Endovascular Revascularization of External Carotid Artery Occlusion Causing Tongue Infarction
—Case Report—

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Abstract
A 62-year-old man with diabetes and a history of ischemic coronary disease visited the emergency department complaining of acute pain and swelling of the tongue. Physical examination found subtle swelling and pallor of the right side of the tongue, and he was initially diagnosed with glossitis. However, his symptoms were progressive, and the tongue had sustained serious tissue damage before the correct diagnosis was established. Digital subtraction angiography of the cervical vessels revealed occlusion of the right external carotid artery (ECA) and lingual artery without collateral circulation to the right side of the tongue from the contralateral ECA or ipsilateral vertebral artery (VA). Endovascular revascularization was performed to restore blood flow to the tongue using balloon angioplasty of the proximal segment of the right ECA followed by deployment of a self-expanding stent. Tongue pain subsided shortly after the procedure, and configuration of the tongue returned to normal 4 months after intervention. Tongue infarction is rare and usually associated with systemic vasculitides. Tongue infarction due to unilateral occlusion of the ECA is extremely rare because of the rich collateral circulation to the tongue from the ipsilateral VA and contralateral ECA. Atherothrombotic unilateral occlusion of the ECA should be included in the differential diagnosis of tongue infarction. Revascularization of the occluded ECA is worth attempting despite substantial tissue damage because of the viability of the tongue muscles and the minimal risk of complications in experienced hands.

Key words: endovascular revascularization, external carotid artery, lingual artery, occlusion, tongue infarction

Introduction
Tongue infarction is a rare medical condition, and is almost invariably associated with systemic vasculitides such as giant cell arteritis.13,12 Tongue infarction caused by unilateral occlusion of the external carotid artery (ECA) is extremely rare because of the rich collateral circulation to the tongue from the ipsilateral vertebral artery (VA) and contralateral ECA.14,16 We describe a case of tongue infarction secondary to atherothrombotic unilateral occlusion of the ECA. Despite delayed diagnosis and treatment, the tongue infarction was resolved by endovascular revascularization.

Case Report
A 62-year-old man visited the emergency department of our institution complaining of acute pain and swelling of the tongue. His medical history included diabetes mellitus and ischemic coronary disease for which percutaneous coronary intervention had been performed and low-dose aspirin was prescribed. On arrival, the patient was alert and oriented, and no focal neurological deficits were noted except for tenderness and paresthesia of the right side of the tongue. Tongue movement was normal. He was afebrile and his vital signs were stable, and there were no signs of cardiac arrhythmia. On physical examination, an emergency medicine physician noted subtle swelling and pallor of the right side of the tongue (Fig. 1A). Laboratory data revealed mildly elevated white blood cell count and C-reactive protein level compatible with glossitis. Furthermore, his blood glucose level exceeded 200 mg/dl, indicating that his diabetes was not controlled optimally. He was prescribed analgesics and antibiotics, and allowed to return home. However, his symptoms aggravated and he returned to the emergency department 2 days after the initial visit.

On second physical examination, swelling of the tongue had worsened (Fig. 1B), and he was admitted to the Ear, Nose and Throat Department for further evaluation. Although the right superficial temporal artery was palpable, the size was significantly diminished compared with
Fig. 1 Serial photographs of the tongue. A: Slight swelling of the right side of the tongue at the time of initial presentation. B: Marked swelling of the right side of the tongue 3 days after onset. C: Discoloration and ulceration in the right side of the tongue 6 days after onset. D: Marked improvement 3 days after intervention. E: Configuration and color of the tongue were normal 4 months after intervention.

Fig. 2 Pre-procedural diagnostic imaging studies. A: T2-weighted magnetic resonance image showing a diffuse high intensity signal in the right side of the tongue (arrowhead). B: Computed tomography scan with contrast medium of the neck showing no opacification of the right external carotid artery (ECA), but opacification of the ipsilateral internal carotid artery (ICA). C: Carotid ultrasound image showing the occluded right ECA with thrombus (arrowhead) as well as the intact ICA (filled with green).

Fig. 3 Digital subtraction angiography of the neck vessels. A: Lateral view of the right common carotid artery (CCA) showing an occluded right external carotid artery (ECA) and intact internal carotid artery (ICA). The ECA was occluded after thin antegrade flow in the proximal portion (arrowhead). B: Anteroposterior view of the right CCA. C: Anteroposterior view of the left CCA showing intact left ECA and ICA. D: Superselective angiogram of the left lingual artery showing little collateral circulation to the right side of the tongue. E: Anteroposterior view of the right vertebral artery showing little collateral flow to the tongue.

the left. Diagnostic imaging studies were performed. Magnetic resonance imaging exhibited marked change in signal intensity of the right side of the tongue (Fig. 2A), and computed tomography with contrast medium of the neck revealed occlusion of the proximal segment of the right ECA (Fig. 2B). Opacification of the distal segment of ECA was delayed, indicating that only the proximal segment was occluded. Doppler ultrasonography also confirmed occlusion of the right ECA with thrombus (Fig. 2C). Tongue infarction secondary to occlusion of the right ECA and lingual artery was suspected. He was referred to us for further evaluation and treatment.

Digital subtraction angiography of the cervical vessels revealed occlusion of the right ECA despite thin antegrade flow in the proximal segment (Fig. 3A, B). The right internal carotid artery (ICA) was only mildly stenotic (Fig. 3A, B). Although the left ECA and the lingual artery were intact, collateral flow to the right side of the tongue was unrecognizable (Fig. 3C, D). Furthermore, the right VA did not supply collateral circulation to the tongue (Fig. 3E). Endovascular revascularization of the right ECA and lingual artery was planned to restore blood flow to the tongue after obtaining informed consent from the patient. The swelling had worsened further and ulcers had developed on the right side of the tongue at the time of intervention (Fig. 1C).

Endovascular revascularization was performed on the 7th day after onset in an angiography suite under local anesthesia. After systemic heparinization, a 9-Fr guiding catheter was inserted through the transfemoral route and positioned in the right common carotid artery. Subsequently, Angioguard® (Johnson & Johnson, Langhorne, Pennsylvania, USA), a protective filter device, was placed into the right ICA, and a Transit® microcatheter (DePuy, Raynham, Massachusetts, USA) was advanced into the right ECA, which easily penetrated over the occluded segment (Fig. 4A). After confirming that deployment of the protective filter device in the ICA did not cause neurologic deficits, pre-dilation was performed using a Gateway Monorail® 3.5 mm × 20 mm balloon (Boston Scientific, Natick, Massachusetts, USA) inflated at 10 atm for 30 seconds under transcranial Doppler surveillance (Fig. 4B). Subsequently, a self-expanding stent (Precise® 6 mm × 20 mm; Johnson & Johnson) was deployed into the ECA. Because of the mildly stenotic ipsilateral ICA, care was taken so that proximal end of the stent did not protrude into the common carotid artery (CCA). The distal end of the stent was located at the level of the C3-4 disk space (Fig. 4C). The stent deployment was followed by post-dilation using a Gateway Monorail® 3.5 mm × 20 mm balloon inflated at 10 atm for 30 seconds. Post-procedural angiography...
showed good recanalization of the lingual artery and facial artery, although the orifice of the ECA remained mildly stenotic (Fig. 4C, D).

Within 24 hours of revascularization, the tongue pain had subsided. Swelling and ulceration of the tongue also improved markedly after the procedure (Fig. 1D). Clopidogrel with low-dose aspirin was prescribed, and the patient was discharged 1 week after the procedure. Four-month follow-up examination found that the configuration of his tongue had returned to normal (Fig. 1E), and imaging studies confirmed patency of the right ECA.

Discussion

Unilateral occlusion of the lingual artery rarely results in tongue infarction because of the presence of rich collateral circulation to the tongue from other branches of the ipsilateral ECA, ipsilateral VA, and contralateral lingual artery. Patients may remain asymptomatic even after bilateral ligation of the lingual arteries. Similarly, intentional unilateral obliteration of ECA rarely results in tongue infarction, even in the setting of polytrauma complicated by profuse hypotension. Tongue infarction may occur only after occlusion of the bilateral ECAs. Most patients with tongue infarction harbor underlying systemic vasculitides, such as giant cell arteritis, temporal arteritis, and Wegener's granulomatosis. The present case was unique in that atherothrombotic unilateral occlusion of the ECA resulted in tongue infarction. Only 3 previous cases were identified in the medical literature based on the internet search engines PubMed/MEDLINE and Ichushi-Web (Japan Medical Abstracts Society, Tokyo) (Table 1). Our review indicates that comorbidity such as hyperlipidemia and diabetes may be a prerequisite for development of tongue infarction after unilateral ECA occlusion. In the present case, absence of collateral circulation to the right side of the tongue together with concomitant uncontrolled diabetes was probably responsible for the infarction. The ipsilateral facial artery, which frequently anastomoses with the lingual artery, was also obliterated, and anastomosis from the contralateral lingual artery was virtually nonexistent.

Recently, endovascular revascularization, consisting of percutaneous transluminal angioplasty and stent deployment, of the stenotic ECA has been shown to be feasible and safe in patients with occluded or severely stenotic ipsilateral ICA. The procedure is intended to increase blood flow to the hemodynamically compromised brain via natural or surgical extracranial-intracranial anastomosis. However, endovascular recanalization of the occluded ECA to restore circulation to the branches of ECA is rarely reported. Although endarterectomy of the proximal ECA segment might have been another option, we chose the endovascular procedure because of the comor-

Table 1 Cases of tongue infarction caused by atherothrombotic unilateral occlusion of the external carotid artery

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Author (Year)</th>
<th>Age (yrs)/Sex</th>
<th>Co-morbidity</th>
<th>Concomitant symptoms</th>
<th>Treatment</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Dixon (1989)</td>
<td>64/M</td>
<td>hyperlipidemia</td>
<td>facial pain, ear cyanosis</td>
<td>conservative with anti-coagulation</td>
<td>improved</td>
</tr>
<tr>
<td>2</td>
<td>Clement and Nordhuus (1993)</td>
<td>49/F</td>
<td>hyperlipidemia</td>
<td>N/A</td>
<td>N/A</td>
<td>N/A</td>
</tr>
<tr>
<td>3</td>
<td>Orita et al. (2000)</td>
<td>67/M</td>
<td>ischemic coronary disease</td>
<td>transient ischemic attack</td>
<td>surgical debridement of the necrotic tissue</td>
<td>improved</td>
</tr>
<tr>
<td>4</td>
<td>Present case</td>
<td>62/M</td>
<td>diabetes, ischemic coronary disease</td>
<td>none</td>
<td>endovascular revascularization</td>
<td>improved</td>
</tr>
</tbody>
</table>

F: female, M: male, N/A: not available.
bidities of uncontrolled diabetes and ischemic coronary disease. The risk of embolic or ischemic complications during endovascular recanalization of the occluded ECA seems to be smaller compared with that for occluded ICA, which is controversial.1,17) During stent deployment, care was taken so that proximal end of the stent did not protrude into the CCA, since the ipsilateral ICA was mildly stenotic and we were concerned that the patient might develop severe ICA stenosis and so require its revascularization in the future. The same size balloon catheter (Gateway Monorail® 3.5 mm × 20 mm) was used for post-dilation for fear that the use of a larger balloon might cause vascular injury.

In the present case, accurate diagnosis was delayed because of rarity of the condition. The tongue had undergone serious tissue damage by the time the patient was referred to us. Considering his comorbidity, ischemia as a cause of tongue pain and swelling should have been suspected earlier. Nevertheless, the configuration and function of his tongue returned to normal shortly after revascularization. The muscles of the tongue have remarkable potential for regeneration,10) and we believe that revascularization of the occluded ECA may worth attempting in patients with severe ischemic damage to the tongue, because the therapeutic window of opportunity is much longer compared with patients with cerebral ischemia.

References

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