Case Report/รายงานผู้ป่วย

Posterior canal-type ocular tilt reaction secondary to vigorous sneeze: A case-report

สิรีธร คูระทอง, พ.บ.

บทคัดย่อ

บทความนี้กล่าวถึงผู้ป่วยที่เกิดภาวะภาพซ้อนทันทีหลังการจามอย่างรุนแรง ตรวจตาพบมี hypertropia ในตาขวา และ extorsion ในตาซ้ายร่วมกับมีศีรษะเอียงไปทางซ้าย ซึ่งเข้าได้กับภาวะ posterior canal-type ocular tilt reaction (OTR) โดยที่ไม่มีอาการผิดปกติอื่นทางประสาทวิทยา โดยสาเหตุเกิดจากเลือดออกในสมองส่วน midbrain ซึ่งอาการแสดงดังกล่าวร่วมกับภาวะความผิดปกติในสมองที่เกิดขึ้นนี้ ไม่เคยพบมีรายงานมาก่อน **จักษุเวชสาร 2017; กรกฎาคม-ธันวาคม 31(2): 73-79.**

คำสำคัญ: ocular tilt reaction, skew deviation, midbrain hematoma, sneeze

ผู้นิพนธ์ทั้งหมดไม่มีส่วนเกี่ยวข้องหรือผลประโยชน์ใดๆ กับผลิตภัณฑ์ที่ได้กล่าวอ้างถึงในงานวิจัยนี้

ภาควิชาจักษุวิทยา คณะแพทยศาสตร์ วชิรพยาบาล มหาวิทยาลัยนวมินทราธิราช กรุงเทพมหานคร

Case Report/รายงานผู้ป่วย

Posterior canal-type ocular tilt reaction secondary to vigorous sneeze: A case-report

Sireedhorn Kurathong, M.D.

Abstract

The author presents an interesting patient who developed diplopia immediately after a vigorous sneeze. Eye examination showed right eye hypertropia and left eye extorsion with left head tilt which characterized posterior canal-type ocular tilt reaction (OTR) that caused from hematoma in the midbrain. Extensive clinical review showed no previous report of such case presentation. **Thai J Ophthalmol 2017; July-December 31(2): 73-79.**

Keyword: ocular tilt reaction, skew deviation, midbrain hematoma, sneeze

No Author has a financial or proprietary interest in material or method mentioned

Department of Ophthalmology, Faculty of Medicine, Vajira Hospital, Navamindradhiraj University. Bangkok

Background

The primary functions of the vestibulo-ocular system are to maintain eye position and stabilize fixation during head movements. In the labyrinth of the inner ear, the semi-circular canals (which include anterior, posterior and lateral canals) sense angular acceleration while the otoliths (saccules and utricles) sense linear acceleration of the head in space[1]. The vestibulo-ocular pathways and graviceptive pathways begin peripherally with sensory organs in the labyrinth of the inner ear and project to the ipsilateral vestibulocochlear nucleus (CN VIII) at the pontomedullary junction via the vestibular portion of CN VIII. This pathway then decussates to the contralateral side at the level of pons to ascend the brainstem in the medial longitudinal fasciculus (MLF) to the supranuclear centers for vertical-torsional eye movements in the rostral midbrain. The rostral interstitial nucleus of the medial longitudinal fasciculus (riMLF) contains the excitatory burst neurons that generate vertical and torsional saccades[2]. Injury to this pathway causes skew deviation, and accompanied by ocular torsion and head tilt, is the result of ocular tilt reaction (OTR)[3].

Case presentation

A 47-year-old man with no known underlying disease, except a history of heavy alcohol drinking for more than 10 years, presented to the eye clinic with a history of sudden onset of diplopia after a vigorous sneeze. He did not have any other neurological abnormalities such as ataxia, loss of sensation or weakness in any parts of the body.

Initial examination revealed his best corrected visual acuity to be 20/25, 20/20. No abnormalities were detected in the fundi. Slit lamp examination revealed subconjunctival hemorrhage in the inferior bulbar conjunctiva of the right eye. Both pupils were 3 mm in diameter and reacted normally to direct light and near stimulus. Ocular motility detected hypertropia of the right eye in all gaze positions (Figure 1). Vestibulo-ocular reflex (VOR) detected by doll's head maneuver was intact. His head tilted to the left and he was unable to correct this tilt

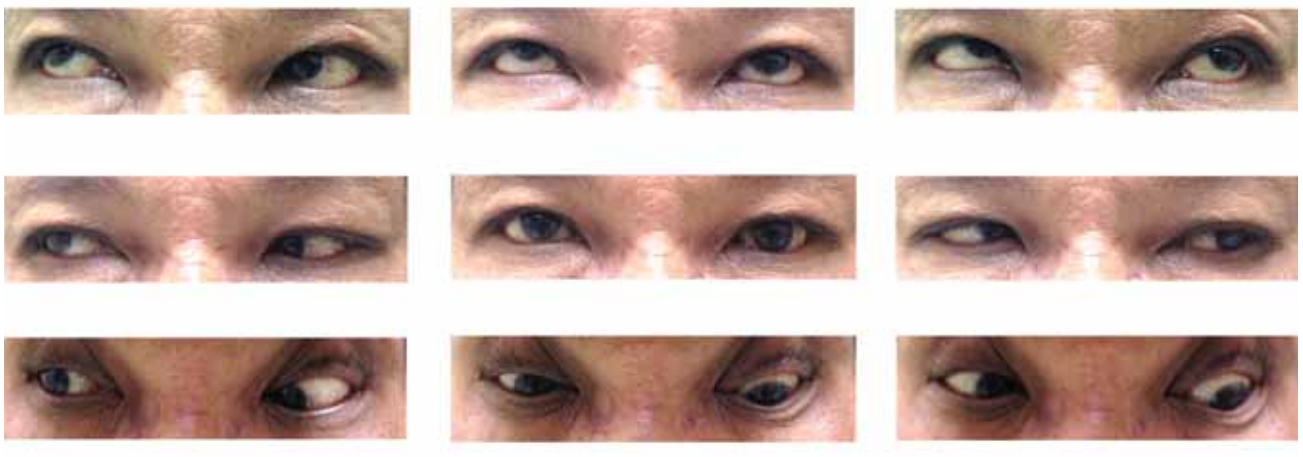

Figure 1 *Hypertropia of the right eye in all gaze positions.*

voluntarily. In the primary position, his eye position was 5 prism diopters exotropia and 10 prism diopters of right hypertropia as detected by prism and cover testing. Double Maddox rod testing demonstrated 5 degrees of left excyclotorsion.

Neurologic examination (mental status, sensation, motor, coordination, gait and deep tendon reflex) did not show any abnormal finding. Other physical examination showed spider nevi and palmar erythema.

Brain CT showed a 0.7-centimeter hematoma at the right paramedian of the rostral midbrain (Figure 2).

Bio-chemical blood examination showed thrombocytopenia with prolonged PT, PTT and abnormal liver function tests compatible with history of prolonged alcohol drinking.

The patient was referred to a hepatologist

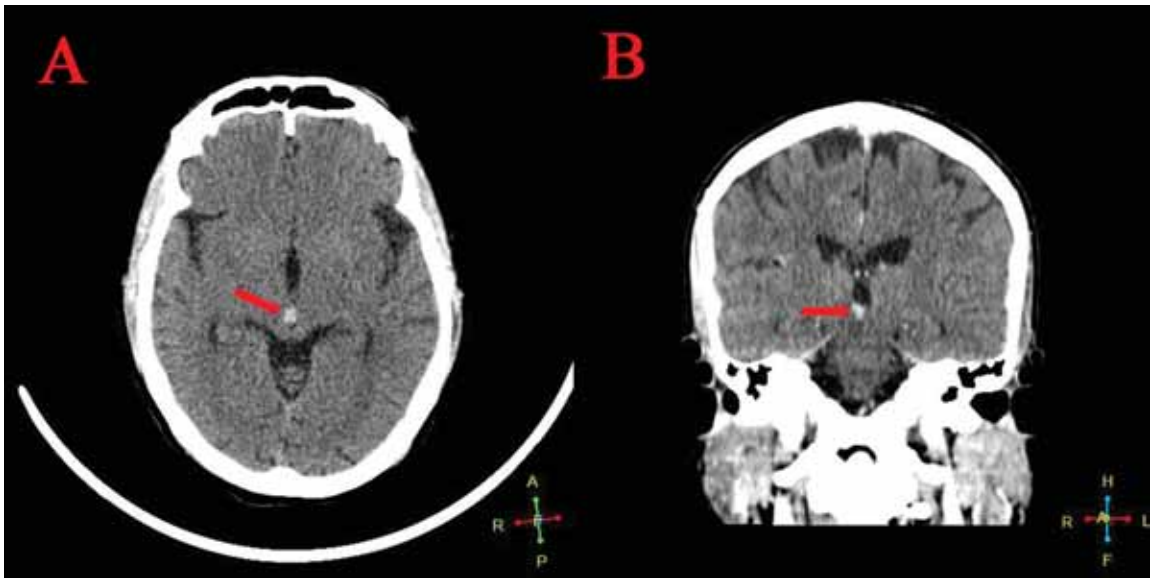

Figure 2 *Computerized tomography demonstrated an increased small-signal density (arrows in A and B) located in the right side of paramedian midbrain. A: axial section. B: coronal section.*

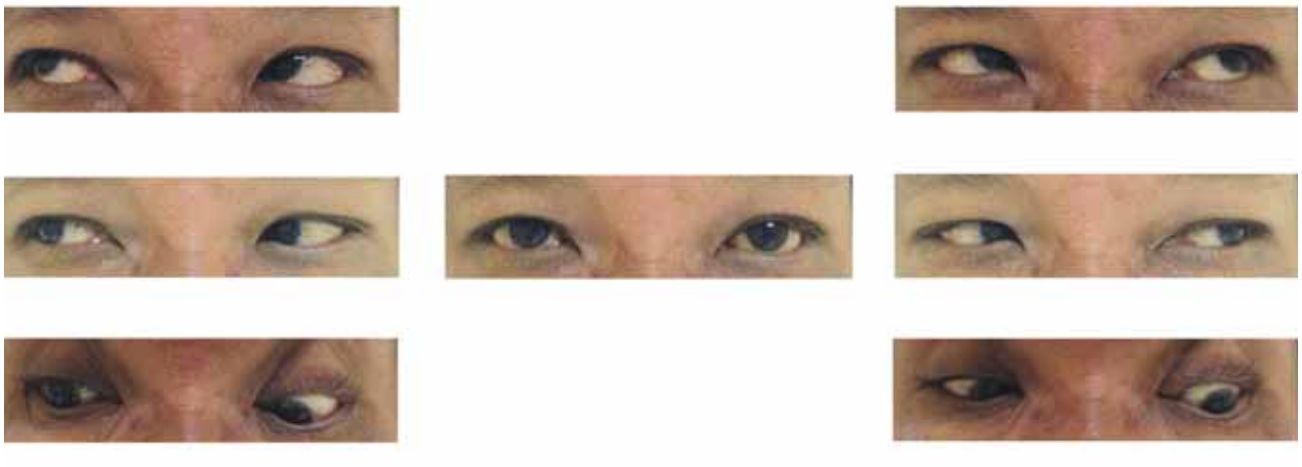

Figure 3 *Improvement of downgaze of the right eye and decreased right hypertropia*

for further management and platelet concentrations were given.

He gradually improved and came back one month later to the eye clinic. The examination revealed improvement in hypertropia and downgaze palsy of the right eye (Figure 3). He was lost to follow up after the last visit.

Discussion

Typically, the ocular torsion in skew deviation is conjugate in both eyes for most lesion locations (i.e. the hypotropic eye excyclotorts and the hypertropic eye intorts). However, if the lesion affected only input from fibers of the anterior (AC) or posterior (PC) semicircular canal to the extraocular eye muscles, monocular OTR may develop[4].

Each posterior semicircular canal provides excitatory input to the ipsilateral superior oblique and the contralateral inferior rectus while inhibiting the ipsilateral inferior oblique and the contralateral superior rectus muscle (Figure 4)[1]. As in this patient, a hematoma at the right midbrain can cause injury of the ascending pathway from the left posterior semicircular canal (crossing the midline at the pontine level) to the left superior oblique (SO) and the right inferior rectus (IR) muscle, moreover the inhibitory input to the left inferior oblique and right superior rectus can be suppressed and result in excyclotorsion of the left eye and hypertropia of the right eye.

Other sign of asymmetric injury to otolithic pathways either anterior or posterior canals is incomitant skew deviation that presents with greater

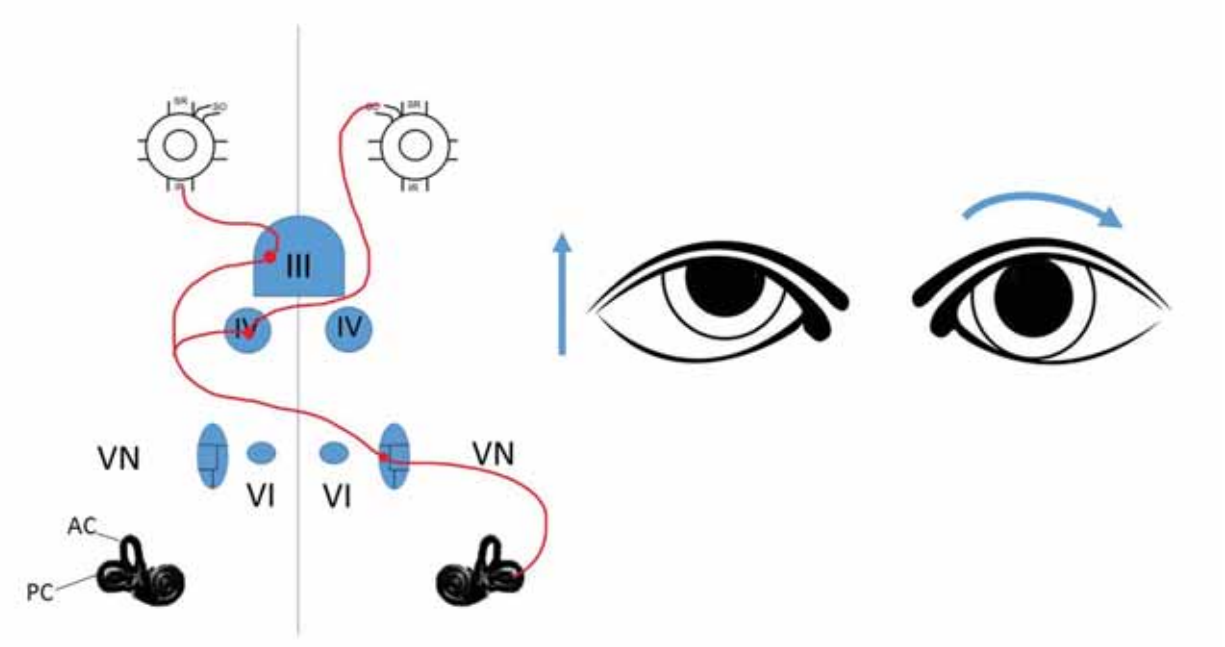

Figure 4 *Diagram showing posterior canal-type skew deviation that can result from selective injury to pathways that begin from the posterior semicircular canal to the contralateral midbrain. (AC: anterior semicircular canal, PC: posterior semicircular canal, VN: vestibular nucleus, III: oculomotor nucleus, IV: trochlear nucleus and VI: abducens nucleus)*

hypertropia in one horizontal field of gaze and minimal in the opposite field of gaze[1]. Like in this patient, the lesion in left posterior canal pathways would inhibit the left superior oblique and right inferior rectus muscles. Because both depressors have a strong vertical action in right gaze, there would be minimal vertical deviation in this position of gaze. In left gaze, however, the left superior oblique has minimal vertical action while the right inferior rectus muscle is still the major depressor. Thus, inhibiting both muscles will result in a right hypertropia in left gaze greater than right gaze (Figure 1).

Posterior canal-type ocular tilt reaction has been previously reported in one case, but unlike the present case-report, it was superimposed by vertical gaze palsy from periaqueductal syndrome and it was caused by unilateral rostral midbrain hemorrhage[5].

Paramedian midbrain contains many ascending fiber tracts, cranial nerve fiber nuclei and the reticular formation nuclei (Figure 5). Lesions in this area usually result in various neurological abnormalities such as paramedian midbrain syndrome (Benedict syndrome)[6]. Most cases of disorder in this area result from infarction, the brain lesion is usually extensive according to vascular distribution; but in the present case-report the primary lesion is a hematoma secondary to thrombocytopenia. There are not many reports of midbrain hematoma; lesion in the midbrain with only ocular abnormalities is also not common[7,8].

A vigorous sneeze can cause many ocular complications such as retrobulbar hematoma[9], orbital fracture[10], periocular ecchymosis[11], orbital emphysema[12-14] and there was a report of secondary

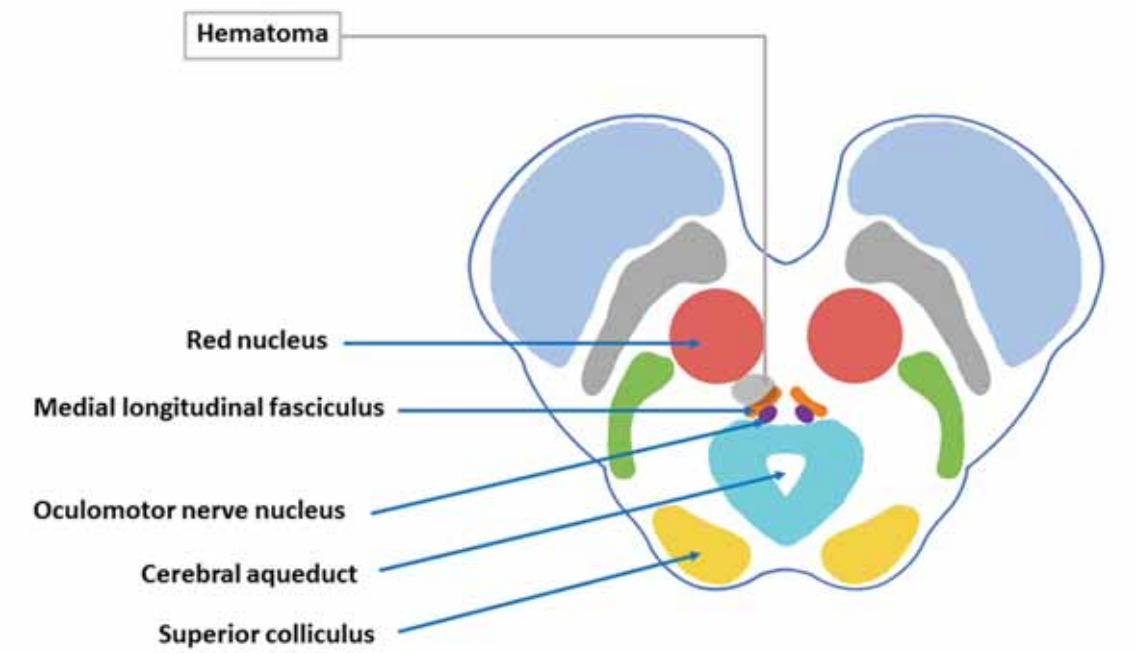

Figure 5 *Cross section at superior colliculus level with presumed site of lesion.*

subarachnoid hemorrhage[15]. We have not found any previous report that resulted from intracerebral hemorrhage after sneeze and resulted in very uncommon eye disorder without any other neurological findings.

In the present case-report, it was due to hematoma at paramedian midbrain which involved the ascending pathway of the posterior canal while sparing other cranial nerve fiber nuclei and the reticular formation nuclei. Extensive clinical review showed no previous report of such case presentation.

Acknowledgement

The author would like to thank Dr. Masuma Vudhivanich from The Radiology Department, Vajira Hospital for her assistance in the brain imaging review.

References

1. Brodsky MC, Donahue SP, Vaphiades M, Brandt T. Skew deviation revisited. Surv Ophthalmol 2006;51(2):105-28.
2. Brandt T DM. Skew deviation with ocular torsion: a vestibular brainstem sign of topographic diagnostic value. Ann Neurol 1993;33:526-34.
3. Wong AM. Understanding skew deviation and a new clinical test to differentiate it from trochlear nerve palsy. J AAPOS 2010;14(1):61-7.
4. Dieterich M, Brandt T, Ann NY. The bilateral central vestibular system: its pathways, functions, and disorders. Acad Sci 2015;1343:10.
5. Kazuya Nokura, Toshihiko Ozeki, Hiroko Yamamoto, Hiroshi Koga, Yoshiaki Shimada, Masayuki Horiguchi. Posterior canal-type ocular tilt reaction caused by unilateral rostral midbrain hemorrhage. Neuro-Ophthalmology 2004;28(5-6): 231-6.
6. Ruchalski K, Hathout GM. A medley of midbrain maladies: a brief review of midbrain anatomy and syndromology for radiologists. Radiol Res Pract 2012; 2012:258524. doi:10.1155/2012/258524.
7. El Ouali O, Messouak O, Belahsen MF. Midbrain hematoma presenting with isolated bilateral palsy of the third cranial nerve in a Moroccan man: a case report. Journal of Medical Case Reports 2012;6:197.
8. Michael J, Link JD, Bartleson D, Glenn F, Frederic B, Meyer B. Spontaneous midbrain hemorrhage: report of seven new cases. Surg Neurol 1993;39:58-65.
9. Mehmet Deveer, Nesat Cullu, Halil Beydilli, Hamdi Sozen, Onder Yeniceri, Selcuk Parlak. Spontaneous retrobulbar haematoma. Case Reports in Radiology 2015; 2015:1. doi:10.1155/2015/796834.
10. Chiu Tor W, Chung C K, Chan H Suk, Ng Wai M, Lam Lai K. Sneezing and orbital fracture. Plastic and Reconstructive Surgery 2006;117(3):1049-50.
11. Maramattom BV. Raccoon eyes following vigorous sneezing. Neurocrit Care 2006;4(2):151-2.
12. Gauguet JM, Lindquist PA, Shaffer K. Orbital emphysema following ocular trauma and sneezing. Radiology Case Reports 2008;3(1):124.
13. Roselle HA, Herman M.A hearty sneeze. Lancet 2010 Nov 27;376(9755):1872.
14. Sen D, Chaturvedi PK. Orbital emphysema after sneezing: a case report. Medical Journal, Armed Forces India 2011;67(3): 282-4.
15. Ali Zohair Nomani, Haris Majid Rajput, Mansoor Iqbal, et al. Subarachnoid hemorrhage secondary to forceful sneeze. Case Reports in Neurological Medicine 2015;2015:896732. doi: 10.1155/2015/896732.