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Tongue Necrosis Secondary to Giant Cell Arteritis

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Abstract

Giant cell arteritis (GCA) is a vasculitis of large and medium sized vessels that commonly affects extracranial branches of the internal and external carotid arteries. It is a diagnosis of the elderly that typically presents temporal tenderness, claudication of the jaw or vision loss. Lingual necrosis is unusual symptom of GCA. We report a case of GCA revealed by a tongue necrosis.

Keywords: tongue necrosis, giant cell arteritis

1. Introduction

Giant cell arteritis is a vasculitis of large and medium sized vessels that commonly affects extra-cranial branches of the internal and external carotid arteries, particularly the temporal artery. Common features include headache, scalp tenderness, visual symptoms and jaw claudication. Lingual necrosis is unusual symptom. We describe a patient who presented with tongue necrosis as initial symptom of GCA.

2. Case Report

An 81-year-old female patient presented with tongue necrosis. She has no pathological history and she's suffering from pain and progressive swelling of the tongue since 3 weeks that was compromising her speech and deglutition. She reported a 2 month history of headache. She did not have vision loss, jaw claudication or scalp tenderness. Physical examination founded edema and a large necrotic ulcer of the tongue (Figure 1) and abolition of left temporal artery pulse. Laboratory analyses showed a raised erythrocyte sedimentation rate (90 mm in the first hour). Anti-neutrophil cytoplasmic antibody was negative. Consultation with the ophthalmologist was normal. Temporal artery biopsy was negative. A tongue biopsy was performed and showed necrotic infiltrate. There are no malignant cells, amyloid deposits, granuloma or signs of vasculitis. The presence of three of five ACR criteria (Table 1) permit a diagnosis of GCA with tongue necrosis to be made. Treatment with high dose corticosteroids (1 mg/kg/day) improved clinical outcome after a 3 month follow-up.

Figure 1. Edema and a large necrotic ulcer of the tongue

Table 1. American College of Rheumatology diagnostic criteria of GCA diagnosis (M. Brodmann, A. Dorr, F. Hafner, T. Gary & E. Pilger, 2009)

American College of Rheumatology diagnostic criteria
1-Age older than 50 years at onset of disease.
2-New onset of localized headache.
3-Abnormal temporal artery with tenderness or decreased pulse.
4-Erythrocyte sedimentation rate higher than 50 mm/hr.
5-A biopsy of the artery showing necrotizing arteritis with predominant mononuclear cell infiltrate or granulomatous process with multinucleated giant cells.

3. Discussion

Tongue necrosis is a rare ischemic complication of GCA. Sixty-two case reports in the literature have been identified in the English language (Grant SW, Underhill HC & Atkin P., 2013). Necrosis commonly affects one side of the tongue like in our observation. In a few cases bilateral necrosis has been described (Jose R. Zaragoza, Natalia Vernon & Gisoo Ghaffari, 2015). Tongue necrosis is no specific of GCA and can appear on other diseases such malignant tumors, chemotherapy, radiation of the neck and calciphylaxis (Table 2). GCA must be evoked in case of unexplained tongue necrosis after exclusion of these appearing reasons specially if typical symptoms of GCA are lacking. In our case the diagnosis of GCA was very likely given the age, the presence of headache, abolition of temporal artery and elevated erythrocyte sedimentation rate despite non-contributory a biopsy of temporal artery. Tongue necrosis is associated with high incidence of concomitant visual loss and is considered as unfavorable prognostic sign. Indeed, tongue necrosis means extensive vessel involvement by the disease, as the tongue area is a well-vascularized region (M. Brodmann, A. Dorr, F. Hafner, T. Gary & E. Pilger, 2009). However, corticosteroids lead frequently to complete healing of the ischemic lesions (C Fongaufier, A Guffroy & J Christophe, 2018), as this was the case in our patient.

Table 2. Differential diagnoses for necrosis of the tongue

Calciphylaxis	Intra-arterial drug injections
Tuberculosis	Syphilis
Radiotherapy	Amyloidosis
Mid-tongue tumour	Floor of mouth abscess
Chemotherapy	Radical neck dissection
Embolism	Hodgkin’s disease

4. Conclusion

In patients presenting with lingual swelling, pain, and discoloration, GCA should be suspected and prompt therapy should be initiated to avoid irreversible complications.

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