Pearls & Oy-sters: Angioplasty and Stenting as New Treatment Method for Cough Headache With Stenotic Internal Jugular Vein: Case Report With 12-Month Follow-up

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We appreciate the consent from the patient/family, and the supports from all physicians and nurses involved in the care of this patient.

Ethics Approval and Consent to Participate
This study was performed with approval from the institutional ethics committee of the Chinese PLA General Hospital (NO: S2019-359-02). The patient and the authorized representative were explicitly informed, and have signed written informed consent of the angioplasty and stenting.
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**Disclosure**

The authors report no disclosures relevant to the manuscript.
PEARLS

- Drainage dysfunction of stenotic internal jugular veins (IJVs) is considered as one etiology of cough headache as a result of fluctuation and imbalance of intracranial pressure.

- Angioplasty and stenting procedure has shown to be a new effective method for treating cough headache caused by severe stenosis of IJVs.

OY-STERS

- It is inaccurate to determine the etiology of cough headache by a singular diagnostic imaging modality, and so emphasizing the need to use multimodal imaging modalities.

CASE REPORT

A 39-year-old male presented with symptoms including recurrent headaches lasting for several hours over the past three years, moderate to severe in intensity, localized on bilateral occipital and temporal areas, and sometimes accompanied by nausea, photophobia, and phonophobia, while no cranial autonomic symptoms. The headache was primarily provoked by sudden body postural changes and partially alleviated by resting, and gradually appeared during coughing, sneezing, and bending down, but was not triggered by sustained physical exercise. The neurological examination was unremarkable and the lumbar opening pressure was 200 mmH\textsubscript{2}O at rest but >350 mmH\textsubscript{2}O after Valsalva maneuver (VM). Magnetic resonance imaging (MRI) scan showed Chiari malformation type I with cerebellar tonsil hernia, with normal enhancement imaging (Figure1 (A)). Magnetic resonance angiography (MRA) and magnetic resonance venous (MRV) imaging revealed no abnormalities. The patient was identified as a cough headache case.

The further color Doppler ultrasound showed normal bilateral IJVs at normal breathing, while bilateral retrograde venous flow at VM (768 ms on the right versus
312 ms on the left). Cerebral vascular angiography located stenoses of both IJVs. Then the patient was recommended indomethacin (75 mg twice a day) to relieve pain and then dabigatran (150 mg twice a day) to prevent thrombosis, but symptoms did not relieve during the 4-month follow-up. Following the third edition of the International Classification of Headache Disorders (ICHD-3), the patient was diagnosed as secondary cough headache rather than primary cough headache based on both his structural cranial abnormalities and clinical features including: age<50 years, primarily provoked by sudden body postural changes rather than coughing, lasting several hours which is much longer than 1 minute, and no response to indomethacin.¹

For this case, surgical treatment of Chiari malformation type I was not suggested by neurosurgeons with the absence of indication. Therefore, we targeted at IJVs stenoses related cough headache by performing angioplasty and stenting as a new treatment approach.

The patient received dual antiplatelet therapy (aspirin 100 mg daily and clopidogrel 75 mg daily) for four days before the procedure. The thromboelastography (TEG) mapping test for antiplatelet drugs was recommended, and the inhibition rates of arachidonic acid and adenosine diphosphate were higher than 50% and 40% respectively.

The angiogram revealed severe stenoses of the bilateral IJVs in the venous phase and slow outflow of the bilateral transverse sinus (Figure 1 (B-D)). Following it, an 8F guiding catheter was inserted to the IJV. A Renegade microcatheter was manually inserted across the stenotic segment. A pressure transducer connected with the Renegade microcatheter was used for functional assessment of IJVs stenoses. The pressure gradient of bilateral IJVs was measured to be both 9 mmHg (distal and proximal pressure of stenosis of the left and right IJVs were 14 and 5 mmHg, 13 and 4 mmHg, respectively).
A 0.014" (300 cm) Transend microwire was manipulated in the IJV. Following balloon inflation, two Precise stents were deployed in the bilateral stenotic IJVs. Angiography showed mild residual postprocedural stenoses of bilateral IJVs, and the VasoCT scan revealed full expansion of the stents (Figure1 (E, F)). Clopidogrel and aspirin were maintained for six months after the operation. During a 12-month follow-up, the headache got mostly relieved from coughing, sneezing, and bending down, but it could appear when bowing his head.

**DISCUSSION**

Chiari malformation type I is the most common cause of secondary cough headache,² which manifests pressure difference between the ventricles and the lumbar subarachnoidal space after a VM.³ IJVs stenosis is also a factor to alter intracranial pressure caused by drainage dysfunction, so it is speculated as another underlying etiology of cough headache.⁴ ⁵ Our study reports a cough headache patient diagnosed with pressure gradient across the IJVs stenosis. This case leads us to investigate the association between cough headache and IJVs stenosis. More importantly, we performed angioplasty and stenting as a new approach to treat IJVs stenosis-induced cough headache with satisfactory outcomes.

This study reveals the importance and necessity of multimodal imaging to diagnose cough headache patients, especially for those with additional headache triggers, higher pain intensities, and diverse headache duration (as observed in the present case).³ For this case, we observed normal MRV and right internal jugular reflux, while the opposite fact from angiography. Because the intra- and extra-cranial venous system is complex with variability between individuals and is often asymmetric, Dolic et al. regarded it almost impossible to determine the relevance of a single structural or hemodynamic venous abnormality and thus emphasized the need to use multimodal imaging modalities.⁶

The diagnostic reasoning of cough headache was based on ICHD-3. Though the
patient has been diagnosed with secondary cough headache, other possible causes of headache were systematically assessed but were eventually precluded.\textsuperscript{7} ICHD-3 diagnosis 6.6 “Headache attributed to cranial venous disorder” could be ruled out because the patient did not present with cerebral venous thrombosis. Moreover, this patient did not fulfill the diagnostic criteria of ICHD-3 diagnosis 7.7 “Headache attributed to Chiari malformation type I” for the following reasons. First, there is no evidence of a clear relation between headache development and Chiari malformation progression, or no surgical treatment for the diagnosis. Second, headache episodes are also not associated with any symptoms and/or clinical signs of brainstem, cerebellar, lower cranial nerve and/or cervical spinal cord dysfunction which do not satisfy the criteria in the ICHD-3.\textsuperscript{7} However, we could not completely exclude this diagnosis because the headache did not completely disappear after treating the stenotic internal jugular vein.

Angioplasty and stenting have been used to treat stenotic IJV since Ryu RK et al. used IJV stent for treating traumatic occlusion in 1997.\textsuperscript{8} Afterward, several IJV stents were performed to treat chronic cerebrospinal venous insufficiency that might induce multiple sclerosis, tinnitus, visual impairment, dizziness, sleep disturbance, and neck discomfort or pain.\textsuperscript{9} In recent years, angioplasty and stenting toward IJV are rarely used because of the complex IJV variability among different individuals. However, a relatively low rate of restenosis and complication have proved stenting as a safe operation.\textsuperscript{10} Donnet et al. performed craniocervical MRV and found stenosis of venous sinus or IJV in 5/7 patients in the cough headache group,\textsuperscript{11} while they didn't perform any surgical intervention for the stenosis. Our previous study has already confirmed that stent implantation or balloon dilatation of cerebral venous sinus can decrease ICP and improve symptoms immediately.\textsuperscript{10} We herein further extended the treatment to IJVs and investigated the efficacy of using angioplasty and stenting to treat the cough headache patient with severe stenotic IJVs.

In this study, we performed stenting to treat cough headache for the first time given
the following conditions. First, the symptoms of cough headache did not improve from the conservative treatment by indomethacin and dabigatran for consecutive 4 months. Second, the pressure difference between the proximal and distal narrow regions of the bilateral IJVs were both >8 mmHg, falling into the range which we usually chose stenting treatment for intracranial venous sinus. Third, the enlarged vertebral venous plexus was present (disappeared after the IJV stent implantation). The rationale to treat this cough headache patient by IJVs stenting has been supported by the final clinical outcomes. Despite the in-depth investigation of stenosis induced cough headache and the new while effective treatment reported by this study, it still had some limitations. Firstly, this is a single case association between cough headache and IJVs stenting which needs to be further confirmed by future study. Moreover, Chiari malformation type I was not treated, so its possibility as a symptomatic cause of cough headache could not be precluded.
## Appendix 1. Authors

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References


Figure 1 Bilateral IJVs stenoses and stenting treatment

A 39-year-old man with bilateral IJVs stenoses and Chiari malformation type I. (A) Sagittal T1-weighted MRI image reveals cerebellar tonsillar herniation (B) Angiography discloses severe stenoses in bilateral IJVs and enlarged vertebral venous plexus (white arrows). (C) Venous angiography reveals severe stenosis at the right IJV (white arrow). (D) The stenotic left IJV shows (white arrow) and enlarged vertebral venous plexus. (E) The image demonstrates mild post-procedural residual stenosis.
and the vertebral vein plexus disappeared after stenting (white arrows). (F) VasoCT scan shows the morphology of the stents after placement.
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