Case Report

Menière's disease and vestibular migraine in a patient with temporomandibular disorder: a case report

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Background: Multiple chronic conditions may affect the craniofacial area, several of which are often rare and the overlapping clinical features of these conditions, often present a diagnostic challenge to health care providers. Thus, a multidisciplinary approach and the central role of the dentist in the treatment of these conditions are of growing importance.

Case Description: An 18-year-old Caucasian male presented on March 27th, 2021 with a chief complaint of pre auricular pain associated with a history of vertigo, dizziness, tinnitus, headache and a recent onset of hearing loss and was evaluated in the Orofacial Pain Clinic. A physical examination was performed, and further investigations were suggested (imaging and serological testing). After a consultation with the neurologist and the otorhinolaryngologist, the diagnosis of probable Menière's disease (MD), vestibular migraine (VM), and temporomandibular disorder (TMD) was made. The treatment was multidisciplinary and included prophylactic pharmacologic management for MD and VM with propranolol, vestibular rehabilitation to improve the vestibular dysfunction, behavioral modification, cognitive behavioral therapy with a psychologist to cope with tinnitus and general psychological distress and finally an occlusal appliance, physical therapy and trigger point injections were suggested for the TMDs. At a three-month follow up, a general improvement of the conditions was reported. To our knowledge, this is the first case of MD associated with VM and TMDs. No other cases with similar features have been published

Conclusions: To the best of the authors' knowledge, this is a rare reported case of MD associated with VM and TMDs. The complex symptomatology presented in the case report demonstrates the importance of the orofacial pain specialist in the diagnosis, management, and treatment of orofacial pain disorders.

Keywords: Menière's disease (MD); vestibular migraine (VM); temporomandibular disorder (TMD); case report

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Introduction

The craniofacial region is characterized by a variety of structures that are the source of the most common acute and chronic pain conditions. The symptomatology manifested has features that are often shared by different diseases. The role of a multidisciplinary approach is vital, considering

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the widespread and overlapping clinical manifestations that are observed in the head and neck region. An example of complex symptomatology is presented in the following case report where the patient had signs and symptoms of probable Menière's disease (MD), vestibular migraine (VM) and temporomandibular disorder (TMD). MD is a poorly understood inner ear disorder characterized by spontaneous episodic vertigo, fluctuating hearing loss, tinnitus, fullness in the ear, sudden fall without loss of consciousness, gait problems, postural instability and nausea. Recent studies show increasing evidence regarding the overlapping features between MD and VM (1). VM is a primary headache that meets the diagnostic criteria for migraine and is associated with spontaneous attacks of vertigo. Moreover, it is well-known that TMDs are comorbidities of primary headaches, such as migraines. However, cases of MD and VM associated with TMDs have not been reported. Here, we describe a case of probable MD and VM associated with TMD. We present the following case in accordance with the CARE reporting checklist (available at https://joma.amegroups.com/article/view/10.21037/joma-22-15/rc).

Case presentation

An 18-year-old Caucasian male was evaluated at the Orofacial Pain Clinic at the Eastman Institute for Oral Health, with a chief complaint of pre auricular pain associated with vertigo, dizziness, tinnitus and headache. The patient reported the first episode of vertigo early in childhood at the age of seven years old. No history of trauma was reported. The episodes of vestibular dysfunction were associated with tinnitus, aural fullness, lightheadedness, rotation, and a floating sensation (mainly directed on the right side), a sense of being off-balance, nausea and occasional vomiting.

The vestibular symptoms usually lasted from minutes to hours and occurred multiple times per day. In the past month, the patient noticed a deterioration of his hearing in the right ear. The vestibular symptoms could be spontaneous or triggered by certain functions or physical activity. The patient reported that engaging in distraction activities such as low-intensity exercise or falling asleep occasionally improved his symptoms. On the other hand, intense exercise, cold, movement and rotation of the head could aggravate the vertigo.

The patient reported mild pain associated with the vestibular dysfunction and was localized bilaterally within the peri temporomandibular joint (TMJ) area with a duration of 30 minutes and occurred every other day. The patient reported that the discomfort was escalating in intensity and frequency, and resulted in a limitation of the physiological range of motion of the TMJ. The patient described the quality of pain as "sore". The pain was aggravated with function of the TMJ, and avoiding this activity alleviated it. The patient reported daily headaches since the age of 15, localized bilaterally in the temporal areas, of mild to moderate intensity. The quality of pain was described as throbbing. Phonophobia and photophobia were associated with the headache. When vestibular symptoms were present, the patient experienced nausea and vomiting with headaches as well. However, usually, the migraines and vertigo were not reported together. Stress and physical activity could aggravate the pain, while non-steroidal anti-inflammatory drugs (NSAIDs) and sleep were reported as alleviating factors.

A comprehensive intraoral, extra oral, and cranial nerve screening was performed. At the examination, the patient showed signs of rotatory vertigo, dizziness, tinnitus, blurred vision, slurred speech, poor coordination of muscle movements, and poor posture and gait. Hyperactive pupil contraction and dilatation to light were also noted. The TMJ exam and palpation of the muscles of mastication resulted in a painful bilateral click and myofascial pain with referral. The familiar mild periauricular pain was reproduced by palpation of the deep masseters. Signs of bruxism were also present, such as linea alba, indentation of the tongue, and signs of wearing on the anterior teeth.

A diagnosis of probable bilateral disc displacement with reduction, arthralgia, and myofascial pain with referral was made. The suspicion of MD associated with the VM was formulated according to the diagnostic criteria for MD and VM (2,3). The patient reported spontaneous episodes of vertigo lasting for more than 20 minutes with fluctuating aural symptoms. Notably, in his description of symptoms, the patient described the vertigo sensation as "rotatory," which is ascribable to MD as well as the pathognomonic sign of pupillary hyperactivity.

The treatment plan was multidisciplinary. An intra oral occlusal appliance, physical therapy, and trigger point injections were proposed for the TMDs. However, the TMD was not the primary source of the chief complaint of the patient. In agreement with the ENT, anesthesiologist and neurologist, the treatment plan recommended included prophylactic pharmacologic-management for MD and VM with propranolol at 40 mg/TID, vestibular rehabilitation to improve the vestibular dysfunction, behavioral

modification, dietary restrictions to avoid migraine triggers and, behavioral measures to improve sleep quality. Low-impact physical activity was also recommended. In addition, cognitive behavioral therapy with a psychologist to cope with tinnitus and general psychological distress was also recommended. At a three-month follow-up, the patient showed a general improvement of his condition with no adverse side effects. The patient had relocated and a long term follow up was not available. No identifiable information was included in the present case report. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013).

Discussion

MD is a rare chronic illness more prevalent in older, white, overweight or obese, lower socio-economic status and females (2:1 female to male ratio). The prevalence of MD is 0.27% and the age of diagnosis between 40 and 60 years old. However, childhood-onset cases have been reported, and a small percentage are diagnosed before 20 years old (1.5–3% of cases) (4).

MD is an idiopathic disorder, and its underlying pathophysiological mechanism is still unclear and controversial. Of the variety of pathologic findings associated with MD that have been identified, endolymphatic hydrops (EH) seems the most reported. The discovery of EH provided a rationale behind episodic vertigo, abnormal cochlear, and vestibular function associated with MD. According to Schuknecht's theory, EH is the cause of the endolymphatic duct distention and the rupture of Reissner's membrane. Secondary to the rupture, there is infiltration of the endolymph in the basal surface where the hair cells and the eighth cranial nerve are exposed to a potentially toxic level of potassium. Moreover, a marked level of inflammation was found in the cochlear scale media in the temporal bone (5). Various hormones have been associated with the endolymphatic homeostasis: antidiuretic hormones (ADH or vasopressin), aquaporin system, ion channel, and epinephrine. Among the contributing factors to EH are allergies, infection, inflammation, autoimmune disease, thyroid dysfunction, anatomical malformations, and genetic predisposition (6).

The spasm of the tensor tympani (TT) can reproduce, to some extent, the symptomatology associated with various inner ear disorders, MD included. The TT is innervated by the TT nerve, purely motor, branch of the mandibular division of the trigeminal nerve. In fact, TT plays a role in the change of perilymphatic pressures because of its function on the stapes. According to Klockhoff, a TT syndrome is characterized by fullness, tinnitus and, hearing impairment or dysacusis due to the variation of the impedance in the middle ear acoustic tympanometry. Therefore, low-frequency conductive hearing loss can be used as an indicator of a TT spasm. Within the etiopathogenesis hypothesis of MD, there is the subject of the dysfunction of the autonomic nervous system. Anomalous pupillary contraction is a symptom of autonomic nervous system dysfunction that also been confirmed in several studies (7).

Concerning the evolution of the symptoms, it has been observed that, when vertigo is the initial symptom, hearing loss follows within a range of 1 month to 13 years (mean 4.5 years) (8).

Among the symptoms, tinnitus is one of the most debilitating, and it can cause emotional distress, concentration problems, and sleep disturbances (9). In 2015, the Equilibrium Committee of the AAO-HNS, the Japan Society for Equilibrium Research, the European Academy of Otology and Neuro-Otology, the Korean Balance Society and the Bárány Society collaborated on a new set of guidelines for the diagnosis and classification of MD. The diagnostic criteria in our case for probable MD include A minimum of two spontaneous vertigo episodes of 20 minutes to 24 hours duration, the affected ear may exhibit fluctuating aural symptoms such as fullness, tinnitus and hearing changes and the condition cannot be attributed to other vestibular diagnosis (10,11).

The diagnostic tests for MD currently available have limited specificity or sensitivity. As of now, laboratory serology tests cannot accurately test for MD, but it is possible to rule out the concomitant conditions. That includes thyroid panels, glucose, and antinuclear antibodies testing, chemistry panels (metabolites), urinalysis for proteinuria or hematuria (oto-renal syndrome), fluorescent treponemal antibody absorption (FTA-ABS) testing and Venereal Disease Research Laboratory (VDRL) tests for syphilis and Lyme disease. VDRL and FTA-ABS testing should be mandatory, considering the similar clinical manifestations of syphilis and MD. Allergy testing should be prescribed in order to evaluate allergy-mediated Menière syndrome (12).

Hearing tests are required for the diagnosis and follow-up of MD. However, it has not been identified as a pathognomonic audiometric pattern because of the

variation encountered within the different stages of the disease. Balance tests can be performed to evaluate vertigo, and stability. Electronystagmography (ENG) can be utilized to assess balance, nystagmus, vertigo, and dizziness by reproducing the vestibulo-ocular reflex. Caloric testing can be part of ENG, which involves the introduction of warm or cold water or air into the affected ear and observation of eye movements. However, the main limitation is that the results may vary based on the stage of the disease (13). Head-shaking nystagmus and vibration nystagmus are tests that evaluate dizziness. In addition to caloric tests, these exams could give a good indication of vestibular abnormalities. Abnormal results are reported for MD patients in 53% to 71% of the cases (14).

Electrocochleography (ECoG) is the most valuable test to assess EH. ECoG results are not correlated to the stage of the disease, duration of symptoms, history of the patient, or audiometric findings. Thus, it is an informative test as a diagnostic and prognostic tool. The Vestibular Evoked Myogenic Potential (VEMP) testing evaluates if the saccule, otoliths, inferior vestibular nerve, and central connections are intact and functioning correctly. The limitation of this test is that it is attack-dependent. Magnetic resonance imaging with gadolinium indicates when EH or hypoplasia in the endolymphatic sac (ES)/ductus is present and pathognomic. This method allows a quantitative analysis and comparison between the affected and contralateral ear complex. As stated by Tyrrell *et al.*, MD patients are twice as likely to report arthritis, chronic fatigue syndrome, irritable bowel syndrome, gastroesophageal reflux disease (GERD), psoriasis, poor mental health, and migraines (15).

The correlation between anxiety, general distress, and MD has been reported in several studies. Thus, MD has a negative impact on the psychological status of the patients (16,17).

Migraines have been reported in 51% of MD patients (18). Furthermore, in the original definition of the disease, Menière proposed a common etiology between migraines and the symptoms of MD. According to his theory, a migraine is a comorbidity concomitant to the disease and not secondary to it (19). Most recently, Dolowitz and Neff *et al.* suggested that headaches should be added to the criteria of MD complaints (1,20).

Multiple studies support the common pathophysiology of migraines and MD (21,22). Migraines usually are characterized by the classic manifestations (photophobia, phonophobia, aggravated by physical activity, etc.) and share common features with MD, such as vertigo, dizziness, and nausea. The Bárány Society and the International Headache Society classified this condition as VM, and they proposed the diagnostic criteria which are listed in the International Classification of Headache Disorders (ICHD), 3rd beta edition. The diagnostic criteria for VM include a minimum of five episodes with a minimum of 50% of the episodes exhibiting at least one of the following features headache with minimum two features (unilateral, pulsating, aggravated by physical activity, moderate-severe intensity), associated with photo or phonophobia, visual aura and cannot be attributed to other ICHD-3 diagnosis, vestibular disorders (2). The diagnosis is based on the symptomatology of the patient, the history, familial history of migraine and MD, and the temporal association of the symptoms. However, discriminating between MD and VM is often a challenge due to the profound fluctuation and overlapping of the clinical features of the two diseases and the lack of specificity/sensitivity of the vestibular tests.

VM is characterized by migraines, moderate to severe headaches, and vertigo episodes with a duration between 5 minutes to 72 hours. Additionally, the vestibular symptoms can occur before the headache (similar to aura) or completely independent. According to Pyykkö *et al.*, when MD and VM are co-occurring, the symptomatology of both conditions is aggravated (23). Patients affected by MD and VM display higher levels of psychological distress, often associated with anxiety. Furman *et al.* has proposed a common pathophysiology called migraine-anxiety related dizziness (MARD) (24). VM affects 3–9 million US citizens, with prevalence estimated around 1% to 3% of the population (25).

Several theories have been proposed for the etiopathogenesis of the association between MD and VM. These theories include shared genetics, channelopathy, the effects of neurotransmitters (serotonin, norepinephrine, and glutamate) or neuropeptides (CGRP) on the vestibular and trigeminal nerves. Another theory is that (EH can cause the headaches through activation of the trigeminal nerve and lead to the abnormal function of the cochlear system (1).

As mentioned before, MD can share some comorbidities with VM. Among migraine concomitant diseases or syndromes, there are TMDs. The comorbidity relationship between headaches (in particular migraines) and TMD has been well established. This correlation has been shown in adults, adolescents, and children as well. The association is so compelling that the International Headache Society has included in its classification TMD as a secondary cause for headache (2).

The common pathophysiology is correlated mainly to three aspects. TMD and headaches share the same nociceptive system, and the stimuli are mediated by the trigeminal nerve and the trigeminocervical complex where the information converges to the brain. Moreover, both conditions are usually chronic and associated with peripheral and central sensitization.

It has been demonstrated that the presence of one condition may not only increase the risk to develop the other disease and vice-versa, but also that the intensity and frequency of the symptomatology are aggravated when TMD and headaches are associated (26).

The management of MD involves different approaches. Pharmacological treatment is determined by the presence of comorbidities, if any. Medications usually prescribed include antihistamines with anticholinergic effects for prevention of antiemetic effect on the vestibular system, benzodiazepines and gamma-aminobutyric acid (GABA) agonists to inhibit effect on the central nervous system (CNS), diuretics, reducing the pressure within the inner ear, and Betahistine, a structural analog of histamine, has shown promising results in the treatment of tinnitus (27,28). Steroid injections of glucocorticoids to improve vertigo and hearing loss are also indicated (29), as well as injection of gentamicin for ablation of hair cells and improve vertigo. However, gentamicin injection can cause hearing loss and should be applied only when significant hearing loss is already present. Prophylactics for migraines such as topiramate, calcium channel blocker, and beta-blockers are also prescribed.

Behavioral modifications are also considered (30). Dietary modification is recommended such as restriction of sodium intake (less than 2,000 mg per day), caffeine reduction, and alcohol being limited to one drink per day. In case of association with migraine, avoid triggers. Stress management and cognitive behavior treatment should be considered. Adequate sleep hygiene and regular exercise should be suggested.

Vestibular rehabilitation with a physical therapist is effective for unilateral peripheral vestibular dysfunction. For complicated or non-remitting cases, surgery should be taken into account. The most common procedures include ES, vestibular neurectomy, and labyrinthectomy (31). In cases of persistent and atypical vestibular manifestations, it is suggested to consider comorbid MD and VM when the treatment plan is defined. Indeed, considering the hybrid diagnosis of MD associated with vestibular migraine (MDVM) can be helpful also in the treatment of the patients. As per MD and VM, the simultaneous treatment of headaches and TMDs is suggested in order to reduce the pain intensity/frequency and increase the quality of life (26). This case report emphasizes on the importance of a multidisciplinary approach in the management of patients with chronic pain. One of the initial challenges was that the patient was unwilling to undergo testing due to high costs and did not have insurance. A good communication between heath care providers and the patient can help develop trust and prevent unnecessary testing and financial burden.

The case presented is relevant because of the unique presentation of the complex clinical manifestation of MD and VM associated with signs and symptoms of TMDs in a young male patient. The multidisciplinary approach required to approach a case of such complexity underlines the importance of collaboration with other health care providers. To our knowledge, no other cases with similar features have been published. If the exact diagnosis is confirmed, the ideal treatment plan would involve prophylactic pharmacologic-management for MD and VM (for example propranolol), behavioral modification such as dietary restrictions, sleep hygiene, low-impact exercise, cognitive behavioral therapy (CBT) to cope with tinnitus and anxiety and treatment of TMD and vestibular rehabilitation to improve the vestibular dysfunction.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at https://joma.amegroups.com/article/view/10.21037/joma-22-15/rc

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://joma.amegroups.com/article/view/10.21037/joma-22-15/coif). MK serves as an unpaid editorial board member of *Journal of Oral and Maxillofacial Anesthesia* from July 2021 to June 2023. The other authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures

performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). The patient had relocated and a long term follow up was not available. No identifiable information was included in the present case report.

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