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Case Report

Angina bullosa hemorrhagica of the oral cavity in a COVID-19–positive patient: a rare case report

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ABSTRACT

Angina bullosa hemorrhagica (ABH) is a benign condition characterized by the sudden onset of painless, blood-filled bullae in the oral mucosa without systemic or hematologic disorders. During the COVID-19 pandemic, both virus-induced coagulopathy and anticoagulant therapy may be etiologies of unusual bleeding manifestations. A 46-year-old male presented with a sudden, painless swelling on the inner cheek that gradually enlarged over three days. The patient had a recent history of severe COVID-19 infection, treated with paracetamol, corticosteroids, and anticoagulants around two weeks before this lesion. Intraoral examination revealed a well-defined bluish-purple lesion on the right buccal mucosa, measuring approximately 3 cm, with no signs of trauma, ulceration, or active bleeding. Hematologic investigations showed normal hemoglobin and platelet counts, mildly prolonged PT and aPTT, slightly elevated INR, and increased D-dimer levels. The lesion was managed conservatively and resolved spontaneously within a week. This case highlights the potential for anticoagulant-associated or post-COVID microvascular fragility leading to oral mucosal hematomas such as ABH. Clinicians should remain vigilant about spontaneous oral hemorrhagic lesions in COVID-19 patients receiving anticoagulant therapy.

Keywords: Angina bullosa hemorrhagica, COVID-19, Oral cavity

INTRODUCTION

Coronaviruses belong to the family Coronaviridae, which includes enveloped viruses with non-segmented, single-stranded, positive-sense RNA genomes.[1] The clinical manifestations of COVID-19 range from mild upper respiratory illness to severe respiratory distress and multiorgan involvement.[2]

Oral manifestations have also been reported, including sore throat, mucosal erythema, xerostomia, swelling, and ulcerations.[3] Importantly, elevated D-dimer levels have been associated with severe COVID-19 and poor outcomes, reflecting the hypercoagulable state induced by the infection.[4,5] This coagulopathy may predispose patients to both thrombotic and bleeding complications.

Anticoagulant therapy during the management of severe symptoms to prevent thromboembolic events may increase the risk of hemorrhagic complications, including spontaneous mucosal hematomas.[6,7]

Angina bullosa haemorrhagica is an idiopathic condition for which the exact etiology is not known.[8] This condition is a benign, subepithelial, blood-filled blister occurring mainly in the oral mucosa. It often arises spontaneously or following mild trauma and typically resolves without intervention. Predisposing factors may include mucosal fragility, corticosteroid use, diabetes, and hypertension.[2,3]

This report describes a case of angina bullosa haemorrhagica of the oral mucosa in a COVID-19–positive patient on anticoagulant therapy.

CASE REPORT

A 46-year-old male presented to our clinic with a complaint of a painless swelling inside his right cheek, opposite the upper second premolar, which had been present for three days. The lesion appeared suddenly and was initially pea-sized and bluish, gradually enlarging to approximately 3 cm in diameter and turning dark purple. There was no history of trauma, pain, or dysphagia.

The patient had tested positive for COVID-19 two weeks earlier and had been hospitalized for one week due to high fever, dyspnea, and fatigue. He received dexamethasone, low-dose heparin, and paracetamol. He was discharged on the 5th day of admission with improvement. His past medical history included hypertension, and he is under Amlodipine 5 mg which was well controlled.

Intraoral examination revealed a well-circumscribed, non-tender, bluish-purple lesion measuring about 3×2 cm in diameter on the right buccal mucosa opposite the upper second premolar (Figure 1). The lesion was soft, compressible, and without signs of ulceration, bleeding, or secondary infection. There was no evidence of mucosal trauma or irritation from opposing dentition. The remainder of the oral cavity appeared normal.

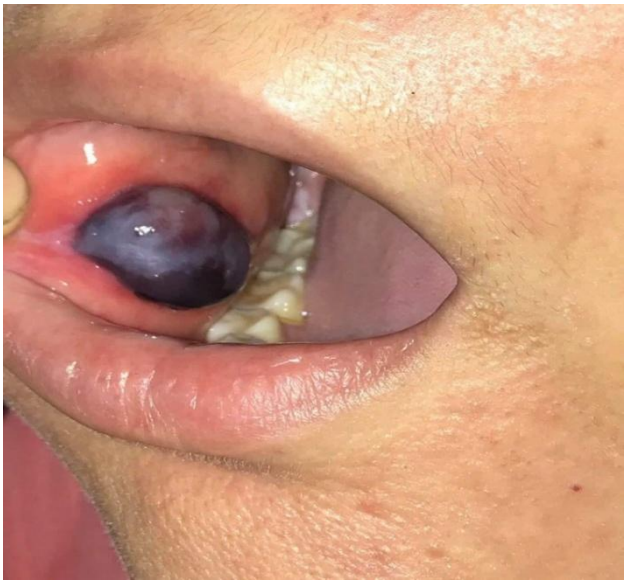

Figure 1: Purplish welling on the right buccal mucosa of around 3×2 cm diameter.

Investigations

Routine hematological tests were performed to assess possible bleeding abnormalities- hemoglobin: 16.8 g/dl, platelet count: 220,000/mm^3, prothrombin time (PT): 14.5 seconds, activated partial thromboplastin time (aPTT): 40 seconds, international normalized ratio (INR): 1.2 and D-dimer: 1112 ng/ml.

These findings indicated no active bleeding disorder or thrombocytopenia but suggested mild residual coagulopathy associated with prior COVID-19 infection.

The patient was managed conservatively, advised to avoid local trauma and maintain oral hygiene. No surgical or pharmacologic intervention was required. The lesion ruptured spontaneously after six days and completely resolved within one week without recurrence or scarring. Follow-up examination after two weeks showed normal mucosal healing

DISCUSSION

It was first described in 'traumatic oral hemophlyctenosis' in 1900.[8] It was renamed angina bullosa haemorrhagica by Badham.[8] It was first described in the late 1900s as 'traumatic oral hemophlyctenosis.[6] Retrospective studies by Grinspan et al and Rosa et al show a prevalence of 0.5% of ABH amongst patients presenting with oral pathology.[9,10]

COVID-19 infection induces widespread endothelial dysfunction and microvascular injury, leading to a prothrombotic state characterized by elevated D-dimer and fibrin degradation products.[4,5] To mitigate thrombosis risk, anticoagulant therapy has become standard practice, but this may paradoxically increase bleeding tendencies, particularly in fragile mucosal tissues.[6,7]

Various factors have been hypothesized to play a role in the pathogenesis of ABH, like diabetes mellitus, hypertension, rheumatoid arthritis, asthma, chronic kidney disease, gastrointestinal disorders, and autoimmune conditions like systemic lupus erythematosus.[11] Beguerie et al reported that 64% of the patients in their study had underlying systemic disease.[12]

In this case, both the anticoagulant therapy and post-COVID vascular changes likely contributed to the spontaneous formation of an oral hematoma. The lesion's benign and self-limiting nature, along with normal platelet counts and absence of systemic bleeding, support a diagnosis of ABH rather than a coagulopathy-related hemorrhage.

The most affected site has been the soft palate. Other areas generally affected include the lateral border of the tongue and the buccal mucosa.[13,14] Rarely, it may occur on the ventral surface of the tongue, lip, the floor of the mouth, pharynx, or esophagus. These regions are more prone to the occurrence of these bullous lesions are they are non-keratinized.[14] Angina bullosa-like lesions are amongst the most uncommon oral manifestations of COVID-19.[14]

COVID-19 is well known to cause systemic immune alterations. This includes a propensity to cause extensive alveolo-interstitial damage, immune dysregulation, and cytokine storms. Even though immune dysregulation has not been proven to play a role in the occurrence of ABH,

the immune dysfunction in COVID-19 may be responsible for this presentation in otherwise healthy individuals.[14]

The differential diagnosis includes mucous cysts, vascular malformations, and vesiculobullous diseases such as pemphigoid or lichen planus. However, the acute onset, isolated lesion, and spontaneous resolution distinguish ABH from these conditions.[14]

CONCLUSION

Clinicians should recognize ABH as a possible oral manifestation in post-COVID or anticoagulated patients to avoid unnecessary investigations or interventions. Conservative management and patient reassurance are generally sufficient.

This case emphasizes the need for clinical awareness of spontaneous oral hematomas such as ABH in patients with recent COVID-19 infection or on anticoagulant therapy. Recognition of this benign condition prevents misdiagnosis and ensures appropriate and non-invasive management.

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