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Quincke's Disease by Alprazolam: A Case Report

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Abstract

AAngioedema corresponds to a self-limited edema, located on the skin or mucosa, which results from leakage of fluid into the interstitium, caused by a loss of vascular integrity. It can occur isolated, as in angioedema of the uvula, or as part of an anaphylactic reaction. Isolated uvula angioedema, or Quincke's disease, is a rare clinical entity with multiple reported etiologies.

The presentation of this clinical case aims to raise awareness of a rare diagnosis that can be deadly. In the case presented it is suspected that the aetiological agent is pharmacological: an allergic reaction to alprazolam.

A 56-year-old man was admitted in the emergency department with oropharyngeal foreign body sensation and dysphagia after taking alprazolam. On examination of the oral cavity there was marked edema in the uvula, with visualization of the posterior wall of the oropharynx. The patient had a history of allergy to aceclofenac. He was treated with an antihistamine (clemastine) and a parenteral corticosteroid (hydrocortisone). After symptomatic improvement, he was discharged with oral corticosteroids.

Quincke's disease is a rare form of isolated angioedema of the uvula in which it is often not possible to discern a cause. Previous intake of unusual drugs should increase the level of suspicion for a pharmacological allergic etiology. Symptoms such as oropharyngeal foreign body sensation and dysphagia should not be underestimated, and patients should be quickly evaluated by the Otorhinolaryngologist.

Keywords: Quincke's disease, Alprazolam

Introduction:

Angioedema corresponds to a self-limited edema, located on the skin or mucosa, which results from leakage of fluid into the interstitium, caused by a loss of vascular integrity. It can occur isolated, as in angioedema of the uvula, or as part of an anaphylactic reaction. Isolated uvula angioedema, or Quincke's disease, is a rare clinical entity with multiple reported etiologies [1,2]. Contrary to other hypersensitivity reactions, it does not progress with skin rash, hypotension and tachycardia, manifesting with a circumscribed, non-pruritic uvula edema, with a quick onset [1,2]. The presentation of this clinical case aims to raise awareness of a rare diagnosis that can be deadly. In the clinical case presented, it is suspected that the aetiological agent is pharmacological, in this case an allergic reaction to taking alprazolam.

Case Report:

A 56-year-old man was admitted in the emergency department with oropharyngeal foreign body sensation and dysphagia, after a single dose of alprazolam. The patient denied cough, dyspnea and fever. Vital signs were within normal parameters. In the examination of the oral cavity there was marked edema of the uvula, with the posterior wall of the

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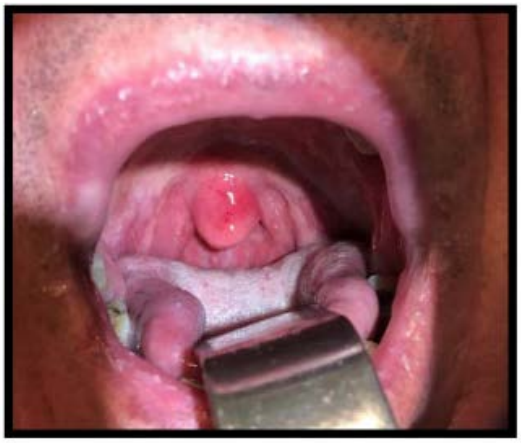

Figure 1: Edema of the uvula, with the posterior wall of the oropharynx visible.

oropharynx visible. In terms of personal history, the patient had arterial hypertension, obesity and chronic gastritis. The usual medication included pantoprazol, valsartan and bisoprolol. He had history of drug allergies, namely to aceclofenac. No family history of hypersensitivity reactions and no personal history of food allergies. The RT-PCR for SARS-CoV-2 was negative. Nasopharyngolaryngoscopy showed a slight edema of the arytenoids and esophageal sphincter, with no other supraglottic and glottic changes. He was treated with antihistamine (clemastine) and parenteral corticosteroid (hydrocortisone). After symptomatic improvement, he was discharged with oral corticosteroids (Figure 1).

Discussion:

Given the likely allergic background in this case (unusual drug in a patient with history of drug allergies), the laboratory investigation should have included a blood count with leukogram, measurement of IgE levels and evaluation of tryptase levels, 1-2h and 24h after the beginning of the symptoms. If a familial cause were suspected, assessment of C1 esterase levels would be helpful. The existence of recurrent attacks should raise suspicion regarding complement diseases, such as hereditary angioedema and acquired C1 esterase inhibitor deficiency [3,4]. Despite the favorable presentation and evolution of this case, this pathology can quickly compromise the airway. In addition to pharmacological treatment, continuous clinical monitoring is essential [4]. The approach to the upper airway (intubation/tracheostomy), oxygen therapy and the use of inhaled epinephrine should be reserved for severe cases [5]. Dexamethasone, due to its anti-inflammatory properties and long duration of action, should be the corticosteroid of choice [6]. Finally, it should be noted that Quincke's disease must be distinguished from infectious uvulitis, which is usually associated with tonsillitis, pharyngitis and epiglottitis [7]. This distinction is important, since the latter entity requires antibiotic therapy. Other differential diagnoses include soft palate/uvula trauma, neoplastic lesions of the uvula and congenital uvula hypertrophy [8].

Conclusion:

Quincke's disease is a rare form of isolated angioedema of the uvula in which it is often not possible to discern a cause. Previous intake of unusual drugs should increase the level of suspicion for a pharmacological allergic etiology. Symptoms such as oropharyngeal foreign body sensation and dysphagia should not be underestimated, and patients should be quickly evaluated by the Otorhinolaryngologist.

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Author Contributions:

PMG: Patient Management with JBC.

Conflict of Interest:

No conflict of interest was declared by the author.

Ethical Approval

The study has been approved by the appropriate ethics. Published with consent of the patient family.

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