CASE REPORT

Ischaemic necrosis of the tongue

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SUMMARY

Necrosis of the tongue is a rare clinical finding. The rich vascularity of the tongue means necrosis is uncommon but it has been reported secondary to giant cell arthritis, radiotherapy and ischaemia. We report the case of a 61-year-old man admitted with an acute abdomen, who later developed gross swelling of the tongue, secondary to ischaemic necrosis, which necessitated tracheostomy placement. The ischaemia was managed conservatively with heparinisation and by allowing the ischaemic area to demarcate and slough off naturally.

BACKGROUND

The tongue is a well vascularised end-organ with its primary blood supply from the lingual arteries. For this reason reports of lingual ischaemia and necrosis are rare. The most common cause for this presentation reported is giant cell arteritis (GCA). This, however, is usually unilateral and associated with other features of that pathology.¹ Other reports of lingual necrosis exist secondary to vasopressor use, radiotherapy and antiphospholipid syndrome (APS).²⁻⁴ Reports of bilateral ischaemia are rare but have been reported in relation to severe hypotension or disseminated intravascular coagulation.⁵⁻⁶ We present a case of bilateral lingual ischaemic necrosis presenting in a 61-year-old patient as an acutely swollen tongue threatening the airway.

CASE PRESENTATION

A 61-year-old man was admitted through the emergency department with severe hypotension (50/30 mm Hg) and an acute abdomen. He had a history of metastatic prostate cancer, deep venous thrombosis and obstructive airways disease. The patient was taken to the operating theatre and underwent sub-total colectomy. The bowel was found to be ischaemic at surgery secondary to emboli. He was later admitted to the intensive care unit (ICU). His admission was further complicated by renal dysfunction requiring haemofiltration, and distal limb ischaemia of toes and fingers requiring heparinisation.

On day 13 of his admission he was noted to develop an acutely swollen tongue, believed initially to be a haematoma. There was increasing concern about the patient's airway and a tracheostomy was placed surgically on day 18. The swelling was later diagnosed as a result of lingual ischaemia. This ischaemia was believed to have resulted from end artery emboli, given the coexistence of other end-organ damage secondary to the same.

DIFFERENTIAL DIAGNOSIS

Lingual ischaemia is a rare clinical presentation; in this setting, potential diagnoses to consider are:

- ▶ Arterial infarction
- ▶ Lingual infarction
- ▶ Profound hypotension
- ▶ Vasopressor use
- ▶ Disseminated intravascular coagulation
- ▶ Giant cell arteritis (most common cause but usually unilateral)

TREATMENT

A conservative treatment approach was decided on. This involved continuing with adequate therapeutic heparinisation of the patient as previously started and awaiting demarcation of the ischaemic area. The area was expected to heal by secondary intention.

OUTCOME AND FOLLOW-UP

The ischaemic area later became necrotic ([figure 1](#)) before sloughing off the tongue. The patient was discharged from ICU to the ward on day 33 to continue supportive care and at this time a clear demarcation between perfused and ischaemic areas was visible on the tongue ([figure 2](#)).

DISCUSSION

The tongue is well vascularised with its primary blood supply emanating from the lingual arteries. The lingual artery is an end artery of the intrinsic

Figure 1 Bilateral ischaemia with necrotic tissue sloughing off the tongue.

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Figure 2 Demarcation between perfused and ischaemic lingual tissue.

muscles of the tongue. It also receives some supply from the facial and pharyngeal arteries. When lingual necrosis presents it is commonly unilateral and related to the vasculitis GCA. Other features of this pathology are usually, though not always, present including a pulsatile temporal artery, jaw claudication and a raised erythrocyte sedimentation rate (ESR).¹ Cases have been reported where *Ergotamine Tartrate*, used to treat migraine, precipitated lingual necrosis. In these cases, it is believed that headaches may have been misdiagnosed as migraine and not GCA. The vasospastic effects of *Ergotamine Tartrate* may then magnify the effect of the vasculitis, an important consideration when prescribing for elderly patients.^{7 8}

Case reports also exist of other causes of lingual necrosis including vasopressor use, radiotherapy, antiphospholipid syndrome, disseminated intravascular coagulation, fungal infection and secondary to carotid artery stenosis.^{2–4 6 9 10} A case series has also been published recently documenting lingual necrosis secondary to severe hypotension in cardiogenic shock. All patients in this series suffered end-organ damage, limb ischaemia and lingual necrosis as a result of hypotension and poor peripheral perfusion.⁵

In our patient, GCA was an unlikely diagnosis given the chronology of the presentation, its bilateral nature and the absence of other features of the pathology. A biopsy could be considered to support this diagnosis if vasculitis was suspected in tandem with characteristic clinical features and an elevated ESR.¹ A biopsy and specific antibodies, anticardiolipin and lupus anticoagulant, can also be used to diagnose lingual lesions relating to antiphospholipid syndrome.⁴ A number of other potential causes listed above could also be reasonably excluded. Our patient was severely hypotensive on admission requiring extensive vasopressor use. His lingual necrosis appeared to be temporally related to the development of other end-organ dysfunction suggesting poor peripheral perfusion or embolic ischaemia as potential causes. The acute swelling may also have suggested possible venous infarction.

The management of lingual necrosis is not well reported but conservative measures are usually adopted. In cases of GCA or APS, it appears that treatment of the underlying pathology also allows for resolution of the lingual necrosis. In the case of GCA this involves high-dose prednisolone once a diagnosis is suspected. Other causes such as APS may require anticoagulant treatment. In previously reported cases of extensive necrosis a conservative approach was adopted, as with our patient.^{5 6} These cases had a high mortality rate related to extensive comorbidities. If surgical treatment is viable, the necrotic area may be debrided to aid with healing and rehabilitation. In cases where a conservative approach is adopted the necrotic tissue will demarcate and autoamputate over time. Healing in this instance is by secondary intention and may lead to considerable deformity and issues with speech and swallowing.² Ultimately, the correlation between extensive lingual necrosis and other severe co-morbidities, resulting in end-organ damage, may highlight a poor overall prognosis.

Learning points

- ▶ Lingual ischaemia with necrosis is a rare presentation.
- ▶ Giant cell arteritis is a common cause of unilateral lingual necrosis.
- ▶ In extensive cases other comorbidities may coexist and lingual necrosis may be a sign of poor overall prognosis.

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