Journal of Otology 16 (2021) 65–70

Contents lists available at ScienceDirect

Journal of Otology

journal homepage: www.journals.elsevier.com/journal-of-otology/

Square wave manoeuvre for apogeotropic variant of horizontal canal benign paroxysmal positional vertigo in neck restricted patients

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ARTICLE INFO

Article history:
Received 1 September 2020
Received in revised form
8 October 2020
Accepted 27 October 2020

Keywords:
Benign paroxysmal positional vertigo
Horizontal canal
Cupulolithiasis treatment

ABSTRACT

Objective: We aimed to describe the clinical features of the apogeotropic variant of horizontal canal benign paroxysmal positional vertigo (HC BPPV-AG) in a cluster of patients with restrictive neck movement disorders and a new therapeutic manoeuvre for its management.
Methods: In a retrospective review of cases from an ambulatory tertiary referral center, patients with HC BPPV-AG in combination with neck movement restriction that prevented any classical manual repositioning procedure or who were refractory to canalith repositioning manoeuvres, were treated with a new manoeuvre comprised of sequential square-wave pattern of head and body supine rotations while nystagmus was being monitored, until either an apogeotropic to geotropic conversion or resolution of the nystagmus was observed.
Results: Fifteen patients were studied. All but one [14/15 cases] showed a positive therapeutic response to the repositioning procedure in a single session. In two cases, a direct relief of vertigo and elimination of nystagmus was observed without an intermediate geotropic phase. Although in three patients the affected ear was not initially identified, it was ultimately identified and successfully treated by the square wave manoeuvre in all of them.
Conclusions: The square-wave manoeuvre is an alternative for HC BPPV-AG treatment in either cases with neck restriction, where the affected side is not well identified at the bedside or when other manoeuvres fail to resolve the HC BPPV-AG.

1. Introduction

Benign paroxysmal positional vertigo involving the horizontal semicircular canal (HC-BPPV) is characterized by positional vertigo and direction-changing horizontal nystagmus induced by head turning to either side while supine. The HC-BPPV is the second most frequent variant of BPPV after the posterior semicircular canal BPPV (Baloh et al., 1993; De la Meilleure, Dehaene et al., 1996; Fife 1998; Yacovino et al., 2009). Two typical forms of HC-BPPV have been recognized according to the nystagmus direction in respect to the dependent ear: Geotropic vs Apogeotropic. The most frequent form, Geotropic HC-BPPV, is characterized by a transient nystagmus beating toward the undermost ear when the head is turned to the side while supine (Baloh et al., 1993; Nuti et al., 1996; Fife 1998). Geotropic HC-BPPV is attributed to free-floating otolithic debris in the endolymph of the horizontal canal (canalolithiasis) (Lempert 1994; De la Meilleure, Dehaene et al., 1996; Nuti et al., 1996). In contrast, in the apogeotropic form of HC-BPPV (HC BPPV-AG), a persistent nystagmus beats toward the uppermost ear. The affected side is presumed to be the side on which the supine roll test produces the least intense nystagmus (McClure 1985).

There are two proposed mechanisms for HC-BPPV AG. The most prevalent theory is that it is produced by otolithic debris attached to the horizontal canal cupula (cupulolithiasis) (Baloh et al., 1995; Nuti et al., 1996; Casani et al., 1997; Fife 1998). Cupulolithiasis in theory, might be on the canal side or utricule side of the cupula, but

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

https://doi.org/10.1016/j.joto.2020.10.003

they are clinically indistinguishable through positional testing (Riga et al., 2013; Shim et al., 2015). Anterior arm HC (close to the cupula) canalolithiasis is also considered a second mechanism for the apogeotropic variant of HC-BPPV (Oron et al., 2015; Shim et al., 2015).

In cupulolithiasis, the attached otoconia need to be detached from the cupula in order to manoeuvre them back to the utricular cavity. Once the otoconia are detached, they move to the lowest part of the lateral canal, and at that point, the nystagmus pattern switches from apogeotropic to geotropic. This detaching procedure is known as a liberatory procedure, a term coined by Semont (1988) based on the cupulolithiasis hypothesis (Semont et al., 1988) and reintroduced by De la Meilleure and associates (1996) (De la Meilleure, Dehaene et al., 1996).

The treatment of the HC BPPV-AG has not been well established, and almost all the reported manoeuvres so far, involve either inertial force with vigorous head oscillation (head shaking), a modified Semont manoeuvre, Brandt-Daroff exercises, or using sequential head rotation - the canalith repositioning manoeuvre (CRM) (De la Meilleure, Dehaene et al., 1996; Casani et al., 2002; Casani et al., 2011; Kim et al., 2012; Zuma e Maia 2016; Ramos et al., 2019). Consequently, appropriate neck mobility range is a necessary pre-condition to perform those procedures. Neck movement restriction due to musculoskeletal and orthopedic conditions or pain can be insurmountable obstacles to properly perform any repositioning manoeuvre (Bhattacharyya et al., 2017).

A large number of medical conditions are associated with restriction of neck movement. In subjects older than 65 years, painful neck and rigidity is highly prevalent, ranging from 14.2% to 71%, with a mean of 48.5% (Cunningham and Kelsey 1984; Aoyagi et al., 1999; Fejer et al., 2006). Neck stiffness and the requirement for substantial neck movement may contribute to the observation that treatment efficacy of CRM is lower in older adults in general and in HC cupulolithiasis in particular (Nahm et al., 2019).

The aim of this study is to report a complementary physical manoeuvre to treat apogeotropic HC-BPPV variant in a physically restricted or refractory subgroup of patients.

2. Methods

A retrospective chart review identified 15 sequential patients evaluated between 2017 and 2019, diagnosed with the apogeotropic form of horizontal positional nystagmus, who also suffered from neck movement restriction that prevented the standard treatment or who were resistant to CRM. The patients satisfied the clinical history and examination typical findings of apogeotropic HC-BPPV according to the consensus statement of the Barany society (von Brevern et al., 2015). Persistence of an apogeotropic, horizontal direction changing positional nystagmus (DCPN) with repetitive performance of the positional test supported the cupulolithiasis diagnosis instead of canalolithiasis according to the above-mentioned consensus statement. Patients were considered to have a resistant (refractory) to CRM condition if 1) the modified Brandt-Daroff (from sitting straight ahead position lying down side to side) exercises or mastoid vibration failed to resolve or convert an apogeotropic type of nystagmus to a geotropic one (von Brevern et al., 2015; Kim et al., 2017; Nahm et al., 2019), or 2) patients with normal neck range of movements, but other specific manoeuvres (i.e. Gufoni manoeuvre and head shaking) failed to convert or resolve the nystagmus or vertigo (Gufoni et al., 1998). Neurological findings were otherwise normal. We identified the affected ear by the side on which the nystagmus was less intense, on supine en bloc rotation to either side (Koo et al., 2006).

The study was approved by the local ethics committee and Institutional Review Board (IRB) of Dr. César Milstein Hospital.

2.1. Intervention

Patients with HC BPPV-AG were treated with the manoeuvre as follows (Fig. 1):

1. With the patient in a straight-ahead supine position on the table wearing Frenzel goggles (or Video oculography), his/her head is lifted 20°–30° from the bed, resting on the examiner's hands.
2. The head and body of the patient was turned 90° en bloc to one side while the building up of apogeotropic nystagmus was monitored.
3. Once the nystagmus reached maximum intensity, the patient was turned 180° en bloc to the other side as fast as consistent with safety.
4. Again, after the nystagmus reversed and built up to a maximum intensity, the patient was briskly rotated 180° en bloc to the contralateral side. The procedure was repeated up to ten times in a single session.

As the head position vs time consisted of abrupt changes of head position from 180° left to 180° right, separated by waiting periods while nystagmus reversed and built up, the head position profile is in a "square wave" in position. Accordingly, we call this manoeuvre the "square wave manoeuvre".

The manoeuvre was concluded once the nystagmus changed its direction from apogeotropic to geotropic DCPN or the nystagmus and vertigo was no longer evoked in any position. If geotropic DCPN was observed after the procedure, the 360° Barbecue rotation from the affected to non-affected ear was subsequently performed to remove the free moving otolith from the horizontal canal (Bhattacharyya et al., 2017).

At the end of the manoeuvre, the position that originally provoked the vertigo and nystagmus was immediately repeated and after 30–60 min repeated to verify that treatment had been effective. No post manoeuvre physical restriction was recommended, other than reasonable safety advice. All the patients were instructed to repeat the positional testing at home (roll over on their bed to both sides) 48 h post procedure to subjectively measure the efficacy and also to reschedule a new clinic visit after a week to objectively evaluate them for positional nystagmus.

3. Results

Among the patients, 11 of 15 were women (73 %), with ages from 23 to 93 years (mean 62.5 + 19 years). The main indication for the square-wave manoeuvre was an irreducible neck movement restriction in 11 (73 %) cases and a refractory to all preceding CRM in 4 (27 %) (Table 1). All but three of the patients presented with a history of BPPV, 10 of the posterior canal and 2 of the horizontal canal. In one case, the apogeotropic nystagmus emerged after a Barbecue rotation performed to treat a left side geotropic HC-BPPV variant. In three patients the affected ear was not initially identified. In two, this was due to symmetrical direction changing nystagmus in left/right side positional testing. In the other patient, there was poor tolerance to the sustained head positional testing necessary to reach the peak and the plateau of the nystagmus required to determine the affected side. The affected ear was ultimately identified after the square wave manoeuvre in all of them. In one patient the square wave manoeuvre was performed while she was wearing a rigid neck collar due to her unstable neck condition (post neck surgery). Overall, the mean number of manoeuvres (cycles) required to resolve or convert apogeotropic to geotropic HC-BPPV was 5 (±2.4). In two patients the square wave manoeuvre was able to resolve the nystagmus and vertigo without conversion to a geotropic variant. In another patient the square wave

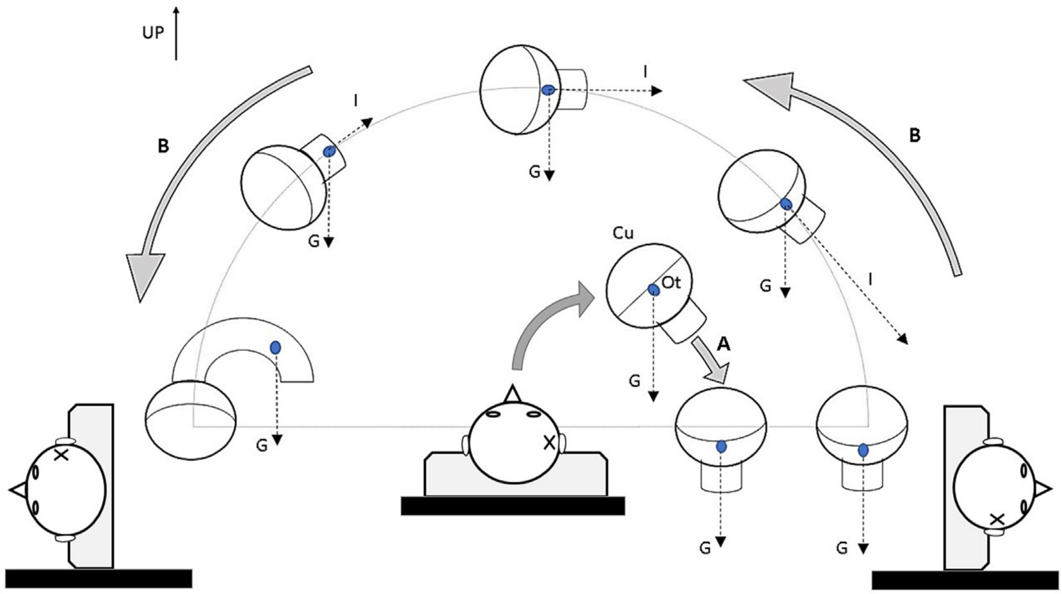

Fig. 1. Square wave procedure for treatment of HC BPPV-AG. Schematic sequences of head rotation in a right-side HC BPPV-AG. Starting at the center, the patient is in supine position and facing up. In Step one; the patient rotates to the affected side (if identified) (A). Gravity (G) provokes cupula deflection, which is simultaneously being monitored by the nystagmus activity. Once the nystagmus velocity increases to approximately the maximum as seen from the supine roll test, the head and body is then rotated 180-degrees to the position 2, on the opposite side (in this case to the left) (B). During the initial whole-body acceleration, inertia (I) and gravity (G) forces contribute otolith detaching force. If the resulting nystagmus is still apogeotropic, when it reaches its maximum velocity, the whole-body rotation is performed again to the other side. This procedure is repeated until apogeotropic nystagmus converts to geotropic, or the positional nystagmus is no longer observed, or 10 cycles have occurred without change. If the procedure was successful, at the end of the rotation the otolith will migrate into the HC canal (shown left part of the graphic). Subsequently, the Barbecue manoeuvre for the HC-BBPV canalolithiasis should be implemented. Cu: cupula; Ot: otolith; X: affected ear.

Table 1
Clinical features of the patients with apogeotropic HC-BPPV that were included in the study. The indication for the square wave (SW) manoeuvre is described. R–BPPV: Refractory BPPV, means that all preceding manoeuvres (Mastoid vibration, modified Brandt Daroff exercises and Gufoni manoeuvre) failed to induce a remission of nystagmus and vertigo. F: female; M: male; L: left; R: right; TBI: traumatic brain injury; CRM: Canalith repositioning manoeuvre; IC: ipsilateral canalolithiasis; RPC: right posterior canal canalolithiasis. UD: Undetermined affected side. *Duration of symptoms is considered from the onset of symptoms to the implementation of the SW therapeutic manoeuvre. ** one cycle is defined as a single 180° side to side movement. Patient 8 underwent an unsuccessful single session (10 cycles) but she was free of vertigo and nystagmus at 1-week the control.

Patient	Age (yrs.)/ gender	Affected side	Etiology of BPPV	Previous BPPV (months)	Days from symptom onset	Cycles performed	Outcome	Indication for maneuver
1	23/M	R	Mild TBI	56	20	6	IC (Barbeque maneuver)	R–BPPV
2	38/F	R	idiopathic/post 1-week CRM for PC BPPV	24	30	8	Complete resolution	R–BPPV
3	43/F	L	Idiopathic	5	15	8	IC (Barbeque maneuver)	Cervical pain
4	53/M	R	Idiopathic	12	23	1	IC (Barbeque maneuver)	R–BPPV
5	55/M	L	Idiopathic	24	10	6	IC (Barbeque maneuver)	R–BPPV
6	60/F	UD	Mild TBI	None	4	4	Switch to RPC (Epley maneuver)	Whiplash-cervical pain
7	60/F	R	Severe TBI	None	2	3	Complete resolution	Cervical collar post TBI
8	61/F	UD	Post-surgical	44	80	10	Complete resolution	Post scoliosis surgery
9	62/F	L	Post-surgical	36	45	3	IC (Barbeque maneuver)	Neck surgery
10	70/M	UD	Superior canal dehiscence	36	30	4	IC (Barbeque maneuver)	Arthrosis with anterocollis
11	71/F	R	Mild TBI (syncopal)	None	55	6	IC (Barbeque maneuver)	Advanced Parkinson disease
12	76/F	L	Idiopathic	8	45	2	IC (Barbeque maneuver)	Ankylosing spondylitis
13	82/F	L	Idiopathic	12	15	4	IC (Barbeque maneuver)	Advanced Parkinson disease
14	91/F	L	Mild TBI	24	8	6	IC (Barbeque maneuver)	Severe arthrosis
15	93/F	R	Mild TBI	12	30	4	IC (Barbeque maneuver)	Severe arthrosis

manoeuvre failed to induce any modification on the nystagmus and vertigo after 10 cycles of body side to side oscillation. In this case, after a week, the positional testing was repeated, happily resulting in an asymptomatic outcome (patients 8). Fig. 2 shows the eye movements observed during the square wave manoeuvre of one case.

To estimate the time required to turns en-bloc 180 degrees of head and body, an analysis of the video recorded of the external camera involving 28 cycles of movement (from 10 patients) was performed. The time spent to turning ranged between 4 and 8 s (mean: 4.9 s), and the velocity of the head and body varied from about 23 to 45 deg/sec.

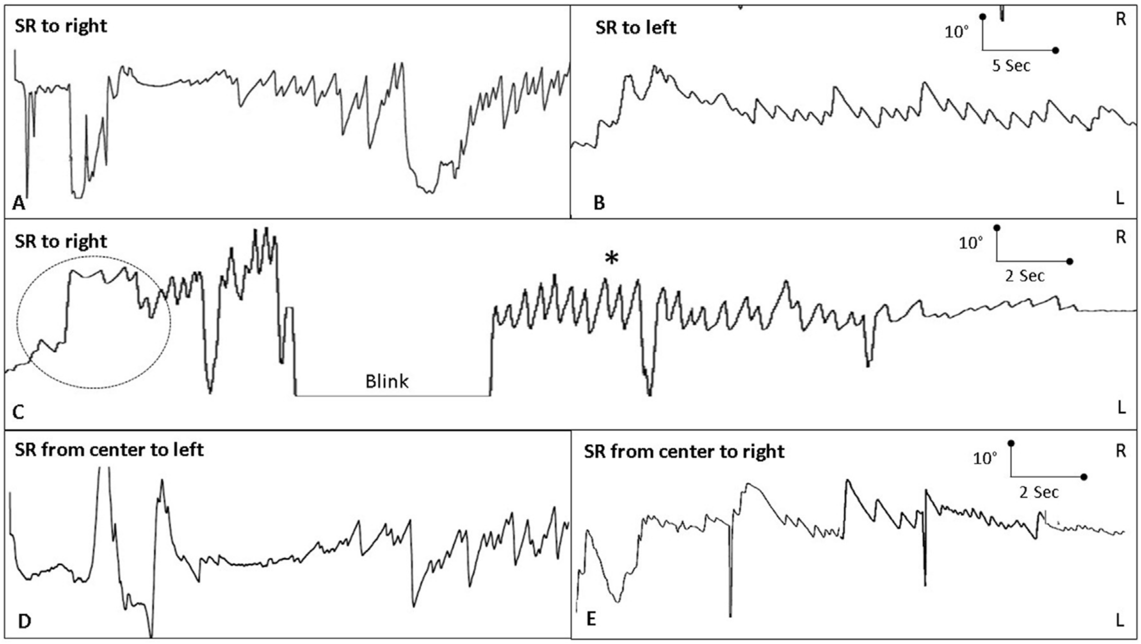

Fig. 2. Square wave procedure recorded by Videonystagmography of case 9. Supine roll (SR) en bloc procedure eyes recorded. Only the horizontal eyes movements are displayed. In the upper panel, figures A and B show a sustained direction changing apogeotropic nystagmus. In C: After a 180° SW manoeuvres, the dashed circle indicates the en bloc body rotation from right to left, followed by an intense burst of short lasting nystagmus to the left (*); presumably the liberatory phase of the procedure. Afterward, in lower panels D and E, a direction changing geotropic nystagmus is observed consistent with canalolithiasis mechanism. After the Barbecue manoeuvre to treat the left HC-BPPV canalolithiasis, the nystagmus and vertigo was no longer observed. R: Right, L: Left.

Although autonomic symptoms, including diaphoresis and nausea were reported in 11 of 15 subjects, in only three of them was of moderate magnitude. Overall, the square wave manoeuvre was quite well tolerated, and no patient required specific assistance to get back home after a recovery period at office.

4. Discussion

Apogeotropic HC-BPPV is more difficult to treat than the geotropic variant. There is only a single randomized controlled trial providing insufficient evidence to recommend a preferred CRM (Kim et al., 2012). The same manoeuvres used to treat the geotropic variant are commonly used to treat the apogeotropic one (e.g. the 360-degree Barbecue manoeuvre) or simply using the same canalith repositioning manoeuvre (CRM) but starting on the opposite side rather than the canalolithiasis variant (Gufoni, Vannucchi-Asprella and Zuma manoeuvres) (Asprella Libonati, Gagliardi et al., 2003; White et al., 2005; Riga et al., 2013; Zuma e Maia 2016; Ramos et al., 2019). The rationale for these procedures is based on the possibility that the debris can be on either the utricular or the canal side of the cupula (or just lodged in the anterior arm of the horizontal semicircular canal) (Ramos et al., 2019). However, those manoeuvres would be effective only when the otoconial debris is dislodged, and the affected ear needs to be identified as well (Tang et al., 2019). A meta-analysis (2015) regarding HC-BPPV with apogeotropic nystagmus showed the efficacy of these manoeuvres ranged between 57 and 65% (Riga et al., 2013). A recent study (2020) failed to find differences in term of efficacy rate between Gufoni's manoeuvre and a sham manoeuvre (64% vs. 69%) for HC-BPPV by cupulolithiasis (Lou et al., 2020).

Furthermore, since all the manoeuvres mentioned involve neck rotations, a considerable range of neck movements is necessary to perform these manoeuvres in the correct form. Pragmatically, one seeks to convert the most treatment-resistant apogeotropic HC-BPPV to the more treatment-responsive geotropic HC-BPPV (Casani et al., 2002; Ciniglio Appiani, Catania et al., 2005). Once the otoliths are detached and free floating into the canal, the 360° or 270° reposition barbecue roll (or supine roll manoeuvre) procedure is indicated (Bhattacharyya et al., 2017; Tang et al., 2019).

In the present study we describe 15 patients with HC-BPPV AG, 14 of whom responded to a single session of the square wave procedure. The remaining case did not display immediate response to the manoeuvre but was asymptomatic at 1-week control. Of course, this successful outcome cannot be certainly linked to the square wave manoeuvre per se. Although this is an uncontrolled case series, these results document a promising new approach to the treatment of HC-BPPV AG in a sub-group of patients with severe neck mobility disorders where other physical treatments are not feasible. Since the square wave procedure involves a symmetric 180° oscillation of head position, this manoeuvre works on unilateral HC-BPPV of either side. Therefore, identifying the affected ear (or the affected side of the cupula) is not necessary.

We propose that a liberatory (detaching otoliths) mechanism, as shown in Fig. 1, is responsible for the effectiveness of this manoeuvre. When the patient is rotated initially 90° to the affected side, a progressive cupular deflection toward the horizontal canal (ampullofugal deflection) is induced by gravity acting on the otoliths. The deflection of the cupula results from an interaction between the force of gravity, the otolith mass and the elastic properties of the cupula. The nystagmus intensity reflects the cupula deflection. As the cupula achieves maximal deflection (and nystagmus), its elasticity is reduced; hence, a further quick rotation of the head (and body) to the opposite side should be associated with tangential and centripetal detaching accelerations resulting in more force on the denser components – the otoliths – than the cupula (Fig. 1). It is noteworthy that only one direction of the square wave procedure cycle has detaching properties. That is, in a left side HC-BPPV cupulolithiasis with otolithic debris attached to the canal side of the cupula, the 180° rotation of the head (and body) to the right should be the liberatory part of the manoeuvre. If, on the

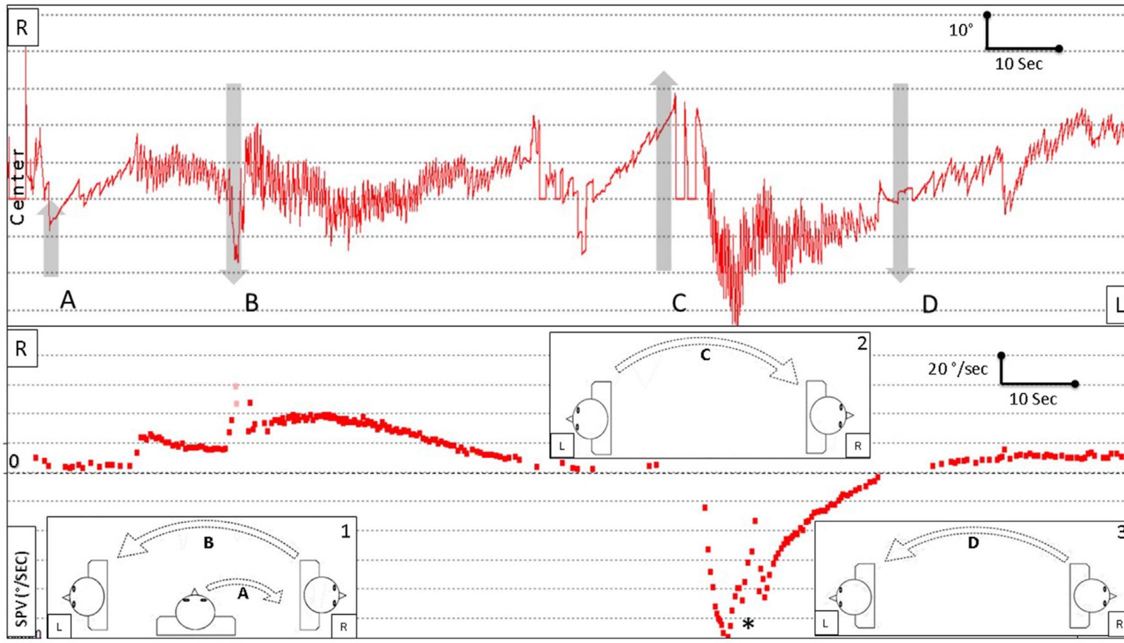

Fig. 3. A continuous Videonystagmography record of a patient (n°4) with apogeotropic DCPN during the supine roll test and SW procedure. The upper part of the graphic shows horizontal nystagmus during the procedure. The lower part of the figure shows synchronized slow phase velocity (SPV) of the nystagmus. °/sec: Degrees/seconds. The short arrow represent 90° head and body rotation from the center to one side. The long arrows symbolizes 180° (side to side) rotation. Up and down directions denote right and left roll directions, respectively. Step by step body and head rotation is represented in the boxes 1,2 and 3. A: The first 90° rotation to the right (first short arrow) is associated with left beating (apogeotropic) nystagmus. B: The 180° head rotation to the left (long arrow going down) evoked the same direction but geotropic nystagmus (left beating nystagmus with left ear down), with a decrescendo SPV profile. Subsequently, in C and D: a direction changing geotropic nystagmus was observed, with more intense SPV with head rotated to the right (*) consistent now with right ear canalolithiasis.

contrary, the otoliths are attached to the utricular side of the left cupula, the head rotation to the left is liberatory. In this case, the liberated otoconia will move directly towards the utricle without a canalolithiasis stage associated. Supporting this theory, in two of the patients the square wave procedure was associated with resolution of nystagmus and vertigo with no intermediate conversion to the geotropic variant, as was described by others (Shim et al., 2015).

One might argue that the mechanism of this manoeuvre need not be one of detaching otoliths from the cupula, but rather it might be an efficient method of dislodging otoconia that are in the anterior arm, but loosely adherent to side of the canals, by adding inertial force to gravity. While this is difficult to disprove, we felt that this was unlikely because in most of the patients, repeated testing during the supine roll test and repositioning procedures performed in all cases at the initial evaluation failed to induce any change on the vertigo and nystagmus. The only exception was one patient in whom a conversion from apogeotropic to geotropic emerged just after a single cycle of head and body rotation, consistent with already free moving debris (canalolithiasis) instead of otoconia adhered on the HC cupula (cupulolithiasis) (Fig. 3).

Although in a strict sense, the procedure described here is considered a detaching manoeuvre, in three cases of this series it also worked as a repositioning manoeuvre. The logical conclusion for these cases is that the otoconia were attached to the utricular side of the HC cupula.

The square wave manoeuvre presented here was quite well tolerated, since the side to side head rotation procedure is performed according to the nystagmus intensity instead of the time (as in other manoeuvres), being the stimulus sub-maximum at all times.

Limitations of this study include, first the small numbers of patients treated and the retrospective cohort of the study. Secondly our results may have potential inclusion biases due to the refractory nature of these patients, therefore the cupulolithiasis mechanism subgroup could be selectively included.

5. Conclusion

The square wave manoeuvre assisted with nystagmus monitoring could be a reasonable therapeutic strategy in a subpopulation of HC-BPPV patients with limited neck mobility that prevents the use of other physical procedures. Nevertheless, there is no logical reason not to use it in patients with normal range neck mobility where other manoeuvres fail to resolve the HC BPPV-AG as observed in four cases here. The procedure could be useful in cases were the affected side cannot be determined. It is desirable however, that more studies are done to confirm our findings.

Declaration of competing interest

On behalf of all authors, the corresponding author states that there is no conflict of interest.

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

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