

Single Case – General Neurology

Early Manifestation of Benign Paroxysmal Positional Vertigo: A Case Report

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Keywords

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Abstract

Introduction: Benign paroxysmal positional vertigo (BPPV) is an inner ear disorder with a heterogeneous etiology, often linked to preceding infections, head injuries, or vestibular neuronitis. While it is commonly observed in the elderly, its occurrence in the pediatric population is rare. To our knowledge, there have been no reported cases of BPPV in patients younger than 5 years. **Case Presentation:** A 4.5-month-old female infant was admitted with episodes of paroxysmal nystagmus. Parents reported fast horizontal eye movements lasting up to 30 s, with one episode accompanied by vomiting. Comprehensive differential diagnosis was considered from epileptic nystagmus to intoxications and both central and peripheral vestibular etiologies. During the observation on ward, connection between the baby's positioning and nystagmus was identified. The diagnostic roll test confirmed a transient positional geotropic nystagmus. The diagnosis aligned with BPPV characteristics, pointing to the right lateral semicircular canal canalolithiasis. A successful Lempert roll maneuver was performed with prompt effect. To further support the diagnosis and research, we introduced a semiautomatic video-oculography method. **Conclusion:** This case highlights a rare instance of BPPV in an infant. The clinical findings combined with the effectiveness of the repositioning maneuvers support the diagnosis of right lateral semicircular canal lithiasis. Despite the rarity of this condition in such a young-age group, the need for thorough diagnostic evaluations is emphasized. In order to document the case, we also present a semiautomatic video analysis pipeline for analyzing abnormal eye movements in a home setting.

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Introduction

Benign paroxysmal positional vertigo (BPPV) is an inner ear disorder, supposedly caused by canalolithiasis of semicircular canals [1]. Etiology is heterogeneous but is oftentimes attributed to preceding infection, traumatic head injury, or vestibular neuronitis. However, over 50% of cases still remain idiopathic [2]. This condition is treatable by repositioning maneuvers [3]. The lifetime prevalence is reported to be as much as 2.4% [4]. Considering vertiginous patients only, the prevalence among elderly patients is reported to be around 30%, whereas in the pediatric population, it is around 10% [5]. To our knowledge, no patient younger than 5 years old has been described in the literature so far.

Case Presentation

A 4.5-month-old female infant was brought to the emergency room after experiencing several episodes of nystagmus. Parents reported horizontal fast eye movements lasting for 30 s and the baby seemed to be unresponsive meanwhile; a single episode was associated with profound vomiting. There were no other problems, no signs or history of recent infection or head trauma, and between the episodes, the infant behaved normally. Parents were initially not aware of any provoking factors, including the changes of head position.

Differential diagnosis for paroxysmal abnormal eye movement in infants was established. Oculogenic etiologies are the most common [6] but less likely, given the paroxysmal nature of the symptoms. Epileptic nystagmus [7] may be a plausible cause, even despite the negative EEG findings during the presumed interictal period. Intoxications [8] either from substances like alcohol, certain medications, or drugs can induce nystagmus. This can be due to the direct cerebellar toxicity or, as in the case of alcohol [9], by altering the specific gravity inside the semicircular canals. Inborn errors of metabolism and leukodystrophies [8] can present with nystagmus, often alongside other symptoms. Any structural abnormalities in the central nervous system, especially within the brainstem, must be thoroughly excluded. Neuro-infections are a potentially treatable cause of nystagmus, warranting the consideration of a spinal tap. Several peripheral vestibular etiologies [10], such as BPPV, vestibular neuronitis, labyrinthitis, perilymphatic fistula, and superior semicircular canal dehiscence, should be considered. Abnormalities in eye movements, including nystagmus, can arise from whiplash-associated disorders due to impaired cervico-oculomotor systems [11]. Thus, a detailed history regarding potential previous injuries is crucial. Functional nystagmus [12] is recognized but improbable in this case due to the patient's age.

Physical examination as well as the acute blood test (ions, CRP, liver and kidney function) were, in presenting case, normal, and transcranial ultrasound showed enlargement of the subarachnoid space. Because of the broad differential diagnosis of nystagmus in infancy, the baby was acutely admitted to the hospital for further evaluation and observation.

The patient's family history was unremarkable. She was delivered by spontaneous prolonged labor at the gestational age of 33 weeks and 3 days, terminated by C-section. The Apgar scale was 5-7-8, nCPAP was used during the first day, and common neonatal jaundice occurred. ATB therapy had been administered for the first 48 h for possible sepsis, finally diagnosed as fetal inflammatory response syndrome. However, the child was well thriving since the discharge from the maternity ward.

Detailed blood tests revealed mild thrombocytosis and neutropenia, which together with a slight increase of the Fahrenaus-Westergren test (34/57) was suspected of the possible parainfectious finding. Both neurological and otorhinolaryngological examinations were normal. BAEP showed slightly prolonged latencies of wave III and V bilaterally. There were no

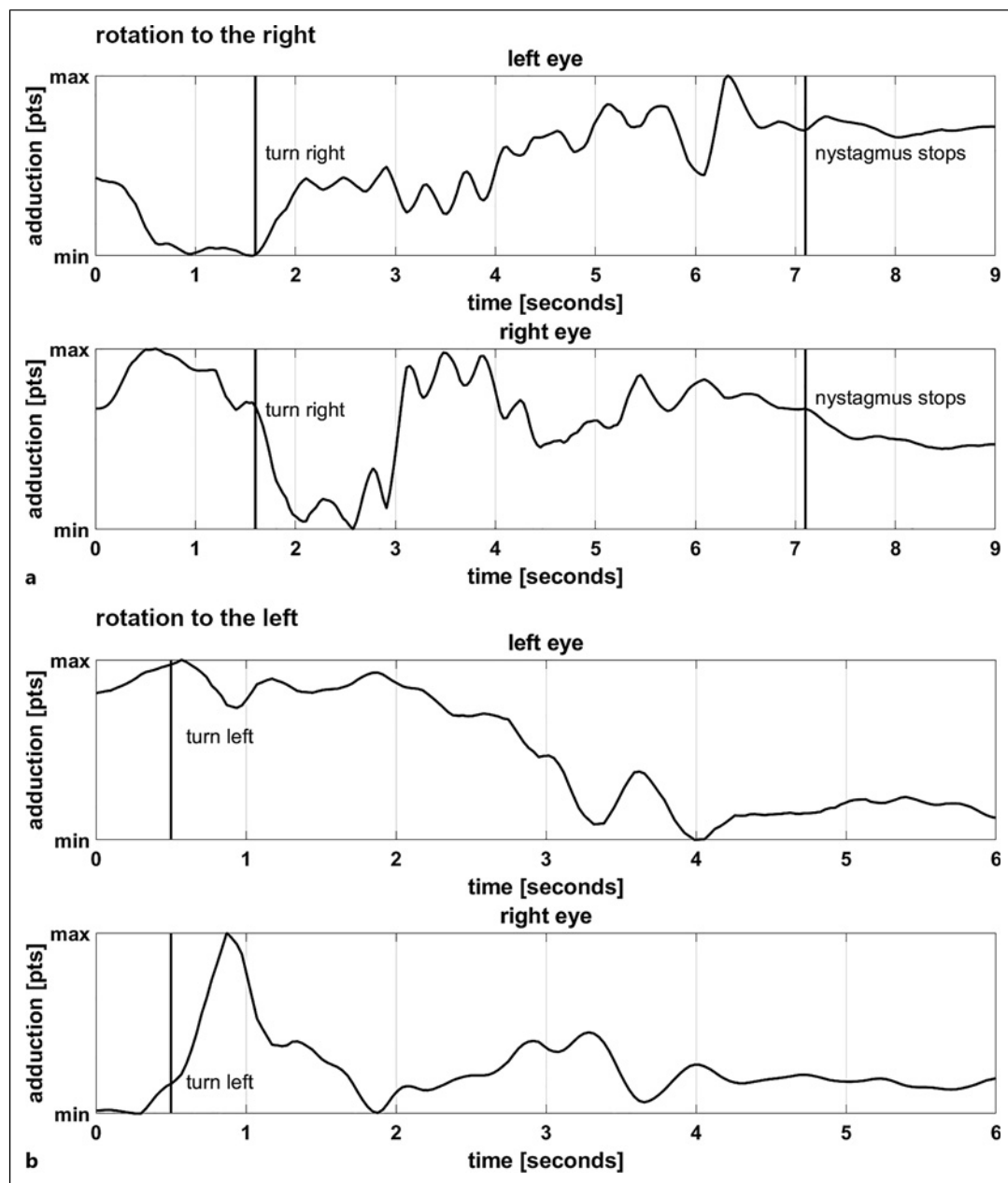


Fig. 1. a, b Video-oculography extracted from the bedside video recording.

epileptiform discharges on EEG. The magnetic resonance of the brain showed enlargement of SA spaces bilaterally (BESS - benign enlargement of subarachnoid space). Screening tests for metabolic disease as well as TORCH were negative.

Meanwhile, during the stay at the hospital, the mother started to report a clear connection between the positioning of the baby and the episodes of nystagmus. The diagnostic roll test was performed, and the subsequent horizontal nystagmus lasting no more than 30 s was disclosed. The diagnosis of transient positional geotropic nystagmus, likely due to right lateral semicircular canal canalolithiasis, was set up. This diagnostic conclusion is congruent with the

characteristics of BPPV. We performed the Lempert roll maneuver (“barbecue roll”) with prompt effect. In case of remission, the parents were instructed to repeat the maneuver at home, which happened once and was quickly resolved this way.

Due to the absence of electrophysiological documentation during the fit, we developed a pipeline to semiautomatically acquire video-oculography from the available video recording [13]. This was done in MATLAB R2021b using the Kanade-Lucas-Tomasi feature-tracking algorithm, which enables motion tracking of the selected points of interest. In this particular case, it was the patient’s pupil and medial palpebral commissures. The Euclidean distance of the pupil to the adjacent one is calculated. In order to correct for the zooming, it is further normalized by the Euclidean distance of the medial commissures. In the online supplementary video 1 (for all online suppl. material, see <https://doi.org/10.1159/000535550>), passive rotation to the right side is immediately followed by horizontal bidirectional nystagmus with frequency 2.5 Hz, which decreases to 1.5 Hz after 4 s and in 3 more seconds ceases completely. Amplitude remains stable during the fit. Nystagmus is absent following the left turning. Dynamics of the eye movements are documented in Figure 1. Sharp change immediately after the head turn attributes to the vestibulo-ocular reflex and rapid change in the feature proportions, which makes a precise assessment of the movement amplitude practically impossible. The CARE Checklist has been completed by the authors for this case report, attached as the online supplementary material.

Discussion

We present a case of a 4.5-month-old infant who was diagnosed with positional nystagmus on the right side. Classical clinical findings and prompt resolution with repositioning maneuvers support the diagnosis of right lateral semicircular canal lithiasis [14]. Pathophysiologically, otoconial debris moves in the direction of gravity causing ampullopetal endolymphatic flow, which stimulates hair cells and consequently induces the nystagmus. Apart from the likely incipient infection and perinatal history, there was no plausible etiology found. Vomiting after an episode could be a vegetative symptom associated with vertigo, making BPPV a possible diagnosis for this case. On the other hand, we note the controversy of diagnosing BPPV without clear signs of vertigo in this age group. This may fulfill the criteria for suspicion of nonbenign paroxysmal vertigo (nystagmus without dizziness in positional tests) [15]. As such, a thorough diagnostic work-up is recommended. In addition, this article presents a video analysis pipeline for analyzing abnormal eye movements in a home setting.

Positional nystagmus may be a rare and treatable occurrence from an early age. A thorough diagnostic workup is nevertheless recommended. Differential diagnosis of positional vertigo and its therapy should, therefore, be a well-mastered skill of pediatric neurologists.

Statement of Ethics

This research was conducted ethically in accordance with the World Medical Association Declaration of Helsinki. The study was approved by the Ethics Committee for Multicenter Clinical Trials at Motol University Hospital (ref. No. EK-869/23). Written informed consent was obtained from the parents of the participant for publication of the details of the medical case and any accompanying images/videos.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Matyas Ebel: conceptualization, methodology, and writing (lead); Alena Jahodova: writing – review and editing and investigation; and Jaroslav Jerabek: investigation and editing.

Data Availability Statement

The data that support the findings of this study are openly available at <https://github.com/MatyasE/eyetracking>. Further inquiries can be directed to the corresponding author.

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