

A Rare Case of Unilateral Tongue Edema with Angiotensin Converting Enzyme Inhibitors



Um Caso Raro de Edema Unilateral da Língua com Inibidores da Enzima de Conversão da Angiotensina

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ABSTRACT

Angiotensin converting enzyme inhibitors (ACEi) are widely used for the treatment of multiple conditions such as hypertension, heart failure and chronic kidney disease. Angioedema is a rare but potentially fatal complication of ACEi use and unilateral tongue edema is a very rare presentation. We report a case of a 55-year-old man, with a history of hypertension, on enalapril for three years, who presented to the hospital with unilateral tongue swelling, without airway compromise. Other causes were excluded and the diagnosis of angioedema due to enalapril was established. The patient was discharged with discontinuation of ACEi with total resolution of symptoms and without relapse after several months. Although very rare, unilateral tongue swelling should be considered in the presentation of angioedema associated with ACEi. Tight surveillance is important to prevent fatal complications such as airway obstruction. ACEi discontinuation is crucial to avoid clinical relapse.

Keywords: Angioedema; Angiotensin-Converting Enzyme Inhibitors; Enalapril

RESUMO

Os inibidores da enzima de conversão da angiotensina (iECAs) são amplamente usados no tratamento de várias patologias como a hipertensão arterial, insuficiência cardíaca e doença renal crónica. O angioedema é uma complicação rara mas potencialmente fatal desta medicação e o edema unilateral da língua é uma apresentação rara desta condição. Reportamos o caso de um homem de 55 anos com hipertensão, medicado há três anos com enalapril, que à admissão hospitalar apresentava edema unilateral da língua sem compromisso da via aérea. Outras etiologias foram excluídas, tendo-se assumido o diagnóstico de angioedema associado ao enalapril. Após suspensão do iECA os sintomas diminuíram progressivamente, sem recorrência do quadro após vários meses. Ainda que raro, o edema unilateral da língua deve ser considerado na apresentação do angioedema associado a iECA. É importante uma vigilância apertada para prevenir complicações fatais, tais como a obstrução da via aérea. A descontinuação do iECA é fundamental para evitar recidiva.

Palavras-chave: Angioedema; Enalapril; Inibidores da Enzima de Conversão da Angiotensina

INTRODUCTION

Angiotensin converting enzyme inhibitors (ACEi) are widely used in the management of several diseases. Angioedema is a self-limited, localized subcutaneous or submucosal swelling, resulting from the increased capillary permeability due to accumulation of bradykinins by ACEi. Unilateral tongue edema is an atypical and rare presentation. In a large clinical trial, angioedema occurred in approximately 0.6% of patients treated with enalapril, and its occurrence decreased over time.¹

We present the case of a 55-year-old man with unilateral tongue edema associated with ACEi.

CASE REPORT

A 55-year-old man with chronic hypertension and receiving treatment with enalapril presented for the first time in the emergency department (ED) with a one-hour history of sudden onset tongue edema. He noticed the edema after waking up with breathing discomfort. Upon admission in the ED, he had left unilateral non tender soft tongue swelling, without lip or palate involvement (Figs. 1 to 3). He was hemodynamically stable, without any abnormal findings in the

pulmonary evaluation. The remaining physical observation was normal, including the neurological examination. The patient denied history of tongue trauma and recent changes in any hygiene products. He had no history of allergies. He also denied dysphagia, hoarseness, rash, fever or pruritus. In the ED, blood tests were performed, without any changes. Cranial computed tomography was normal. During several hours of medical observation, the edema progressively resolved without any complications, such as airway compromise. Due to daily treatment with ACEi for almost three years and given the absence of other causes, the diagnosis of enalapril angioedema was assumed and the patient was discharged with indication to stop enalapril.

A few months after the discontinuation of enalapril, at follow-up, he had a normal volume tongue with no history of angioedema relapse (Figs. 4 and 5).

DISCUSSION

Angioedema is one of the well-known side effects of ACEi treatment. ACEi inhibit angiotensin converting enzyme (kininase II), which is responsible for the degradation

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Figure 1 – Unilateral left tongue edema, left lateral view



Figure 2 – Unilateral left tongue edema, frontal view



Figure 3 – Unilateral left tongue edema, lateral perspective

of bradykinin. The increased concentration of this peptide increases capillary permeability, with fluid extravasation and swelling of tissues, resulting in angioedema.

Angioedema due to ACEi usually occurs in the initial months after starting therapy, although there are described cases when the effect only appears after several years, with a greatly decreasing risk over time.^{1,2} According to the literature, it can develop over minutes to hours, resolving over 24 to 72 hours after medication discontinuation.^{2,3} Previous angioedema history, smoking, age above 65 years, use of non-steroidal anti-inflammatory drugs or female sex are risk factors for angioedema development.^{1,4-6} Our patient only presented a history of smoking and was treated for almost

three years with enalapril, which reveals the importance of considering ACEi side effects during the course of treatment.

In this patient, we observed unilateral tongue edema involvement. This is a very rare angioedema presentation with few cases reported in the literature.⁷⁻¹³ The cause of this specific involvement remains uncertain, but one theory states that unilateral edema might be the progression over time to bilateral edema.¹⁴ Although rare, this presentation should be considered in this diagnosis due to the risk of progression to airway obstruction. The management of these patients involves tight surveillance, with immediate airway evaluation, since its compromise could be potentially fatal. Although there is no specific targeted therapy for angioedema, some patients may require corticosteroids and antihistamines.¹⁴ The effective long-term treatment is ACEi discontinuation. In this case report, after enalapril discontinuation, symptoms resolved without angioedema relapse.

Regardless of the time of use, considering this case with the appearance of angioedema after several years on enalapril, clinicians should always be aware of the possibility of this side effect in patients under treatment with ACEi, no matter how long ago the drug was introduced.

AUTHORS CONTRIBUTION

BGB: Draft of the article.

MC, PN: Draft and critical review of the article.

FP, CM: Critical review of the article.

PROTECTION OF HUMANS AND ANIMALS

The authors declare that the procedures were followed according to the regulations established by the Clinical Research and Ethics Committee and to the Helsinki Declaration of the World Medical Association updated in 2013.

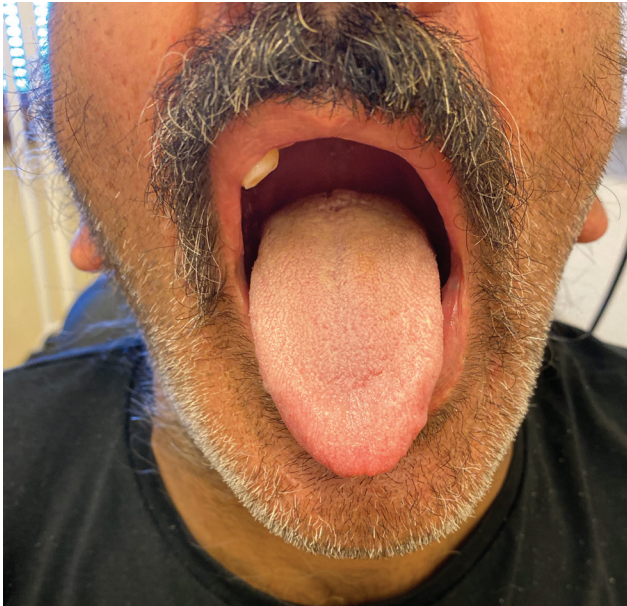


Figure 4 – Follow-up, one and a half months later, frontal perspective



Figure 5 – Follow-up, one and a half months later, left side perspective

DATA CONFIDENTIALITY

The authors declare having followed the protocols in use at their working center regarding patients' data publication.

INFORMED CONSENT

Obtained.

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COMPETING INTERESTS

The authors declare that there are no competing interests.

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