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Clinical Characteristics and Natural Course of Recurrent Vestibulopathy: A Long-Term Follow-Up Study

Hong-Kyoung Lee, MD; Seong-Ki Ahn, MD, PhD; Sea-Yuong Jeon, MD, PhD; Jin-Pyeong Kim, MD; Jung Je Park, MD; Dong Gu Hur, MD; Dae Woo Kim, MD; Seung Hoon Woo, MD; Hung-Soo Kang, MD

Objectives/Hypothesis: To investigate the clinical characteristics and the natural course of recurrent vestibulopathy (RV).

Study Design: Retrospective study.

Methods: During the period April 2002 to February 2008, we reviewed the clinical records of 98 patients diagnosed with RV. All patients were approached by telephone and using a questionnaire. The analysis included age, sex distribution, natural history, pure-tone audiometry, caloric response, age at onset, and the characteristics of vertigo.

Results: Median follow-up was 63.1 months (range, 24–103 months). Patients had a mean age at onset of 39 years and a mean duration of 4.2 years. An obvious female predilection was found, and unilateral caloric paresis ($\geq$25%) was seen in 35%. Of the 98 patients, symptoms resolved in 82% but were unchanged in 12%. RV developed to Ménière's disease in four patients and to migraine in two. No patient with RV developed a central nervous system disease or benign paroxysmal positional vertigo during follow-up.

Conclusions: The study suggests that in the majority of cases, vertigo spontaneously resolves and that the risks of development to Ménière's disease or migraine are low.

Key Words: Vertigo, recurrence, follow-up study.

Level of Evidence: 2b

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INTRODUCTION

Recurrent vestibulopathy (RV) is a disease that displays recurrent symptoms of episodic vertigo that last for several minutes to several hours without auditory or neurologic signs or symptoms. Its clinical characteristics and follow-up results were first reported by Leliever and Barber[1] in 1981. To date, several studies have sporadically examined characteristics of RV, but few have investigated its course in the long-term.[1–3]

At dizziness clinics, patients complain of recurrent episodic vertigo at a much higher frequency than single episodic vertigo. Recurrent vertigo presents variable characteristics, and its frequencies also vary from several times a day to once in several years. The vertigo can last for seconds or several days, and sometimes it is accompanied by symptoms or signs such as hearing difficulty, migrainous headache, neurologic abnormalities, and others. No precise diagnosis of recurrent vertigo can be made by otoneurologic examination, vestibular function tests, or imaging studies because of the diversity of its clinical characteristics, and thus a definite diagnosis cannot be made in some cases.

The etiology of RV remains unclear, but its pathophysiology has been proposed to include endolymphatic hydrops, abnormal vascular compression of the eighth cranial nerve, recurrent viral infection, and others, and RV can develop into migraine, benign paroxysmal positional vertigo, or Ménière's disease.[4,5] However, neither the natural course nor the clinical features of RV have been determined, and therefore, the aim of this study was to document its clinical characteristics and natural course by long-term follow-up.

MATERIALS AND METHODS

We retrospectively reviewed the medical records of 138 consecutive RV patients who visited our tertiary referral neurotology and dizziness clinic with a chief complaint of two or more recurrent attacks of vertigo during the period April 2002 to February 2008. The clinical characteristics of vertigo, such as sex and age distribution, duration and frequency of vertigo, associated symptoms, migraine, and neurologic abnormalities, were reviewed, as were the results of hearing and vestibular function tests and brain magnetic resonance images.

RV was diagnosed as described by Leliever and Barber[1] in 1981. Times from onset of vertigo and attack duration were based on the durations of subjective complaints, and the frequencies of symptoms and improvement were based on patients' subjective answers.

From the Department of Otolaryngology (H.-K.L., S.-K.A., S.-Y.J., J.-P.K., J.J.P., D.G.H., D.W.K., S.H.W., H.-S.K.); and Institute of Health Sciences (S.-K.A.), School of Medicine, Gyeongsang National University, Jinju, South Korea.

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Send correspondence to Seong-Ki Ahn, MD, PhD; Department of Otolaryngology, School of Medicine, Gyeongsang National University, Jinju, 660-702, South Korea. E-mail: skahn@gnu.ac.kr

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TABLE I.
Dizziness Questionnaire.

Registration Number: _____ Name: _____ Gender/Age: ___

Did you have any attacks of vertigo since the last outpatient visit? Yes/No

– If yes, How many attacks did you have?

Has the number of attacks decreased, increased or remain unchanged?

Which of these best describes your dizziness?

A sensation of movement of yourself or the room: spinning, tilting, or wave-like movement

Lightheadedness or feeling that you are going to faint

Loss of balance

Disassociation or disorientation with the world

Is vertigo provoked by change in head position? Yes/No

Do you have any of the following symptoms?

Difficulty in hearing? Right/ Left

Noise in your ears? Right/ Left

Fullness or stuffiness in your ears? Right/ Left

Have you experienced headache during an attack of vertigo? Yes/No

– If yes,

Have you been diagnosed with migraine or tension headaches?

How would you describe your pain?

Tight band around your head?

Pounding pain in your head?

On one side of your head?

Over your eyes?

How long have you been having headaches?

Do you experience auras prior to the onset of your migraine?

___ Flashing lights ___ Zigzag lines

___ Facial numbness Other _____

Do you have any history of a neurological disease such as stroke? Yes/No

– If yes,

Have you experienced any of the following symptoms?

Yes No Double vision, blurred vision or blindness.
Yes No Numbness of face.
Yes No Numbness of arms or legs.
Yes No Weakness in arms or legs.
Yes No Clumsiness of arms or legs.
Yes No Confusion or loss of consciousness.
Yes No Difficulty with speech.
Yes No Difficulty with swallowing.
Yes No Pain in the neck or shoulder.

Patients with at least two recurrent vertigo attacks were included, but patients ≥65 years old were excluded to rule out presbyastasis. In addition, patients with sensorineural hearing loss, audiologic symptoms such as tinnitus and aural fullness, a migrainous headache, or neurologic abnormalities were excluded, as were patients with vertigo induced by a positional change, to rule out benign paroxysmal positional vertigo. Patients provided informed consent before taking part in the study. In February 2011, an otolaryngologist conducted telephone interviews with patients using a dizziness questionnaire, which contained questions about the presence of vertigo, accompanying symptoms (Table I), the characteristics of vertigo, hearing difficulties, tinnitus, aural fullness, and migraine. In addition, we also checked whether patients had been examined at other hospitals. Patients with persistent vertigo or suspected audiologic symptoms or migraine visited the outpatient clinic of the department of otorhinolaryngology and underwent an otoneurologic examination and pure-tone audiometry. Patients that had experience of vertigo during the past year were checked for the development of Ménière's disease, migraine, or benign paroxysmal positional vertigo. In addition, patients with recurrent episodic vertigo caused by RV without a change in clinical pattern over the previous year were defined as having active RV, and those with no experience of vertigo over this period were defined as having inactive RV.

All data were collected retrospectively. Statistical analyses were performed using SPSS software version 13.0 (SPSS Inc., Chicago, IL). The Student t test was used to compare age at onset and vertigo frequency in the active RV and inactive RV groups. Fisher exact test was used to compare age distributions and caloric paresis in both groups. $P < .05$ was considered statistically significant.

TABLE II.
Clinical Characteristics of Recurrent Vestibulopathy.

Parameter	Total, n = 98
Duration of follow-up, mo, mean (range)	63.1 (24–103)
Duration of disease, yr, mean (range)	4.2 (1–18)
Age of onset, yr, mean (± SD)	39.0 (± 14.0)
Sex, no. (%)	
Male	22 (22.4)
Female	76 (77.6)
Duration of attack, no. (%)	
5–10 min	6 (6.1)
10 min – 1 hr	8 (8.2)
1–6 hr	2 (2.0)
6–24 hr	60 (61.2)
>24 hr	22 (22.4)
Frequency, no. per yr, mean (± SD)	1.4 (± 0.9)
Caloric paresis, ≥25%, no. (%)	35 (35.7)

SD = standard deviation.

RESULTS

Clinical Characteristics

Of the 138 patients with a diagnosis of RV who had undergone follow-up for at least 3 years, 98 (71%) responded to the telephone interview. For these patients, mean follow-up was 63.1 months (range, 24–103 months), and mean time elapsed since disease onset was 4.2 years (range, 1–18 months). Our clinical series consisted of 22 men (22.4%) and 76 women (77.6%), and mean age at onset was 39 (±14) years. The duration of a vertigo attack was 5 to 10 minutes in six patients, 10 to 60 minutes in eight, 1 to 6 hours in two, 6 to 24 hours in 60, and more than 24 hours in 22 patients. The mean annual frequency of a vertigo attack was 1.4 (±0.9), and 35 (35.7%) of the 98 patients showed unilateral caloric paresis of over 25% in a caloric test (Table II).

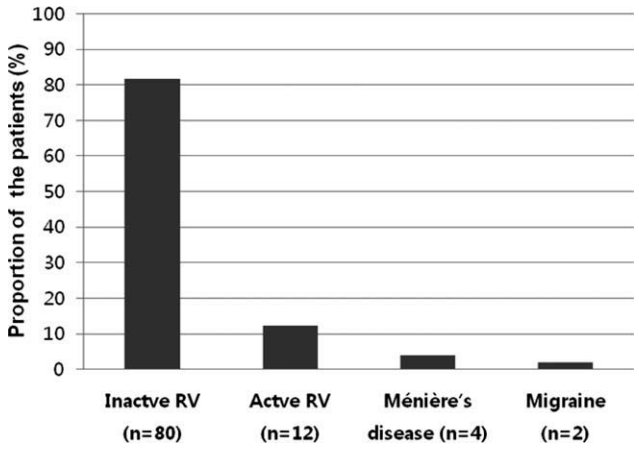

Fig. 1. Summary of natural course for patients with recurrent vestibulopathy (RV) during a mean follow-up period of ≈5.3 years.

Natural Course

During follow-up, vertigo symptoms disappeared in 80 patients (82%) (inactive RV), continued in 12 (12%) (active RV), and developed into Ménière's disease in four (4%) and into migraine in two (2%) (Fig. 1).

Prognostic Factors

We investigated whether any relevant variables were significantly different in the active RV (n = 12) and inactive RV (n = 80) groups. A significant intergroup difference was found for the frequency of vertigo (2.1 ± 1.0 vs. 1.3 ± 0.7 times per year) ($P < .05$) (Table III). However, no significant differences were found in terms of sex and age distributions or unilateral canal paresis by the caloric test.

DISCUSSION

RV is a recurrent episodic vertigo not accompanied by auditory or neurologic symptoms and not induced by a change in head position. Migraine-related vasoconstriction, viral infection, and endolymphatic hydrops have been suggested to be causes, but its pathogenesis and optimal treatment have not been determined. The clinical characteristics of RV have been described in the literature, but few studies have addressed its natural course in the long-term.[6–11]

The cause of RV is uncertain, and it may be a provisional diagnosis before differentiation in cases of suspected peripheral vestibular disorder. In patients with recurrent vertigo, a diagnosis cannot be made easily, as RV has no definite or clear diagnostic criteria, and an initial temporary diagnosis of RV may be changed to benign paroxysmal positional vertigo, migraine, or Ménière's disease.[4,5]

Previous studies have reported male-to-female ratios of 1:1.3 to 1:1.7 for RV,[1,12] but in the present study, the male-to-female ratio was 1:3.5, indicating a clear female predilection. The annual frequency of vertigo attack in the present study was 1.4 (±0.9) and was significantly higher in the active RV group than in the inactive RV group. In terms of duration of vertigo attacks, 61.2% of the 98 patients reported a duration of 6 to 24 hours. In an earlier study, the proportion of patients with unilateral caloric paresis was 17% to 22%.[1,12] It has also been reported that unilateral caloric paresis is less frequent in patients with a vertigo improvement than in those with continued vertigo.[12] However, in the present study, unilateral caloric paresis was not found to be related to the continuance of vertigo. Further studies are warranted to examine the association between the characteristics and presence of caloric paresis and RV.

Some authors have prescribed medications for Ménière's disease and migraine or administered vestibular suppressants to alleviate symptoms in patients with a diagnosis of RV, and others have sometimes prescribed medications for vestibular rehabilitation.[12,13] In the present study, depending on symptom duration and severity, vestibular suppressants, when administered, were only prescribed short-term. In four cases, RV developed into Ménière's disease, and symptoms were controlled by a low-salt diet and diuretics. In the two patients diagnosed with migraine, beta-blockers were administered, and symptoms improved accordingly.

Regarding diseases related to RV, Leliever and Barber[1] argued that RV differs from Ménière's disease, in the following ways. First, patients with RV do not have tinnitus and aural fullness as often as those diagnosed with vestibular Ménière's disease. Second, most RV

TABLE III.
Comparison of the Patient Data.

Parameter	Active RV, n = 12	Inactive RV, n = 80	Total, n = 92	*P* Value
Age of onset, yr, mean (± SD)	39.5 (± 10.2)	39.2 (± 14.9)	39.2 (± 14.3)	.9442*
Sex, no. (%)				
Male	4 (19.1)	17 (80.9)	21 (100.0)	.4598†
Female	8 (11.3)	63 (88.7)	71 (100.0)	
Frequency, no. per year, mean (± SD)	2.17 (± 1.25)	1.25 (± 0.73)	1.37 (± 0.87)	.0288*
Caloric paresis (≥25%), no. (%)	2 (6.3)	30 (93.7)	32 (100)	.2049†

*Student *t* test.
†Fisher exact test RV = recurrent vestibulopathy; SD = standard deviation.

cases do not develop into Ménière's disease; finally, RV does not show a change in the direction of nystagmus during each attack, as is observed in Ménière's disease. In the present study, only four of 98 (4%) patients developed Ménière's disease. Slater reported that a diagnosis of benign recurrent vertigo should be made if recurrent vertigo lasts for several hours to several days without impaired hearing or a neurologic abnormality.[14] However, this initial case series of seven patients showed a high prevalence of migraine. Subsequent reports also supported an association between recurrent vertigo and migraine,[14,15] but in the present study, only two of 98 (2%) patients developed migraine. The natural course of RV disease has been reported to be favorable, and the majority of patients with a diagnosis of RV reportedly recover from vertigo.[1–3,12] In the third review report issued on the subject, Rutka and Barber[3] found, over a mean follow-up period of 8.5 years, that initial diagnoses were changed to Ménière's disease in 14% of RV cases and to benign paroxysmal positional vertigo in 9% of cases and that in 70%, initial diagnoses were unchanged. In addition, the symptoms disappeared spontaneously in 90% and continued in only 10%. Furthermore, no case has been reported of RV developing into an abnormality of the central nervous system. In a report issued by van Leeuwen and Bruintjes,[12] 89 RV patients were followed up for a mean of 31 months. Of the 89, 62% experienced symptom improvement, 35% experienced no improvement, and 2% and 1%, respectively, developed into migraine or Ménière's disease. In the present study, during a mean follow-up period of ≈5.3 years, the initial diagnosis was changed to Ménière's disease in 4% of all cases and to migraine in 2%. Furthermore, no patient developed a central nervous system disease or benign paroxysmal positional vertigo during follow-up. Therefore, we speculate that RV is a separate pathophysiologic entity and not a precursor of migraine or Ménière's disease.

The limitations of the present study are as follows: Although telephone interviews provide a convenient means for collecting data, they suffer from recall bias; for example, in the present study, patients recalled their vertigo histories. However, no accurate, objective means of assessing vertigo history is currently available. As shown by the present study, many patients with RV believe that symptoms have improved over time. Accordingly, there may be a need to check by continuous follow-up whether RV has developed into other diseases in patients with persistent symptoms.

CONCLUSION

The present study indicates that most RV patients have a favorable prognosis, even in the absence of specific treatment. It also shows that only a small percentage of cases develop into migraine or Ménière's disease. In the present study, no RV patient developed a central nervous system disease or benign paroxysmal positional vertigo during follow-up. We suggest that meta-analysis be conducted to clarify the outcome and the mechanism underlying the pathogenesis of RV.

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