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Case Report

Unilateral Tongue Angioedema Caused Bay Lopril: A Case Report

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A B S T R A C T

Introduction: ACE inhibitors are widely prescribed for anti-proteinuria, nephroprotection, cardio protection or treatment of hypertension. Angioedema, one of the adverse effects of ACE inhibitors, is not uncommon. It can sometimes be life-threatening. Unilateral angioedema of the tongue due to ACE inhibitors is rarely described.

We report in this study a case of a hemodialysis patient treated with an ACE inhibitor lopril for 2 years. He presented with a severe attack of unilateral angioedema of the tongue.

Case presentation: This is a 57-year-old patient (S. A.) with a history of chronic renal failure on hemodialysis for two years and hypertensive, initially treated with an ARB II for two years then replaced by an ACE inhibitor, lopril. Six months after the introduction of lopril, the patient presented with slight edema of the left half of the tongue with moderate glossitis 2 days after a CT scan injected with an iodinated contrast agent. The symptoms were linked to a probable allergy to iodinated PDC and the patient was treated with an antihistamine and intravenous corticosteroids with rapid regression in two days. Six months later, the patient presented with significant edema of the right half of the tongue with difficulty speaking and swallowing saliva with a state of anxiety and agitation, a hypertensive peak at 190/90 mm Hg and a correct arterial oxygen saturation in room air at 98%.

He was taken into emergency care with injection of hydrocortisone hemi succinate and nebulizations with adrenaline with the prescription of an antihistamine. The complete blood count did not show hypereosinophilia and specific IgE was negative for food products. The evolution was marked by a progressive regression of symptoms until disappearance in three days. The diagnosis was unilateral angioedema of the tongue due to ACE inhibitors and the causal treatment was permanently discontinued.

Discussion: ACE inhibitors are usually well tolerated by the majority of patients. However, 0.7% of patients taking ACE inhibitors develop angioedema. The time to onset of angioedema due to ACE inhibitors ranges from a few hours to several years after initiating treatment. Angioedema can also occur after treatment discontinuation. Bradykinin is considered the main factor contributing to the development of angioedema. The combination of an ACE inhibitor with certain drugs that increase bradykinin concentrations increases the risk of developing bradykinin-induced angioedema. Unilateral tongue involvement is rare and only a few case reports have been published documenting this condition. The reason for unilateral tongue edema is not well understood. The basic treatment of ACE inhibitor-induced angioedema consists of securing the upper airway and preparing for artificial ventilation if necessary. Corticosteroids and antihistamines are widely used in the management of these cases.

Conclusion: Unilateral angioedema of the tongue, which is less frequently described in the literature, among other things, should consider the use of ACE inhibitors.

Keywords: ACE inhibitor; Lopril; Unilateral angioedema of the tongue

Introduction

Angioedema in general may result from a mast cell-mediated reaction or be associated with an increase in bradykinin concentration, linked to a defect in its catabolism[1].

It is clinically difficult to differentiate between the two mechanisms, particularly in the presence of a poor presentation or one made exclusively of angioedema, especially since there is currently no reliable biomarker to differentiate the two diagnoses. Angioedema most often presents as non-pruritic, sudden-onset hypodermal edema, located mainly on the face, tongue or upper airways[2].

It can also be localized to the digestive mucosa and present in the form of abdominal pain of varying intensity. Angioedema can be of hereditary origin (associated or not with a deficiency in C1 inhibitor) or of drug origin by inhibition of enzymes which degrade bradykinin, in particular: the converting enzyme for angiotensin converting enzyme inhibitors (ACE) or angiotensin II receptor antagonists (ARB II) or dipeptidyl peptidase for gliptins. Other causes of angioedema, such as pollen-food allergy syndrome which is a cross-reaction mediated by immunoglobulin E type I to an antigen of plant origin, as it can be caused by an infection[3,4].

ACE inhibitors are widely prescribed for anti-proteinuria, nephroprotection, cardio protection or treatment of hypertension. Angioedema, one of the adverse effects of ACE inhibitors, is not uncommon. It can sometimes be life-threatening. Angioedema due to ACE inhibitors is often localized to the ENT area. Unilateral angioedema of the tongue due to ACE inhibitors is rarely described.

We report in this study a case of a hemodialysis patient treated with an ARB II for 2 years and then with the ACE inhibitor lopril for 2 years. He presented with a severe attack of unilateral angioedema of the tongue with difficulty speaking and swallowing saliva, due to the adverse effect of lopril, requiring permanent discontinuation of the ACE inhibitors.

Case Presentation

This is a 57-year-old patient (S. A.) with a history of chronic renal failure due to familial nephropathy on hemodialysis for two years and hypertensive, initially treated with an ARB II for two years then replaced by an ACE inhibitor, lopril. Six months after the introduction of lopril, the patient presented with slight edema of the left half of the tongue with moderate glossitis 2 days after a CT scan injected with an iodinated contrast agent. The symptoms were linked to a probable allergy to iodinated PDC and the patient was treated with an antihistamine and intravenous corticosteroids with rapid regression in two days **(Figure 1)**.

Six months later, the patient presented with significant edema of the right half of the tongue with difficulty speaking and swallowing saliva with a state of anxiety and agitation, a hypertensive peak at 190/90 mm Hg and a correct arterial oxygen saturation in room air at 98%.

He was taken into emergency care with injection of hydrocortisone hemi succinate and nebulization's with adrenaline with the prescription of an antihistamine. The complete blood count did not show hyper eosinophilia and specific IgE was negative for food products. The evolution was marked by a progressive regression of symptoms until disappearance in three days. The diagnosis was unilateral angioedema of the tongue due to ACE inhibitors and the causal treatment was permanently discontinued.

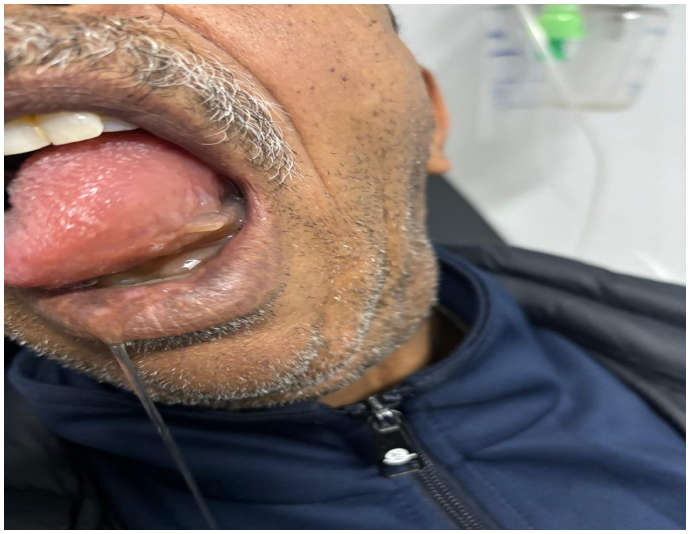

Figure 1: Pateint with antihistamine and intravenous corticosteroids with rapid regression.

Discussion

ACE inhibitors are usually well tolerated by the majority of patients. However, 0.7% of patients taking ACE inhibitors develop angioedema[5].

Other studies suggest that the incidence of angioedema in patients treated with ACE inhibitors is estimated at 0.20%[6]. Estimates of cross-reactions between ACE inhibitors and ARB II in different studies range from 7.7% to 50%[3].

The time to onset of angioedema due to ACE inhibitors ranges from a few hours to several years after initiating treatment, but the majority of cases occur within the first three months following treatment initiation[7].

Cases of angioedema have been reported after 23 years of continuous treatment[6]. In our patient, angioedema appeared after two years of ACE inhibitor treatment and the second attack was more severe than the first. Angioedema can also occur after treatment discontinuation. In another published case, angioedema developed after 32 years of continuous ACE inhibitor treatment and its recurrence was observed 2 years after discontinuation[8].

The attacks can become more and more frequent and/or more and more severe as in our case, always with a predilection for the ENT sector.

The combination of an ACE inhibitor or sartan with certain drugs that increase bradykinin concentrations increases the risk of developing bradykinin-induced angioedema. We noted in the literature the publication of a case of unilateral periorbital angioedema induced by contrast media, as well as another case of intestinal angioedema also induced by contrast media[9,10].

Another case of unilateral angioedema of the tongue induced by paracetamol (Acetaminophen) has been published[11].

Aspirin has also been described as an inducer of angioedema of half the tongue in another publication[12].

We wonder whether the injection of contrast agent in our patient's case plays an additional role in the onset of ACE inhibitor-induced angioedema during the first attack.

ACE inhibitor-induced angioedema is due to the effects of ACE inhibitors on the renin-angiotensin-aldosterone system, which results in increased angiotensin I and bradykinin levels due to a failure of bradykinin degradation by inhibition of enzymes that degrade bradykinin by ACE inhibitors, particularly the angiotensin-converting enzyme[1].

Bradykinin is considered the main factor contributing to the development of angioedema, as it causes vasodilation and swelling[1]. Unlike histamine-releasing angioedema, bradykinin angioedema spontaneously regresses despite continued use of the causative drug, often leading to the drug hypothesis not being considered. However, the symptoms improved with the administration antihistamines; therefore, an associated histamine mechanism could not be excluded[1].

Unlike hereditary or acquired forms of bradykinin-induced angioedema, C1 inhibitor (weight or functional) and C4 levels are normal.

The diagnosis of ACE inhibitor-induced angioedema is generally based on clinical symptoms. Typical symptoms are localized angioedema of the face, erythema without pruritus persisting for 24 to 72 hours, followed by spontaneous remission[2].

Unilateral tongue involvement is rare and only a few case reports have been published documenting this condition. Our case represents the second case of unilateral ACE inhibitor-induced angioedema published in Africa after that of Julian Robert Paul Eloff despite the frequency of hypertension and frequent use of ACE inhibitors suggesting the rarity of the involvement or underestimation due to non-reporting of the cases encountered[13]. Quincke's edema resolved within two days in the majority of cases even in our presented case. The reason for unilateral tongue edema is not well understood; however, it is assumed that unilateral edema precedes bilateral angioedema and that the asymmetry of the lingual nerve results in a left-right difference in the chemical microenvironment, resulting in unilateral sensitivity to the action of inflammatory mediators[12,14].

Recurrence can be observed if the treatment is not stopped by error of diagnosis as in our case or as the case published by et all[15], as it can recur even after stopping the drug supposed to be in question (8) and this must review the diagnosis and rediscuss the causal agent especially since the published cases of unilateral angioedema of the tongue reveal more and more different causes such as paracetamol, sartan, iodinated contrast agent, aspirin or recombinant tissue-type plasminogen activator[3,9,11,12,16].

Recurrence can be observed if the treatment is not stopped by error of diagnosis as in our case or as the case published by Mlynarek A, et al.[15] as it can recur even after stopping the drug supposed to be in question[8] and this must review the diagnosis and rediscuss the causal agent especially since the published cases of unilateral angioedema of the tongue reveal more and more different causes such as paracetamol, sartan, iodinated contrast agent, aspirin or recombinant tissue-type plasminogen activator[3,9,11,12,16]. Patients should be informed that angioedema can occur or reappear several weeks after stopping treatment with ACE inhibitors. Although the incidence of angioedema induced by ARBs II is much lower than that induced by ACE inhibitors, cross-reactivity may occur in less than 10% of cases[13]. Therefore, the initiation of an ARB II in patients who have experienced angioedema induced by ACE inhibitors should be carefully evaluated based on the benefit/risk ratio.

Treatment of an ACE inhibitor-induced angioedema attack is urgent because life is compromised, particularly if the edema is significant and affects the upper respiratory tract. The basic treatment of ACE inhibitor-induced angioedema consists of securing the upper airway and preparing for artificial ventilation if necessary. Corticosteroids and antihistamines are widely used in the management of these cases. Epinephrine was also prescribed in our case, as in the case of Yuki Matsuhisa, et al.[8].

The offending drug must be discontinued.

Although not yet approved by the U.S. Food and Drug Administration, fresh frozen plasma (FFP) has been used successfully in some patients. FFP contains ACE and some patients have reported marked improvement after administration of two units[17].

Although its efficacy remains controversial, tranexamic acid (TXA) is used to treat hereditary angioedema caused by inherited C1 esterase inhibitor deficiency in some countries.

One study evaluated TXA as an on-demand and prophylactic treatment in patients with hereditary angioedema. The majority of studies (80%) demonstrated the ineffectiveness of on-demand TXA for cutaneous, abdominal or laryngeal swelling. In prophylaxis, approximately 50% of case series, case reports and observational studies reported beneficial effects of TXA[18].

Newer therapies, such as icatibant and pdC1-INH, have been shown to be more effective[19].

Some studies consider prescribing ATX in cases of recurrence of angioedema[8].

However, there are no conventional recommendations on the use of TXA in the treatment of drug-induced angioedema or in the prevention of recurrence.

Conclusion

ACE inhibitor-induced angioedema is a potentially serious adverse effect of ACE inhibitors and can be exacerbated by the use of other medications (aspirin, paracetamol, iodinated contrast agents that activate palsminogen, etc.).

Furthermore, unilateral angioedema of the tongue, which is less frequently described in the literature, should raise this diagnosis and, among other things, should consider the use of ACE inhibitors. Recurrence after treatment discontinuation has also been observed, requiring patient education. The mechanism of unilateral tongue involvement, as well as that of recurrence after treatment discontinuation, remains unclear.

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