INTRODUCTION

When evaluating dizzy patients, the presence of spontaneous nystagmus suggests to the physician an acute vestibulopathy, either temporary or permanent, such as vestibular neuritis or Meniere’s disease [1]. In contrast, a diagnosis of benign paroxysmal positional vertigo (BPPV) is suggested by a position-induced nystagmus that changes its direction according to head position. As such, spontaneous nystagmus while in the sitting position is not considered as a typical sign of BPPV [2].

However, the occurrence of spontaneous nystagmus in lateral semicircular canal BPPV (LC-BPPV) patients has been reported previously, and recently a new physiological mechanism has been suggested [2-6].

The occurrence of spontaneous nystagmus in BPPV cases has been explained by two pathophysiologic mechanisms. First, when otolithic particles jam in the narrow semicircular canals (‘canalith jam’) [5], the resulting nystagmus does not show characteristic directional changes in different head positions, but rather a constant direction regardless of head position. The affected patients show a typical nystagmus related to unilateral vestibulopathy, and a decreased caloric response on the affected side. Second, recent studies report the occurrence of spontaneous nystagmus in patients with LC-BPPV in the absence of unilateral vestibulopathy—a condition which has been named ‘pseudo’-spontaneous nystagmus, based on the physiologic mechanism of its origin [2-4]. However, the reported incidence of this nystagmus in LC-BPPV is widely variable (40-96%), and its clinical
The patient’s head to the opposite direction. When LC-BPPV was confirmed, the direction of nystagmus in the supine roll test denoted the type of BPPV: geotropic or apogeotropic. The nystagmus directions for the supine position with the head centered (lying-down nystagmus or leaning nystagmus) and for the head bowing position (bending-over nystagmus or bowing nystagmus) were used to confirm the side of involvement according to the relative strength of nystagmus for right and left head turns in the supine roll test [7,8].

The bithermal caloric tests were performed in 9 of the 16 cases in the PSN group. For all nine, canal paresis was less than 20% in both affected and unaffected sides. The remaining 7 patients of the PSN group did not undergo the caloric tests, because the PSN disappeared immediately after the canalith repositioning maneuver and vertiginous symptoms improved dramatically.

For all patients, the barbecue maneuver was performed and instructed about the forced prolonged position with minimal restriction. For cases of the apogeotropic type, the barbecue maneuver was done with the application of vibration on the mastoid tip of the affected side. Subsequently, patients were followed up twice a week with a repetition of the barbecue maneuver (with or without vibrator) until the positional nystagmus completely disappeared. Symptom duration is defined here as the period from the date when the first symptom was self-reported to the date of diagnosis. Treatment duration was taken as the period from the date of diagnosis to the date when positional nystagmus completely disappeared.

Statistical analyses were performed using SPSS ver. 12 (SPSS Inc., Chicago, IL, USA) to compare the gender, age, symptom duration, and treatment duration between groups. The Mann-Whitney U-test and linear regression (stepwise) analysis were used to assess significance, and the criterion for significance was \( P<0.05 \).

**RESULTS**

Among the 95 patients, PSN was observed in 16 (PSN group, 16.8%, 3 men and 13 women), among these, 6 were of the geotropic type and 10 were of the apogeotropic type. The involved side was the left in 6 and the right in 10 patients of the PSN group. The other 79 cases did not reveal PSN (non-PSN group, 17 men and 62 women). Of these, the distribution of BPPV type was geotropic in 34 and apogeotropic in 45. The involved side was the left in 44 and the right in 35 cases. Sex \( (P=0.085) \) and the ratio of geotropic to apogeotropic type \( (P=0.553) \) did not differ significantly between the two groups. The mean age (± standard deviation) was 57.81±10.80 in the PSN group and 49.53±13.87 in the non-PSN group. The age difference between these groups was significant \( (P=0.023) \).

Symptom duration prior to the first visit was 8.00±9.36 days in the PSN group and 7.52±11.73 days in the non-PSN group; these differences were not significant \( (P=0.481) \) (Table 1). How-
Fig. 1. Video nystagmograms showing pseudo-spontaneous nystagmus (PSN) and positional nystagmus in representative cases of LC-BPPV. (A) A case of geotropic type involving the right ear. Left-beating PSN (7 degree/second) is recorded. In supine roll test, geotropic nystagmus is recorded in the head right (18 degree/second, directing to the right side) and head left (13 degree/second, direction to the left side) position. When the positional test is performed in the pitch plane, lying-down nystagmus and bending-over nystagmus are directed to the same (left-beating, 8 degree/second) and to the opposite direction (right-beating, 8 degree/second) as the PSN, respectively. (B) A case of apogeotropic type involving the left ear. Left-beating PSN (8 degree/second) is recorded. Apogeotropic type nystagmus is examined in the supine roll test (left-beating, 146 degree/second with the head turned to the right; right-beating, 57 degree/second to the left). In the pitch plane, the direction of evoked nystagmus was the same as the direction for PSN in the lying down position (left-beating, 79 degree/second), and to the opposite direction in the bending over position (right-beating, 47 degree/second). LC-BPPV, benign paroxysmal positional vertigo involving the lateral semicircular canal; LH, horizontal component of nystagmus measured on the left eye; LB, nystagmus of which fast component directing to the left side of the patient; RB, nystagmus of which fast component directing to the right side of the patient.
In 2001, Bisdorff and Debatisse [6] first reported spontaneous nystagmus in LC-BPPV patients without canalith jam from 2 cases of the cupulolithiasis type. Towards the end of that decade, its occurrence in both geotropic and apogeotropic types of LC-BPPV had been reported and studies investigating its clinical value performed to a limited extent [2,4,6]. The mechanism of spontaneous nystagmus in LC-BPPV was explained by the fact that maintaining the vertical axis of one’s head earth-vertical in a seated posture causes the lateral semicircular canal to be upwardly inclined by about 30°, which results in movement of otolith particles according to the direction of gravity. Since this nystagmus is developed by the specific head position that evokes gravitational movement of otolith particles, and not by imbalance of spontaneous discharge from vestibular afferents, it was called “pseudo”-spontaneous nystagmus [3,4]. The reason why PSN is observed in BPPV involving only the lateral semicircular canal lies in the anatomical features of the canals. While otolith particles within the vertical semicircular canals hardly produce endolymphatic flow in a seated position, endolymph movement within the lateral semicircular canal can be easily obtained by changing the degree of head inclination.

In diagnosing peripheral vestibular disorders, spontaneous nystagmus has been considered the characteristic feature of acute vestibular imbalance, such as in vestibular neuritis and the ictal phase of Meniere’s disease. In rare cases of BPPV, spontaneous nystagmus has been observed when a canalith jam occurs. Canalith jam is the state in which otolith particles directly block endolymphatic flow in the canal, or attached to the cupula causing a fixed position of the cupula regardless of gravity [5,9]. Differently from PSN, spontaneous nystagmus observed due to canalith jam may occur for jams of any semicircular canal, regardless of a change in posture. However, the PSN discussed here is limited to only BPPV involving the lateral semicircular canal. PSN refers to the ocular movement only while in the seated position, and the nystagmus can disappear or change direction according to head posture.

In theory, the direction of PSN is toward the contra-lesional side in the geotropic type and directed ipsi-lesionally in the apogeotropic type, which is the same for lying-down nystagmus. This is because the physiologic mechanism of PSN is identical to that of lying-down nystagmus, and it is only differentiated by the inclination of the lateral semicircular canal [7,8]. Direction of PSN and other positional nystagmus in LC-BPPV is summarized in Table 2. The PSN should disappear with the patient’s head inclined forward at 30°, and the direction of the nystagmus change to the opposite side if the head is inclined further forwardly in the pitch plane. Consequently, PSN has been proposed as a clinical sign to determine the involved side of LC-BPPV [3,4]. As the direction of PSN examined in a seated position is always opposite to the direction of the bending-over nystagmus (Table 2), authors suggest to examine nystagmus in the head bowing position when encountering dizzy patients with spontaneous nystagmus to rule out the possibility of LC-BPPV, espe-
Table 2. Direction of PSN and positional nystagmus in LC-BPPV

| Type                      | Involved ear | Direction of the nystagmus |
|---------------------------|--------------|----------------------------|
| Geotropic type (canalolithiasis) | Right        | LB, RB, LB, RB, LB         |
|                           | Left         | RB, LB, RB, RB, LB         |
| Apogeotropic type (cupulolithiasis) | Left         | RB, LB, RB, LB, RB         |

PSN, pseudo-spontaneous nystagmus; LC-BPPV, benign paroxysmal positional vertigo involving the lateral semicircular canal; BO, bending-over nystagmus examined in the head bowing position; LD, lying-down nystagmus examined in the supine position with the head centered; HR, nystagmus examined in the supine position with head turned to the right; HL, nystagmus examined in the supine position with head turned to the left; RB, nystagmus of which fast component directing to the right side of the patient; LB, nystagmus of which fast component directing to the left side of the patient.

... particularly when the patient appeals aggravation/relief of dizziness by positional changes.

In previous studies, the frequency of PSN in LC-BPPV has been reported to vary from 40.1% to 76%, and to increase to 56.2-96% after mild head shaking [3,4,10]. In the present study, this ratio for patients showing PSN was only 16.8%, which is considerably lower than in previous reports. Two situations at the outpatient clinic can account for this difference. First, in the present study, the time devoted at the clinic to examining spontaneous nystagmus was relatively short, although it was sufficient to detect apparent nystagmus (minimum 10 seconds). Second, and more important, the head position of the patient was not precisely controlled. The existence and direction of PSN were observed immediately after a patient sat on a chair with his/her head roughly earth vertical. Patients with LC-BPPV naturally prefer to bend their head slightly forward, since this posture minimizes movement of otolith particles in the lateral semicircular canal. Otherwise, patients favor at least having the head fixed in the pitch plane. The occurrence of PSN and its direction were probably affected by head position immediately before and during the examination, which was not precisely controlled in this study. The discordant direction of PSN with lying down nystagmus in three cases might be attributed to this uncontrolled head inclination during the examination. Previous research also supports the idea that the frequency of PSN is subject to head position and movement during the examination [3,4]. Considering the busy clinical setting in medical institutes, we consider the current result more realistic.

Results of the present study confirm that PSN can accompany LC-BPPV, and the presentation of PSN may be associated with longer follow-up duration before cure. Previously reported factors resulting in poorer treatment outcome of BPPV are a history of head trauma, inner ear disorders (e.g., Meniere’s disease, vestibular neuritis), gender and age [11,12]. After correcting for these outcome variables, we find that the presence of PSN extends treatment duration. One possible explanation for this is that patients who developed PSN might have relatively more otolithic load in the lateral semicircular canal than those without PSN. In this context, abnormal endolymphatic flow might be produced in patients with PSN with enough strength to evoke nystagmus, even with a small degree of inclination of the head in a seated position. More lithiasis particles might require longer treatment duration to be cleared out from the lateral semicircular canal. Though this explanation is plausible, the small adjusted R-square from the linear regression analysis suggests caution in attributing any prognostic value to this observation, particularly since a previous study revealed no effect of PSN on treatment outcome [10].

In conclusion, horizontal nystagmus in the upright position can be examined in patients with LC-BPPV, and this can possibly be misinterpreted as spontaneous nystagmus representing acute vestibular imbalance. When spontaneous nystagmus is observed in patients suspected to have peripheral vertigo, performing the positional test immediately in the clinic would help provide a correct diagnosis of the cause. Examination of positional nystagmus only in the pitch plane would suffice for screening purposes, and will save time and money by avoiding further vestibular laboratory testing. The prognostic importance of PSN suggested by the current study is still an issue to be clarified in future research.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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REFERENCES

1. Hamid M. Medical management of common peripheral vestibular diseases. Curr Opin Otolaryngol Head Neck Surg. 2010 Oct;18(5):407-12.
2. Asprella Libonati G. Diagnostic and treatment strategy of lateral semicircular canal canalolithiasis. Acta Otorhinolaryngol Ital. 2005 Oct;25(5):277-83.
3. Califano L, Melillo MG, Mazzone S, Vassallo A. “Secondary signs of lateralization” in apogeotropic lateral canalolithiasis. Acta Otorhinolaryngol Ital. 2010 Apr;30(2):78-86.
4. Asprella-Libonati G. Pseudo-spontaneous nystagmus: a new sign to diagnose the affected side in lateral semicircular canal benign paroxysmal positional vertigo. Acta Otorhinolaryngol Ital. 2008 Apr;28(2):73-8.
5. Epley JM. Human experience with canalith repositioning maneuvers. Ann NY Acad Sci. 2001 Oct;942:179-91.
6. Bisdorff AR, Debatisse D. Localizing signs in positional vertigo due to lateral canal cupulolithiasis. Neurology. 2001 Sep;57(6):1085-8.
7. Koo JW, Moon IJ, Shim WS, Moon SY, Kim JS. Value of lying-down nystagmus in the lateralization of horizontal semicircular canal benign paroxysmal positional vertigo. Otol Neurotol. 2006 Apr;27(3):367-71.
8. Choung YH, Shin YR, Kahng H, Park K, Choi SJ. ‘Bow and lean test’ to determine the affected ear of horizontal canal benign paroxysmal positional vertigo. Laryngoscope. 2006 Oct;116(10):1776-81.
9. von Brevern M, Clarke AH, Lempert T. Continuous vertigo and spontaneous nystagmus due to canalolithiasis of the horizontal canal. Neurology. 2001 Mar;56(5):684-6.
10. De Stefano A, Kulamarva G, Citraro L, Neri G, Croce A. Spontaneous nystagmus in benign paroxysmal positional vertigo. Am J Otolaryngol. 2011 May-Jun;32(3):185-9.
11. Brandt T, Huppert D, Hecht J, Karch C, Strupp M. Benign paroxysmal positioning vertigo: a long-term follow-up (6-17 years) of 125 patients. Acta Otolaryngol. 2006 Feb;126(2):160-3.
12. Del Rio M, Arriaga MA. Benign positional vertigo: prognostic factors. Otolaryngol Head Neck Surg. 2004 Apr;130(4):426-9.