Angina Bullosa Hemorrhagica: A Case Report

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INTRODUCTION

Angina bullosa hemorrhagica (ABH) is a rare oral mucosal disorder characterized by blood blisters which is not related to hematologic or immunopathologic abnormalities. ABH is most common in middle-aged and elderly people and associated with local trauma, diabetes and long-term use of inhaled steroids. Diagnosis is generally based on reviewing history and clinical presentation and it is important to distinguish it from other serious disorders. Usually, ABH is benign condition which requires no treatment. We present a case of 81-year-old female with ABH.

Key Words: Angina bullosa hemorrhagica; Blood blister; Oral mucosa

CASE REPORT

An 81-year-old female presented to the Department of Orofacial Pain and Oral Medicine of Wonkwang University Daejeon Dental Hospital (Daejeon, Korea) complaining of a lesion on tongue which had appeared a month ago after taking a meal. In clinical examination, an ulcerative lesion, which measured around 8 mm in diameter, was observed on the right lateral border of tongue (Fig. 1). She was taking medications for diabetes and hyperlipidemia. A biopsy was taken due to recurrence in the same site and delayed healing. In the result of biopsy, subepithelial cleft and...
infiltration of chronic inflammatory cells were observed (Fig. 2, 3).

After a month, the patient revisited complaining of black discoloration and ulceration in the same site after a pea-sized blister had broken, which had appeared a week before her visit. The ulcerative lesion was in healing state at clinical examination. Lesions occurred twice during a month of follow-up. When she came the Department of Orofacial Pain and Oral Medicine, clinical examination showed two intact blisters of bright red color and 8 mm each in diameter at the same site (Fig. 4). She stated that there was no pain on blisters. However, she reported mild pain as the ulcerative lesion appeared after the breakdown of blister. Aspiration biopsy and blood test including herpes simplex virus antibody test were done. Aspiration biopsy showed hemorrhagic fluid with a few inflammatory cells (Fig. 5). Except for decreased white blood cell count and increased glucose, blood test results were within normal range. Through these clinical and laboratory results, she was diagnosed as ABH.

**DISCUSSION**

Clinically, ABH presents a single or multiple dark red blisters of 1 to 3 cm in diameter. The majority of cases reported previously were single lesion.\(^*\) Usually, multiple blisters present two or three small-sized blisters\(^*\) but there was

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*Fig. 1.* Clinical photograph shows shallow ulcerative lesion on right lateral border of tongue.

*Fig. 2.* Microscopic examination shows ulceration with chronic inflammatory cell infiltration in the submucosa (H&E staining, × 200).

*Fig. 3.* Microscopic examination reveals a subepithelial cleft filled with blood and fibrinous materials. Underlying lymphoplasmacytic inflammatory cell infiltration was observed in lamina propria (H&E staining, × 200).

*Fig. 4.* Two blisters of bright red color and 8 mm each in diameter on right lateral border of tongue.

*Fig. 5.* Aspiration biopsy shows hemorrhagic fluid with a few inflammatory cells (H&E staining, × 200).
a report of numerous blisters in ABH. This report presents two blisters of 8 mm in diameter with bright red color and it is different from the previous reports in terms of color.

Soft palate is the most common site. Stephenson et al. reported that the lesions were found on soft palate in 93% of patients. Involvement on non-keratinized oral mucosa including buccal mucosa, tongue, lips, mouth floor, and pharynx was also reported by several researchers. There has been no reported case with ABH in keratinized mucosa and other mucosal surfaces or skin. In this case, lesions were recurred only on right lateral border of the tongue.

The degree of pain is different according to patients. According to da Rosa et al., pain was evoked in 36.1% of patients. Burning sensation was frequently reported in patients who suffered pain. In cases reported by Rai et al., the researchers found lesions by coincidence while the patient had not been recognizing. Generally, ABH breaks spontaneously within some minutes or hours and heals without scarring within 1 to 2 weeks. Due to these clinical features, ABH is considered to be more common than previously reported.

Grinspan et al. reported recurrence rate of 30% and da Rosa et al. reported recurrence of lesion within 12 months in 27.6% of patients. Persistent recurrence over 15 years was observed in a report by Edwards et al. Recurrence interval was varied from a few weeks to several months and recurred condition was similar but not same as first lesion. Present report showed the similar lesions recurred three times with 1 to 3 weeks intervals during a month.

Until now, the etiology of ABH has been unclear; however, mucosal alteration due to local trauma and systemic condition is a suspected cause. The weakened attachment between oral epithelium and corium may induce separation in non-keratinized mucosa, even in subclinical trauma. This results in bleeding from capillary vessels and the consequent formation of subepithelial blood blisters.

Trauma seems to be a major provoking factor of ABH, and many cases were reported after various intraoral traumas. Especially, many studies reported ABH cases related to the ingestion of crispy, hard and hot food. In this case, the patient mentioned that recurred lesion had appeared after meal.

Long-term use of inhaled steroids is referred to another common cause of ABH as well. It is thought that long-term use of inhaled steroids is associated with alterations in collagen synthesis, atrophy of mucosa and decrease in submucosal elastic fiber. Senile atrophy occurred in middle-aged people is similar as these alterations. This similarity can be a reason of why ABH is common in middle-aged and elderly people and relatively rare in young people.

Grinspan et al. assumed that diabetes, hyperglycemia and family history of diabetes were associated with ABH but further study would be needed. In this case, the patient had history of diabetes. Hyperglycemia was often observed during follow up visits. Especially on the visit with existence of intact blisters, 200 mg/dL of blood glucose level was measured. Based on this, uncontrolled hyperglycemia might be associated with frequent recurrence although additional research is needed.

Generally, ABH is diagnosed by excluding other diseases with the history and clinical symptoms. Disorders which need differential diagnosis from ABH are bullous diseases including pemphigus, pemphigoid, bullous lichen planus, erythema multiforme, linear IgA disease, amyloidosis and dermatitis herpetiformis and hematological disorders such as thrombocytopenia. In ABH, the blisters occur only in intraoral non-keratinized mucosa and the Nikolsky sign is negative. These clinical features are distinct from other bullous diseases. Biopsy can be used if necessary. Blood-filled subepithelial bulla and ulcerative lesion
with chronic inflammation are observed in the histological appearance, respectively before\textsuperscript{8,11} and after the breaking of blister.\textsuperscript{9} Generally, non-specific results appear in the immunofluorescence tests.\textsuperscript{10}

ABH generates blood-filled blister, however, hematologic abnormalities may exhibit similar lesion as well. Hematological disorders might exhibit epistaxis, ecchymosis and other signs. Thus clinicians should pay attention to these signs. In case of ABH, evaluation of complete blood count and hemostatic function result within normal range.\textsuperscript{8}

In the patient of our report, the biopsy of ulcerative lesion was done and subepithelial cleft with chronic inflammation was observed in the results. Hemorrhagic fluid with a few inflammatory cells was observed in the aspiration biopsy of intact blister before breaking. In combination of thorough review of medical history, lab test and biopsy, we diagnosed the patient as ABH.

Management of ABH includes reassuring patient, relief of pain, promotion of healing, and preventive treatment of secondary infection. Use of chlorohexidine gluconate was recommended as well.\textsuperscript{6,9,12} In this case, ABH was managed by topical steroid mouthwash.

**CONFLICT OF INTEREST**

No potential conflict of interest relevant to this article was reported.

**REFERENCES**