Treatment of Mal de Debarquement Syndrome in a Computer-Assisted Rehabilitation Environment

LTC Carrie W. Hoppes, SP, USA[*]*; Michael Vernon*[†]*; MAJ Rebecca L. Morrell, SP, USA*[‡]*; Susan L. Whitney DPT, PhD*[§]

ABSTRACT Individuals with mal de debarquement syndrome (MdDS) describe symptoms of swaying, rocking, and/or bobbing after sea or air travel. These symptoms may be because of maladaptation of the vestibulo-ocular reflex (VOR) to roll of the head during rotation. Dai and colleagues have developed a treatment paradigm that involves passive roll of the patient's head while watching optokinetic stripes, resulting in adaption of the VOR and improvement of MdDS. The purpose of this case report is to describe replication of this treatment paradigm in a virtual reality environment with successful resolution of symptoms in two visits. A 39-year-old female reported swaying and rocking after returning from a 7-day cruise. The patient was treated with two sessions in a computer-assisted rehabilitation environment (CAREN). The patient canceled her third visit because of complete resolution of her symptoms. Her Global Rating of Change was +7 (on a 15-point scale of −7 to +7). She had returned to her prior level of function. This case report is the first to describe use of the CAREN for effective treatment of MdDS by replicating the treatment paradigm developed by Dai and colleagues.

INTRODUCTION

Mal de debarquement is characterized by constant symptoms of swaying, rocking, and/or bobbing and commonly occurs after sea travel.[1] In a survey of 234 healthy crew members, 73% of the naval personnel reported having experienced mal de debarquement.[2] For 93% of those naval personnel, the symptoms of mal de debarquement lasted less than 6 hours.[2] Some individuals do not spontaneously recover, and symptoms can last from months to years. Mal de debarquement syndrome (MdDS) describes a motion-induced onset of mild unsteadiness and anxiety symptoms that are minimally relieved with antivertiginous medication, last months to years, and occur in the presence of normal neurologic and vestibulo-ocular examinations.[3] Hain et al. conducted a survey of individuals with MdDS and found that nearly all were middle-aged women who had returned from a cruise.[4] Meclizine or scopolamine was not helpful in alleviating their symptoms.[4]

The mechanism of MdDS is unclear. Changes in resting state metabolism, functional connectivity, and gray matter volume have been found in individuals with MdDS.[5,6] Mal de debarquement syndrome subjects have hypermetabolism in the left entorhinal cortex and amygdala and relative hypometabolism in the left superior medial gyrus, left middle frontal gyrus, right amygdala, right insula, as well as in clusters in the left superior, middle, and inferior temporal gyri.[5] Mal de debarquement syndrome subjects also had increased functional connectivity between the entorhinal cortex/amygdala cluster and posterior visual and vestibular processing areas and decreased functional connectivity to multiple prefrontal areas.[5] Mal de debarquement syndrome subjects had increased gray matter volume in the left inferior parietal lobule, right inferior occipital gyrus, right temporal pole, bilateral cerebellar hemispheric lobules VIII/IX, and left cerebellar hemispheric lobule VIIa/VIIb.[6] Mal de debarquement syndrome subjects had decreased gray matter volumes in the bilateral inferior frontal, orbitofrontal, pregenual anterior cingulate cortex, and left superior medial gyri.[6] Illness duration–dependent changes in gray matter volume were also observed (notably, decreased volume in the pregenual anterior cingulate cortex and increased volume in the left inferior frontal gyrus/anterior insula with longer illness duration).[6] These structural and functional changes may affect central integration of visual, vestibular, and somatosensory information. Citing evidence from monkeys and humans, Dai et al. proposed that MdDS is due to maladaptation of the vestibulo-ocular reflex (VOR).[7] The velocity storage integrator may be a possible explanation of the development of MdDS.[8,9]

Dai et al. have proposed a treatment paradigm that involves passive roll of the patient's head while watching full-field optokinetic stripes, resulting in adaption of the VOR and improvement of MdDS.[7] The frequency of perceived rocking was determined by either of two means: (1) using an accelerometer attached to the wrist, subjects made arm movements synchronously with their perceived internal sense of

[*]U.S. Army Medical Center of Excellence, Army-Baylor University Doctoral Program in Physical Therapy, Fort Sam Houston, TX 78234, USA

[†]Center for the Intrepid, Department of Rehabilitation Medicine, Brooke Army Medical Center, Fort Sam Houston, TX 78234, USA

[‡]Physical Therapy Services, Department of Rehabilitation Medicine, Brooke Army Medical Center, Fort Sam Houston, TX 78234, USA

[§]Department of Physical Therapy, University of Pittsburgh, Pittsburgh, PA 15219, USA

The views and information presented are those of the authors and do not represent the official position of the U.S. Army Medical Center of Excellence, U.S. Army Training and Doctrine Command, Department of the Army, Defense Health Agency, Department of Defense, or U.S. Government.

doi:10.1093/milmed/usab077

motion and (2) via postural sway recorded on a force plate. The direction of the optokinetic stripes was determined by one of three means: (1) video-oculography recording of vertical nystagmus during passive head roll at the previously determined frequency of perceived rocking, (2) rotation in the Fukuda stepping test,[10] or (3) based on the direction of perceived rotation. The subject's head was passively rolled ±20° at the frequency of their perceived rocking while watching the optokinetic stripes. The velocity of the stripes started at 10°/s but was manipulated according to the subject's response. Treatments lasted for 3–5 minutes, with 1–8 treatments per day for up to 5 days over a 1-week period. In 24 subjects, 70% were asymptomatic following the intervention described above.[7] One year later, significant improvement was maintained in 52% of subjects and 27% had complete remission of symptoms.[11] As the first and only successful treatment for MdDS, it is known as the Dai treatment for the MdDS.[12] Improvements in symptoms and postural stability following the Dai treatment were supported by a sham-controlled study.[13]

The positive results reported by Dai et al.[7] inspired the idea to replicate full-field optokinetic stripes and use passive roll motion to treat a patient with MdDS who presented to our clinic. The hope was to offer the patient relief and demonstrate the capability to replicate the intervention. The purpose of this case report is to describe application of the Dai et al.[7] treatment paradigm in a virtual reality environment with successful resolution of symptoms in two visits.

CASE REPORT

A 39-year-old African American female (1.6 m, 74.4 kg, 27.28 body mass index) returned from a 7-day cruise. She was seen by her primary care physician, who referred her to a physical therapist specializing in vestibular rehabilitation. The patient was treated with two sessions in a computer-assisted rehabilitation environment (CAREN) (Motek Medical, Houten, Netherlands) at the Center for the Intrepid, Fort Sam Houston, TX. The CAREN has a dual-belt instrumented treadmill on a 6-degree-of-freedom moveable platform within a full field-of-view virtual and augmented (surround sound) reality environment. The course of the patient's care is described below.

Primary Care Evaluation

The patient was an active duty U.S. Army human resources officer. She was physically active, engaging in 150 minutes of moderate-intensity exercise per week and muscle strengthening activities at least two times per week. The patient presented to her primary care physician 1 week after returning from a cruise with a chief complaint of dizziness and imbalance. She reported dizziness, lightheadedness, spinning, and imbalance. The patient also reported nasal discharge (runny nose), nasal passage blockage (stuffiness), and mucous in the back of her throat. She denied any nausea or visual disturbance.

The patient's past medical history included hypertension, low back pain, and headaches. Her current medications included 10 mg cetirizine daily; 80 mg/25 mg hydrochlorothiazide daily; 30 mg nifedipine extended release daily; 500 mg naproxen twice daily as needed; 1.5 tablets of 100 mg sertraline hydrochloride at bedtime; 1–2 tablets of 25 mg hydroxyzine at bedtime as needed for insomnia; 10 mg buspirone twice daily; 1 tablet of 50/325/40 mg acetaminophen/butalbital/caffeine every 4–6 hours as needed; 10 mg cyclobenzaprine at bedtime as needed for muscle spasm; analgesic balm as needed; and an over-the-counter multi-vitamin daily.

The physical examination was unremarkable, except for fluid in both middle ears and an erythematous left tympanic membrane. Nasal discharge was also seen. The patient did not have a fever (temperature was 36.8°C).

She was diagnosed with left ear otitis media and instructed to increase her fluid intake and take acetaminophen as needed. She was prescribed 1 tablet 125 mg amoxicillin/clavulanate potassium 875 twice daily for 10 days. She was also prescribed a sinus rinse with instructions to wash her sinuses every day and as needed. Additionally, the patient was referred to physical therapy with a diagnosis of bilateral benign paroxysmal positional vertigo for a trial of repositioning maneuvers. She was prescribed 1 tablet 25 mg meclizine three times daily as needed for dizziness. A comprehensive metabolic panel was ordered as part of the work-up for dizziness; only serum glucose was noted to be high. The patient was instructed to follow up with her primary care physician in 3 weeks or sooner if there were problems.

Physical Therapy Evaluation

The patient, referred for bilateral benign paroxysmal positional vertigo, presented to her physical therapist 26 days after returning from a cruise with a chief complaint of an internal sense of swaying and rocking. She described this mostly as medial–lateral but also anterior–posterior. She stated that the symptoms began on the second day of the 7-day cruise and persisted. She felt like she was going to fall off the treadmill when on the ship and felt off balance when walking on the ship. This had never occurred on previous cruises and she denied a history of motion sickness. She reported intermittent, bilateral tinnitus that she believed was unrelated to her symptoms of swaying and rocking. After returning from the cruise, running on the treadmill aggravated her symptoms, while focusing on a point when driving eased her symptoms. She reported an intermittent sense of self-motion and imbalance that was variable in intensity and mild nausea. She affirmed that she was recently treated for otitis media with an antibiotic and sinus rinse; she had consumed all 30 tablets of meclizine. She denied a headache at the time of examination but did have a history of headaches. Additionally, she had a history of depression. Her patient goal was to resolve her symptoms of swaying and rocking, stating that she wanted to "fix [the] imbalance issues in my head."

Her Dizziness Handicap Inventory[14] score was 30 (indicating mild dizziness[15]), and her Hospital Anxiety and Depression Scale score was 13 for anxiety and 4 for depression. A cutoff score of 8 on the Hospital Anxiety and Depression Scale was used to identify anxiety and depression.[16] Her Vestibular Activities Avoidance Instrument score was 237 (total score ranges from 0 to 484 with higher scores indicating greater avoidance beliefs/anxiety). The patient marked "strongly agree" for the item "I am aware of bodily sensations much of the time". She also agreed with several items relating to kinesiophobia. Most notably, the patient marked "strongly agree" for the item "Being careful that I do not make any unnecessary movements is the safest thing I can do to prevent my dizziness/unsteadiness from worsening". She marked "agree" for the item "I am afraid that I might make myself dizzy or unsteady if I exercise" and "I should not do physical activities, which might make my dizziness/unsteadiness worse". She also marked "somewhat agree" for the item "I cannot do physical activities, which might make my dizziness/unsteadiness worse".

She rated her dizziness/unsteadiness as moderately bothersome in the last 2 weeks. She reported completely missing at least 1 day and limiting her participation at least 4 days during scheduled activities in social or community events in the last 2 weeks because of her dizziness.

Her Functional Gait Assessment[17] and modified Clinical Test of Sensory Interaction in Balance[18] scores were normal. Her cervical spine range of motion was normal and pain free. Bedside assessment of the vestibular system was normal. In room light, there was no spontaneous, end gaze, or gaze-holding nystagmus. Ocular range of motion, vergence, smooth pursuit, and saccades were normal. The cover/uncover test was unremarkable. The VOR and ability to cancel the VOR were intact. The head impulse test was negative bilaterally. With Frenzel goggles, bilateral Dix-Hallpike and roll tests were negative for nystagmus and symptoms. The patient was diagnosed with MdDS. Up to eight sessions in the CAREN were planned.

Treatment Session 1

The patient completed the Motion Triggered Questionnaire.[19] Her pretreatment severity of MdDS was rated as 1 (0, asymptomatic, to 10, most severe).[7] She reported that walking from the parking garage to the clinic had aggravated her symptoms of swaying and rocking. Two trials of the Fukuda stepping test[10] were positive to the right. Although 1-minute Fukuda stepping tests were planned, each test was terminated early to avoid the patient walking into a clinic cabinet. Each time, the patient veered 90° to the right and advanced 1 m.

The patient wore a safety harness and was accompanied by the physical therapist on the CAREN platform. The patient sat in a chair in the middle of the CAREN, and the physical therapist was in tall-kneeling behind her. The patient struggled to identify her perceived rocking frequency; therefore, the physical therapist selected the speed of roll motion based on clinical experience. Her head was passively rolled at 50 bpm while watching optokinetic stripes in yaw moving at 1.5°/s (speed 2) from right to left for 5 minutes (Fig. 1). The patient had a seated rest break outside of the CAREN until she felt comfortable with proceeding. She then walked on the right-hand sidewalk of a virtual reality city scene at 0.85 m/s for 10 minutes, with head turns to identify objects in the scene (Fig. 2). After walking in the CAREN, she reported "a little blurriness" (a new symptom) and she felt like when she looked to the right she moved to the right. Her posttreatment severity of MdDS was rated as 2 (immediately after treatment); within this session, her symptoms had increased slightly from baseline to posttreatment. The patient tolerated the treatment session well without adverse effects.

Treatment Session 2

The following day, the patient's pretreatment severity of MdDS was again rated as 1. The patient and therapist were in the same position as the previous session. Her head was passively rolled at 60 bpm while watching optokinetic stripes in yaw moving at 3.0°/s (speed 5) from right to left for 5 minutes. The patient had a seated rest break outside of the CAREN

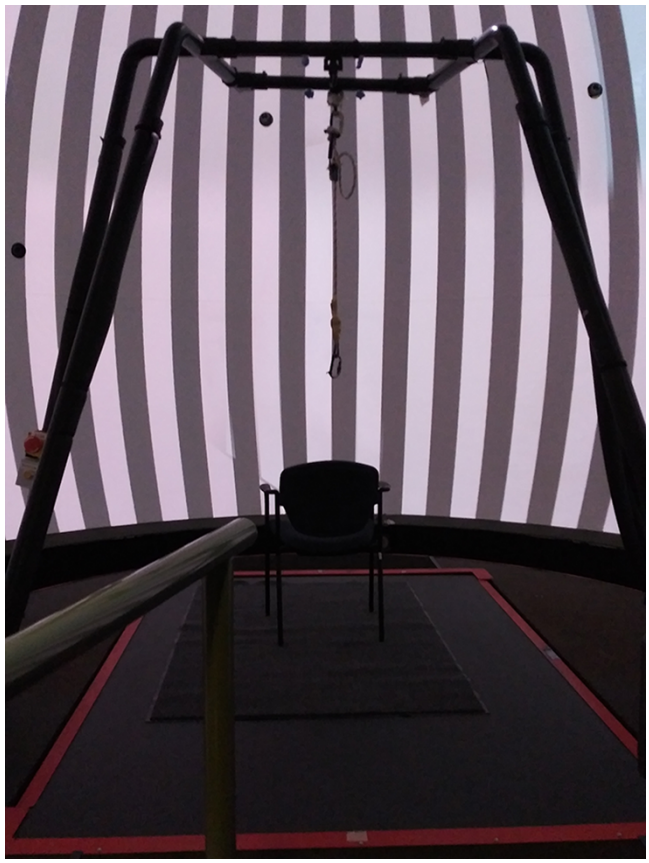

FIGURE 1. Optokinetic stripes in a computer-assisted rehabilitation environment for use during treatment of mal de debarquement syndrome. The patient is seated in the chair during treatment.

FIGURE 2. Virtual reality city scene in a computer-assisted rehabilitation environment for use during treatment of mal de debarquement syndrome.

until she felt comfortable with proceeding. She then walked on the right-hand sidewalk of a virtual reality city scene at 0.85 m/s for 10 minutes, with head turns to identify objects in the scene. Medial–lateral perturbations of the visual surround (visual field translations in the *x*-axis) were introduced during this walking task. The medial–lateral visual oscillations were driven by our standard pseudo-random algorithm [(SIN(TIME) + 0.8 × SIN(TIME × 1.3) + 1.4 × SIN(TIME × 1.5) + 0.5 × SIN(TIME × 3.1)) × 0.5] (note: time in seconds and distance in meters) and further scaled down by 0.25% and 0.50% overall as necessary for the patient. Her posttreatment severity of MdDS was rated as 1 (immediately after treatment); within this session, her symptoms had remained the same from baseline to posttreatment. The patient tolerated the treatment session well without adverse effects.

RESULTS

The patient elected to cancel her third visit because of complete resolution of her symptoms (36 days after returning from the cruise). Her Global Rating of Change was +7 (on a 15-point scale of −7 to +7[20]), her severity of MdDS was rated as 0, and her Dizziness Handicap Inventory score was 0. She had returned to her prior level of function.

DISCUSSION

The patient experienced MdDS after sea travel, which was the most common mode of triggering event reported by Arroll et al.[21] in their survey of 66 persons with MdDS. The patient met Arroll et al.'s[21] criteria for MdDS; her primary symptom was chronic rocking that started within 2 days of getting off a boat and lasted at least 2 hours plus other diagnoses were excluded based on physician examination. Other authors have required the person to have had symptoms for at least 1 month for diagnosis of MdDS.[4,5,22,23] A systematic review has proposed diagnostic guidelines for motion-triggered MdDS.[24]

Unlike other reports, our patient was diagnosed quickly. Macke et al. reported that the mean number of visits to receive a diagnosis of MdDS was 19.[22] The quick diagnosis of our patient was unusual and may have been why she recovered so quickly. She returned to her prior level of function following resolution of her MdDS, while return to work was only reported by 61% of persons with MdDS surveyed by Arroll et al.[21] This case report supports direct access to physical therapy, as the time to diagnosis could have been further reduced if she had presented directly to physical therapy instead of first seeing her primary care physician for her complaint of dizziness and imbalance.

In Arroll et al.'s report of 66 persons with MdDS, 91% of the subjects were women with a mean age of 52 ± 12 years.[21] Our patient was a woman who fell within 1 SD of subjects in the Arroll et al. report. The patient described the classic symptom of rocking. Like our patient, others have reported postural instability associated with MdDS.[7,25,26] Although she reported symptoms, her Functional Gait Assessment and modified Clinical Test of Sensory Interaction in Balance scores were normal. Otologic symptoms, like our patient's complaint of tinnitus, have been reported in 89% of persons with MdDS.[4] Migraine has been associated with MdDS.[27] Our patient reported a history of headache but no migraines. Clark et al. reported that persons with MdDS demonstrated kinesiophobia and reported fatigue but not depressive symptoms in comparison to control subjects.[26] Similarly, our patient reported agreement with several items related to kinesiophobia on the Vestibular Activities Avoidance Instrument. Our patient reported a history of depression before the development of her symptoms but no additional mood concerns.

The positive results reported by Dai et al.[7] inspired the idea to replicate full-field optokinetic stripes and use passive roll motion to treat the patient with MdDS who presented to our clinic. Replicating the treatment paradigm in a virtual reality environment was not without its challenges. A CAREN engineer operator (M.V.) had to build the optokinetic stripe scenario from the details provided by Dai et al.[7] A limitation to delivering this treatment for MdDS is the cost and availability of the CAREN, in addition to the need for a specialized engineer operator to build the scenario and run the system.

We did not have the capability to determine the frequency of perceived rocking using an accelerometer attached to the

wrist or by recording postural sway on a force plate. Additionally, the patient struggled to identify her perceived rocking frequency. Therefore, the physical therapist used her clinical expertise to select the speed of roll motion. Further research is needed to determine a clinically feasible means to identify the frequency of perceived rocking and if treatment success is predicated upon individualized treatment parameters for frequency of passive roll motion. We were able to determine the direction of the optokinetic stripes based on the patient's rotation during the Fukuda stepping test. The patient's head was passively rolled $\pm 20^{\circ}$ at the selected frequency while watching the optokinetic stripes. The velocity of the stripes started at 1.5°/s (speed 2) and was manipulated according to the patient's response by increasing to 3.0°/s (speed 5) for the second treatment session. The patient tolerated treatment for 5 minutes without adverse effects. Remarkably, only one treatment per day for 2 days was required for complete symptom resolution. Her Global Rating of Change score of +7 indicated she felt completely recovered. Mucci et al.[13] found that the greatest postural changes occurred within the first 3 days of treatment, supporting a shorter course of treatment than first proposed by Dai et al.[7]

CONCLUSIONS

This case report is the first to demonstrate the capability to apply the Dai treatment for the MdDS[7] using the CAREN. Two sessions resolved her symptoms and improved her quality of life.

ACKNOWLEDGMENT

The authors wish to thank the individual with MdDS for allowing us to share her story.

FUNDING

No funding was received for this work.

CONFLICT OF INTEREST STATEMENT

No conflicts of interest.

REFERENCES

1. Darwin E: *Zoonomia*. J Johnson; 1796.
2. Gordon CR, Spitzer O, Shupak A, Doweck I: Survey of mal de debarquement. BMJ: Br Med J 1992; 304(6826): 544.
3. Brown JJ, Baloh RW: Persistent mal de debarquement syndrome: a motion-induced subjective disorder of balance. Am J Otolaryngol 1987; 8(4): 219–22.
4. Hain TC, Hanna PA, Rheinberger MA: Mal de Debarquement. Arch Otolaryng Head Neck Surg 1999; 125(6): 615–20.
5. Cha Y-H, Chakrapani S, Craig A, Baloh RW: Metabolic and functional connectivity changes in mal de debarquement syndrome. PLoS One 2012; 7(11): e49560.
6. Cha Y-H, Chakrapani S: Voxel based morphometry alterations in mal de debarquement syndrome. PLoS One 2015; 10(8): e0135021.
7. Dai M, Cohen B, Smouha E, Cho C: Readaptation of the vestibulo-ocular reflex relieves the mal de debarquement syndrome. Front Neurol 2014; 5: 1–6.
8. Raphan T, Matsuo V, Cohen B: Velocity storage in the vestibulo-ocular reflex arc (VOR). Exp Brain Res 1979; 35(2): 229–48.
9. Cohen B, Yakushin SB, Cho C: Hypothesis: the vestibular and cerebellar basis of the mal de debarquement syndrome. Front Neurol 2018; 9: 1–11.
10. Fukuda T: The stepping test: two phases of the labyrinthine reflex. Acta Otolaryngol 1959; 50(1–2): 95–108.
11. Dai M, Cohen B, Cho C, Shin S, Yakushin SB: Treatment of the mal de debarquement syndrome: a 1-year follow-up. Front Neurol 2017; 8: 1–10.
12. Cohen B: Dedication to Mingjia Dai, Ph.D. for discovery of the first successful treatment of the mal de debarquement syndrome. Front Neurol 2019; 10: 1–3.
13. Mucci V, Perkisas T, Jillings SD, et al: Sham-controlled study of optokinetic stimuli as treatment for mal de debarquement syndrome. Front Neurol 2018; 9: 1–13.
14. Jacobson GP, Newman CW: The development of the Dizziness Handicap Inventory. Arch Otolaryng Head Neck Surg 1990; 116(4): 424–7.
15. Whitney SL, Wrisley DM, Brown KE, Furman JM: Is perception of handicap related to functional performance in persons with vestibular dysfunction? Otol Neurotol 2004; 25(2): 139–43.
16. Bjelland I, Dahl AA, Haug TT, Neckelmann D: The validity of the Hospital Anxiety and Depression Scale. An updated literature review. J Psychosom Res 2002; 52(2): 69–77.
17. Wrisley DM, Marchetti GF, Kuharsky DK, Whitney SL: Reliability, internal consistency, and validity of data obtained with the functional gait assessment. Phys Ther 2004; 84(10): 906–18.
18. Cohen H, Blatchly CA, Gombash LL: A study of the clinical test of sensory interaction and balance. Phys Ther 1993; 73(6): 346–51.
19. Canceri JM, Brown R, Watson SR, Browne CJ: Examination of current treatments and symptom management strategies used by patients with mal de debarquement syndrome. Front Neurol 2018; 9: 1–13.
20. Stratford PW, Binkley JM, Riddle DL: Health status measures: strategies and analytic methods for assessing change scores. Phys Ther 1996; 76(10): 1109–23.
21. Arroll MA, Attree EA, Cha Y-H, Dancey CP: The relationship between symptom severity, stigma, illness intrusiveness and depression in mal de debarquement syndrome. J Health Psychol 2016; 21(7): 1339–50.
22. Macke A, LePorte A, Clark BC: Social, societal, and economic burden of mal de debarquement syndrome. J Neurol 2012; 259(7): 1326–30.
23. Cha Y-H, Cui Y: Rocking dizziness and headache: a two-way street. Cephalalgia 2013; 33(14): 1160–69.
24. Van Ombergen A, Van Rompaey V, Maes LK, Van De Heyning PH, Wuyts FL: Mal de debarquement syndrome: a systematic review. J Neurol 2016; 263(5): 843–54.
25. Nachum Z, Shupak A, Letichevsky V, et al: Mal de debarquement and posture: reduced reliance on vestibular and visual cues. Laryngoscope 2004; 114(3): 581–6.
26. Clark BC, LePorte A, Clark S, et al: Effects of persistent mal de debarquement syndrome on balance, psychological traits, and motor cortex exctiability. J Clin Neurosci 2013; 20(3): 446–50.
27. Cha YH, Brodsky J, Ishiyama G, Sabatti C, Baloh RW: Clinical features and associated syndromes of mal de debarquement. J Neurol 2008; 255(7): 1038.