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What is This?
A Case of Dysphonia and Cough Caused by Spontaneous Intracranial Hypotension

Mirabelle Sajisevi, MD¹, Gina Vess, MA, CCC-SLP¹, and David L. Witsell, MD, MHS¹

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Keywords
hoarseness, dysphonia, spontaneous intracranial hypotension

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Introduction
Spontaneous intracranial hypotension (SIH) occurs due to spontaneous cerebrospinal fluid (CSF) leaks, the cause of which is unknown.¹ The annual incidence is 5 per 100,000, affecting females more commonly.¹ The most common symptom is orthostatic headache.¹ Posterior neck pain, nausea, and vomiting are also frequently found.¹ Less common symptoms include facial numbness or weakness, dysgeusia, and visual field defects.¹ There have been no reports to our knowledge of dysphonia or cough as a symptom of SIH. The goal of this article is to describe a rare, previously unidentified etiology of voice change and cough. Exempt status was granted by the Duke University Health System Institutional Review Board.

Case Presentation
A 54-year-old female presented to our clinic with cough, hoarseness, and throat discomfort for 5 months. She first developed the symptoms after an episode of bronchitis. She had been evaluated by pulmonology, but inhaled corticosteroids, prednisone, and antibiotics did not significantly improve her symptoms; therefore, she was referred to our clinic for further evaluation.

Videolaryngostroboscopy was performed, revealing acute adenoiditis, fungal laryngitis, and supraglottic hyperfunction. She was treated with antibiotics and antifungals and underwent voice therapy, which initially improved her cough and voice. However, she was worse 3 months later, with hoarseness characterized by a strained, shaky voice with glottal fry. Repeat videolaryngostroboscopy showed resolution of fungal laryngitis, but twitching and involuntary spasms of the larynx were seen. Manometry and pH studies were negative for esophageal dysmotility or reflux. Laryngeal electromyography showed no evidence of neuropathy. She also had a videofluoroscopic modified barium swallow study, revealing no aspiration. She continued with voice therapy and subsequently reported headaches, neck stiffness, and dizziness 3 months later. This prompted a computed tomography (CT) guided lumbar puncture with myelogram, which showed contrast extravasation into the transforaminal regions at bilateral C7-T1 and left T11-T12 (Figure 1A). TISSEEL dural patch treatments were performed at these sites (Figure 1B). She noted immediate improvement in her headaches, neck stiffness, cough, and voice problems and continued to do well at 1-month follow-up with no shakiness in her voice or pitch instability. She then developed recurrence of headaches, neck discomfort, cough, and voice changes 5 months later after a heavy coughing episode for which she underwent repeat autologous blood and TISSEEL dural patch treatments. She again had immediate improvement of her voice and cough with continued resolution 8 months later.

Discussion
Cerebrospinal fluid leak leading to intracranial hypotension occurs as a result of small tears in the dura, which can be secondary to trauma, spinal surgery, and lumbar puncture.² Intracranial hypotension that is idiopathic is termed SIH.² Our patient demonstrated the criteria commonly used to diagnose SIH, including headaches and neck stiffness, evidence of CSF leak on CT myelography, and no history of dural puncture or spinal trauma, and her headache resolved after dural patch treatment.¹ Headache is the most common symptom, but various other presentations may occur such as cranial nerve (CN) palsies. Traction or compression of cranial nerves may result in visual nerve deficits (CN II), diplopia (CN III, IV, VI), facial numbness (CN V) or weakness (CN VII), dysgeusia (CN IX), and tinnitus (CN VIII).¹ The patient presented to our clinic with symptoms of cough and hoarseness with extensive negative work-up. The turning point in the case that led to accurate diagnosis with CT myelography occurred when the patient reported headaches and neck stiffness. Her voice and cough immediately improved after dural

¹Division of Otolaryngology–Head and Neck Surgery, Department of Surgery, Duke University School of Medicine, Durham, North Carolina, USA

Corresponding Author:
Mirabelle Sajisevi, MD, Division of Otolaryngology–Head and Neck Surgery, Department of Surgery, Duke University School of Medicine, DUMC 3805, Durham, NC 27710, USA.
Email: mirabelle.sajisevi@duke.edu
patch, which is the mainstay of treatment. Other options are conservative management with caffeine, hydration, or bed rest, whereas surgery is reserved for when the previous methods fail. She did develop recurrence of her symptoms, which can be seen in 10% of patients. Her cough and voice complaints resolved after repeat dural patch treatment. Rebound intracranial hypertension may occur after successful treatment.

Conclusion
Spontaneous intracranial hypotension has the potential for widely varying presentations. Voice changes and cough are frequently encountered by otolaryngologists, with gastroesophageal reflux, allergies, or asthma being the most common causes. In cases refractory to medical management, further symptoms such as headaches and neck stiffness should be sought out to assess for the possibility of SIH. If present, a CT myelogram may be diagnostic.

Author Contributions
Mirabelle Sajisevi, acquisition and interpretation of data, drafting and revising the article, final approval; Gina Vess, acquisition of data, revising and final approval of article; David L. Witsell, conception, acquisition, interpretation of data, revising and final approval of data.

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