis will present with pruritic rash, erythema multiforme will present with target lesions. (3) (4) (5) It can also occur in leukemia, hematologic disorder and vasculitis. In amyloidosis bullae are persistent and other clinical features are present. In Epidermolysis bullosa, there is bullous skin lesions (6) (7). Genetic syndromes like Kindler syndrome or Ehler danlos syndrome also must be ruled out. (8)

Diagnosis of angina bullosa hemorrhagica is clinical. Microscopic picture shows subepithelial bulla filled with blood with mild mononuclear inflammatory cell infiltrate in lamina propria. It heals spontaneously without scar (9). Topical corticosteroids, oral antibiotics, anti-inflammatory, Chlorhexidine gluconate mouthwash to prevent superinfection and oral ascorbic acid to prevent recurrence can be given. Secondary infection in ulcerated area is not uncommon. So patient should be adequately counselled and followed up. Patients with lesions of soft palate or other parts of oral cavity should be started on antibiotics. Oral Chlorhexidine gluconate should be given as it is important to prevent infection of the raw area after rupture of blister. Large lesions are ruptured or surgically decompressed as there is risk of upper airway obstruction. (10). It can recur in 12 – 24 months. (11) Our patient did not have any previous history of similar episode. Diabetes mellitus, glucose metabolism abnormality and hypertension may be associated factor (12).

CONCLUSION

Angina bullosa hemorrhagica is a rare condition of oral cavity. It should be differentiated from other severe diseases of the oral cavity as this has a benign course. Proper history and examination will help in diagnosis and reassurance of patient about the benign course. The good prognosis should also be explained to the patient.

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