



# The prevalence of isolated otolith dysfunction in a local tertiary hospital

Kenneth Wei De Chua<sup>a, b, \*</sup>, Heng Wai Yuen<sup>a</sup>, David Yong Ming Low<sup>a</sup>, Savitha Hosangadi Kamath<sup>a</sup>

<sup>a</sup> Department of Otorhinolaryngology-Head and Neck Surgery, Changi General Hospital, Singapore

<sup>b</sup> The American Institute of Balance (AIB), Largo, FL, USA

## ARTICLE INFO

### Article history:

Received 29 May 2021

Received in revised form

21 June 2021

Accepted 22 June 2021

### Keywords:

Vestibular disorders

Balance

Equilibrium

Otolith dysfunction

Vestibular-evoked myogenic potential

## ABSTRACT

**Objective:** Patients with dizziness may present with symptoms of tilting, swaying, rocking, floating or with disequilibrium. This may be suggestive of an isolated otolithic dysfunction yet, there is little emphasis on this emerging clinical entity. To characterize and describe the prevalence of isolated otolith dysfunction in a local tertiary hospital and correlate them with clinical diagnosis.

**Methodology:** Retrospective medical chart review of patients who presented with dizziness to the specialist outpatient Otolaryngology clinic, who required vestibular laboratory investigation.

**Results:** Of the 206 patients, more than half of them (52.4%) fulfilled the criteria for either probable or definite isolated otolith dysfunction. When there are clinical symptoms of otolith dysfunction reported, there is a 1.62 odds of a remarkable laboratory otolith finding. The most common clinical finding was “no clear diagnosis” (65.5%) followed by Vestibular Migraine (13.6%).

**Conclusion:** The prevalence of isolated otolith dysfunction is quite high. Laboratory tests of otolith function should be performed more routinely. This can be done in a sequential way to optimize cost effectiveness in countries with no insurance reimbursement. Prospective cohort studies on isolated otolith dysfunction, will lay the groundwork for achieving diagnostic consensus and formulating rehabilitation plans to aid this group of patients.

© 2021 PLA General Hospital Department of Otolaryngology Head and Neck Surgery. Production and hosting by Elsevier (Singapore) Pte Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

## Author contributions

Chua KWD wrote the manuscript. Yuen HW, Low YM D and Kamath S reviewed, edited and confirmed the final draft of the manuscript. All authors took part in the study design, with Chua KWD carrying out the procedures.

## 1. Introduction

Some patients with dizziness may have unremarkable laboratory findings of the semi-circular canals but have abnormal otolith function tests (Park et al., 2019). Patients may report symptoms of tilting, rocking, floating, translational movements in the roll or pitch planes or disequilibrium with drop attacks (Murofushi et al.,

2013; 2015). It is questionable whether these patients have an isolated otolith dysfunction (iOD), as there is currently no diagnostic criteria nor consensus. If we only focus on symptoms of otolith dysfunction (SxOD), some studies have reported a correlation with abnormal findings on the Vestibular Evoked Myogenic Potential (VEMP) tests (Farrell and Rine, 2014; Cleworth et al., 2017). Similarly, patients with reported abnormal laboratory otolith test findings note symptoms of swaying or rocking when they complain of dizziness (Pelosi et al., 2013). In addition to having no consensus on defining otolith symptoms and diagnosing otolith dysfunction (OD) in isolation, there is a further lack of standardized assessment of laboratory otolith function (Kumar et al., 2017). Hence, there is a need for diagnostic criteria with structured definitions of iOD.

There has been several synonyms of OD described in the past. For example, some authors have described otolith organ-specific vestibular dysfunction (OSVD) defined as abnormal VEMP responses with unremarkable caloric and video head impulse test (vHIT) in each semi-circular canal (SCC) plane (Fujimoto et al., 2018). Others have described iOD as “idiopathic otolith vertigo” (IOV) defined as tilting or translational movement in the anterior-

\* Corresponding Author. Department of Otorhinolaryngology-Head and Neck Surgery, Otolaryngology Balance and Hearing Implant, Audiology, Changi General Hospital 2 Simei St 3, S529889, Singapore.

E-mail address: [kenneth\\_chua@cgh.com.sg](mailto:kenneth_chua@cgh.com.sg) (K.W.D. Chua).

Peer review under responsibility of PLA General Hospital Department of Otolaryngology Head and Neck Surgery.

posterior, lateral or up-down planes (Murofushi et al., 2015). Isolated utricular dysfunction has also been reported as abnormal ocular VEMP with normal caloric response. Many studies agreed that the gold standard for guaranteeing normal SCC function was with an unremarkable caloric test (Park et al., 2019; Pelosi et al., 2013; Fujimoto et al., 2018). However, caloric results can be false negative due to central compensation and is an “aphysiological” test of vestibular function. Hence, past reports of OD may not have been truly isolated if the patients also had SCC abnormalities due to an otologic history (Roberts et al., 2005) but have centrally compensated for the deficit. The most common symptom of OSVD reported was rotational vertigo (Fujimoto et al., 2018). As vertigo is usually a symptom of dysfunction in the SCC, looking at laboratory results alone would pose risks of overestimating iOD. When considering diagnosis of iOD, symptoms descriptors of OD should be taken into consideration in addition to laboratory-based findings. To our knowledge, there are no local studies on iOD. Given that there may be a significant prevalence of this patient type, it is important to perform our own study contextualized to our local demographics.

The aim of this study was to classify patients with dizziness who present for laboratory vestibular investigations into two groups: probable iOD proven by laboratory findings only (Lab based) without symptoms of OD, or SxOD (Symptom based) without laboratory findings; and definite iOD proven by both laboratory findings and symptoms in the patients’ reported history. We looked at the clinical profiles and characteristics of these two groups and the association between Videonystagmography (VNG), VEMP and symptoms of OD. This study may help us to further understand a novel disease entity that is underappreciated clinically, because the underlying mechanism of dizziness behind these patients presenting with otolith symptoms and laboratory findings, cannot be fully explained by classic vestibular function test or vestibular diagnoses.

## 2. Methods

### 2.1. Study design and participants

Retrospective case series review of patients with dizziness who presented to the Otolaryngology Clinic over a three-year period and required laboratory vestibular investigation. 1038 patients who were referred to the vestibular laboratory were identified. Of the 1038 patients, only 258 patients who had available vestibular evoked myogenic potential (VEMP) test results were selected. All personal identifiers were stripped before data was populated onto an excel sheet for analysis. This study was exempted from centralized institute review board (CIRB) approval, as the data obtained had no identifiers that can be traced back to the patients in any way. Demographic and clinical characteristics such as age, gender, race, clinical symptoms, laboratory findings and clinical diagnosis are described. When patients had symptoms of disequilibrium, floating, rocking, swaying or a combination of more than one symptom, they were considered remarkable for symptoms of otolith dysfunction (SxOD). Non-specific symptoms of dizziness or vertigo were not considered and excluded as SxoD. When laboratory findings of asymmetry in the VEMP were noted, the patients were “positive” for laboratory-based otolith dysfunction (LabOD). If VEMP were bilaterally symmetrical or absent, they will be “negative” for LabOD. Bilaterally absent VEMP were equivocal as it could be due to insufficient contractions or fatigue of the sternocleidomastoid or ocular muscles. VEMP were also expected to be degraded with age and hence, only asymmetrical VEMP were considered pathological. In the absence of any other remarkable vestibular findings, when either SxOD or LabOD was positive, the

patients were identified as probable iOD. When both SxOD and LabOD were positive, patients were identified as definite iOD. We looked at the categories of presenting clinical symptoms and correlated them with the actual clinical diagnosis and diagnosis of iOD. We also looked at the odds ratio of an abnormal VEMP finding when VNG was normal, compared to the odds ratio of an abnormal VEMP finding when clinical symptoms are of an otolith origin. The prevalence of iOD was described, in accordance to the previous proposal of definite or probable iOD (Park et al., 2019).

### 2.2. Vestibular function testing

All included patients had undertaken videonystagmography (VNG) evaluation that included oculomotor, gaze, spontaneous, positional, Dix-Hallpike, post-headshake and bi-thermal caloric test. Participants were also screened with the video head impulse test (vHIT) and/or bedside head thrust of the lateral canals. Additional VEMP results were also obtained and correlated with clinical findings and diagnosis. Nystagmus was analyzed quantitatively with the VisualEyes system (Micromedical, Chatham, IL, USA). Unilateral vestibular hypofunction was confirmed when canal paresis was  $\geq 22\%$ . Cervical VEMP was recorded from the ipsilateral sternocleidomastoid (SCM) muscle with the Eclipse system (Interacoustic A/S Middlefart, Denmark) in a 30° supine position with elevation and rotation of head. Using the insert earphones, 500 Hz alternating air conducted tone-burst sounds was delivered between 90 and 95dbnHL. The rise/fall time, plateau time and repetition rate was 1 ms, 2 ms and 5.1 Hz respectively. Optimal techniques for eliciting VEMP was used (McCaslin et al., 2011) and the cut-off for asymmetry of 33% was based on our local unpublished data. Ocular VEMP was recorded in similar semi-recumbent position with eyes gazing upwards (30°). The same repetition rate of 5.1 Hz was used with a different rise/fall time of 1.5 ms with no plateau. The inter-aural difference (IAD) was considered abnormal if measurement value was  $>0.33$  (Rosengren et al., 2019). For the vHIT, either eye was recorded with the Eyeseecam (Interacoustics A/S, Middlefart, Denmark). The subjects were instructed to gaze at a target of 1.5 m. Eye position was calibrated using laser targets projected from the goggles. Head impulse delivered was unpredictable and abrupt in timing and direction. At least 10 impulses were recorded for each direction with peak-velocity of head rotation of more than 150°/s. The system uses instantaneous gain and gain in the horizontal canal  $<0.79$  at 60 ms was considered abnormal (Blodow et al., 2015). All vestibular function assessments were conducted primarily by a fellowship trained Senior Vestibular Audiologist and all results were reviewed by either of the two consultant neurotologist who scrutinized the raw data and endorsed the clinical reporting.

### 2.3. Statistical analyses

Descriptive analysis of age, gender and race was performed. Statistical analysis was performed with SPSS version 21.0 (IBM Corp., Armonk, NY, USA). Parametric T-tests and Chi-square test (or Fischer’s exact test) were used for analysis of continuous and categorical variables respectively. A p-value of less than 0.05 indicated statistical significance.

## 3. Results

Of the 1038 patients, only 24.9% (258/1038) had additional c-VEMP testing with o-VEMP only performed in five patients. Of the 258 patients, 119 were females and 139 were males with age ranging from 17 to 82 years. There was no significant difference in the mean age between genders ( $P = 0.07$ ). 168 (65.8%) were

Chinese, 25 (9.5%) were Indians, 37 (13.9%) were Malays and 28 (10.7%) were of Other Races. (Table 1). 59.7% (154/258) had normal VNG and VEMP, 20.2% (52/258) had normal VNG but abnormal VEMP, 15.1% (39/259) had abnormal VNG but normal VEMP and 5.0% (13/258) had both abnormal VNG and VEMP (shown in Fig. 1). We excluded 52 patients with abnormal VNG and/or vHIT as they did not fulfil the criteria for iOD. All remaining 206 patients had unremarkable VNG, vHIT and bedside head thrust test of the lateral canal. They also had no significant neurotologic, orthopedic or non-corrected visual deficits as determined by an otolaryngologist.

Of the 52 patients with normal VNG and abnormal VEMP, almost half of them (25/52; 48.1%) had both clinical SxOD and LabOD fulfilling the criteria for definite iOD. More than half of them (27/52; 51.9%) had abnormal VEMP but were unremarkable for symptoms of OD (probable iOD). Similarly, of the 154 patients with unremarkable VNG and VEMP, 36.4% (56/154) only reported SxOD (probable iOD) while 63.6% of patients (98/154) had neither laboratory or clinically reported OD and hence did not fulfil criteria for iOD (shown in Fig. 2). Taken together, of the 206 patients, only 47.6% (98/206) did not fulfil criteria for iOD while majority 52.4% (108/206) fulfilled criteria for either probable or definite iOD (shown in Fig. 3).

When VNG results were normal, there is a 1.02 times odds of getting an abnormal VEMP results with a 95% confidence interval between 0.67 and 1.12,  $p = 0.18$  (Table 2). However, when clinical symptoms of OD were reported (SxOD “YES”), there is a 1.62 times odds of an abnormal laboratory otolith finding (LabOD “YES”), with a moderate effect size and 95% confidence interval between 1.25 and 4.13,  $p = 0.01$  (Table 2).

We described the distribution of the clinical diagnoses and correlated them with the category of iOD (Table 3). The sub-types of Vestibular Migraine and Menieres Disease are further elucidated in Fig. 4. The most common clinical finding was no clear clinical diagnosis (135/206; 65.5%) followed by 13.6% with Vestibular Migraine (28/206), 8.3% with Menieres Disease (17/206), 4.4% with Benign Paroxysmal Positional Vertigo, other diagnoses, 2.4% with vestibular neuronitis/labyrinthitis (5/206) and 1.4% with post-concussion symptoms (3/206) (Table 4). Diagnosis of Menieres and Vestibular Migraine were in accordance with the International Classification of Vestibular Disorders (ICVD).

Most of the patients with no clear clinical diagnosis had either probable iOD (62/135; 46%) or definite iOD (17/135; 12.6%), while 41.4% (56/135) did not fulfil the diagnostic criteria for iOD. For patients with BPPV, more than half of them had definite iOD (5/9; 55.6%) while the rest were not identified as iOD (4/9; 44.4%). Majority of patients with vestibular migraine as a diagnosis did not fulfil criteria for iOD (19/28; 68%), leaving six patients with probable iOD (6/28; 21.4%) and three patients with definite iOD (3/28; 10.6%). None of the patients diagnosed with Menieres Disease had definite iOD and most did not fulfil the proposed diagnostic criteria for iOD (10/17; 58.8%). Less than half the patients with Menieres Disease (7/17; 41.2%) had probable iOD. All patients with vestibular

neuronitis/labyrinthitis had no iOD and all patients with post-concussion symptoms had probable iOD. More than half the patients with other diagnoses had probable iOD (5/9; 55.6%) with the remaining patients not fulfilling diagnostic criteria for iOD (4/9; 44.4%).

#### 4. Discussion

It is not uncommon to find patients presenting clinically with complains of swaying, rocking, tilting, floating and disequilibrium. Such symptoms though not pathognomonic of OD, can suggest an isolated deficit of the otolith organs especially when confirmed by laboratory findings. A significant number of patients had either probable or definite iOD, despite VEMP not done routinely as part of the vestibular test battery. If VEMP were deferred, diagnosis of probable or definite iOD in more than half of the 206 patients would have been missed. If VEMP were routinely performed in all patients referred for vestibular laboratory investigation, there may be an increase in incidence rates of iOD. However, the medical system in different countries may not be based on an insurance reimbursement model. Hence, clinicians have to be prudent with selection of procedures to minimize the patients’ out-of-pocket cost. It is therefore advisable in our local context to sequentially carry out vestibular assessment based on the test results obtained at each stage to optimize cost-effectiveness and time (shown in Fig. 5). When most tests including the caloric test results of the VNG are unremarkable, the odds ratio of getting an abnormal VEMP is not significant. However, when there are clinical symptoms of OD, there is a significant 1.62 times odds of obtaining an abnormal VEMP. This suggests that in the absence of caloric weakness, we should obtain information about the otoliths with VEMP testing, especially when clinical symptoms of OD are present. If VEMPS cannot be obtained, a quick test of subjective visual vertical (SVV) should be included as VEMPS are produced by only type I hair cells of the striola, which is about 5% of each otoconia receptor. SVV could measure a different function of the utricular hair cells and value-add, as it is still uncertain if the acoustic-induced reflex in oVEMP involves the otoconia. This could explain the 36% of patients who had normal VNG and VEMPS but who reported symptoms of OD.

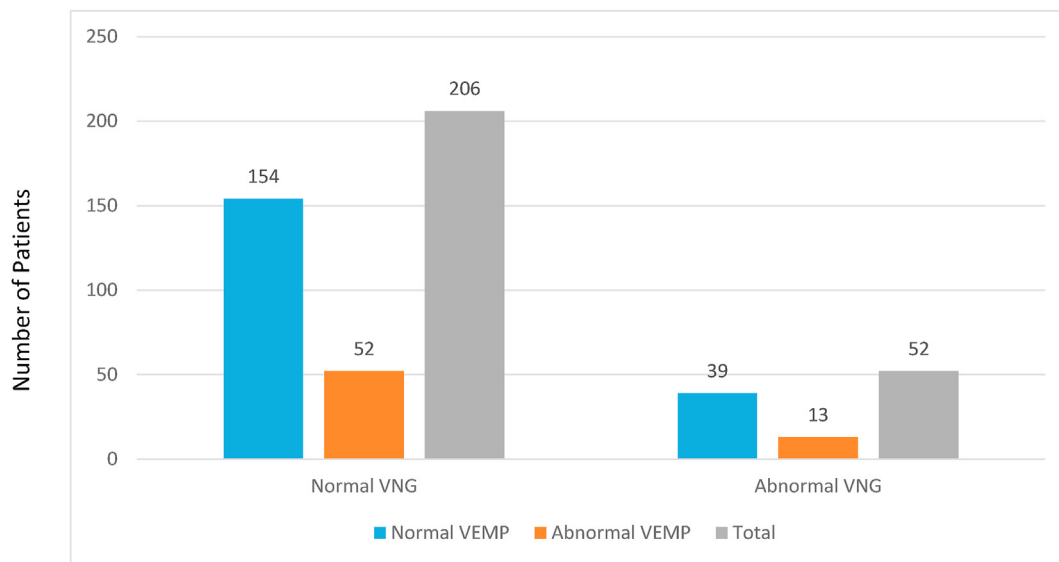
However, when there are no symptoms of OD, it may be more prudent to exercise caution with performing VEMP to settle for probable iOD as a diagnosis or unremarkable laboratory vestibular findings. Given the current diagnostic climate, there is yet a formal diagnostic consensus on iOD, much less rehabilitation and/or management plans. Hence, adding VEMP to the vestibular test battery is additional cost to the patients that has to be carefully weighed against the value it adds to clinical management.

Most of the patients with probable or definite iOD also had no clear clinical diagnosis, which makes iOD appealing as a possible diagnosis. When there is a clear clinical diagnosis such as Benign Paroxysmal Positional Vertigo (BPPV), depending on whether the presenting symptoms are vertiginous, it may be definite or probable iOD. It is of note that the vertical canals, though uncommon to be affected have to be screened before concluding on iOD. It is also interesting to note that majority of the patients with Vestibular Migraine (VM) and Menieres Disease (MD) did not fulfil the criteria for iOD. This could be because both VM and MD are fluctuating conditions and at the point of assessment, most of the patients could be in the interictal stage with no symptoms nor VEMP weakness. For those that met the criteria, it could be because not all patients with VM or MD strictly fulfilled the diagnostic criteria by the ICVD and may be atypical variants without vertigo.

All the patients with a history of vestibular neuronitis or labyrinthitis had unremarkable laboratory vestibular finding, with complains of non-specific giddiness but no symptoms of OD. This is

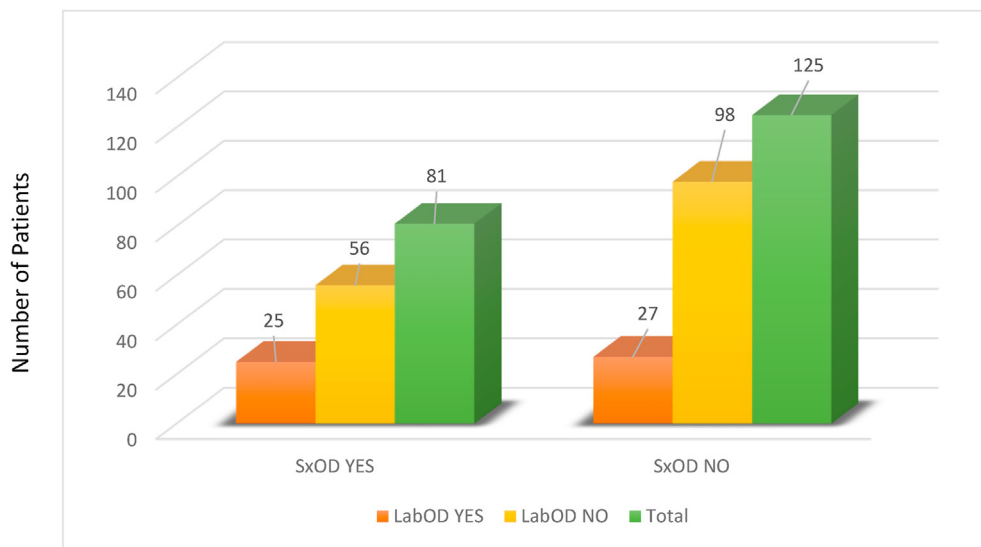
**Table 1**  
Demographics of patients seen for vestibular investigation.

Demographic	Males	Females	Overall
<b>Gender</b>	139	119	258
<b>Age range (Mean)</b>	17-82 (46.2)	20-81 (50.5)	17-82
<b>Race</b>	Males	Females	Total
Chinese	81	87	168
Indian	20	5	25
Malay	19	18	37
Others	19	9	28
	139	119	258



Category of Videonystagmography (VNG) results versus Vestibular evoked myogenic potential (VEMP) results

Fig. 1. The association between Videonystagmography (VNG) and Vestibular-evoked myogenic potential (VEMP) test results. (N = 258).



Category of Symptom-based Otolith Dysfunction (SxOD) versus Laboratory-based Otolith Dysfunction (LabOD)

Fig. 2. The association between clinically reported otolith dysfunction (SxOD) and laboratory findings (LabOD). (N = 206).

either suggestive of a very efficient central vestibular compensation or a misdiagnosis as only about up to half of the patients with previous neuronitis/labyrinthitis have documented recovery in the caloric function (Schmid-Priscoveanu et al., 2001). All patients with history of concussion had either symptoms of OD or VEMP weakness. This could be due to labyrinthine concussion that is isolated to the otolith or non-specific symptoms of cortical concussion that is masquerading as OD. As there are no pathognomonic SxOD, it may be hard to distinguish the non-specific symptoms of OD from cortical concussion, especially when it is more commonly associated with labyrinthine concussion (Wallace and Lifshitz et al., 2016).

The authors that proposed for this new diagnosis also explained that iOD could be further categorized as primary idiopathic or secondary to a known cause (Park et al., 2019). We think that primary idiopathic definite/probable iOD is easier to consider when there is no clear clinical diagnosis and patients present with SxOD (rocking, swaying, tilting, disequilibrium, floating etc ...) and/or with laboratory-based confirmation. Although a counter-argument here is that pathognomonic otolith symptoms are not well understood and hence may be confused with descriptors of non-specific dizziness not of an otolith origin, it is nevertheless still worth to consider iOD in the absence of a clinical diagnosis.

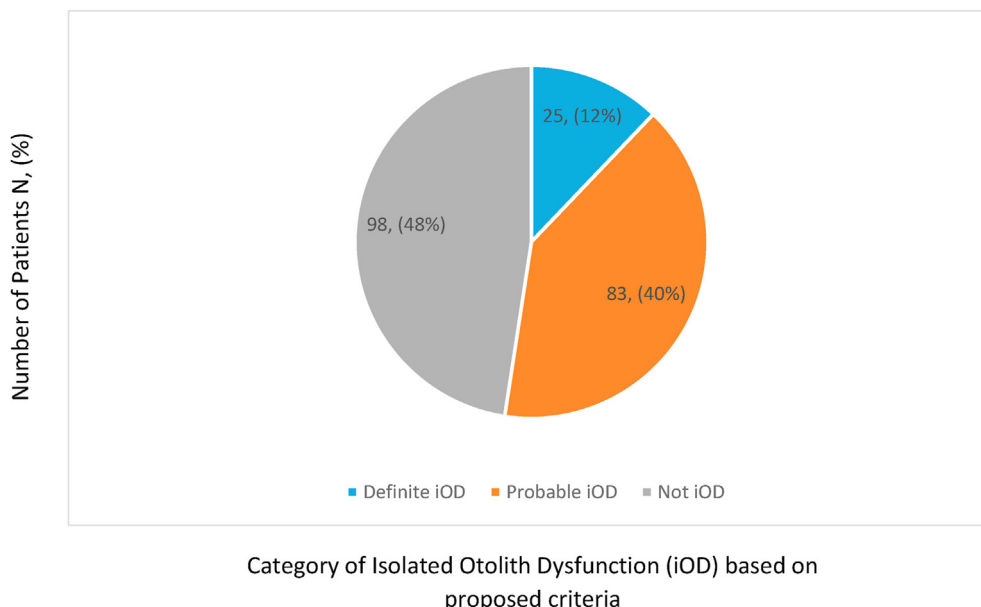


Fig. 3. Incidence of isolated Otolith Dysfunction (iOD) fulfilling diagnostic criteria. (N = 206). Number and percentage representation.

**Table 2**  
The association of videonystagmography with vestibular-evoked myogenic potential

	Normal Vestibular evoked Myogenic Potential	Abnormal Vestibular evoked Myogenic Potential	Total	Odds Ratio (Abnormal Vestibular evoked Myogenic Potential)	P-Value
Normal Videonystagmography	154	52	206	1.02	0.18
Abnormal Videonystagmography	39	13	52		

**Table 3**  
The association between symptom and laboratory based findings of otolith dysfunction

	Lab-based Otolith Dysfunction (YES)	Lab-based Otolith Dysfunction (NO)	Total	Odds Ratio (Abnormal Vestibular Evoked Myogenic Potential)	P-Value
Symptoms of Otolith Dysfunction (YES)	25	56	81	1.62	0.01
Symptoms of Otolith Dysfunction (NO)	27	98	125		

However, when considering secondary iOD with known or suspected vestibular diagnosis, it may be more challenging especially for fluctuating conditions such as MD and VM. Central compensation may also result in “false negative” laboratory test results and further limit the diagnosis of a truly iOD (Chua et al., 2021).

#### 4.1. Meniere Disease

For example in MD, there may be a sequential progressive lesion of the labyrinthine that eventually includes other structures, although the otoliths have been reported to be more sensitive than semi-circular canals to hydropic expansion (Pender, 2015). If laboratory test suggests that the otoliths are the only structures affected, it does not rule out the involvement of other structures. As MD progresses and fluctuates, the patients may find themselves in a dynamic state of compensation-decompensation with variable laboratory test results. In fact, some authors are suggesting that the discordant video-head impulse test (normal) with calorics (abnormal) happens in 60–70% of patients with MD (Hannigan

et al., 2019) and hence may not fit the clinical picture of iOD. When active MD and OD are both present, the symptoms of MD should dominate the clinical presentation and hence OD is neither isolated (other labyrinthine structures affected) nor definite (presence of vertigo). However, if MD has been inactive for a period, symptoms of OD may now be more prominent, the diagnosis may change to definite OD.

#### 4.2. Vestibular migraine

If we look at VM as a differential diagnosis due to the overlapping symptoms with MD, we are also considering that migraine is a neurovascular event that undeniably involves central pathways. The stability of VEMP latencies may argue against a central pathology but does not rule out non-destructive changes in brainstem nuclear sensitivity (Zuniga et al., 2012). Hence, abnormal vestibular evoked myogenic potentials (VEMP) may not always suggest a peripheral pathology.

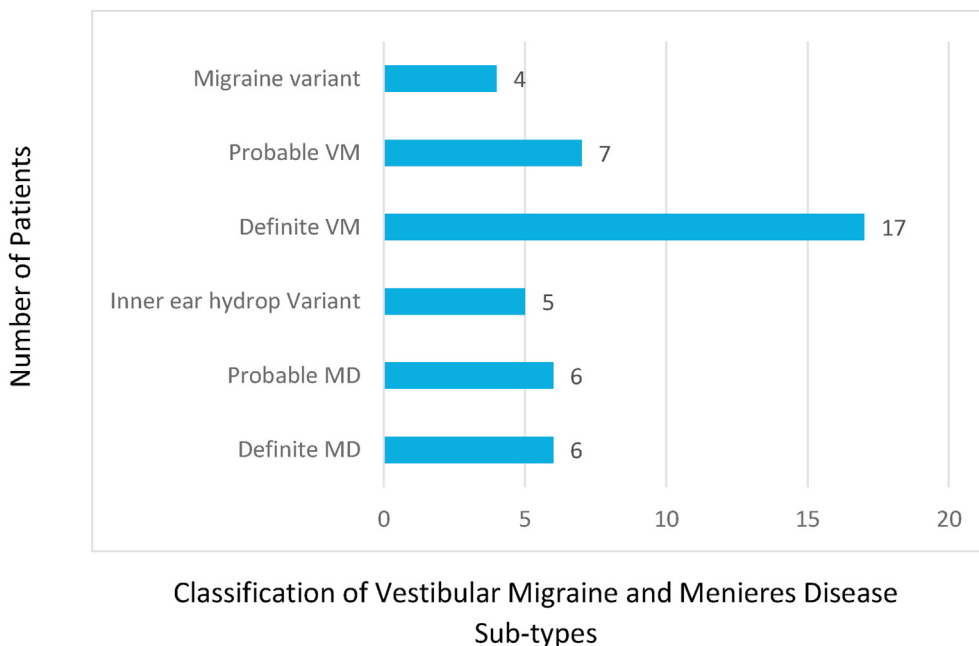


Fig. 4. Distribution and sub-types of vestibular migraine and menieres disease.

**Table 4**  
Correlation of clinical diagnoses with isolated otolith dysfunction.

Clinical Diagnosis	Definite iOD	Probable iOD	Not iOD	Total
No clear clinical Diagnosis	17	62	56	135
Benign Paroxysmal Positional Vertigo (BPPV)	5	0	4	9
Vestibular Migraine	3	6	19	28
Menieres Disease	0	7	10	17
Vestibular Neuritis/labyrinthitis	0	0	5	5
Post-Concussion Symptom	0	3	0	3
Others <sup>a</sup>	0	5	4	9
<b>Total</b>	<b>25</b>	<b>83</b>	<b>98</b>	<b>206</b>

<sup>a</sup> Others: Superior Semi-Circular Canal Dehiscence, Vestibular Schwannoma, Sudden Hearing loss, Multi-Factorial Dizziness and Hyperventilation Vertigo.

### 4.3. Benign Paroxysmal Positional Vertigo

Where BPPV is concerned, it is commonly understood that vascular ischemia or neural denervation/degeneration can be a cause of both OD and BPPV (Lee et al., 2014). Vascular damage may produce substantial detachment of the otoconia in the denervated and/or degenerated area (utricle) and lead to BPPV. Although BPPV is not the cause of OD, it is also not a coincidental entity. Hence, diagnosis of OD is neither primary idiopathic nor secondary to BPPV.

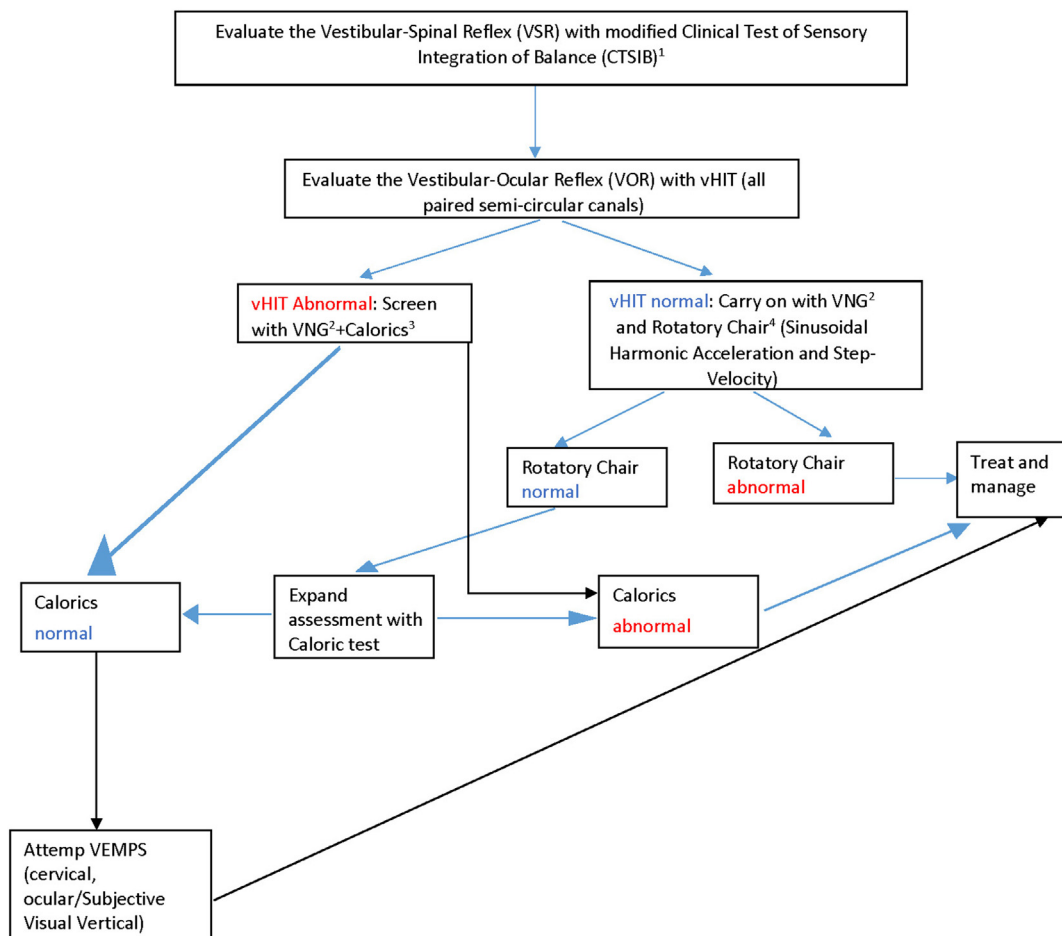
With greater awareness and knowledge of OD, treatment strategies should change depending on the predominant symptoms. For example, if OD symptoms are dominating, treatments devised should focus on OD more than MD and intra-tympanic dexamethasone or gentamicin may not be applicable here.

### 4.4. Limitations of study

VEMPS only activate the type I haircells with irregular resting discharge which is approximately 5-10% of the otolith function. Haircells and neurons with regular resting discharge are not activated by air conducted stimulation of bone conducted vibration even at maximal levels used (Curthoys et al., 2018). This means that assessing VEMPS alone may be insufficient as SVV and or ocular counter roll assess different functions of the otoliths and allow for

the reflection of different otolith pathologies (Hosli and Straumann, 2021). However, as oVEMP and SVV clinical protocols are not standardized, most clinicians were not comfortable with carrying out the test leaving limited information about utricular function. When cVEMP results are unremarkable, regardless of whether there are symptoms of OD present, patients may still fulfil criteria of at least a probable iOD if oVEMP and/or SVV are remarkable for an asymmetry. Future prospective studies should include information of the utricle with at oVEMP, subjective visual vertical (SVV) or ocular counter roll test; otherwise the prevalence of OD will be underestimated. Nevertheless, even without oVEMP information in this study, there is still a significant number of patients with possible OD with either cVEMP asymmetry of subjective symptoms reported. One must be careful with the presumptuous interpretation of possible OD based on just cVEMPS alone, given these limitations. Furthermore, the vertical canal should also be screened for any deficits with the vHIT before any diagnoses of iOD can be made. Rotatory chair information assessing the mid-frequencies should also be included in at least a truncated sinusoidal harmonic acceleration (SHA) protocol known to be most sensitive (Maes et al., 2011; Ahmed et al., 2009) at 0.025 Hz, 0.01 Hz, 0.05 Hz, 0.25 Hz and 0.5 Hz with Step-Velocity (SV) test to rule out further involvement of the semi-circular canals and determine status of compensation. Information about the vestibular-spinal reflexes (VSR) should also be objectively quantified with computerized

### Vestibular Assessment: Sequential Protocol



<sup>1</sup> CTSIB: Six conditions. Eyes opened or closed on firm ground in normal stance. Eyes opened or closed in semi-tandem on firm ground. Eyes opened or closed on dynamic foam surface.

<sup>2</sup>VNG includes Gaze/Spontaneous, Oculomotor (pendular, saccades, OPK), High-frequency Headshake, Positional (head right and left) and Dix-Hallpike.

<sup>3</sup>Caloric irrigation; use monothermal warm screen with conservative 20% cut-off.

<sup>4</sup>Sinusoidal Harmonic Acceleration (SHA) at 0.01Hz, 0.025Hz, 0.05Hz, 0.25Hz and 0.5Hz with Step-Velocity (SV) test

Fig. 5. Proposed sequential vestibular assessment, flowchart.

dynamic posturography (CDP) or evaluated with a bedside equivalent such as the GANS Standard Operating Protocol (SOP) (Roberts, 2018). Such information on whether there are vestibular, somatosensory, or visual deficits will aid in the rehabilitation plans.

#### 5. Conclusion

iOD is an emerging concept that needs to be worked on, to develop an international consensus. iOD symptoms may overlap greatly with symptoms of Persistent Perceptual Postural Dizziness (PPPD) described by the Barany Society (Stabb et al., 2017). PPPD symptoms may also include a false sense of swaying, rocking or bobbing and will need to be carefully differentiated from iOD. Finally, the management plans for iOD remains elusive as there are no current rehabilitation techniques for improving otolith function. Nevertheless, continuous and further refinement of iOD diagnosis

will lay the groundwork for the eventual conceptualisation and development of rehabilitation strategies for patients with OD. While objective laboratory vestibular test results remain important, patients' subjective complains and functional impairments should be the focus (Chua, 2020) of rehabilitation plans to assist recovery of activities of daily living. Further refinement of iOD should also include the concept of isolated otoconia loss as a possible explanation. Hypofunction of otolith haircells may not be the only reasons of OD, with loss of otoconia mass recently described (Hegemann and Bockisch, 2019; Hegemann et al., 2020). Subjective visual vertical and/or ocular counter roll tests should be included to optimize assessment of otolith function in future studies.

#### Statement of ethics

This study was exempted from centralized institute review

board (CIRB) approval, as the data obtained had no identifiers that can be traced back to the patients in any way and is in accordance with the Helsinki declaration.

### Funding sources

None

### Declaration of competing interest

The authors declare that no conflicts of interest exist. No sponsorship or funding was received for this study.

### References

- Ahmed, M.F., Goebel, J.A., Sinks, B.C., 2009. Caloric test versus rotational sinusoidal harmonic acceleration and step-velocity tests in patients with and without suspected peripheral vestibulopathy. *Otol. Neurotol.* 30, 800–805.
- Blodow, A., Blodow, J., Bloching, M.B., Helbig, R., Walther, L.E., 2015. Horizontal VOR function shows frequency dynamics in vestibular schwannoma. *Eur. Arch. Oto-Rhino-Laryngol.* 272, 2143–2148.
- Chua, K.W.D., Yuen, H.W., Low, D.Y.M., Kamath, S., 2021. Letter to the editor: proposal on the diagnostic criteria of definite isolated otolith dysfunction. *J. Audiol. Otol.* 25, 1–4.
- Curthoys, I.S., Grant, J.W., Burgess, A.M., Pastras, C.J., Brown, D.J., Manzari, L., 2018. Otolithic receptor mechanisms for vestibular-evoked myogenic potentials: a review. *Front. Neurol.* 9, 366.
- Chua, K.W.D., 2020. The focus on functional impairments in the rehabilitation of patients with unexplained functional dizziness. *Int. Phys. Med. Rehab. J.* 3, 122–123.
- Cleworth, T.W., Carpenter, M.G., Honegger, F., Allum, J.H.J., 2017. Differences in head impulse test results due to analysis techniques. *J. Vestib. Res.* 27, 163–172.
- Farrell, L., Rine, R.M., 2014. Differences in symptoms among adults with canal versus otolith vestibular dysfunction: a preliminary report. *ISRN Rehabilitation* 2, 1–11.
- Fujimoto, C., Suzuki, S., Kinoshita, M., Egami, N., Sugawara, K., Iwasaki, S., 2018. Clinical features of otolith-organ specific vestibular dysfunction. *Clin. Neurophysiol.* 129, 238–245.
- Hannigan, I.P., Welgampola, M.S., Watson, S.R.D., 2019. Dissociation of caloric and head impulse tests: a marker of Meniere's disease. *J. Neurol.* 268 (2), 431–439. <https://doi.org/10.1007/s00415-019-094>.
- Hosli, S., Straumann, D., 2021. Independent measures of utricular function: ocular vestibular evoked myogenic potentials do not correlate with subjective visual vertical or fundus photographic binocular cyclorotation. *Front. Neurol.* 12, 658419. <https://doi.org/10.3389/fneur.2021.658419>.
- Hegemann, S.C.A., Bockisch, C.J., 2019. Otoconial loss or lack of otoconia- an overlooked or ignored diagnosis of balance deficits. *Med. Hypotheses* 128, 17–20.
- Hegemann, S.C.A., Weisstanner, C., Ernst, A., Basta, D., Bockisch, C.J., 2020. Constant severe imbalance following traumatic otoconial loss: a new explanation of residual dizziness. *Eur. Arch. Oto-Rhino-Laryngol.* 9, 2427–2435.
- Kumar, L., Thakar, A., Thakur, B., Sikka, K., 2017. Sensitivity and specificity of clinical and laboratory otolith function tests. *Otol. Neurotol.* 38, e378–e383.
- Lee, S.K., Kim, S.J., Park, M.S., Byun, J.Y., 2014. Otolith organ function according to subtype of benign paroxysmal positional vertigo. *Laryngoscope* 124, 984–988.
- Maes, L., Vinck, B.M., Wuyts, F., D'haenens, W., Bockstael, A., Keppler, H., et al., 2011. Clinical usefulness of the rotatory, caloric and vestibular evoked myogenic potential test in unilateral peripheral vestibular pathologies. *Int. J. Audiol.* 50, 566–576.
- McCaslin, D.L., Jacobson, G.P., Grantham, S.L., Piker, E.G., Verghese, S., 2011. The influence of unilateral saccular impairment on functional balance performance and self-report dizziness. *J. Am. Acad. Audiol.* 22, 542–549.
- Murofushi, T., Komiya, S., Yoshimura, E., 2013. Do patients who experience episodic tilting or translational sensations in the pitch plane have abnormal sacculo-colic reflexes? *Neurosci. Lett.* 553, 95–98.
- Murofushi, T., Komiya, S., Hayashi, Y., Yoshimura, E., 2015. Frequency preference in cervical vestibular evoked myogenic potential of idiopathic otolith vertigo patients. Does it reflect otolith endolymphatic hydrops? *Acta Otolaryngol.* 135, 995–999.
- Park, H.G., Lee, J.H., Oh, S.H., Park, M.K., Suh, M.W., 2019. Proposal on the diagnostic criteria of definite isolated otolith dysfunction. *J. Audiol. Otol.* 23, 103–111.
- Pelosi, S., Schuster, D., Jacobson, G.P., Carlson, M.L., Haynes, D.S., Bennett, M.L., et al., 2013. Clinical characteristics associated with isolated unilateral utricular dysfunction. *Am. J. Otolaryngol.* 34, 490–495.
- Pender, D.J., 2015. Membrane stress in the human labyrinth and Meniere disease: a model analysis. *Int. Arch. Otorhinolaryngol.* 19, 336–342.
- Roberts, R.A., Gans, R.E., Kastner, A.H., Listert, J.J., 2005. Prevalence of vestibulopathy in benign paroxysmal positional vertigo patients with and without prior otologic history. *Int. J. Audiol.* 44, 191–196.
- Roberts, R.A., 2018. Management of recurrent vestibular neuritis in a patient treated for rheumatoid arthritis. *Am. J. Audiol.* 27, 19–24.
- Rosengren, S.M., Colebatch, J.G., Young, A.S., Govender, S., Welgampola, M.S., 2019. Vestibular evoked myogenic potentials in practice: methods, pitfalls and clinical applications. *Clinical Neurophysiology Practice* 4, 47–68.
- Schmid-Priscoveanu, A., Bohmer, A., Obzina, H., Straumann, D., 2001. Caloric and search-coil head-impulse testing in patients after vestibular neuritis. *J. Assoc. Res. Otolaryngol.* 2, 72–78.
- Stabb, J.P., Eckhardt-Henn, A., Horii, A., Jacob, R., Strupp, M., Brandt, T., et al., 2017. Diagnostic criteria for persistent postural perceptual dizziness (PPPD): consensus document of the committee for the Classification of Vestibular Disorders of the Barany Society. *J. Vestib. Res.* 27, 191–208.
- Wallace, B., Lifshitz, J., 2016. Traumatic Brain injury and vestibulo-ocular function: current challenges and future prospects. *Eye Brain* 8, 153–164.
- Zuniga, M.G., Janky, K.L., Schubert, M.C., Carey, J.P., 2012. Can vestibular-evoked myogenic potentials help differentiate Meniere's disease from vestibular migraine? *Otolaryngol. Head Neck Surg.* 146, 788–796.