CASE REPORT

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Angina Bullosa Hemorrhagica: A Case Report

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ABSTRACT

Angina bullosa hemorrhagica (ABH) is a self-limited oral blistering disorder that heals spontaneously within a few weeks. ABH has a benign nature and clinical diagnosis is usually straightforward. However, it should be differentiated from other bullous disorders affecting oral mucosa in some cases. In this case report, we presented a 57-year-old male patient diagnosed with ABH and revisited this rare entity with its diagnostic and clinical features.

Keywords: Oral mucosa, Hemorrhagic blister, Sudden onset, Bullous lesion

Introduction

Angina bullosa hemorrhagica (ABH) is a benign disorder characterized by sudden onset hemorrhagic bullous lesions that heal spontaneously within 1-2 weeks [1,2]. In this paper, we report a case of ABH presented to our clinic with recurrent hemorrhagic blisters in oral mucosa. Informed consent was obtained from the patient for this study.

Case Report

A 57-year-old man presented to our clinic with blood-filled blisters on the left side of his tongue. His lesions appeared as hemorrhagic bullae and slight erythema a few days ago without subjective symptoms. He had experienced similar lesions for the last 10 years, occurring 1-2 times per year and healing spontaneously. His lesions were triggered by hot drinks and smoking in previous episodes. In his past medical history; hypertension, ear eczema and left-sided direct inguinal hernia was present. Dermatologic examination revealed multiple hemorrhagic ulcers on the left side of tongue (Figure 1). To exclude other diagnoses; complete blood count, biochemistry, coagulation parameters, anti-nuclear antibody profile, erythrocyte sedimentation rate, complement levels, urinalysis tests were performed. His laboratory values were within normal range.

Based on his typical clinical history and examination findings, we diagnosed the patient as ABH. We prescribed benzydamine hydrochloride mouthwash twice a day for 1 week and advised him

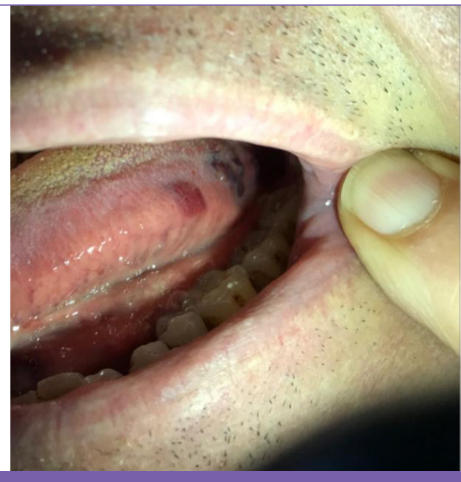

Figure 1. Ruptured hemorrhagic bullae on the left side of tongue

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to avoid hot, spicy, crispy foods and quit smoking. Ten days later, his lesions healed completely (Figure 2).

Discussion

ABH is a self-limited disorder characterized by sudden onset hemorrhagic bullous lesions. Soft palate is the most commonly affected area; followed by buccal mucosa, lateral side of tongue and lip [1,2,3]. Very rarely, gingiva can be affected [4]. Lesions are usually painless; whereas, secondary to rupture of bullae, ulceration and pain may occur [2,5]. Oral cavity floor, esophagus, pharynx, epiglottis involvement can also be seen [5,6,7]. İncision of blisters may be necessary to prevent airway obstruction [2,8]. ABH is seen mainly in middle-aged people. It is reported almost equally in both genders, mean age of diagnosis is 54 [7,8].

Etiology of ABH is obscure. Loosening of cohesion between epithelium and mucosal dermis, and mucosal vascular abnormalities are proposed to play a role in pathogenesis. Hot, spicy foods; dental trauma, intubation, local anesthesia application, endoscopy, air travel are reported as triggering factors [3,5,7,8]. ABH is also more commonly reported in the premenstruation period of women [7].

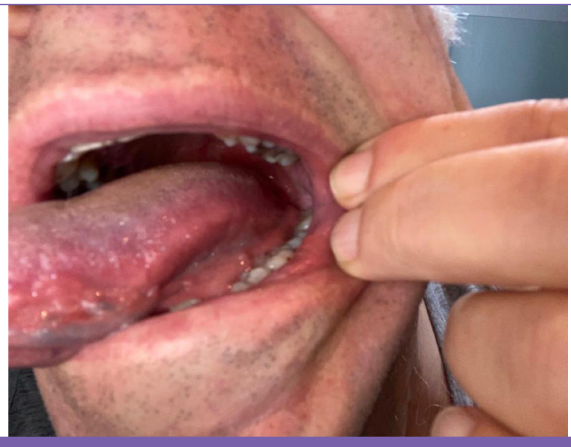

Figure 2. Completely healed lesions

Table 1. Angina bullosa hemorrhagica diagnostic criteria

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

To diagnose angina bullosa hemorrhagica, at least 6 of 9 criteria positivity is required, with the presence of both 1st and 2nd criteria positivity are required

Inhaled steroid use (especially more than 5 years), hypertension, diabetes mellitus are major predisposing factors of ABH [7,8]. Long term use of inhaled corticosteroids can disrupt collagen and elastin formation, cause epithelial atrophy; and this can cause weakening and breaking down of capillaries [2,3].

Diagnosis can be made easily by typical clinical history. In some situations, exclusion of other disorders presenting with bullous lesions is necessary. Sudden onset, blood-filled tense blisters in soft palate are very typical findings of ABH. Most of the time, biopsy is unnecessary [1]. In order to diagnose ABH, nine criteria are proposed by Ordoni et al. [3] (Table 1) [9].

If biopsy is performed from blood-filled bulla, most common anatomic localization of detachment is at the subepithelial area. However, intramucosal and intradermal detachments are also reported in the literature [4]. At the surrounding tissue parakeratosis can be shown. In direct immunofluorescence immunoglobulin (Ig) G, IgA and C3 staining is not observed, which is helpful in differentiating from autoimmune bullous disorders [6].

There is no specific treatment of disorder. Mouthwashes or sprays containing chlorhexidine can be used for symptomatic relief. Ascorbic acid and citroflavonoid containing tablets can also be helpful [3,7].

Conclusion

ABH should be considered in patients presenting with sudden onset bullous lesions that heal spontaneously. Although this may be perceived as a fearful condition at first glance for both patient and clinician, ABH has a benign nature and heals spontaneously. Clinicians should be aware of this benign disorder and differentiate it from other oral blistering disorders.

Ethics

Informed Consent: Informed consent was obtained from the patient for this study.

Peer-review: Internally and externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: O.A., M.E.T., E.A., Concept: O.A., M.E.T., E.A., Design: O.A., M.E.T., E.A., Data Collection or Processing: O.A., M.E.T., İ.A.K., E.A., Analysis or Interpretation: O.A., M.E.T., İ.A.K., E.A., Literature Search: O.A., M.E.T., İ.A.K., E.A., Writing: O.A., M.E.T., E.A.

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References

1. Horie N, Kawano R, Inaba J, Numa T, Kato T, Nasu D, Kaneko T, Kudo I, Shimoyama T. Angina bullosa hemorrhagica of the soft palate: a clinical study of 16 cases. J Oral Sci 2008;50:33-36.
2. Yamamoto K, Fujimoto M, Inoue M, Maeda M, Yamakawa N, Kirita T. Angina bullosa hemorrhagica of the soft palate: report of 11 cases and literature review. J Oral Maxillofac Surg 2006;64:1433-1436.
3. Alberdi-Navarro J, García-García A, Cardona-Tortajada F, Gainza-Cirauqui ML, Aguirre-Urizar JM. Angina bullosa hemorrhagica, an uncommon oral disorder. Report of 4 cases. J Clin Exp Dent 2020;12:e509-e513.
4. Kurban M, Kibbi AG, Ghosn S. Expanding the histologic spectrum of angina bullosa hemorrhagica: report of one case. Am J Dermatopathol 2007;29:477-479.
5. Beguerie JR, Gonzalez S. Angina bullosa hemorrhagica: report of 11 cases. Dermatol Reports 2014;6:5282.
6. Giuliani M, Favia GF, Lajolo C, Miani CM. Angina bullosa haemorrhagica: presentation of eight new cases and a review of the literature. Oral Dis 2002;8:54-58.
7. Grinspan D, Abulafia J, Lanfranchi H. Angina bullosa hemorrhagica. Int J Dermatol. 1999;38:525-528.
8. Shoor H, Mutalik S, Pai KM. Angina bullosa haemorrhagica. BMJ Case Rep 2013;2013:bcr2013200352.
9. Ordioni U, Hadj Saïd M, Thiery G, Campana F, Catherine JH, Lan R. Angina bullosa haemorrhagica: a systematic review and proposal for diagnostic criteria. Int J Oral Maxillofac Surg 2019;48:28-39.