



“Sitting-up vertigo as an expression of posterior semicircular canal heavy cupula and posterior semicircular canal short arm canalolithiasis”

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ABSTRACT

Background: Vestibular symptoms on sitting-up are frequent on patients seen by vestibular specialists. Recently, a benign paroxysmal positional vertigo (BPPV) variant which elicits vestibular symptoms with oculomotor evidence of posterior semicircular canal (P-SCC) cupula stimulation on sitting-up was described and named sitting-up vertigo BPPV. A periaimpullar restricted P-SCC canalolithiasis was proposed as a causal mechanism.

Objective: To describe new mechanisms of action for the sitting-up vertigo BPPV variant.

Methods: Eighteen patients with sitting-up vertigo BPPV were examined with a pre-established set of positional maneuvers and follow-up until they resolved their symptoms and clinical findings.

Results: All patients showed up-beating torsional nystagmus (UBTN) and vestibular symptoms on coming up from either Dix-Hallpike (DHM) or straight head-hanging maneuver. Sixteen out of 18 patients presented a sustained UBTN with an ipsitorsional component to the tested side on half-Hallpike maneuver (HH). A slower persistent contratortional down-beating nystagmus was found in eleven out of 18 patients tested on nose down position (ND).

Conclusions: Persistent direction changing positional nystagmus on HH and ND positions indicative of P-SCC heavy cupula was found in 11 patients. A sustained UBTN on HH with the absence of findings on ND, which is suggestive of the presence of P-SCC short arm canalolithiasis, was found on 5 patients. All patients were treated with canalith repositioning maneuvers without success, but they resolved their findings by means of Brandt-Daroff exercises. We propose P-SCC heavy cupula and P-SCC short arm canalolithiasis as two new putative mechanisms for the sitting-up vertigo BPPV variant.

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1. Introduction

Vestibular symptoms on sitting-up are common in the general population (Baloh et al., 2010) and in patients who consult vestibular medicine specialists (Büki et al., 2011). Many mechanisms are potentially related to such symptomatology and

Abbreviations: BPPV, benign paroxysmal positional vertigo; CRM, canalith repositioning maneuvers; DBTN, down-beating torsional nystagmus; DHM, Dix-Hallpike maneuver; HH, half-Hallpike maneuver; HYT, head yaw test; ND, nose down position; SCC, semicircular canal; SHH, straight head hanging; UBTN, up-beating torsional nystagmus.

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differentials include: hemodynamic orthostatic dizziness/vertigo (Kim et al., 2019), structural abnormalities, central and peripheral vestibular syndromes (Baloh et al., 2010). In the last group, the BPPV, which is the most common cause of vertigo in the general population, will be found. Its accepted pathophysiological mechanism involves either dislodged otoconia from the utricular macula that enter and flow freely in the semicircular canals (SCC) on canalolithiasis type BPPV, or a gravity sensitive cupula that has changed its flotation characteristics in relation to the endolymph on the heavy/light cupula type BPPV. In canalolithiasis, a change in head position elicits movement of the otoconial debris inside the SCC mediated by gravity. That movement will push or pull the endolymphatic column as a piston, which ultimately deflects the cupula to the vestibulum or SCC side respectively. Cupula deflection generates a paroxysmal positional nystagmus whose characteristics will depend on the involved canal and the direction of deflection. In heavy/light cupula type BPPV, the density ratio between the cupula

and surrounding endolymph determines its flotation characteristics and defines it as either heavy or light (von Brevern et al., 2015). Cupulolithiasis is considered the commonest type of heavy cupula, where dense otoconia are assumed to remain attached to it, thus making it heavier. On the other hand, alcohol and light otoconia fragments attached to the cupula have been ascribed as light cupula causes (Bergeniuss and Tomanovic, 2006). Migraine has been associated with heavy as much as with light cupula (Asprella Libonati, 2011). Typically, graviceptive cupula syndrome findings are persistent direction changing positional nystagmus with neutral positions, in which the plane of the cupula is in the earth vertical position, preventing its deflection. Lateral canal heavy/light cupula variants were the first and the most commonly described in the literature (Hiruma and Numata, 2004). There are few reports of graviceptive posterior canal cupula, all of them belonging to the heavy cupula variant (Asprella Libonati, 2011; Ichijo, 2013; Imai et al., 2009). They describe a persistent up-beating torsional nystagmus (UBTN) with torsional component towards the side of the lowermost ear when the ipsilateral to the involved cupula Dix-Hallpike maneuver (DHM) is performed, indicative of persistent excitatory ampullofugal posterior cupula deflection. A less intense down-beating torsional nystagmus (DBTN) with inverted torsional direction is described when rotating the head to the opposite side or in nose-down position (ND) (180° to the opposite side from DHM), indicative of a persistent inhibitory ampullopetal posterior cupula deflection. The most sensitive maneuver to detect graviceptive posterior cupula is the half-Hallpike (HH) maneuver, where gravity is orthogonal to the posterior cupula axis (Epley, 2001). Neutral positions have been described on further extending the head from DHM (Imai et al., 2009; von Brevern et al., 2015) or in DHM itself (Büki, 2014). An ampullopetal inhibitory cupula deflection on DHM eliciting a contratorsional DBTN has also been suggested (Büki, 2014; Büki et al., 2014) and ascribed to variations in cupula angulation (Fig. 1). Cupulolithiasis (Schuknecht, 1969) and migraine (Asprella Libonati, 2011) have been proposed as etiologies of this variant.

The term subjective BPPV has been proposed for vestibular symptoms without oculomotor findings suggestive of vestibular activation evoked by positional maneuvers (Balatsouras and Korres, 2012). Frequently, these patients have the antecedent of BPPV or uncharacterized positional vertigo. When this symptomatology starts after canalith repositioning maneuvers (CRM), it is generally called residual dizziness. This is a common finding in clinical practice and, most of the time, it is evoked by sitting-up (Büki et al., 2011). It is also generally accompanied with persistent non-positional disequilibrium and spatial disorientation. Büki et al. have described a subgroup of patients with subjective BPPV on sitting-up in whom symptoms are lateralized, and present with posturographic documented anteroposterior truncal oscillations simultaneously with vertigo or retropulsion but without oculomotor findings. They propose that these patients probably represent a definite BPPV variant that is defined as Type 2 BPPV and suggest a P-SCC short arm canalolithiasis as a putative mechanism (Büki et al., 2011).

Recently Scocco et al. (2019) presented a retrospective cohort of 15 patients with vestibular symptoms on sitting-up from DHM accompanied by a transient up-beating nystagmus with torsional component towards the tested side, suggesting an ipsilateral P-SCC cupula stimulation. These patients also described persistent non-positional spatial disorientation and disequilibrium. A posterior canal canalolithiasis limited to the periampullary portion by means of an anatomical restriction of distal movement of the otoconial debris was proposed as a causal mechanism. They also described a sustained inhibitory posterior cupula deflection in most patients on ND manifested by a persistent, less intense, DBTN with opposite

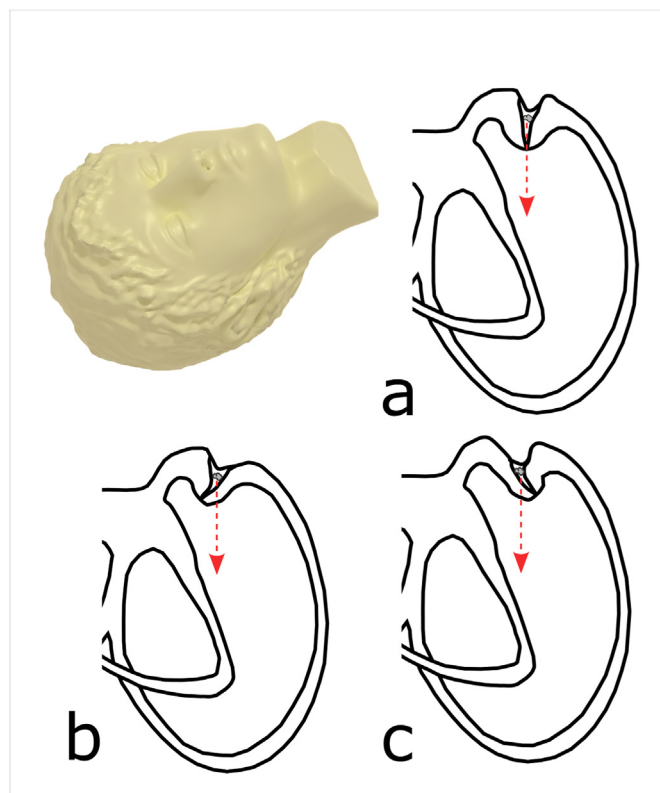


Fig. 1. Possible effects of right posterior semicircular canal cupula plane orientation variants in the context of a heavy cupula on Dix-Hallpike maneuver. Different scenarios could be expected. a, neutral. The plane of the posterior cupula is in the earth vertical plane on Dix-Hallpike maneuver. The cupula is not stimulated; b, a vestibulum angulated cupula (the posterior cupula plane is closer to the earth vertical in sitting position than the neutral configuration) elicits a persistent ampullofugal deflection in the context of a heavy cupula; c, a canal angulated cupula (the posterior cupula plane is farther from the earth vertical in sitting position than the neutral configuration) elicits a persistent ampullopetal deflection in the context of a heavy cupula. The red arrow represents the influence of the gravity force over the cupula.

torsional component. Most patients had recently been treated for ipsilateral typical P-SCC canalolithiasis by means of CRM. No patient resolved their sitting-up nystagmus by means of different CRM intents on the first attempt, but they were all resolved in terms of days or weeks by means of domiciliary Brandt-Daroff exercises.

We present here a series of 18 patients that presented vestibular symptoms and transient UBTN when sitting-up, compatible with sitting-up vertigo BPPV. They were studied by a set of pre-established positional maneuvers in order to better characterize the mechanism of the P-SCC cupula modulation.

2. Materials and methods

At the neuro-otology unit of the Institute of Neuroscience of the Favaloro Foundation University Hospital, Buenos Aires, Argentina, we retrospectively collected a group of 18 patients that were evaluated during the period from June 2018 to June 2021 and that presented with recent vestibular symptoms and showed a transient UBTN on sitting-up from positional maneuvers. Information related to previous medical history, previous history of vestibular symptoms or diagnosis, antecedents of migraine, time elapsed since the beginning of the current vestibular symptoms, history of immediate previous vestibular diagnosis or CRM performed before the inclusion evaluation was recorded. Detailed neurological and

neuro-otological evaluation was performed on all patients. All subjects were examined by means of video-oculography (Chartt, Otometrics, Denmark). Video recordings of the examination sessions were analyzed. Gaze with and without fixation, saccades, smooth pursuit, spontaneous, and head-shake nystagmus were tested. The set of positional tests was pre-established in our clinic for patients that presented with sitting-up vertigo and consisted in bilateral DHM, straight head hanging (SHH), head yaw test (HYT), bilateral half Hallpike maneuver (HH), nose-down position contralateral to the side of the torsional direction of the UBTN evoked by sitting-up (ND). When nystagmus was observed, head position was maintained for at least 1 min or until nystagmus vanished. In six patients, it was not possible to complete the set of maneuvers given particular clinical situations such as intolerance. All patients were followed in person and on a weekly basis for 3 weeks: only then, if the condition continued, was it carried out every two or three weeks until they resolved their symptoms and the positional nystagmus. Some patients underwent brain magnetic resonance imaging (MRI) to rule out central origin of atypical positional nystagmus if clinically considered adequate. Only patients who had video recordings of the positional testing on each visit and a complete follow up were included in the series. Informed consent was obtained in all patients whose video recordings were used as supplemental digital content.

3. Results

Eighteen patients (aged 47–78; median 62; females 13; males 5) were analyzed. Twelve out of 18 patients had a personal documented or suggestive history of recurrent BPPV. Four patients had a personal history of migraine. Four patients had been treated with Epley maneuver at our clinic for P-SSC canalolithiasis. On three of them, the inclusion evaluation was an early re-test in which less than an hour had passed from original CRM. The other patient was tested two months after CRM. On three patients, CRM was performed for positional vertigo outside our clinic and we have only partial information regarding its findings. Time elapsed from the beginning of vestibular symptoms range from 2 days to 60 days with a median of 14 days.

In the description of the findings, we will assign the label “ipsilateral (i)” to the side of the patient shoulder toward which the torsional component of the UBTN evoked by sitting-up was directed. We will assign “contralateral (c)” to the opposite side. All except one patient showed UBTN with its torsional component directed toward the tested side on sitting-up from DHMi. The other patient presented UBTN when sitting-up from SHH. Nystagmus starts after a short latency or no latency at all and then shows a slow crescendo-decrescendo velocity profile different from the typical paroxysmal P-SSC canalolithiasis. It lasts from 15 s to more than 1 min and is accompanied by vestibular symptoms. UBTN on different positional maneuvers was also found: sitting-up from DHMc in ten out of 15 patients tested; sitting-up from SHH in eleven out of 12 patients tested. Sixteen out of 18 patients presented a sustained UBTN with ipsilateral torsional component on HHi for at least 1 min. A slower persistent DBTN with contralateral torsional component was found in eleven out 18 patients tested on ND and in 10 out of 18 patients tested on HYTc. Five patients exhibited a subtle geo or apogeotropic horizontal nystagmus component on HYT. Findings on each patient are depicted in Table 1.

The four patients who had had a documented typical P-SSC canalolithiasis and were treated in our clinic by means of CRM presented UBTN with ipsilateral torsional component when sitting-up from the previously affected side. Most patients described vertiginous symptoms when sitting-up from bed and most

described severe persistent non-positional disequilibrium.

Epley and Semont maneuvers were attempted on all patients without success for the treatment of the sitting-up nystagmus in the initial evaluation. All patients were prescribed domiciliary Brandt-Daroff exercises (2 sets of ten maneuvers to each side a day). All of them resolved their symptoms between 2 and 45 days (median 14 days). Some of them persisted with much less significant asymptomatic positional nystagmus in positional testing but without UBTN on sitting-up. No patient transformed to another BPPV variant during the follow-up. No patient showed evidence of central vestibular compromise in the neurologic and oculomotor examination. MRI imaging on patients on whom it was performed did not show any lesion suggestive of central vestibular or oculomotor compromise.

4. Discussion

In this cohort of patients, we found an UBTN suggestive of stimulatory-ampullofugal deflection of the P-SCC cupula when sitting-up from DHM or SHH. This atypical finding was previously reported (Scocco et al., 2019) and a model of periampullar restricted P-SCC canalolithiasis was proposed. Sixteen out of 18 patients of our cohort showed a sustained UBTN with ipsilateral torsional component on HHi suggestive of persistent ampullofugal P-SCC cupula deflection. Eleven patients presented a less intense sustained contratorsional DBTN on NDc; that is a head position in which the gravity would deflect P-SCC cupula in an inhibitory ampullopetal direction. The persistent and direction-changing nature of the nystagmus and its provocative positions are suggestive of the presence of an ipsilateral P-SCC heavy cupula in these patients (patients 1–11, Fig. 2A, Video 1).

Supplementary video related to this article can be found at <https://doi.org/10.1016/j.joto.2022.02.001>.

Five patients presented UBTN with ipsilateral torsional component on HHi, but did not present nystagmus on NDc (Patients 12–16). We think that those patients could have had a P-SCC short arm canalolithiasis given that they presented signs of P-SCC canal heavy cupula in positions where the weight of dislodged otoconia trapped in the utricular side of P-SCC ampulla would persistently deflect cupula ampullofugally (ie: HHi), but not in ND, where the detached otoconia would fall away toward the ampulla opening or towards the vestibule, thus relieving the stimulation over cupula (Fig. 2B).

It has been speculated that otoliths directly dislodged from the utricular macula or moved from SCC, spontaneously or after a CRM, are prone to fall on the P-SCC short arm given that its opening is in the more dependent position of the vestibule to then elicit a P-SCC short arm canalolithiasis (Oas, 2001). As had been previously proposed, it is probable that otoconia in the short arm of the P-SCC attach to the cupula (Schuknecht, 1969) and thus change its clinical expression to a heavy cupula pattern (Buckingham, 1999; Büki et al., 2014; Schuknecht, 1969) or a mixed pattern with characteristics of heavy cupula and P-SCC short arm canalolithiasis (von Brevern et al., 2015). It is also possible that some of these free floating otoliths on the vestibule simultaneously enter into the H-SCC, eliciting a horizontal nystagmus, as was the case with five out of all patients.

Various mechanisms could account for the sitting-up nystagmus on the heavy cupula or on the short arm canalolithiasis scenarios that we propose here. As mentioned, it is possible that, both in the P-SCC short arm canalolithiasis and also in the heavy cupula secondary to cupulolithiasis scenarios, some fragments of otoconia move freely on the P-SCC short arm or on the vestibulum. On performing positional maneuvers (maximally on DHi and SHH), this free floating otoconia would tend to move away from posterior

Table 1
Patients findings.

	Sex/Age	Side	DHMi	↑DHMi	DHMc	↑DHMc	SHH	↑SHH	HHi	HYTc	Nose down
1	F 73	L	No	↑ti	No	No			↑ti	No	↓tc
2	M 57	R	↓tc	↑ti	↓tc	↑ti	No	No	↑ti	↓tc	↓tc
3	F 60	R	No	No	No	No	↓tc	↑ti	↑ti	↓tc	↓tc
4	M 61	R	↓tc	↑ti	No	↑ti	No	↑ti	↑ti	↓tc	↓tc
5	F 56	L	H geo	↑ti	↓tc	↑ti			↑ti	↓tc	↓tc
6	F 48	R	↓tc	↑ti	↓tc	No	↓tc	↑ti	↑ti	↓tc	↓tc
7	M 78	L	↑ti	↑ti	↓tc	↑ti			↑ti	↓tc + H apo	↓tc
8	F 68	L	No	↑ti	↓tc	No	No	↑ti	↑ti	↓tc	↓tc
9	F 70	R	↓tc	↑ti	↓tc	↑ti	↓tc	↑ti	↑ti	↓tc	↓tc
10	F 72	L	↓tc + H geo	↑ti	↓tc	↑ti	↓tc	↑ti	↑ti	↓tc + H apo	↓tc
11	M 65	R	No	↑ti	No	↑ti	No	↑ti	↑ti	H geo	↓tc
12	F 67	R	H geo	↑ti	No	↑ti	No	↑ti	↑ti	↑ti	H apo
13	F 59	R	H geo	↑ti					↑ti		H geo
14	F 60	R	No	↑ti	↓tc	↑ti	No	↑ti	↑ti	No	No
15	F 61	R	No	↑ti	↓tc	No	↓tc	↑ti	↑ti	↓tc	No
16	F 63	L	No	↑ti	No	↑ti	No	↑ti	↑ti	No	No
17	M 72	R	↓tc	↑ti					H apo	H apo	H apo
18	F 47	R	No	↑ti	No				No	No	No

c indicates contralateral; DHM, Dix-Hallpike maneuver; H apo, apogeotropic horizontal nystagmus; H geo, geotropic horizontal nystagmus; HH, half Hallpike position; HYT, Head Yaw Test; i, ipsilateral; L, Left; No, no nystagmus; R, Right; SHH, straight head-hanging maneuver; ↑ti, up-beat nystagmus with ipsitorsonal component; ↓t, down-beat nystagmus with contratorsonal component ; ↑, sitting-up from.

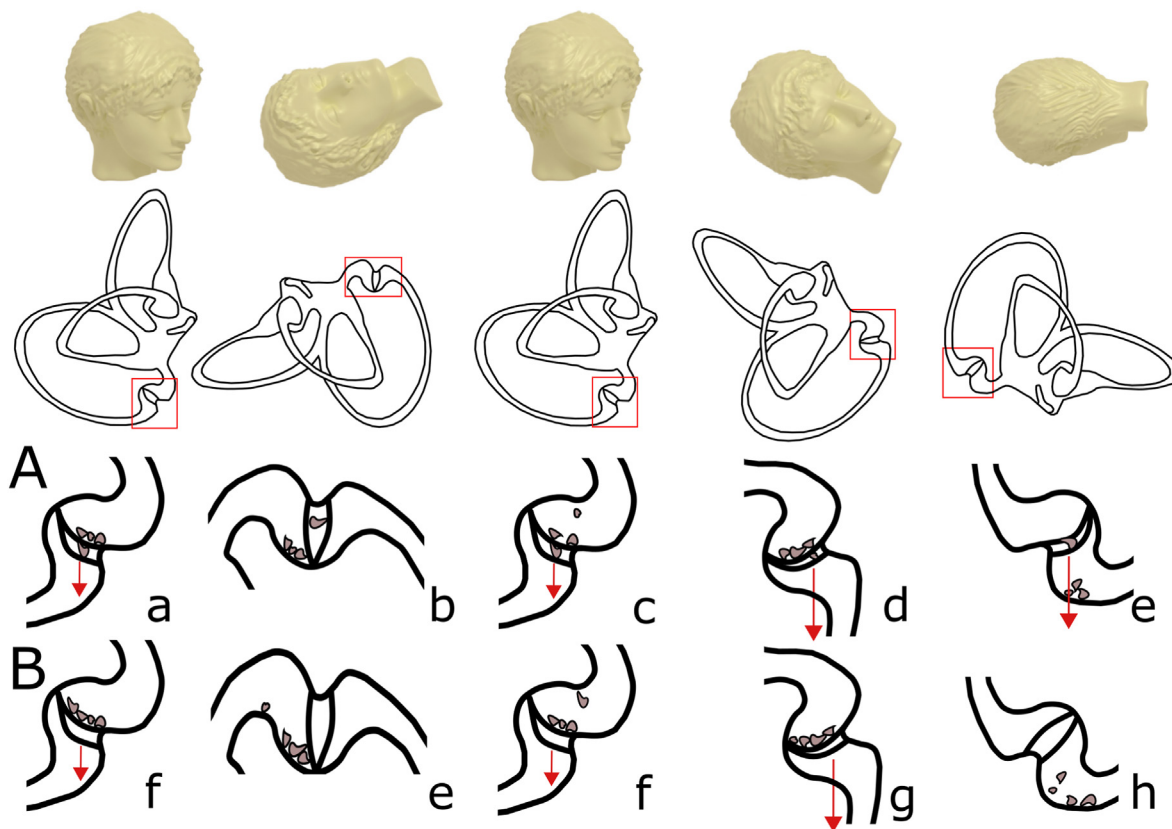


Fig. 2. Right posterior semicircular canal cupula stimulation on different positions (upper and second row) in the context of a heavy cupulolithiasis plus short arm canalolithiasis (A) and a short-arm posterior canal canalolithiasis (B). A sustained ampullofugal deflection is expected on sitting position (a,f) and on sitting-up from Dix Hallpike maneuver (c,h) in both scenarios. A neutral deflection on Dix-Hallpike maneuver is shown in both scenarios (b,g). A maximal ampullofugal deflection on half-Hallpike position is expected on both scenarios (d,i). The nose-down position elicits an ampullopetal deflection on the heavy cupula scenario (e) but a neutral stimulation on short-arm canalolithiasis scenario (j). Red arrow, gravity force influence over cupula.

cupula towards the P-SCC short arm opening to the vestibule. When returning to the sitting position, otoconia would fall by action of gravity towards the posterior cupula and then elicit an ampullofugal stimulation (Fig. 2B). As the mechanism of that stimulation differs from typical canalolithiasis, a different pattern of nystagmus

should also be expected. The long lasting non-paroxysmal crescendo-decrescendo nystagmus pattern that we found in our cohort could be due to that mechanism. This hypothesis has been previously suggested as a putative mechanism for Type 2 BPPV, but without oculomotor findings that support it (Büki et al., 2011).

On the other hand, a P-SCC heavy cupula could alter the appropriate estimation of cephalic rotation speed and orientation due to interference at various points in the central vestibular processing. In the sitting position, the incidence of the gravity vector on P-SCC cupula is not maximal but it is not neutral either, and a sustained ampullofugal stimulatory deflection should be expected (Büki, 2014) (Fig. 2). Since a spontaneous nystagmus on sitting position associated to P-SCC heavy cupula has not been reported, and we have not observed it either, a process of set-point adaptation should probably take place (Büki, 2014; Jareonsettasin et al., 2016) on such natural position in order to avoid a nystagmus that disrupts visual stabilization. It is possible that the stimulation on performing positional maneuvers transiently disrupt such adaptation, thus making the stimulation of the posterior heavy cupula on sitting position in the form of the sitting-up nystagmus visible – that is, till the adaptation takes place again.

There are no reports in the literature regarding adaptation characteristics or vestibulo-ocular reflex (VOR) dynamics in the context of graviceptive cupula. VOR changes in a previously deflected normal cupula have been studied by means of video head impulse in post-rotatory nystagmus (Mantokoudis et al., 2015) and after caloric stimulation (Tamás et al., 2018, 2020). Findings differ between paradigms since, in the former, they did not find influence of the post-rotatory nystagmus on the VOR gain and, in the latter, a decrease in cupula sensitivity was found after an inhibitory or a stimulatory cupula deflection was induced by caloric stimulation. It is uncertain if the dynamics of a heavy cupula should be comparable with these models and also if all the different types of heavy couples proposed (cupulolithiasis, migraine induced) behave in the same way. Also, the dynamic change of the gravity force over the cupula during positional maneuvers was not explored in these studies.

In normal conditions, the sitting-up movement from DHM elicits a P-SCC cupula ampullopetal deflection mediated by the endolymph inertia. When maneuver stops, a transient cupular ampullofugal deflection is produced by the moving endolymph in a phenomenon that is known as canal rotation aftereffect. Normally, a corrective rotation signal of angular velocity opposite to the one produced by the canal rotation aftereffect is produced by the rotation feedback mechanism on the velocity storage, and the rotation canal aftereffect is cancelled (Laurens and Angelaki, 2011). We hypothesize that, on sitting-up from DHM in the context of a heavy posterior canal cupula, the gravity influence on cupula should elicit an abnormal and variable stimulus. Initially, at DHM position, a neutral or slightly inhibitory P-SCC deflection is expected. The ampullopetal deflection expected during the head rotation could be altered by the simultaneous ampullofugal force mediated by gravity or by a different cupular dynamic. During the head rotation, gravity would exert an increasing force over cupula that will tend to deflect it ampullofugally till it reaches its maximum when the head is flexed by 30°. From there, a decreasing but still ampullofugal gravity mediated force is expected till the head reaches the sitting position. When the rotation stops, the gravity induced ampullofugal excitatory deflection will add to the ampullofugal deflection mediated by the moving endolymph to then deliver an enhanced and abnormally persistent canal rotation aftereffect signal to the velocity storage. That anomalous rotational aftereffect would cause a large difference between the direction of gravity estimated by the integration of SCC signals and the otolith signal surpassing the correction capabilities of the rotation feedback mechanism, thus producing a nystagmus in the direction of the inhibited cupula during positioning, here an UBTNi, and a forwardly tilted estimated gravitational direction with a consequent retropulsion (Fig. 3). A similar mechanism has been proposed for central paroxysmal positional nystagmus, where the

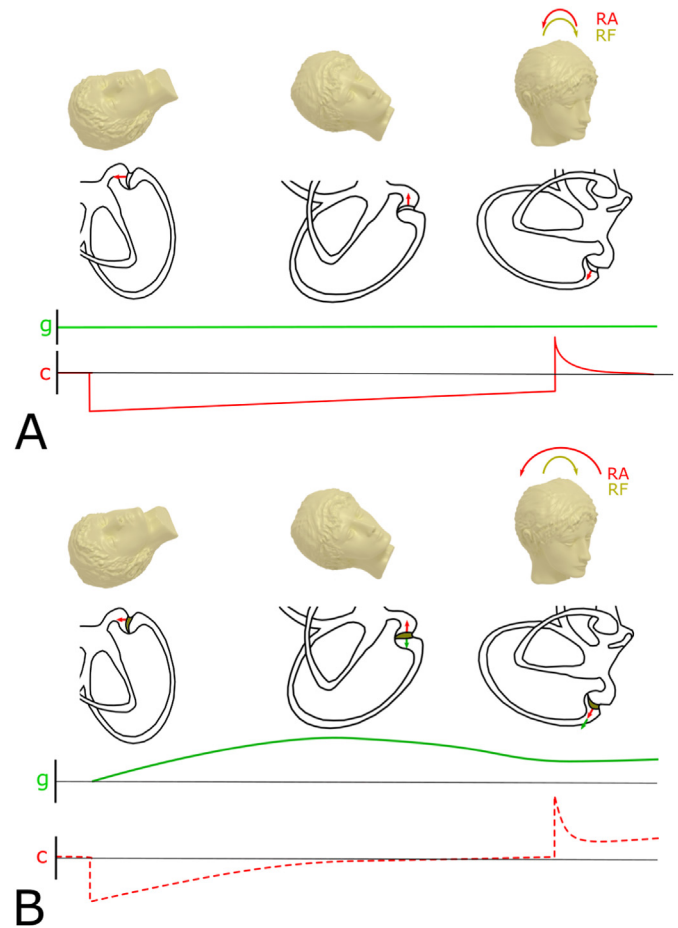


Fig. 3. Right posterior cupula deflection during sitting-up from Dix-Hallpike maneuver. A, a normal cupula is ampullopetally deflected during positioning. When rotation stops, a transient ampullofugal deflection arises as an inertial rotation aftereffect. Gravity has no influence on the cupula. The rotation feedback mechanism of the velocity storage produces a virtual rotation that cancels the inertial rotation aftereffect. B, in a heavy cupula scenario, gravity force increases its influence until the half-Hallpike position is reached. From there until sitting position, the gravity influence is decremental but remains positive. The hypothetical cupular deflection during sitting-up mediated by the interaction between the inertial and gravity forces is depicted by the dotted line (c). An enhanced and abnormally persistent rotation aftereffect is expected given the summation influence of the inertial rotation aftereffect and the gravity force. The rotation feedback mechanism is unable to compensate for the abnormally enhanced rotation feedback. c, cupular deflection (upward ampullofugal, downward ampullopetal). g and green arrow, gravity force influence over cupula (upward ampullofugal, downward ampullopetal). RA, rotation aftereffect. RF, rotation feedback mechanism. Red arrow, influence of endolymph inertia over cupula.

abnormally enhanced rotation aftereffect is elicited by the disinhibition of the irregular vestibular afferents, usually in the context of lesions involving the nodulus and uvula (Choi et al., 2015).

Nine out of eleven heavy cupula pattern patients and 2 out of five short arm canalolithiasis pattern patients exhibited subtle sustained and asymptomatic DBTNc on DHMi, DHMc or SHH. This finding could be the result of an ampullopetal cupular deflection mediated by gravity if this heavy cupula was utricularly angulated, as has been previously suggested (Büki, 2014) (Fig. 1). Another mechanism behind the DBTNc could be the expression of a P-SCC cupula adaptation to a persistent ampullofugal deflection on sitting position that is relieved or diminished on neck extension during positional maneuvers. Clinicians should be alerted of these variations since the presence DBTN on DHM or SHH could be erroneously interpreted as other vertical canal BPPV variants, such as apogeotropic P-SCC or anterior canal BPPV, or central positional

nystagmus, if graviceptive cupula signs were not looked for or were overlooked. Just one patient with a heavy P-SCC cupula pattern in our series presented a persistent UBNTi on DHMi, as is commonly reported (Asprella Libonati, 2011; Ichijo, 2013; Imai et al., 2009). That could imply that our heavy cupula pattern patients could have had a more utricularly angulated cupula axis than those reported elsewhere and that such characteristic makes them prone to sitting-up BPPV.

The benefit of Brandt-Daroff exercises in our patients and the lack of response to CRM are probably due to the fact that this therapeutic approach has a better potential for detaching otoconia from cupula and for disaggregating free floating otoconia on the vestibulum and on the P-SCC short arm. On the other hand, spontaneous resolution in 14 days is reported in migraine-associated heavy cupula (Asprella Libonati, 2011).

Sitting-up nystagmus can be very subtle and should be specifically looked for (Video 2). Likewise, signs of graviceptive cupula could be missed if not specifically looked for on HH and ND positions. It is also possible that a subtle heavy cupula or short arm P-SCC canalolithiasis may exceed threshold to cause spatial disorientation, but not to evoke nystagmus, as could be the case of Type 2 BPPV.

A part of the patients reported in the Scocco et al. series (Scocco et al., 2019) probably share the same pathophysiology as patients reported here, but as sustained head positions were not tested, the presence of a graviceptive cupula is not certain. Phenomenologically, the periampullar restricted canalolithiasis model proposed here predicts similar findings to the one of heavy cupula proposed here. An exception is that ampullofugal cupular deflections and hence UBNTi should be transitory on assuming HHi position after NDC, as shown in two patients of that series, and not sustained as this model proposes.

The same nystagmus pattern on sitting-up, with a down beating contra-torsional nystagmus on DHM was recently reported and a distal P-SCC long arm canalolithiasis with restriction in its proximal and distal movement proposed as putative mechanism (Shigeno and Kitaoka, 2020). Unfortunately, the authors do not provide findings on other positional maneuvers in order to find out whether their patients presented findings suggestive of a graviceptive posterior cupula. Findings on patient 17 in our series, who presented neither signs of heavy cupula nor short arm canalolithiasis, could eventually correspond to that mechanism.

Limitations of this work include the small number of patients in our series given that sitting-up vertigo is an infrequent and transient finding. Also, having quantitative records is essential in order to better analyze nystagmus parameters that will help to clarify its origin. Unfortunately, this is a transient finding and 3D video-nystagmography or scleral search coils are not easily available. Future work involves affected cupula VOR characterization by means of video head impulse test and the testing of a customized positional treatment.

5. Conclusion

We propose graviceptive heavy P-SCC cupula and P-SCC short arm canalolithiasis as other pathophysiologic mechanisms to sitting-up vertigo BPPV. The relation of these mechanisms to subjective BPPV, BPPV type 2 and residual dizziness post CRM is suggested. The close attention to ocular movements on sitting-up and the importance of the half Hallpike maneuver and nose-down position in order to disclose this BPPV variant is underlined.

Conflicts of interest and source of funding

None.

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