

**Bilateral ischemic necrosis of the tongue due to disseminated intravascular
coagulation**

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Abstract

Ischemic necrosis of the tongue is a rare condition because the tongue has a rich blood supply. Temporal arteritis appears to be the most frequent cause of tongue necrosis. We present the case of an 82-year-old man who developed bilateral ischemic necrosis of the tongue. The necrosis was considered as a sequela of thrombosis of the lingual artery due to disseminated intravascular coagulation. To our knowledge, this is the first reported case of necrosis of the tongue secondary to disseminated intravascular coagulation.

Keywords: tongue, necrosis, DIC

Introduction

Ischemic necrosis of the tongue is a rare condition because the tongue has a rich supply of blood from the bilateral lingual arteries; from branches of the facial, pharyngeal and palatine arteries; and due to collateral circulation. Temporal arteritis appears to be the most frequent cause of tongue necrosis, with approximately 60 cases having been reported in the literature.⁶ Necrosis of the tongue has also been reported subsequent to intra-arterial vasopressin therapy, transient ischemic attack and intestinal infarction.^{4,5,10} Recently, we encountered a patient with rapid development of bilateral ischemic necrosis of the tongue secondary to thrombosis of the lingual artery due to disseminated intravascular coagulation (DIC). To our knowledge, this is the first reported case of necrosis of the tongue secondary to DIC.

Case report

An 82-year-old obese (body mass index, 25.7 kg/m²) Japanese man was referred to our hospital due to bloody stools. His past medical history included Meniere's disease, lacuna infarction, diabetes mellitus and lung hernia. Examination after admission revealed hepatocellular carcinoma, abdominal aortic aneurysm, adrenal tumor, haemorrhoids, tubular adenoma of the colon and prostate cancer. The patient

underwent partial liver resection and extirpation of the adrenal gland. Nine days after the operation, he had a re-operation due to suture failure. Anaesthesia was induced with midazolam, and it was maintained with sevoflurane and fentanyl. The airway was secured with a 7.5-mm oral endotracheal tube via direct laryngoscopy, and no compression of the tongue by the oral endotracheal tube was noted during the operation. After the second operation, aspiration pneumonia was observed, and the patient was transferred to the intensive care unit. Laboratory examination at 5 days after the second operation indicated leukocytosis with neutrophilia, high levels of C-reactive protein (CRP), fibrinogen/fibrin degradation products (FDP), D-dimer, activated partial thromboplastin time (APTT) and prothrombin time (PT), low platelet count and low activity of anti-thrombin-III (AT-III) (data not shown). However, the erythrocyte sedimentation rate (ESR) was within the normal range, and negative results were obtained in the tests for anti-nuclear antibodies, anti-DNA antibodies and rheumatoid factors. A tracheostomy was performed 10 days after the second operation under intravenous sedation and local anaesthesia. On the occasion of extubation of the oral endotracheal tube, necrotic tissue was detected on the bilateral anterior one-third of the tongue (Fig. 1). Infarction was suspected; however, the patient did not undergo angiography because of his general condition was poor.

Although the mouth was cleaned, the signs of necrosis were not diminished. Therefore, the non-haemorrhagic necrotic tissues were resected. Histological examination revealed thrombosis of the peripheral tongue artery and vein, and atrophy of the muscle with fatty degeneration (Fig. 2). The final diagnosis was ischemic necrosis of the tongue due to thrombosis of the artery subsequent to DIC. Although aggressive medical treatment with intravenous administration of antibiotics, anticoagulants and other supportive therapies was started, the patient's status rapidly deteriorated. He finally died on day 19 after the second operation.

Discussion

DIC is a complex, systemic, thrombohaemorrhagic disorder characterized by an imbalance in the haemostatic process, resulting in extensive thrombosis and haemorrhage complications. Laboratory evidences of DIC are procoagulant activation, fibrinolytic activation, inhibitor consumption and biomechanical evidence of end-organ damage.^{1,2} DIC is frequently associated with obstetrical disorders, malignant diseases and severe infections.⁹ Pathological findings in patients with DIC include major and small vascular thrombosis, and hemorrhage; vascular thrombosis is frequently observed in the kidney, skin, brain, lung and heart.⁸ In DIC, most of the

thrombosis develops formed in the systemic circulation, and it eventually impinges upon smaller vessels.⁷ Since the distal portion of the infarct has no blood supply, it does not undergo inflammatory changes and gets mummified. In this case, based on the laboratory and pathological findings, DIC was the most probable cause of the ischemic tongue necrosis. We believe that thrombosis of the tongue is common but that it does not result in clinical infarction because of rich collateral circulation, which might have been deficient in this particular patient for some reason.

Tongue necrosis is almost always unilateral, and reports of bilateral necrosis are rare.⁶ The most frequent cause of tongue necrosis appears to be temporal arteritis, which is a common manifestation of systemic arteritis (giant-cell arteritis). In 70% of patients, the physical symptoms included loss of arterial pulsation and a tender, thickened and nodular temporal artery. Other symptoms, such as ischemic ulcers of the scalp, hair loss and jaw claudication may also be observed.⁴ Estimation of the ESR and temporal artery biopsy are generally the most informative diagnostic steps.⁶ In the present case, although arterial biopsy was not performed, a diagnosis of temporal arteritis was not considered because of the normal level of ESR, palpable pulsation of the external carotid arteries and temporal arteries and no histological findings of lingual arteritis.

Endotracheal intubation is a rapid, simple, safe and non-surgical technique that achieves all the goals of airway management. The technique protects the lungs from aspiration and permits leak-free ventilation during mechanical ventilation. Direct airway injury by an endotracheal tube should be considered as a cause of tongue necrosis.³ Although the patient gagged and coughed, no bleeding from the tongue was observed, and his tongue was not injured during laryngoscopy. Prolonged pressure to the base of the tongue by an endotracheal tube and the use of mouth packs and oral airways should also be considered as causes. However, in the present case, the position of the endotracheal tube was changed for several times everyday, and the patient did not use mouth packs or oral airways. Therefore, we considered the thrombosis of the lingual artery due to DIC as the cause for the tongue necrosis.

Although ischemic tongue necrosis by causes other than the complication of giant-cell arteritis is extremely rare, the possibility of tongue necrosis in patients with DIC should be considered by physicians.

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Figure legends

Fig. 1: Anterior and bilateral lingual ischemia with necrosis.

Fig. 2: Thrombosis of the peripheral artery (A) and vein (B). Hematoxylin and eosin staining ($\times 100$).

Fig. 1



Fig. 2

