**INTRODUCTION**

Ischaemic necrosis of the tongue is rare. Temporal arteritis is most common cause, which often affects the tongue unilaterally. We report a case of an 80-year-old woman who developed bilateral ischaemic necrosis of the tongue due to an atypical presentation and delayed diagnosis of temporal arteritis.

**CASE REPORT**

An 80-year-old woman presented to her general practitioner (GP) with a 10-day history of bilateral frontal and occipital headache associated with neck pain, throat pain and low-grade fever. She was given oral antibiotics, but on review 4 days later, tongue pain and swelling was noted and low-dose oral prednisolone was prescribed. 8 days after initial GP review, the patient was admitted to her local hospital for suspected cerebrovascular event. CT brain and angiogram were normal. In this admission, she developed grey-coloured plaques on her tongue. She experienced increasing tongue pain, odynophagia and dysphagia to oral fluids. This was suspected to be due to a fungal infection. She developed acute painless visual loss in her right eye 2 days later, which prompted urgent transfer to the Royal Victorian Eye and Ear Hospital.

On examination, there were friable grey-yellow plaques involving majority of the dorsal anterior two-thirds of her tongue (Figure 1). Ophthalmic exam confirmed absent right temporal artery pulse and right eye retinal changes suggestive of temporal arteritis.

Initial investigations revealed white cell count of 15.6 x10^9/L with neutrophilia, ESR of 34mm/h and CRP of 69mg/L. Sphyllis serology and connective tissue disease screen were negative.

Histopathology of tongue revealed extensive inflammation, ulceration and necrosis. These features were consistent with ischaemic necrosis (Figure 2). Tissue culture of tongue was negative for microbial growth. Histopathology of temporal artery confirmed temporal arteritis.

The patient received 3 days of intravenous methylprednisolone. There was no further extension of the tongue necrosis. A portion of the necrotic tongue was demarcated and sloughed off (Figure 3). Her pain and oral intake gradually improved. However, her right eye vision experienced minimal improvement (from hand movements to 6/60). The patient was discharged on oral prednisolone, omeprazole and amoxicillin.

On review 3 weeks later, there was significant improvement in the appearance of her tongue, which was healing with minimal ulceration remaining (Figure 4). She described mild paraesthesia over the left lateral aspect of her tongue where most of the necrosis occurred, which was causing her mild dysphagia.

**DISCUSSION**

Temporal arteritis is the most common cause of ischaemic tongue necrosis, typically affecting it unilaterally. Other causes include shock, medium-large vessel vasculitis such as polyarteritis nodosa and granulomatosis with polyangiitis, infection (fungal, syphilis, tuberculosis), pro-thrombotic states and radiation therapy.

In our case, the diagnosis of temporal arteritis was delayed due to an atypical presentation of what was misdiagnosed as antibiotics. We considered fungal infection of the tongue. The classic symptoms of temporal arteritis are visual loss, temporal tenderness and jaw claudication. However, 30% of patients may exhibit neurological symptoms instead, including central neuropathies, peripheral neuropathies or stroke. When temporal arteritis is suspected, steroid therapy should be commenced as soon as possible. In our institution, patients receive 3 days of intravenous methylprednisolone (1mg/kg).

The rarity of tongue necrosis makes it difficult to establish definitive management guidelines. Other than identifying and treating the underlying cause, most case reports describe conservative measures. This involves allowing necrotic sections of the tongue to demarcate and autuminate over time, with aim of allowing healing via secondary intention. Prophylactic oral antibiotics have been suggested to cover oral flora, with surgical debridement of necrotic tissue if infection develops. Even after resolution of the underlying cause, necrosis of the tongue may affect the patient’s speech and swallowing. The role of surgical interventions for such issues or in those with extensive loss of lingual tissue remains unclear.

We suggest that, in the absence of typical symptoms of temporal arteritis, any patient with lingual pain and evidence of tongue swelling or necrosis should be treated with a high index of suspicion of temporal arteritis, especially in patients above the age of 50.

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**REFERENCES**

2. Mccollinick DM, Khan I, Cotter CJ. Ischaemic necrosis of the tongue. BMJ Case Reports. 2011;2011: