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# Calcification following tongue necrosis induced by vasopressor use in a nonintubated patient with septic shock: a case report

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## Abstract

**Background** Tongue necrosis is a rare and relatively uncommon condition, usually caused by vasculitis, thrombosis, severe hypotension due to septic or cardiogenic shock, vasopressor use, or intubation. Following damage such as necrosis, dystrophic calcification, a type of soft tissue calcification, can occur.

Case presentation: Herein, we present a unique case of bilateral tongue necrosis in a patient with nonintubated septic shock. A 70-year-old East Asian man with no significant medical history presented to the emergency department with postprandial epigastric pain. The patient was admitted to the intensive care unit with hypotension due to septic shock and disseminated intravascular coagulation. After a short course of vasopressors, the patient developed tongue discoloration and swelling without limb ischemia. Computed tomography was performed to observe the tongue necrosis, and calcification of the tongue was found. The patient was successfully treated by wiping the area with a hexamidine-soaked gauze.

**Conclusion** Tongue necrosis remains a rare finding, and its occurrence as a complication of vasopressor use is even rarer. Therefore, even with relatively short courses of vasopressors in the intensive care unit, daily visualization of the tongue to check for discoloration, along with daily inspection and pulse checks of the limbs, can help identify vasospasms. These measures allow for prompt intervention, minimizing permanent damage and shortening the recovery time.

**Keywords** Case report, Tongue, Necrosis, Septic shock, Vasopressor

## Introduction

The tongue is a well-vascularized organ that rarely undergoes ischemic necrosis. It is supplied primarily by the lingual artery [1]. The lingual artery originates from the external carotid artery and branches into the dorsal lingual, sublingual, and deep lingual branches, forming a rich anastomosis that provides an arterial supply to the tongue and floor of the mouth. Venous drainage of the tongue was performed through the lingual vein, which subsequently drains into the internal jugular vein.

The most common etiology of tongue necrosis is vasculitis, such as giant cell arteritis (GCA) [2–4], with a few reports linked to severe hypotension, vasopressor use, and thrombosis. Vasopressors are commonly used in patients with septic/cardiogenic shock and may lead to peripheral ischemia. However, ischemic necrosis of a well-vascularized tongue is extremely rare. Therefore, we present a rare case of a patient who developed ischemic

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tongue necrosis without intubation. We present this article in accordance with the case report guidelines (CARE) reporting checklist.

## Case report

A 70-year-old East Asian male with no significant underlying medical history except for benign prostatic hyperplasia presented to the emergency department with postprandial epigastric pain. Abdomen–pelvis computed tomography (CT) revealed an enlarged mass in the liver, along with dilatation of the intrahepatic bile ducts, suggesting cholangiocarcinoma or cholangitis (Fig. 1). The patient was admitted to the general ward. Initially, the patient's vital signs were relatively stable (blood pressure of 167/99 mmHg, heart rate of 95 beats per minute, respiratory rate of 20 per minute, body temperature of 36.4 ℃, saturation of 97% in room air); however, after 24 hours, the patient suddenly exhibited hypotension (80/55 mmHg) and an increased oxygen requirement (5 L/minute). Blood tests showed decreased platelet count (45,000/μL), elevated fibrin degradation products (99.39 μg/mL), significantly elevated D-dimer (> 3,200 ng/mL), and prolonged activated partial thromboplastin time (44.7 seconds) and prothrombin time (24.0 seconds). After percutaneous transhepatic biliary drainage insertion, *Candida* grew in the drainage fluid. The patient was suspected of having septic shock and disseminated intravascular coagulation (DIC). He was transferred to the intensive care unit (ICU) and was started on norepinephrine, reaching a maximum dose of 0.15 mcg/kg/min, used over a relatively short duration of 34 hours (1.5 days). The patient was treated with a high-flow nasal cannula (flow rate of 40 L/minute and fraction of inspired oxygen of 0.6) and continuous renal replacement therapy (CRRT) for approximately 2 weeks without intubation and showed gradual improvement of the septic condition.

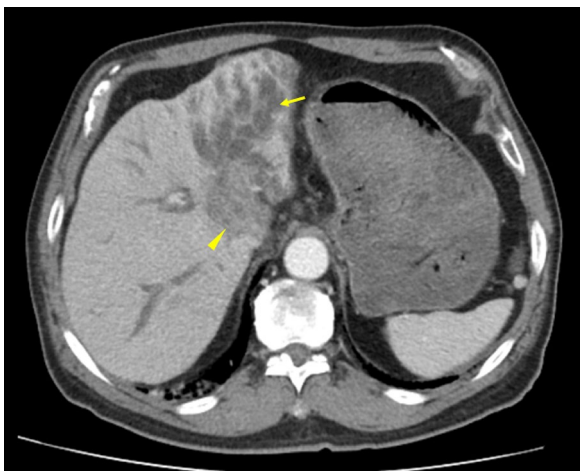

**Fig. 1** Abdomen–pelvis computed tomography of the patient. The abdomen–pelvis computed tomography revealed an enhancing mass in the central part of the liver (arrowhead) along with dilatation of intrahepatic bile ducts (arrow), suggesting cholangiocarcinoma or cholangitis

About 10 days after admission, 7 days following 1.5 days of norepinephrine use, we observed a dark discoloration with significant swelling predominantly affecting the central and posterior regions of the tongue. This condition, which covered approximately thirty percent of its area, was not apparent upon admission (Fig. 2 A, B). The patient complained of pain and discomfort in the tongue. After consulting an oral and maxillofacial surgeon, the patient's tongue was diagnosed with multiple ulcers and a necrotic surface. The treatment consisted of swabbing the area with a 0.12% chlorhexidine gluconate solution (hexamedine, Buk-wang Pharmaceutical, Seoul, South Korea)-soaked gauze twice a day and monitoring the progress. However, 1 week later, the nearly entire surface of the tongue had worsened, and the swelling had increased bilaterally (Fig. 2C, D). To investigate the potential primary causes of tongue necrosis, such as vasculitis and carotid artery stenosis, various diagnostic tests were performed, including autoimmune and lipid laboratory tests and brain computing tomography (CT) angiography. Laboratory tests showed weakly positive anticardiolipin antibody immunoglobulin M, but other tests, including antinuclear antibodies, antineutrophil cytoplasmic antibodies, and lupus anticoagulant, were all negative. A rheumatologist concluded that these findings are not indicative of rheumatoid diseases and suggested a low likelihood based on clinical symptoms, as the patient did not exhibit other vasculitis symptoms such as skin lesions, arthritis, or pain, aside from the condition of the tongue. Lipid laboratory results were unremarkable, and brain CT angiography revealed no severe atherosclerosis with well-detected lingual arteries. However, precontrast CT showed signs of tongue calcification due to ischemic changes (Fig. 3). After exclusively using 0.12% chlorhexidine gluconate solution-soaked gauze continuously, the patient's tongue began to show gradual improvement 2 weeks later. Approximately 1.5 months after ICU admission, the tongue had almost returned to its normal state (Fig. 2E, F). The patient no longer reported tongue pain or discomfort while eating.

## Discussion

Tongue necrosis is a rare occurrence. The tongue is a highly vascular organ that is generally tolerant of injury [5]. Few cases have been documented in literature, and most are typically caused by vasculitis such as GCA or polyarteritis nodosa [2–4, 6]. Some studies have described tongue necrosis caused by septic

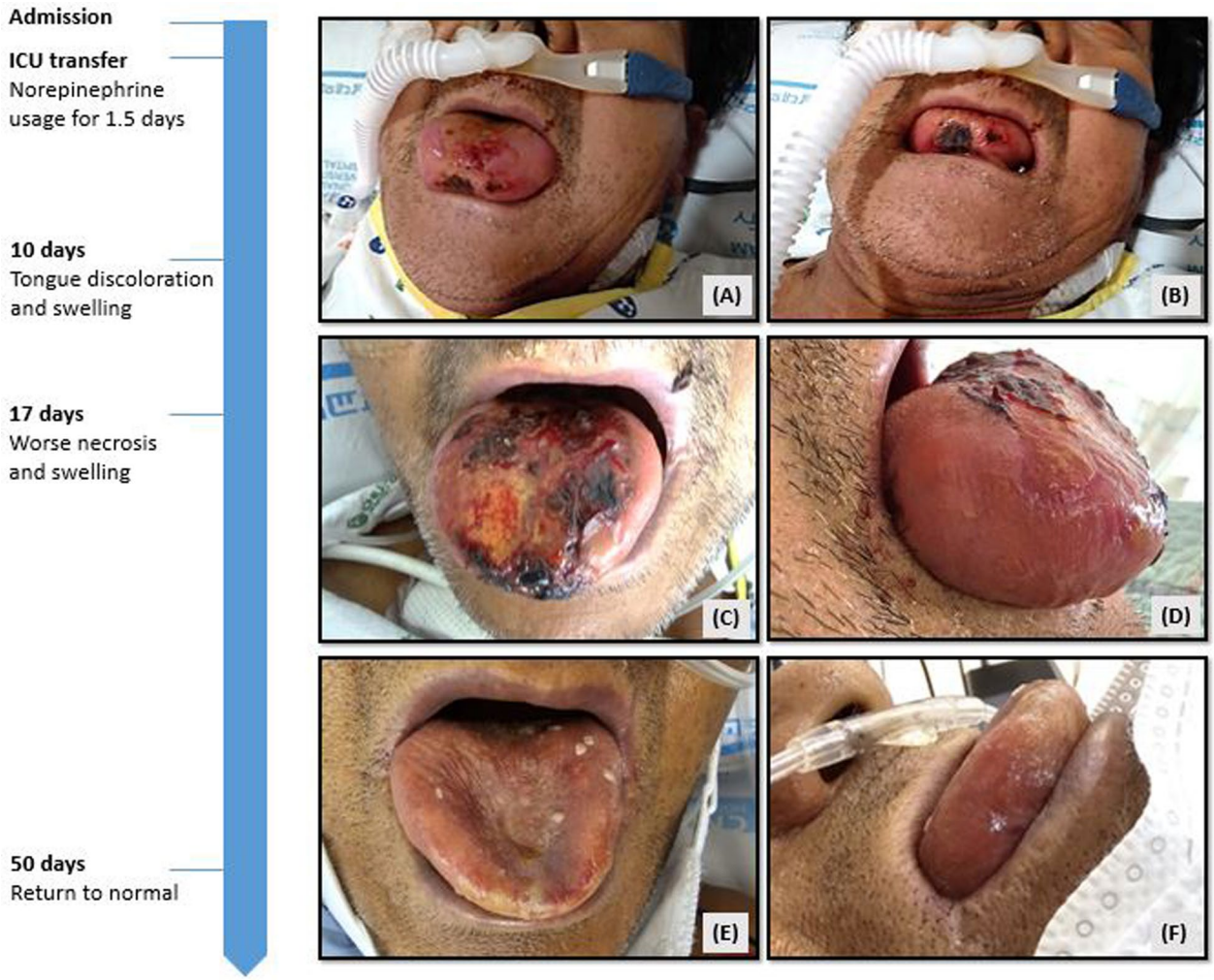


**Fig. 2** Serial images of tongue necrosis. **A**, **B** Necrotic change on tongue 10 days after admission. A darkish discoloration was observed in the center and back of the tongue, with significant swelling. **C**, **D** More severe necrotic change at 17 days. The surface of the tongue worsened, and the swelling intensified bilaterally. **E**, **F** Improvement of necrotic change of tongue at 50 days. Discoloration and swelling improved, and the patient reported no pain or discomfort

or cardiogenic shock. To date, only about ten cases of tongue necrosis due to septic shock and cardiogenic shock have been reported [7–12]. Other studies have reported other causes of tongue necrosis, such as prolonged oral intubation [13–16] and carotid artery stenosis [1, 17].

In patients with vasculitis, tongue necrosis is often unilateral and limited to the anterior half or tip of the tongue [1]. Involvement of the base or both sides of the tongue is rare. In this case, necrosis occurred in both the bilateral and posterior aspects of the tongue. Vasculitis and other rheumatic diseases were excluded based on autoimmune laboratory tests and expert assessments of clinical manifestations. Moreover, arterial stenosis or infarctions were excluded based on normal lipid laboratory test results and angiographic CT scans.

At the time of admission to the ICU, the blood pressure of the patient was low, and laboratory results indicated DIC. We initiated CRRT due to metabolic acidosis. To maintain the blood pressure, norepinephrine, a vasopressor, was administered for 1.5 days. After being transferred to the general ward, the patient continued dialysis because of kidney damage resulting from previous septic shock. Due to cancer progression, the lab data of the patient were similar to DIC condition, with low platelet counts and elevated prothrombin time and D-dimer levels. Additionally, while in the general ward, the patient experienced another infection, resulting in septic shock with low blood pressure. However, the family did not want to life-sustaining treatments anymore, and thus, vasopressors were not administered. Despite these conditions, the patient's tongue remained in an improved

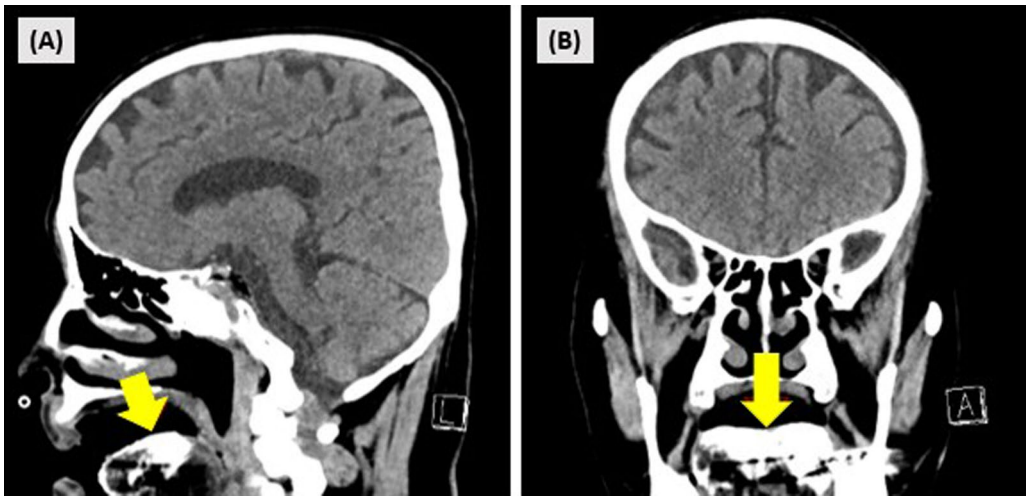


**Fig. 3** Head and neck computed tomography angiography of the patient. Calcification in the tongue (arrows) due to ischemic changes, with no steno-occlusive vessels observed in the sagittal (**A**) and coronal (**B**) views

state, clearly indicating that the changes in his tongue were caused by the short-term use of vasopressors, rather than by DIC or septic shock. Norepinephrine is structurally classified as a catecholamine, and the addition of a methyl group to amine group results in the formation of epinephrine. Norepinephrine binds to α- and β-adrenergic receptors in different tissues. In the blood vessels, it triggers vasoconstriction, which increases blood pressure [18]. This also results in decreased blood volume in dialysis patients, potentially causing poor perfusion or ischemia [19, 20].

To our knowledge, this is one of the rare reported cases of bilateral tongue necrosis in a patient with non-intubated septic shock without a significant surgical history. All previously reported cases of tongue necrosis due to cardiogenic or septic causes involved patients who underwent surgical procedures, such as coronary artery bypass or intra-aortic balloon pumps. And they were all intubated [8, 10]. In contrast, in this study, the patient had septic shock but did not undergo any surgical or medical procedures. Moreover, intubation, a possible cause of tongue necrosis, was not performed. Endotracheal intubation is associated with many oral complications, such as damage to the lips, teeth, gums, and tongue [13]. Instead, the patient used a high-flow nasal cannula. This could avoid potential damage or pressure effects associated with the endotracheal tube. In other studies, patients with septic shock who were treated with vasopressors developed not only tongue necrosis but also peripheral limb ischemia [7, 10, 12]. However, in the present case, no signs of peripheral limb ischemia or tongue necrosis were observed.

In addition, this study reported the use of CT to observe tongue necrosis and complete healing in patients with sepsis. In previous studies, CT was not used to evaluate tongue necrosis in patients with cardiogenic or septic shock. In our case, CT imaging was used to evaluate the condition of the tongue, which showed intact vessels and calcified tongues despite normal calcium and phosphorus levels.

Oral ulcers are common lesions of the oral mucosa resulting from various mechanical or chemical injuries. Bacterial contamination of these lesions can adversely affect the healing process, leading to prolonged healing times and the formation of exuberant granulation tissue [21, 22]. Chlorhexidine gluconate solution has been found to reduce the incidence, duration, and severity of many types of oral ulcers [23]. In this case, we propose that the use of chlorhexidine gluconate solution reduced bacterial contamination of the tongue ulcer and accelerated the healing process.

Dystrophic calcification is a type of soft tissue calcification that occurs in damaged or necrotic tissue when serum levels of calcium and phosphorus are normal [24, 25]. There are some reports of patients who were diagnosed with ischemic stroke and developed calcification in the brain tissue [26–29]. Moreover, calcification can be observed in the oral cavity [30], muscles [31], and subcutaneous tissues [32] after damage. Common sites of soft-tissue calcification in the intraoral area include the gingiva, tongue, lymph nodes, and cheeks. To our knowledge, this is the first study of calcified tongues after tongue necrosis. Similar to heterotopic ossification, which is the presence of bone in soft tissues where bone is not normally found, these calcifications

can range from small, clinically insignificant foci to large deposits that cause pain and limit function [33, 34]. In our patient, the ischemic damage to the tongue likely led to calcification, potentially causing pain and dysfunction.

The prognosis for the tongue in previously known cases of tongue necrosis caused by septic or cardiogenic shock has generally been poor. This case is exceptionally rare because the tongue had almost completely returned to normal. Owing to the rich blood supply to the tongue, it has been suggested that the tongue gradually recovers over time following transient ischemic damage. Despite the patient's tongue returning to normal, he was eventually diagnosed with cholangiocarcinoma and passed away due to cancer progression 3 months after admission. Consequently, a follow-up CT for tongue calcification could not be performed. However, considering the apparent normalization of tongue appearance, it is likely that CT imaging would have shown an improvement in calcification.

## Conclusion

Tongue necrosis is mainly associated with vasculitis, but it remains a rare finding, and its occurrence as a complication of vasopressor use is even rarer. However, clinicians should be aware that even with relatively short courses of vasopressors in the ICU, daily visualization of the tongue during physical examinations to check for discoloration, along with daily inspection and pulse checks of the limbs, can help identify vasospasms. These measures allow for prompt intervention, minimizing permanent damage and shortening recovery time.

### Abbreviations

| | |
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| CT | Computed tomography |
| DIC | Disseminated intravascular coagulation |
| GCA | Giant cell arteritis |
| ICU | Intensive care unit |
| CRRT | Continuous renal replacement therapy |

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### Author contributions

Conception and design: SYK, SHK, and SIL. Administrative support: SHK, HL, and SIL. Provision of study materials or patients: HL. Collection and assembly of data: SYK and SIL. Data analysis and interpretation: SYK and SHK. Manuscript writing: all authors. Final approval of manuscript: all authors.

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### Availability of data and materials

Not applicable.

## Declarations

### Ethics approval and consent to participate

The study was approved by the local ethics review board in accordance with the Declaration of Helsinki.

### Consent for publication

Written informed consent was obtained from the patient's next-of-kin for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

### Competing interests

Not applicable.

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## Publisher's Note