

ORIGINAL RESEARCH

Posterior canal benign paroxysmal positional vertigo with long duration: Heavy or light cupula?

Chang-Hee Kim MD, PhD  | Jung Eun Shin MD, PhD | Hansol Kim MD | Joon Yong Park MD

Department of Otorhinolaryngology-Head and Neck Surgery, Konkuk University Medical Center, Research Institute of Medical Science, Konkuk University School of Medicine, Seoul, Republic of Korea

Correspondence

Chang-Hee Kim, Department of Otorhinolaryngology-Head and Neck Surgery, Konkuk University Medical Center, Konkuk University School of Medicine, 120-1 Neungdong-ro (Hwayang-dong), Gwangjin-gu, Seoul 143-729, Republic of Korea.
Email: ryomachang@naver.com

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Abstract

Objective: To investigate the characteristics of positional nystagmus in posterior semicircular canal (PSCC) benign paroxysmal positional vertigo (BPPV) patients with longer durations, and to discuss the possible underlying mechanism of this nystagmus.

Methods: We conducted a retrospective review, and enrolled 118 consecutive patients with unilateral PSCC BPPV. The duration of nystagmus during a Dix-Hallpike test was classified into short (<1 min) and long (≥ 1 min) durations. For the identification of a neutral point in PSCC BPPV patients with long durations, the patient's head was turned 45° to the lesioned side to set the affected PSCC on the sagittal plane, and the disappearance of positional nystagmus was investigated in a pitch plane.

Results: Among 118 patients with PSCC BPPV, positional nystagmus during a Dix-Hallpike test showed short durations (<1 min) in 112 patients and long durations (≥ 1 min) in 6 patients. Of 6 PSCC BPPV patients with a long duration, a neutral point was identified in 5 patients whose nystagmus lasted for longer than 2 min; interestingly, a neutral point was observed when the patient's head was slightly tilted backward in all 5 patients.

Conclusion: Considering that a neutral position was identified when the patient's head was slightly tilted backward while keeping the head turned 45° to the right or left, we assume that the light cupula condition of the ipsilateral PSCC or the contralateral anterior semicircular canal, and not PSCC BPPV cupulolithiasis, could be responsible for the occurrence of persistent torsional-upbeating nystagmus in a Dix-Hallpike test.

Level of Evidence: 4

KEYWORDS

benign paroxysmal positional vertigo, Canalolithiasis, Cupulolithiasis, light cupula, posterior semicircular canal

1 | INTRODUCTION

Benign paroxysmal positional vertigo (BPPV) is characterized by paroxysmal vertigo elicited by head position changes, and the

posterior semicircular canal (PSCC) is known as the most frequently involved canal. Gravity-dependent movement of otoconial particles within the PSCC is accepted as a possible mechanism of the canalolithiasis type of PSCC BPPV. PSCC BPPV can be diagnosed when

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upbeating and torsional nystagmus, which lasts for less than 1 min in most cases, is induced by a Dix-Hallpike maneuver.¹⁻³ The concept of PSCC BPPV cupulolithiasis has been recently introduced,⁴⁻⁹ and diagnostic criteria for PSCC BPPV cupulolithiasis were proposed by the Barany Society.^{3,10} Cupulolithiasis in PSCC BPPV is distinguished from canalolithiasis in that the duration of positional nystagmus is longer than 1 min. Otoconial particles attached to the cupula have been proposed as a cause of cupulolithiasis of PSCC BPPV.¹¹⁻¹³ Epley assumed that a half Dix-Hallpike test could elicit greater deflection of the PSCC cupula, producing stronger positional nystagmus in PSCC BPPV cupulolithiasis.¹⁴

However, because the exact anatomical orientation of the cupula in the PSCC with respect to the direction of gravity has not yet been identified, the presence of cupulolithiasis involving the vertical semicircular canals has not been clearly understood. In the present study, we report on 118 patients who exhibited positional upbeat nystagmus with a torsional component beating toward the lowermost ear by a Dix-Hallpike test. In particular, characteristic positional nystagmus profiles in PSCC BPPV patients with longer durations are described, and the possible mechanism is discussed.

2 | SUBJECTS AND METHODS

We conducted a retrospective review and enrolled 118 consecutive patients with unilateral PSCC BPPV who were diagnosed at our outpatient dizziness clinic between January 2021 and September 2022. All of the patients complained of positional vertigo. The diagnosis of PSCC BPPV was made when paroxysmal positional upbeat nystagmus with a torsional component beating toward the lowermost ear was evoked by the ipsilateral Dix-Hallpike test.¹⁻³ Patient eye movement was examined using goggles installed with an infrared camera (EasyEyes, SLMED, Seoul, Republic of Korea) by otolaryngologists in the clinic. In the present study, the duration of nystagmus during the Dix-Hallpike test was classified into short (<1 min) and long (≥1 min) durations. Only idiopathic PSCC BPPV patients were included, and those with multicanal involvement, posttraumatic BPPV, central positional nystagmus or secondary BPPV were excluded from the study. No cochlear symptoms, including hearing loss, tinnitus or aural fullness, were found. Neurological examination, including a cerebellar function test, was performed on all patients and did not reveal abnormalities. A modified Epley maneuver was performed to treat PSCC BPPV.

This study was conducted in accordance with the 1964 Helsinki Declaration for ethical standards and was approved by the Institutional Review Board (approval no. 2022-10-053).

3 | RESULTS

3.1 | Clinical characteristics of PSCC BPPV patients with short and long durations

One hundred eighteen patients (77 women and 41 men; age range, 22-88 years) were included (Table 1). Among 118 patients with PSCC

BPPV, positional nystagmus during the Dix-Hallpike test showed short durations (<1 min) in 112 patients and long durations (≥1 min) in 6 patients. The male-to-female ratios were 38:74 and 3:3 in patients with short and long durations, respectively, indicating no significant difference ($p = 0.406$, Fisher's exact test). The ages ranged from 22 to 88 years old with an average age of 57 ± 16 years old and from 44 to 78 years old with an average age of 62 ± 12 years old in patients with short and long durations, respectively, indicating no significant difference ($p = .251$, Fisher's exact test). The involved side was the right in 60 of 112 patients with short durations and 2 of 6 patients with long durations, and was not significantly different ($p = 0.421$, Fisher's exact test).

3.2 | Characteristics of positional nystagmus in PSCC BPPV patients with long duration

The 112 patients with short duration (<1 min) can undoubtedly be diagnosed with PSCC BPPV canalolithiasis.¹ They showed a latency period between the completion of the Dix-Hallpike maneuver and the onset of nystagmus, and a reversal of the nystagmus was observed during the return to the upright position. Of six patients with a long duration (≥1 min), the nystagmus duration was shorter than 2 min in one patient and longer than 2 min in five patients (Table 2). Recent studies have suggested that cupulopathy of the lateral semicircular canal (LSCC) is characterized by the identification of the neutral point, at which the nystagmus disappears because the long axis of the LSCC cupula is aligned with the direction of gravity.¹⁵⁻²⁰ Therefore, we sought to identify the neutral point in PSCC BPPV patients with a long duration. Although the location of the LSCC cupula has been reported,²¹ the exact orientation of the PSCC cupula has not been clearly identified. Therefore, for the identification of a neutral point, the patient's head was turned 45° ipsilaterally to set the affected PSCC on the sagittal plane, and positional nystagmus was investigated in a pitch plane. First, upbeat and ipsilateral torsional nystagmus with a long duration was observed during a Dix-Hallpike test to confirm a diagnosis of PSCC BPPV cupulolithiasis (Video S1). Then, downbeat and contralateral torsional nystagmus was observed in a bow position while keeping the head turned 45° to the affected side (Video S2). Subsequently, the presence of a neutral point, at which

TABLE 1 Clinical features of posterior semicircular canal benign paroxysmal positional vertigo (PSCC BPPV) patients with short and long durations.

	PSCC BPPV with short duration	PSCC BPPV with long duration	<i>p</i> value
Number of patients	112	6	
Sex, M:F	38:74	3:3	.406
Age, years	57 ± 16 (range, 22-88)	62 ± 12 (range, 44-78)	.251
Side, R:L	60:52	2:4	.421

Abbreviations: F, female; L, left; M, male; R, right.

TABLE 2 Identification of a neutral point in posterior semicircular canal benign paroxysmal positional vertigo (PSCC BPPV) patients with long durations.

Patient no.	Sex/age	Side	Nystagmus duration	Identification of a neutral point
1	M/60	L	>2 min	+
2	F/44	L	>2 min	+
3	M/69	R	>2 min	+
4	F/65	L	>2 min	+
5	F/54	L	>2 min	+
6	M/78	R	<2 min	–

TABLE 3 Treatment outcomes of posterior semicircular canal benign paroxysmal positional vertigo (PSCC BPPV) patients with short and long durations.

	1 session	2 sessions	3 or more sessions	p value
Short duration (n = 112)	73	29	10	<.001
Long duration (n = 6)	0	1	5	

Abbreviation: SN, spontaneous nystagmus.

the nystagmus ceases and changes its direction with further head movement, was investigated (Video S3). Of six PSCC BPPV patients with a long duration, a neutral point was identified in five patients whose nystagmus lasted for longer than 2 min (Table 2). Interestingly, a neutral point was observed when the patient's head was slightly tilted backward in all five patients (Video S3) (Table 3).

4 | DISCUSSION

Cupulolithiasis has been demonstrated in human pathologic studies, and suggested as one of the types of BPPV.^{11–13} A nystagmic characterization of PSCC BPPV cupulolithiasis has been recently demonstrated,^{4,6} and PSCC BPPV cupulolithiasis is receiving increasing attention in differential diagnosis for positional vertigo. The incidence of PSCC BPPV cupulolithiasis is considered much lower than that of canalolithiasis, and the proportion of cupulolithiasis cases in PSCC BPPV has been reported to be 6.4%–7.2%,^{6,8} consistent with the proportion of 5.1% in the present study. Although many studies have supported the efficacy of canalith repositioning maneuvers (CRMs) in PSCC BPPV canalolithiasis, immediate resolution of BPPV after CRMs was not achieved in any of the patients with PSCC BPPV cupulolithiasis.^{9,22} Despite the lack of immediate efficacy of CRMs, the long-term prognosis is good in most cases.^{4,5}

Ichijo examined the difference between positional nystagmus in the head-hanging position and that in the nose-down position to confirm the validity of Ewald's third law in patients with PSCC BPPV cupulolithiasis fulfilling the following conditions⁴: (1) a Dix-Hallpike test elicits upbeat/torsional nystagmus, and it lasts for longer than

1 min in a head-hanging position; (2) no nystagmus or mild vertical (downbeating) nystagmus is observed in a sitting position; (3) torsional/vertical (downbeating) nystagmus, which lasts for longer than 1 min, is observed in a nose-down position; and (4) torsional/vertical (upbeating) nystagmus, which lasts for longer than 1 min, is observed in a supine position. He comprehensively compared the intensity of nystagmus by calculating the sum of the maximal slow-phase velocity of horizontal, vertical and torsional components and observed no significant difference between the head-hanging position and the nose-down position. That study did not support Ewald's third law in cases with PSCC BPPV cupulolithiasis, although this law has been well supported in cases with canalolithiasis.²³ It was postulated that the reason for this outcome was a difference in the intensity of stimulation.⁴ Recently, Ichijo reported another study on the clinical characteristics of 30 patients with PSCC BPPV cupulolithiasis.⁵ The affected side was determined according to the direction of the torsional component of nystagmus in the supine position, and the onset time of vertigo was the time of awakening (n = 14), in the afternoon (n = 10), in the morning (n = 2), during sleep (n = 2), and unknown (n = 2). Most patients were cured within 1 month without CRPs or vestibular rehabilitation.⁵

Imai et al. divided 111 PSCC BPPV patients into two groups; one with a time constant lasting longer than 40 s (8 patients) and another with a time constant lasting shorter than 20 s (103 patients), and those with a longer time constant were diagnosed with PSCC BPPV cupulolithiasis.^{6,24} These authors sought, for the first time, to identify the position of a neutral point in 8 patients with PSCC BPPV cupulolithiasis. A neutral point, in which a vertical-torsional nystagmus disappeared, was identified in the ipsilateral head hanging position with further extension of the neck in 8 patients with long durations. Furthermore, the presence of PSCC BPPV cupulolithiasis was additionally examined by confirming that vertical-torsional positional nystagmus elicited direction-changing characteristics when the patient's head position was changed from the contralateral head-hanging position to the ipsilateral head rotation in the supine position. The authors reported that the axis angles of positional nystagmus were parallel to the ipsilateral PSCC and the contralateral anterior semicircular canal (ASCC).⁶

Wang et al. investigated whether the half Dix-Hallpike test induces more conspicuous nystagmus than the classical Dix-Hallpike test in PSCC BPPV cupulolithiasis, and reported that the occurrence rate of torsional-upbeating nystagmus was higher in the classical Dix-Hallpike test than in the half Dix-Hallpike test.⁸ The authors concluded that the classic Dix-Hallpike test is a better diagnostic test than the half Dix-Hallpike test for the diagnosis of PSCC BPPV cupulolithiasis because the half Dix-Hallpike test, although feasible and simple, could elicit a higher false negative rate.

In the present study, the half Dix-Hallpike test was also not performed in any patients because we had often observed that the half Dix-Hallpike test did not evoke torsional-upbeating nystagmus as robustly as the Dix-Hallpike test in PSCC BPPV patients with long duration (unpublished personal experience). Instead, for the diagnosis of PSCC BPPV cupulolithiasis, we sought to identify a neutral point in

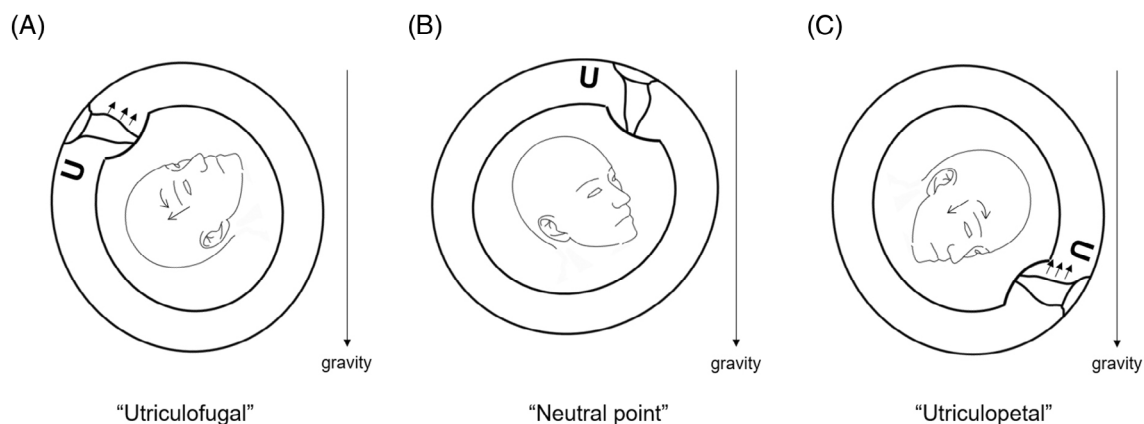


FIGURE 1 Representative case with persistent positional nystagmus in the right Dix-Hallpike test. (A) Considering that the right Dix-Hallpike test evokes persistent right torsional-upbeating nystagmus, the light cupula condition is assumed to cause utriculofugal deflection of the PSCC cupula, eliciting excitation of the right PSCC. (B) A neutral point, in which the cupula is aligned in the direction of gravity, is identified when the patient's head is slightly tilted backward while keeping the head turned 45° to the right. (C) Persistent left torsional-downbeating nystagmus is observed when the patient's head is bent forward while keeping the head turned 45° to the right. PSCC, posterior semicircular canal; U, utricle.

a pitch plane while keeping the head turned 45° to the affected side to align the PSCC on the sagittal plane (Videos S1, S2, and S3). A neutral point refers to the position where the long axis of the PSCC cupula is assumed to be aligned with gravity.⁶ A neutral point was identified when the patient's head was slightly tilted backward in all five PSCC BPPV patients with long durations (>2 min), which might be consistent with the findings of the previous study that no nystagmus or mild downbeating nystagmus was observed in the sitting position.⁴ Because the anatomical orientation of the PSCC cupula is not clearly understood, we attempted to conjecture about the axis of the PSCC cupula based on the findings of positional nystagmus (Figure 1). For a neutral point to be identified when the head was slightly tilted backward while keeping the head turned 45° to the affected side in the right PSCC BPPV cupulolithiasis patient, the axis of the right PSCC cupula was aligned in the direction of gravity (Figure 1B). Persistent right torsional-upbeating nystagmus was observed in the right Dix-Hallpike position, caused by utriculofugal deflection of the right PSCC cupula (Figure 1A). Under the assumption of anatomical orientation of the PSCC cupula conjectured from a neutral point (Figure 1B), the light cupula condition, rather than heavy cupula (cupulolithiasis), is more relevant in explaining the occurrence of this positional nystagmus (Figure 1A). In a bow position while keeping the head turned 45° to the right side, the right PSCC cupula is deflected utriculopetally, eliciting left torsional-downbeating nystagmus (Figure 1C). Thus, the light cupula condition in the ipsilateral PSCC could be responsible for the occurrence of persistent torsional-upbeating nystagmus in a Dix-Hallpike test. The position of a neutral point was not consistent with that in a previous study, in which a neutral point was observed in the ipsilateral head hanging position with further extension of the neck,⁶ and we believe that further study with a larger sample size is required to clarify the reason for this discrepancy. Conversely, not only the right PSCC (Figure 2A) but also the left ASCC (Figure 2B) is placed on the sagittal plane when the head is turned 45° to the right. Assuming that

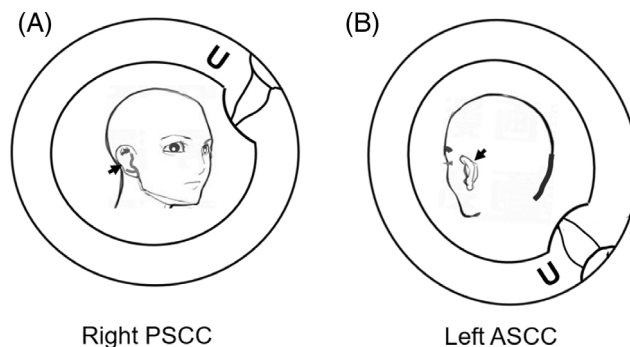


FIGURE 2 Suggested anatomical orientation of the right PSCC cupula (A) and the left ASCC cupula (B), which are conjectured based on the position of a neutral point in the representative case depicted in Figure 1. Note that a neutral point is identified when the patient's head is slightly tilted backward while keeping the head turned 45° to the right. ASCC, anterior semicircular canal; PSCC, posterior semicircular canal.

the left ASCC cupula is oriented as depicted in Figure 2B, a neutral point can be identified at which the patient's head was slightly tilted backward while keeping the head turned 45° to the right (Figure 3B). Considering that right torsional-upbeating nystagmus was observed during the right Dix-Hallpike position, utriculopetal deflection of the ASCC cupula due to the light cupula condition could be a relevant explanation for this positional nystagmus (Figure 3A). In a bow position while keeping the head turned 45° to the right, the left ASCC cupula is deflected utriculofugally, eliciting left torsional-downbeating nystagmus (Figure 3C). Because Ewald's third law stands on the premise that either excitatory or inhibitory stimulation, as pointed out in a previous study,⁴ is equally applied, we did not compare the slow-phase velocity at each position.

The limitations of the present study are as follows: (1) Owing to the small sample size of PSCC BPPV cupulolithiasis patients in this

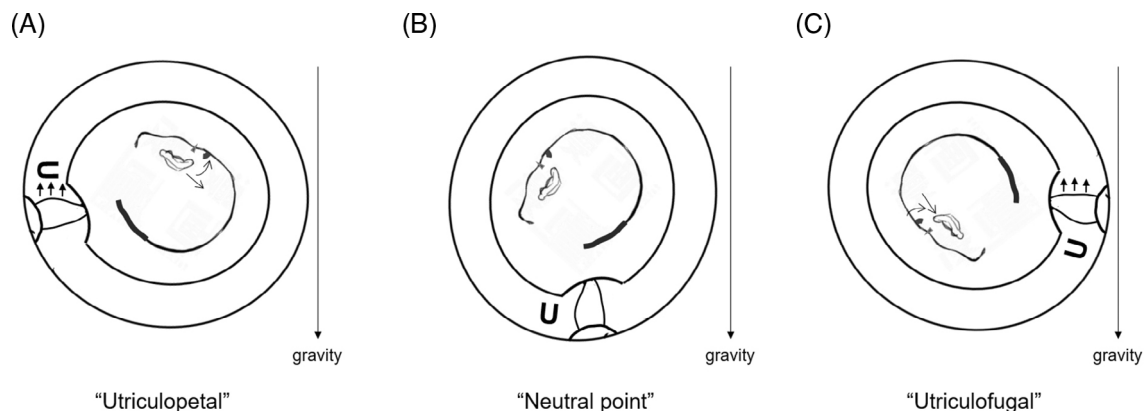


FIGURE 3 Same case as Figure 1 showing persistent positional nystagmus in the right Dix-Hallpike test. (A) Considering that the right Dix-Hallpike test evokes persistent right torsional-upbeating nystagmus, the light cupula condition in the contralateral ASCC is assumed to cause utriculopetal deflection of the left ASCC cupula, eliciting inhibition of the left ASCC. (B) A neutral point, in which the left ASCC cupula is aligned in the direction of gravity, is identified when the patient's head is slightly tilted backward while keeping the head turned 45° to the right. (C) Persistent left torsional-downbeating nystagmus is observed when the patient's head is bent forward while keeping the head turned 45° to the right. ASCC, anterior semicircular canal; U, utricle.

study, the identification of a neutral point should be validated in more patients with PSCC BPPV cupulolithiasis in future studies. (2) The slow-phase velocity of positional nystagmus could not be calculated in the present study because we used a video Frenzel goggles without an eye movement interpretation program. Comprehensive interpretation of SPV of torsional, horizontal, and vertical components may support the diagnosis of cupulopathy of ASCC or PSCC BPPV. (3) Lacking knowledge on the anatomical orientation of the cupulae of vertical canals made it difficult to determine whether the involved canal is the PSCC or the contralateral ASCC.

5 | CONCLUSION

This study demonstrated that 5 of 118 patients with PSCC BPPV showed persistent torsional-upbeating nystagmus in a Dix-Hallpike test. A neutral position was identified when the patient's head was slightly tilted backward while keeping the head turned 45° to the right or left in all five patients. It is postulated that the light cupula condition of the ipsilateral PSCC or the contralateral ASCC might be responsible for the occurrence of persistent torsional-upbeating nystagmus in a Dix-Hallpike test.

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CONFLICT OF INTEREST STATEMENT

None.

ORCID

Chang-Hee Kim  <https://orcid.org/0000-0001-5667-861X>

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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