

Horizontal canal cupulolithiasis presenting with horizonto-torsional pseudospontaneous nystagmus toward the affected ear: a case report (with video)

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Case Report

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Abstract

Background: Horizontal semicircular canal cupulolithiasis is a rare form of BPPV, typically characterized by persistent apogeotropic positional nystagmus. We report an unusual case associated with persistent spontaneous horizontal-torsional nystagmus beating toward the affected ear.

Case presentation: A 79-year-old woman presented with persistent rotational vertigo for one week. Examination revealed left-beating spontaneous horizontal-torsional nystagmus. The supine roll test showed bilateral apogeotropic nystagmus, with greater vertigo during rightward rotation, consistent with left horizontal canal cupulolithiasis. An adapted Gufoni maneuver converted the apogeotropic into a geotropic pattern, followed by a Lempert maneuver. Five minutes later, positional and spontaneous nystagmus had completely resolved, with no recurrence at one-week follow-up.

Conclusions: This rare presentation suggests an otolithic origin. Gravitational effects of otoconial debris attached to the cupula may increase its sensitivity. In the upright position, the canal's anatomical inclination may induce utriculopetal cupular deflection, producing excitatory nystagmus toward the affected side by Ewald's second law.

Background

Benign paroxysmal positional vertigo (BPPV) is the most common peripheral vestibular disorder and results from displacement of otoconial debris from the utricle into a semicircular canal [1]. Depending on the location of these particles, two principal pathophysiological mechanisms have been described: canalolithiasis, in which free-floating otoconia move within the endolymph, and cupulolithiasis, first proposed by Schuknecht in 1969, in which otoconial debris adheres to the cupula, rendering it abnormally sensitive to gravity [2].

Horizontal canal cupulolithiasis typically presents with persistent apogeotropic direction-changing positional nystagmus during the supine roll test and may exhibit a null point that helps identify the affected side [3]. Several cases of patients presenting with a spontaneous horizontal-torsional nystagmus beating toward the healthy ear have been reported [4, 5]. This phenomenon could be explained by the attachment of otoconial debris to the inferior part of the horizontal cupula, where their weight would induce a constant gravity-dependent utriculofugal cupular deflection in the sitting position, corresponding to an inhibitory stimulation of the affected horizontal canal [5]. Libonati emphasized that this pseudo-spontaneous nystagmus may sometimes be mistaken for vestibular neuritis [6].

We report a case of apogeotropic horizontal canal BPPV, consistent with cupulolithiasis, associated with a spontaneous horizontal-torsional nystagmus beating toward the affected ear. We discuss the pathophysiological mechanisms underlying this unusual clinical presentation.

Case presentation

The patient gave consent for the diffusion of their case report to the scientific community.

We report the case of a 79-year-old woman who was referred for persistent true rotational vertigo lasting one week. Symptoms were exacerbated in the supine position, particularly when lying in bed, although the patient was unable to identify a clearly more symptomatic side. She reported associated nausea without vomiting, with no auditory symptoms, headaches, or other neurological complaints. Her medical history was unremarkable, with no significant past medical conditions.

At initial bedside examination, the HINTS examination was negative. A grade II left-beating horizontal-torsional spontaneous nystagmus according to Alexander's law was observed. In the supine position with the head elevated at 30°, a strong left-beating horizontal-torsional nystagmus was present. During the supine head roll test, the direction of the nystagmus reversed depending on the side tested: the fast phase was directed to the right during the left supine roll test and to the left during the right supine roll test, consistent with a bilateral apogeotropic horizontal nystagmus pattern. The patient reported greater vertigo intensity during the right supine roll test.

A diagnosis of left horizontal semicircular canal cupulolithiasis was made. An adapted Gufoni maneuver was performed, resulting in conversion of the apogeotropic positional nystagmus into a geotropic pattern, supporting the transformation of cupulolithiasis into canalolithiasis. A subsequent Lempert maneuver was performed to treat the resulting left horizontal canal canalolithiasis.

At reassessment five minutes after the maneuvers, both positional vertigo and positional nystagmus had completely resolved. Notably, the previously observed spontaneous horizontal-torsional nystagmus had also disappeared. At one-week follow-up, the patient remained asymptomatic, with no spontaneous nystagmus and no recurrence of positional vertigo or positional nystagmus during diagnostic testing.

The clinical findings and the evolution of the nystagmus pattern throughout the diagnostic and therapeutic process are illustrated in the accompanying video.

Discussion and Conclusions

Our case highlights an unusual presentation of apogeotropic horizontal canal BPPV consistent with cupulolithiasis, characterized by a persistent spontaneous horizontal-torsional nystagmus beating toward the affected ear. This observation raises questions regarding the mechanisms responsible for sustained cupular deflection.

The pathophysiological basis of spontaneous horizontal-torsional nystagmus in horizontal semicircular canal cupulolithiasis remains incompletely understood. The most widely accepted explanation relies on the gravitational effect of otoconial debris attached to the cupula. In the upright position, the horizontal semicircular canal is anatomically tilted by approximately 30° relative to the earth-horizontal plane [7]. This orientation, combined with the specific axis of the horizontal cupula, allows gravity to exert a constant force on a cupula whose density is increased by attached otoconia [3,6,7]. This gravitational

force is thought to induce an utriculopetal cupular deflection (figure 1), which is excitatory for the horizontal canal according to Ewald's second law, resulting in a spontaneous nystagmus beating toward the affected ear [3,6,7].

A disappearance of the spontaneous horizontal-torsional nystagmus could be observed when the head is tilted 30° forward, positioning the horizontal canal in a neutral orientation perpendicular to the axis of gravity and thereby reducing the gravitational influence exerted on the otoconial debris [6]. This phenomenon could represent an equivalent of the **null point**, classically sought during the *supine roll test* to identify the affected side in apogeotropic forms of horizontal semicircular canal BPPV [8]. Further forward flexion of the head up to 60° may result in a reversal of the nystagmus direction [6], corresponding to the principle of the **bow test**, which is used clinically to lateralize both geotropic and apogeotropic forms of horizontal canal BPPV [9].

In conclusion, this case illustrates an unusual presentation of apogeotropic horizontal canal BPPV consistent with cupulolithiasis, associated with a persistent spontaneous horizontal-torsional nystagmus beating toward the affected ear. The resolution of nystagmus after repositioning maneuvers supports an otolithic origin and highlights the need to recognize this atypical presentation to avoid misdiagnosis of acute vestibular disorders.

Abbreviations

BPPV

Benign paroxysmal positional vertigo

Declarations

- **Ethics approval and consent to participate:** Ethics approval was obtained from the Data Protection Officer of the University of Toulouse. Written informed consent to participate was obtained from the patient.
- **Consent for publication:** Written informed consent for publication of this case and the accompanying images was obtained from the patient. A copy of the signed consent form will be made available for review by the Editor-in-Chief of the journal upon request.
- **Availability of data and materials:** The data that supports the findings of this study are available on request from the corresponding author.
- **Competing interests:** The authors have no competing interests to declare that are relevant to the content of this article
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- **Authors' contributions:** Quentin Legois: Conceptualization, Analysis, Writing—original draft; Simon Fargeas: Writing—review & editing; Salim Ismail: Writing—review & editing; Mikhael Makhoul: Conceptualization, Writing—review & editing

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Figures

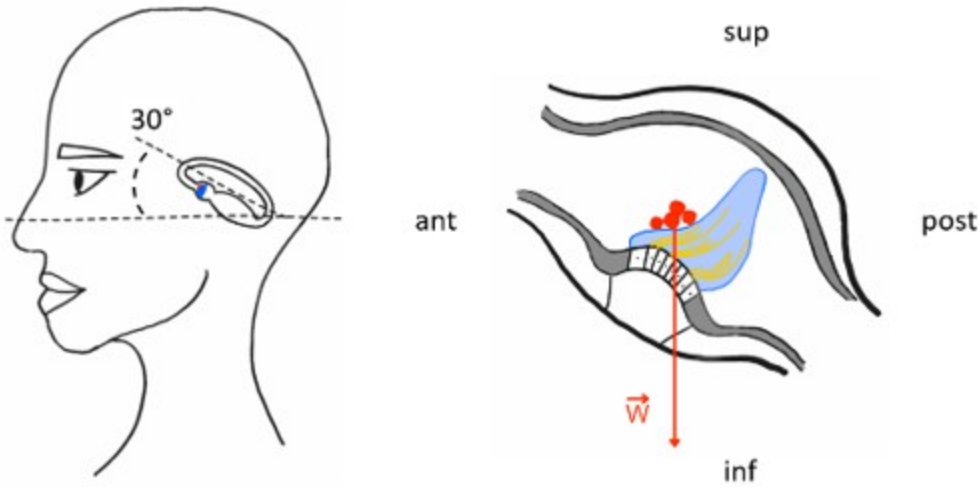


Figure 1

Gravitational cupular deflection induced by otoconial debris in horizontal semicircular canal cupulolithiasis (left: anatomical orientation of the horizontal semicircular canal relative to the earth-horizontal plane; right: gravitational deflection of the cupula by attached otoconial debris)

Supplementary Files

This is a list of supplementary files associated with this preprint. Click to download.

- [Video.mp4](#)