Letters to the Editor

To the Editor

Hemilingual Edema in Acute Ischemic Stroke

Unilateral tongue edema has rarely been reported in cases of acute ischemic stroke treated with tissue plasminogen activator (tPA).[1,2] We present a case of hemilingual edema in acute stroke not associated with tPA.

An 82-year old male with no prior history of stroke presented with forehead sparing right facial droop, mild weakness and ataxia of the right upper limb, dysarthria, dysphagia and significant right hemilingual edema. Electrocardiogram (EKG) showed atrial fibrillation and the international normalized ratio (INR) was 1.4. He was on warfarin, simvastatin, ramipril, metoprolol, levothyroxine, furosemide, and potassium chloride supplements. Head computed tomography (CT) showed an old lacune in the left lentiform nucleus and small hypodensity in the right internal capsule.

The tongue edema and dysphagia took two and four days, respectively, to resolve. Examination 12 days post-stroke revealed improved right facial droop, and he was slow though not ataxic with finger-nose finger testing on the right. The right hemiparesis and gait difficulties had resolved. Tongue, palate and gag were normal. A mild right Horner's syndrome was noted (not commented on previously), which was later confimed by neuro-ophthalmology. Carotid dopplers showed no significant stenosis, with antegrade flow in both vertebral arteries. Brain magnetic resonance imaging (MRI) seven months post-stroke showed diffuse atrophy and non-specific white matter changes consistent with microvascular disease. There was no evidence of acute, subacute or chronic infarct on T1, T2, FLAIR or diffusion weighted images. Brain magnetic resonance angiogram (MRA) showed normal vertebral, basilar, internal and middle carotid arteries. Echocardiogram revealed mild bi-atrial enlargement and possible inferior septal hypokinesia, but no significant valvular abnormalities.

His presentation was most consistent with a brainstem event,[3] likely from a cardioembolic source given his history of atrial fibrillation and subtherapeutic INR. Horner's syndrome ipsilateral to the limb and facial weakness is puzzling; it was not commented on initially but was likely missed.

Lingual angioedema in acute ischemic stroke treated with tPA may be bilateral or unilateral, often contralateral to the affected hemisphere, with a greater risk in strokes affecting the insula or anterior frontal cortex.[1] All but one of the cases reported by Hill et al.[1] resolved within 1-24 hours. In our case, the lingual edema resolved within 48 hours. Interestingly, five of nine patients with angioedema were on both an angiotensin converting enzyme (ACE) inhibitor and a beta-blocker,[1] as was our patient.

The mechanism of tPA associated lingual edema likely involves genetic factors such as bradykinin metabolism, and infarction location, as basal cardiac sympathetic tone was found to be maintained in the insular cortex.[1] Through tPA hydrolysis of plasminogen to plasmin, a bradykinin precursor is released, resulting in vasodilation.[1] ACE inhibitors also increase the bradykinin level and may result in orolingual and, rarely, gut edema.[4,5,6] This patient was on ramipril at the time of stroke, which may have predisposed him to angioedema. One cannot exclude that the lingual edema was simply a side effect of the ACE inhibitor. Coumadin-induced lingual hemorrhage presenting as sudden-onset angioedema has been reported[7] but our patient's INR was only 1.4 and his symptoms spontaneously resolved. His lingual edema was likely not caused by a hereditary[4,8,9] or acquired angioedema; however, complement and C1 esterase inhibitor levels were not measured. He also had no manipulations of the airway that would predispose him to an acute angioedema.[5,9]

Hemilingual edema may be a rare presenting feature of acute ischemic stroke not associated with tPA. The mechanism for this remains unclear. The combination of ACE inhibitors and beta-blockers may be predisposing factors.

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