Isolated Hypoglossal Neuropathy due to Cervical Spondylosis Mimicking Lingual Angioedema

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A woman in her 80s presented to the emergency department with a 4-month history of “tongue swelling,” mild drooling, difficulty in swallowing, and slurred speech that worsened during 1 day. Asymmetric lingual angioedema predominantly of the right side of the tongue secondary to lisinopril was suspected. The patient was treated with diphenhydramine, dexamethasone, famotidine, and tranexamic acid, then admitted to the hospital for observation. Lisinopril was discontinued after 4 years of treatment. Alternative causes were considered when symptoms persisted. Neurologic examination found left-sided tongue paresis with atrophy and flaccid dysarthria (Figure A). Magnetic resonance imaging of the brain excluded a cerebral or

FIGURE. Clinical image with corresponding neuroimaging study. A, Patient protruding tongue on command, demonstrating leftward tongue deviation and atrophy ipsilateral to the hypoglossal neuropathy (Supplemental Video). B, Coronal computed tomography of the head shows a distinctive bird-beak appearance of jugular tubercle overlying the left hypoglossal canal, which is narrowed by hypertrophic C0-C1 joint osteoarthropathy (arrows). C, Axial computed tomography of the head shows obstruction of the left hypoglossal canal (arrows).
brainstem infarction and demonstrated atrophy of the left side of the tongue. Computed tomography imaging studies of the head and neck demonstrated degenerative hypertrophic arthropathy of the left atlanto-occipital joint compromising the hypoglossal canal (Figure B, C). Electromyography confirmed neurogenic abnormalities confined to the left genioglossus muscle.

Hypertrophic cervical spondylosis of the cranio cervical junction is a cause of hypoglossal neuropathy with a predilection for elderly women.¹ The hypoglossal canal may be compromised by degenerative juxta-articular cysts, retro-odontoid fibrous pseudotumors, and osteophytes.² Management in our patient was conservative with speech therapy.² Although asymmetric angioedema of the tongue has rarely been reported,³ the absence of clinical or imaging evidence of edema and prominent tongue deviation with hemiatrophy better supported a primary neurologic cause (Supplemental Video, available online at http://www.mayoclinicproceedings.org).

ACKNOWLEDGMENTS
We acknowledge Thomas Poterucha, MD, and John C. Matulis III, DO, MPH, for interpretation of clinical data and manuscript review.

SUPPLEMENTAL ONLINE MATERIAL
Supplemental material can be found online at http://www.mayoclinicproceedings.org. Supplemental material attached to journal articles has not been edited, and the authors take responsibility for the accuracy of all data.

Potential Competing Interests: The authors report no competing interests.

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