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Original Article

Co-existence of Benign Paroxysmal Positional Vertigo and Meniere's Syndrome

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OBJECTIVE: Recent studies indicate interrelation of benign paroxysmal positional vertigo (BPPV) and Meniere's disease (MD). These two entities may have different clinical characteristics.

MATERIALS and METHODS: Five hundred thirty patients with BPPV evaluated between 2009–2015 were enrolled in the study. 351 patients who had no clear problem associated with BPPV (idiopathic) and 17 patients with MD were analyzed in detail. The age, sex, site of involvement, type of BPPV, symptom duration, and treatment outcome were compared.

RESULTS: Meniere's disease + BPPV was more common in the female population (2/15; 7.5 v 127/224; 1.8, $p<0.05$). Comparative analysis of average age was not statistically significant (42.82±9.94 v. 40.29±1.65, $p=0.601$). There was no difference in right and left ear involvement between groups. Lateral canal involvement was more common in the BPPV + MD group (9/17; 53% v. 100/351; 28%, $p<0.05$). BPPV was ipsilateral to the ear with MD in 75% of patients and it was present before the diagnosis of BPPV in 82.3% of patients. Comparative analysis of cure rate between idiopathic BPPV and BPPV + MD after one session was significant (64.7% v 78%, $p<0.05$).

CONCLUSION: Benign paroxysmal positional vertigo associated with MD presented a divergent picture. It was more frequent in females. Lateral canal involvement was higher. Patients had MD before the development of BPPV and they had prolonged symptoms, which raised a question of diagnostic delay since these two problems were in the same ear in majority of patients. Finally, relief of symptoms required more attempts of repositioning maneuvers.

KEYWORDS: Benign paroxysmal positional vertigo, nystagmus, Meniere's syndrome

INTRODUCTION

Patients with benign paroxysmal positional vertigo (BPPV) frequently experience a sense of spinning during sudden head motion [1]. The pathophysiological mechanism that induces the positional vertigo is assumed to be due to freely floating otoconia inside the semicircular canals or those adhering to the cupula that make the labyrinth sensitive to gravitational forces. The origin of these deposits is claimed to be due to degeneration of utricular neuroepithelium, although the causative pathology is obscure [2]. In most cases, there is no clear cause of BPPV and it occurs spontaneously. However, BPPV can be associated with various other conditions such as trauma, viral neurolabyrinthitis, Meniere's disease (MD), and vertebral-basilar ischemia. Inner ear problems that may cause detachment of the otoconia could have an impact on the development of BPPV. The treatment outcome and recurrence in idiopathic and secondary cases of BPPV may have different aspects.

Fluctuating low frequency hearing loss and episodic bouts of vertigo and tinnitus are well-known symptoms of MD. MD and BPPV are two of the most common peripheral diseases seen in outpatient clinics of neurology and otolaryngology. Recent studies report the association of these two entities and even indicate an interrelation [3]. Maekawa et al. [4] have reported a patient with normal hearing who had been treated for BPPV previously. The patient developed Meniere-like episodic attacks one year later. Psillas et al. [5] have reported a patient with acute attack of MD who developed posterior canal BPPV in a few days. Very few studies have focused on secondary BPPV, which may be an underestimated entity. The clinical aspects of patients with BPPV associated with MD are documented in this study. Pathophysiological mechanisms are discussed and the treatment outcome is analyzed and compared with those having idiopathic BPPV.

MATERIALS and METHODS

Five hundred thirty patients with BPPV, confirmed by videonystagmography (VNG) (Micromed., Inc, Chatham, IL 62629, USA), who had been evaluated between 2009–2015 during outpatient clinical visit were enrolled in the study. Verbal informed consents were obtained from each patient. The procedures were in accordance with the ethical standards of the declaration of Helsinki and of the institutional review board. Distribution of sex, age, and affected side were reviewed. Patients were grouped as those with posterior canal BPPV (PC-BPPV), lateral canal

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BPPV (LC-BPPV), geotropic or apogeotropic type, and superior canal BPPV (SC-BPPV). Associated problems were noted. Five main subjects that might be directly related to otoconial degeneration were specifically questioned. These were migraine, vestibular neurinitis, MD, story of trauma, and otosclerosis. Chronic problems like diabetes, hypothyrodism, hypertension, high cholesterol, anemia, and vascular disease were also documented. Those who had no clear problem associated with BPPV were defined as "idiopathic". Patients with MD were analyzed in detail. Diagnosis of MD was based on guidelines from the committee on hearing and equilibrium, American Academy of Otolaryngology-Head and Neck Surgery Foundation [6].

Diagnosis of LC-BPPV with canalolithiasis or cupulolithiasis was based on medical history and the presence of geotropic or apogeotropic nystagmus (bi-directional, horizontal) during roll-on maneuver. The affected side for LC-BPPV was determined according to the severity of nystagmus as seen on the VNG recording, which was severe on the affected side in patients with geotropic nystagmus and was severe on the healthy side in patients with apogeotropic nystagmus. Diagnosis of PC-BPPV was based on medical history and the presence of clockwise or counter-clockwise rotatory nystagmus (torsional, up-beating) during the head-hanging maneuver. Diagnosis of SC-BPPV was based on medical history and the presence of clockwise or counter-clockwise rotatory nystagmus (torsional, down-beating) during the head-hanging maneuver. Patients with combined problems on the same or both ears were defined as multiple canal BPPV (MC-BPPV). Patients with PC-BPPV were treated with the Epley maneuver. Patients with SC-BPPV were treated with the Li or reverse-Epley maneuver. Patients with LC apogeotropic or geotropic nystagmus were treated with Barbeque, Semont, or Gufoni maneuvers. Therapeutic maneuvers were repeated during one session unless patients had no induced nystagmus. All patients were re-evaluated, at maximum, 7 days after each session. They were also requested to admit themselves to the clinic whenever they felt imbalance. Difference in sex, affected side, and treatment outcome was compared between patients with idiopathic BPPV and those associated with MD. Mean values and standard deviations (± SD) were calculated. A one-way ANOVA test was used for multiple variance analysis of the groups (Statistical Package for the Social Sciences 17.0 version, IBM, Chicago, IL, USA). Statistical significance was set at $p<0.05$.

RESULTS

Three hundred fifty patients with no chronic or other problems associated with BPPV were identified (Idiopathic group; 66.2%, 351/530). Of those, 127 were male and 224 were female. The Female to male ratio was 1.8 (224/127). The age ranged between 12 and 87. There were 17 patients with MD and BPPV (Meniere group; 3.2%, 17/530). All MD patients had low frequency neurosensorial hearing loss ranging from 25 to 45 dB. They all had at least two episodes of true vertigo. Their symptoms ranged from six months to nine years. Of those, two were male and fifteen were female. MD + BPPV was more common in the female population (7.5; 15/2). A comparative analysis of the sex ratio between BPPV with MD and idiopathic BPPV was statistically significant (7.5 v 1.8, $p<0.05$). Age ranged between 30 and 60 in BPPV patients associated with MD. A comparative analysis of average age between MD + BPPV and idiopathic BPPV was not statistically significant (MD + BPPV; 42.82±9.94 v. Idiopathic; 40.29±11.65, $p=0.601$). Eight patients had right-sided and eight patients had left-sided MD. One patient had bilateral disease. However, BPPV was on the left side in 10 and on the right side in 7 patients in the BPPV + MD group. BPPV was on the right ear in 156 patients and on the left ear in 144 patients with idiopathic BPPV. There was no difference in side of involvement between the groups.

Table 1. Co-existence of MD + BPPV in reported studies

	Study by	Year	Patients	Incidence
MD among BPPV	Katsarkas et al.	1978	255	0.8%
	Baloh et al.	1987	240	2.1%
	Hughes et al	1997	151	31%
	Karlberg et al.	2000	2847	0.5%
	Monobe et al.	2001	62	3.2%
	Tanimato et al.	2008	145	15%
	Lee et al.	2010	718	2.8%
	Balatsouras et al.	2012	345	8.4%
	This study	2016	530	3.2%
BPPV among MD	Gross et al.	2000	162	5.5%
	Proctor et al.	2000	122	44%
	Perez et al.	2002	90	10%

MD: Meniere's disease; BPPV: benign paroxysmal positional vertigo

Table 2. Comparative analysis of clinical characteristics of patients with idiopathic BPPV and BPPV associated MD

		BPPV + MD	Idiopathic BPPV	p
Sex	Male	2/17 (12%)	127/351 (36%)	
	Female	15/17 (88%)	224/351 (64%)	
	Ratio	7.5 (2/15)	1.8 (127/224)	p<0.05
Age	Mean	42.82±9.94	40.29±11.65	p=0.601
	Range	30–60	12–87	
Symptoms >2 months		12/17 (75%)	85/351 (24%)	p<0.05
Side of BPPV	RE	10/17 (59%)	156/300 (52%)	p=0.803
	LE	7/17 (41%)	144/300 (48%)	p=0.765
SCC involved	PC	8/17 (47%)	192/351 (55%)	p=0.354
	LC	9/17 (53%)	100/351 (28%)	p<0.05
	SC	-	8/351 (2%)	
	Multiple	-	51/351 (15%)	
Relief after one session		11/17 (64.7%)	273/351 (78%)	p<0.05

BPPV: benign paroxysmal positional vertigo; MD: Meniere's disease; PC: posterior canal; LC: lateral canal; SC: superior canal; RE: right ear; LE: left ear

Eight patients with MD had PC-BPPV. Five patients had apogeotropic and four patients had geotropic-type LC-BPPV. Excluding the patient with bilateral MD, BPPV was ipsilateral to the ear with MD in twelve patients (12/17; 75%). Of those, five were PC, four were apogeotropic LC and three were geotropic LC-BPPV. Fourteen patients had documented MD before the diagnosis of BPPV (82.3%; 14/17). However, three patients with BPPV later developed sudden low frequency hearing loss and episodic vertigo of MD. Twelve patients having BPPV associated with MD had symptoms for more than two months (12/17; 75%). Eleven patients were cured after a single session (64.7%; 11/17). 273 patients with idiopathic BPPV were cured after one session (78%). Comparative analysis of the cure rate between idiopathic BPPV and BPPV associated with MD after one session was significant (64.7% v 78%, $p<0.05$). Relevant comparative data and literature analyses are presented in Table 1 and 2.

DISCUSSION

Inner ear problems in patients with BPPV have been the subject of many studies. The incidence of coexistence of BPPV and MD ranges from 0.5% to 44% [7-9]. The diversity between the reported incidences could be due to differences in selected patient population, study method, or follow-up criteria (Table 1). Proctor reported the highest incidence. However, their MD patients were those who had serial vestibular testing and frequent follow-up [9, 10]. One of the largest series was reported by Lee et al. [11], and they found an inner ear disease in 9% of 718 patients with BPPV. Almost one third of them were MD (2.8%). The incidence of BPPV associated with MD in this series was 3.2% and it is close to the previous reports [11-15]. Differential diagnosis of peripheral vestibular problems is primarily based on the patient's story. Duration and frequency of symptoms are the main keys to eliminate the other problems. However, diagnostic and treatment difficulties may arise especially when two balance problems occur in the same patient, simultaneously. Taura et al. [16] have reported that 32.8% of patients with MD actually had BPPV-like attacks. Yetişer [17] reported a video of simultaneous presentation of positional nystagmus of PC-BPPV and spontaneous nystagmus of MD in the same patient. These reports support the possibility of a diagnostic delay of BPPV in patients with MD since many clinicians avoid testing the positional nystagmus if the patient has spontaneous nystagmus at the primary gaze position.

Meniere's disease and BPPV were on the same side in 75% of patients in this series. Predominance of ipsilateral occurrence of BPPV and MD have been reported in the majority of studies that indicate a causal relationship between the two disorders. Three basic assumptions may apply. For patients presenting symptoms of MD and BPPV simultaneously, it is likely to hypothesize that these two clinical disorders may share a common pathophysiological etiology. Circulating debris inside the membranous labyrinth, leading to positional nystagmus of BPPV, could also cause blockage of the endolymphatic ductus. However, 82% of patients having BPPV and MD in this series had MD symptoms long before the diagnosis of BPPV. Therefore, another hypothesis could be proposed for the co-existence of these two vestibular problems. Repeated episodes of hydrops may eventually result with sacculo-utricular degeneration and detachment of the otoconia. This assumption may also explain the necessity of several attempts for relief of BPPV symptoms in those patients. Recurrence of BPPV may be related with the episodic degenerative process of MD. The final assumption could be the sleeping habit. Patients with unilateral hearing loss generally prefer to sleep lying on the ear with hearing loss to keep the better hearing ear in the open environment. Shim et al. [18] and Sato et al. [19] have reported that sleeping habit is closely related with the effected side in BPPV. Otoconial debris dislodged from the utricle may fall into the lateral or posterior semicircular canals of the undermost ear during sleep. This may explain more the common lateral canal involvement and ipsilateral occurrence of MD and BPPV.

Benign paroxysmal positional vertigo secondary to the inner ear problem is prone to recur and the rate of symptomatic relief in one session is significantly low compared to those with idiopathic cases. Rupa [20] reported that all patients with persistent vertigo following particle repositioning maneuvers had coincidental pathology. Monobe et al. [12] have reported that 9.7% of patients with PC-BPPV had no resolution and they had secondary problems. Dornhoffer and Colvin [21] have reported that recurrence rates appeared to be correlated with the presence of MD. Del Rio and Arriaga [22] have reviewed the prognostic factor in BPPV. They have reported 22.6% recurrence, and the endolymphatic hydrops was the leading one. Tanimato et al. [23] have reviewed risk factors in recurrent BPPV. They have found that 75% of them have endolymphatic hydrops and all were in the same ear with BPPV. Perez et al. [24] have proposed intratympanic gentamycin associated with repositioning procedures in those patients. All these reports focused on prognostic factors in BPPV patients with persistent or recurrent vertigo after repositioning maneuvers, and comparative data with other group of patients with BPPV were lacking. Comparison of the number of patients who had relief of symptoms after one session of treatment in idiopathic and BPPV + MD group was statistically significant in this series.

In conclusion, BPPV associated with MD presents divergent aspects compared to idiopathic BPPV. It was more frequent in females and lateral canal involvement was much higher. The majority of patients had MD before the development of BPPV and they had prolonged symptoms before the diagnosis of BPPV, which raised a question of diagnostic delay since these two problems were on the same ear in the majority of patients. Finally, relief of symptoms required more attempts of repositioning maneuvers.

Ethics Committee Approval: Ethics committee approval was received for this study from the ethics committee of Anadolu Medical Center.

Informed Consent: Verbal informed consent was obtained from the patients who participated in this study.

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