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Mouth-Opening-Induced Vertigo and Downbeat Nystagmus

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Dear Editor,

Episodic vertigo and downbeat nystagmus can be observed in rotational vertebral artery syndrome, Ménière's disease (MD), episodic ataxia, and Chiari malformation.^{1,2} Here we describe a patient who experienced paroxysmal vertigo and downbeat nystagmus during mouth opening.

A 37-year-old male with hypothyroidism presented with recurrent vertigo while eating and yawning for 2 months. He had been diagnosed elsewhere 6 years previously with probable MD due to recurrent vertigo and fluctuating tinnitus in the left ear.

A neurological examination showed subtle right-beating nystagmus without visual fixation. Video head impulse tests revealed decreased gain of the vestibulo-ocular reflex (VOR) in the left horizontal and posterior canals. Downbeat nystagmus was observed 10 s after the patient opened his mouth wide (Fig. 1A, Supplementary Video 1 in the online-only Data Supplement), was maintained during mouth opening, and ceased immediately after mouth closure. Downbeat nystagmus was also observed during neck extension (Fig. 1B, Supplementary Video 1 in the online-only Data Supplement). He denied headache, tinnitus, ear fullness, and hearing impairment during mouth opening or neck extension. Nystagmus was not evoked by applying vibratory stimuli, during horizontal head shaking, when turning the head in either direction while sitting, or during mastication, Valsalva maneuvers, or swallowing.

Bithermal caloric tests showed a canal paresis of 84% in the left ear. The rotatory chair test documented decreased gain of the VOR without asymmetry or phase leads. The results for cervical vestibular-evoked myogenic potentials and pure-tone audiometry were normal. A neurological examination also revealed no limb ataxia, motor weakness, or sensory changes. Brain magnetic resonance imaging revealed no discernible abnormalities except basilar artery fenestration on angiography (Fig. 1C and D). Computerized tomography of the cervical spine showed loss of the lordotic curvature (Fig. 1E).

Our patient exhibited downbeat nystagmus exclusively during mouth opening and neck extension. This may be explained by a transient decrease in blood flow to the inferior cerebellum due to vertebral artery compression during mouth opening. Downbeat nystagmus could also be ascribed to pre-existing MD in our patient. The pressure in the middle ear during mouth opening or neck extension may have played a role in his MD attacks. Indeed, downbeat nystagmus can be observed during MD attacks due to asymmetry in the vertical VOR or saccular dysfunction.² However, torsional components are mostly accompanied by downbeat nystagmus from MD, and our patient also denied any associated auditory symptoms during or before the development of downbeat nystagmus. In addition, disappearance of the downbeat nystagmus immediately after mouth closure contrasts with the typical clinical picture of an MD attack. Moreover, the Eustachian tubes are mostly collapsed during mouth opening or neck extension, and yet they open transiently during swallowing and in the presence of a positive pressure. Furthermore, our patient also did not develop downbeat nystagmus during the Valsalva maneuver.

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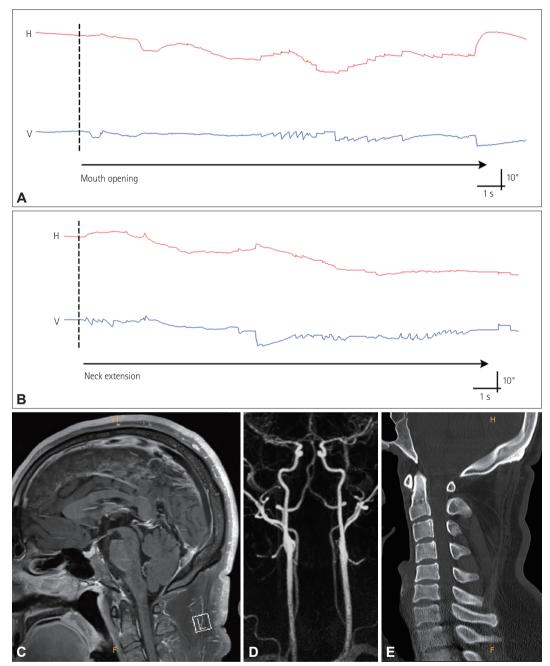


Fig. 1. Oculography, magnetic resonance imaging, and computerized tomography of the cervical spine. Downbeat nystagmus evoked by mouth opening (A) and neck extension (B) with a 10 s latency. Esotropia is also observed. C: Gadolinium-enhanced T1-weighted image showing unremarkable findings. D: Magnetic resonance angiography showing basilar artery fenestration. E: Computed tomography showing straightening of the cervical spine. H, horizontal position of the left eye; V, vertical position of the left eye.

In rare cases, patients can experience vertigo related to eating. It is therefore important for a patient to recount the circumstances in detail because vertigo and nystagmus can occur at any stages of mastication,³ swallowing,⁴ or digestion.⁵ In particular, mastication can also give rise to nystagmus in MD due to trigeminal modulation of the vestibular system.³ In such instances the nystagmus gradually increases during mastication and then decreases slowly after ceasing mastication. In

contrast to these situations, the intensity of the downbeat nystagmus in our patient peaked approximately 10 s after mouth opening and disappeared immediately when the mouth was closed. This temporal profile may be explained by a transient decrease in blood flow to the inferior cerebellum due to vertebral artery compression during mouth opening, rather than to trigeminovestibular connections causing mastication-induced nystagmus.⁶



Downbeat nystagmus was induced during both neck extension and mouth opening with similar latencies. Given that the head posture changes posteriorly at maximal jaw depression,7 we speculate that mouth opening reversibly occluded the vertebral artery in our patient (Supplementary Fig. 1 in the online-only Data Supplement). Indeed, neck extension is an integral part of complete mouth opening (due to trigeminocervical coupling),8,9 and normal conscious subjects usually extend the neck by 26° during full mouth opening.8,10 This neural connection may avoid compression of the soft tissue behind the mandible during full mouth opening. In addition, the muscles responsible for mouth opening (lateral pterygoids, digastrics, geniohyoids, and mylohyoids) may function better when the neck is in extended position compared with a neutral position.¹⁰ Straightening of the cervical spine can compromise the hemodynamics of the vertebral artery during neck extension, which may also contribute to downbeat nystagmus.11 Indeed, both vertebral arteries can be simultaneously compressed during neck extension without any stenosis or hypoplasia.¹² However, digital subtraction angiography and sonographic monitoring were unfortunately not available to confirm our hypothesis. The rarity of this peculiar phenomenon also needs further investigation.

In conclusion, the present case highlights the importance of detailed history-taking and neurotological evaluation dedicated to the provoking condition in patients presenting with episodic vertigo.

Supplementary Video Legend

Video 1. Predominant downbeat nystagmus developed approximately 10 s after mouth opening and neck extension, with associated esotropia. RH, horizontal position of the right eye; RV, vertical position of the right eye.

Supplementary Materials

The online-only Data Supplement is available with this article at https://doi.org/10.3988/jcn.2021.17.4.607.

Ethics Statement

This study was approved with a waiver of informed consent by the institutional review board of Soonchunhyang University Bucheon Hospital.

Availability of Data and Material

All data generated or analyzed during the study are included in this published article (and its supplementary information files).

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Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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REFERENCES

- 1. Franz B, Anderson C. The potential role of joint injury and eustachian tube dysfunction in the genesis of secondary Ménière's disease. Int Tinnitus J 2007;13:132-137.
- 2. Lee SU, Kim HJ, Choi JY, Kim JS. Ictal downbeat nystagmus in Ménière disease: a cross-sectional study. Neurology 2020;95:e2409-e2417.
- 3. Park SH, Kim HJ, Kim JS, Koo JW, Oh SW, Kim DU, et al. Mastication-induced vertigo and nystagmus. J Neurol 2014;261:480-489.
- 4. Lee SU, Park JH, Kim HJ, Deriu F, Kim JS. Swallowing-induced vertigo and downbeat nystagmus. J Neurol 2014;261:2457-2459.
- 5. Luciano GL, Brennan MJ, Rothberg MB. Postprandial hypotension. Am J Med 2010;123:281.e1-281.e6.
- 6. Choi KD, Choi JH, Kim JS, Kim HJ, Kim MJ, Lee TH, et al. Rotational vertebral artery occlusion: mechanisms and long-term outcome. Stroke 2013;44:1817-1824.
- 7. Dugailly PM, Salgado C, Rooze M, Feipel V. Cervical spine motions during mandible depression. Comput Methods Biomech Biomed Engin 2005;8(Suppl 1):85-86.
- 8. Eriksson PO, Zafar H, Nordh E. Concomitant mandibular and headneck movements during jaw opening-closing in man. J Oral Rehabil
- 9. Marfurt CF, Rajchert DM. Trigeminal primary afferent projections to "non-trigeminal" areas of the rat central nervous system. J Comp Neurol 1991;303:489-511.
- 10. Calder I, Picard J, Chapman M, O'Sullivan C, Crockard HA. Mouth opening: a new angle. Anesthesiology 2003;99:799-801.
- 11. Bulut MD, Alpayci M, Şenköy E, Bora A, Yazmalar L, Yavuz A, et al. Decreased vertebral artery hemodynamics in patients with loss of cervical lordosis. Med Sci Monit 2016;22:495-500.
- 12. Yun SY, Lee JY, Kwon EJ, Jung C, Yang X, Kim JS. Compression of both vertebral arteries during neck extension: a new type of vertebral artery compression syndrome. J Neurol 2020;267:276-278.