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Light cupula phenomenon: a systematic review

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Abstract

Geotropic direction-changing positional nystagmus (DCPN) is transient in lateral semicircular canal (LSSK) canalolithiasis; it usually cuts off within a minute and fatigues. However, a "light cupula" mechanism has been described for patients with positional vertigo who exhibit a persistent geotropic DCPN without delay or fatigue. When the cupula becomes lighter than the surrounding endolymph, deflection may occur in the cupula under the influence of gravity. The person experiences dizziness and persistent positional nystagmus can be observed while remaining in that position.

In this review, studies investigating the "light cupula phenomenon," which is a newly defined phenomenon in the literature, were compiled. A systematic literature search was conducted on the light cupula phenomenon in PubMed and Google Scholar databases to illuminate the clinical side of this new phenomenon and reveal its distinctive features. Turkish and English articles published between 2010 and 2021 were scanned; the thesis, reviews, and books were excluded from the study. Forty-eight articles were included in the study.

Mechanisms underlying light cupula has been explained as the "heavier endolymph hypothesis" with increased endolymph specific gravity due to an acute attack such as labyrinth hemorrhage, insufficient inner ear perfusion, or inflammation in the inner ear; "lighter cupula hypothesis" based on alcohol acting on the cupula earlier and making it lighter than the endolymph; "light particle hypothesis" due to the buoyancy of light debris, which are degenerative, swollen, and inflammatory cells in the endolymph adhering to the cupula and the "altered endolymph/perilymph density ratio hypothesis" that the difference in density between perilymph and endolymph causes light cupula. The pathophysiology of the light cupula phenomenon is still unclear, but it can be thought that all the different hypotheses may be effective in this phenomenon. Therefore, nystagmus characteristics and clinical course should be considered in patients for a more effective diagnosis and treatment process. In addition, the results of the studies show that light cupula may not be an uncommon disease and that some patients with geotropic DCPN can often be misdiagnosed as canalolithiasis LSCC-BPPV.

Keywords: Light cupula, Null plane, Lateral semicircular canal, Cupulolithiasis, Positional vertigo

Background

Benign paroxysmal positional vertigo (BPPV) is the most common peripheral vestibular disorder characterized by transient episodes of vertigo triggered by head position changes. The most accepted theory for its pathophysiology is that otoconia leave the utricle and move into the semicircular canals (SCC). Lateral SCC (LSCC) BPPV accounts for 5–30% of all BPPV cases [1]. Positional nystagmus in BPPV involving LSSC is characterized as direction-changing positional nystagmus (DCPN) on the head roll test (HRT). DCPN is caused by the gravitational movement of otolith particles within the LSCC (canalolithiasis); if particles gravitate within the non-ampullary arm of the LSC, the direction of nystagmus is toward the underlying ear (geotropic). Conversely,

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paroxysmal DCPN is apogeotropic on HRT if debris is located within the ampullary arm of the LSC. In addition, it is transient and usually cuts and fatigues within a minute. However, patients with positional vertigo who exhibited a persistent geotropic DCPN without delay or fatigue have been reported, and the "light cupula" mechanism has been described to explain this variant [2].

The cupula is a gelatinous, wavy structure found in each SCC of the vestibular organ. It does not move unless head movements such as shaking the head mean "no" or "yes" or tilting the head to the side [3]. The movement of fluid known as endolymph pushes or deflects the cupula to "excite" or "inhibit" each canal. Normally, the density of the cupula is equal to the surrounding endolymph. When the head is at rest, it remains motionless, and gravity does not affect it. However, deviation of the cupula under the influence of gravity can occur when the cupula becomes lighter (light cupula) than the surrounding endolymph. Under these conditions, persistent positional nystagmus can be observed with the influence of gravity.

This study, it is aimed to compile studies examining the pathophysiology, nystagmus features, prognosis, and treatment methods of the "light cupula phenomenon," which is a newly defined phenomenon in the literature, to illuminate the clinical side of this new phenomenon and to reveal its distinctive features.

Methodology

In PubMed and Google Scholar databases, a systematic literature review on "light cupula phenomenon" was conducted with "light cupula," "density difference and cupula," "direction changing positional nystagmus," "persistent geotropic positional nystagmus," and "lateral semicircular canal and positional vertigo" keywords. While articles published between 2010 and 2021 were scanned, theses, reviews, books, and articles in languages other than Turkish and English were excluded from the study. 596 articles whose abstract or full text were read were excluded because they were not relevant to the subject, and 48 articles were included in the study (Fig. 1).

Main text

Pathophysiological mechanisms

Studies investigating the morphological and functional properties of the cupula have found that changes occur in cupula density and elasticity under different conditions

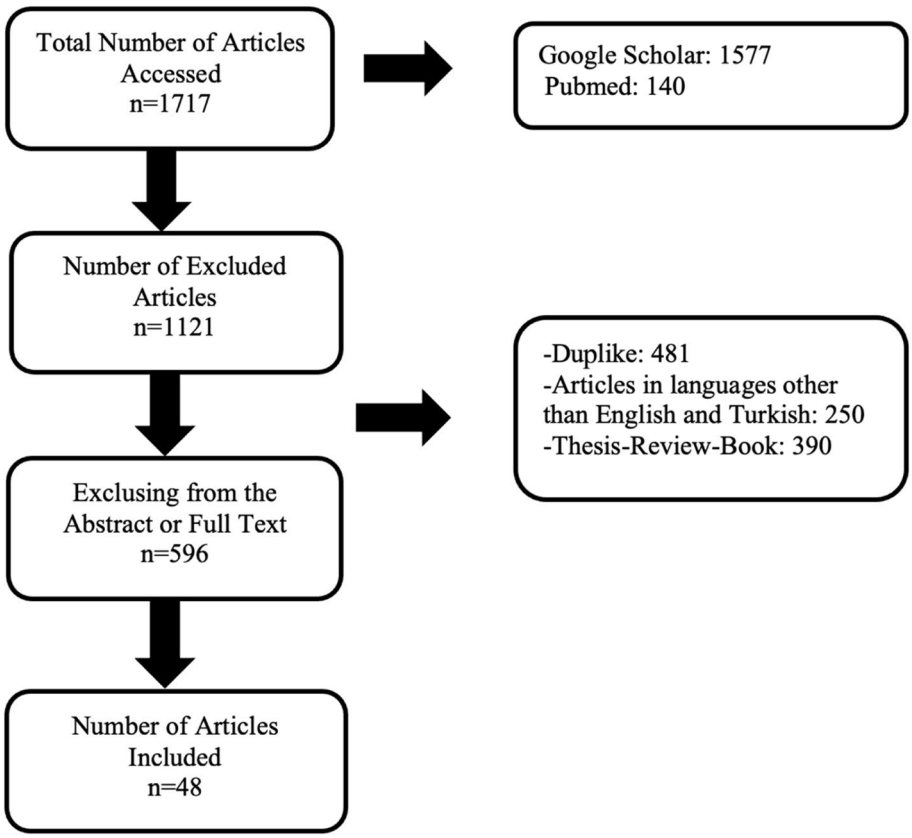

Fig. 1 Flowchart of articles through screening and selection process

[4, 5]. Although more work is needed to elucidate the pathophysiology of the light cupula phenomenon, the "lighter cupula," "heavier endolymph," "light debris," and "density" hypotheses have been proposed [6].

The "lighter cupula" hypothesis

The lighter cupula occurs due to decreased density of the cupula and has been attributed to the mechanism of positional alcohol nystagmus (PAN). Alcohol has a lower density than endolymph, and it is thought to transiently affect the density relationship between the cupula and endolymph by converting SCCs into gravity-sensitive receptors [7]. In the early stage after alcohol intake, alcohol in the bloodstream, due to its proximity to the capillary, enters the cupula faster than the endolymph and makes the cupula lighter than the endolymph [8]. When the affected side is down, the cupula density is lower than that of the endolymph, resulting in ampullopetal flow, resulting in persistent geotropic nystagmus (PAN 1) [7]. Over time, as the blood alcohol concentration decreases, the direction of alcohol diffusion reverses, making the cupula heavier than the endolymph, and apogeotropic nystagmus occurs (PAN 2) [8, 9].

In a study, it was stated that alcohol would act on all cupulas at the same time, and therefore, the vertical pattern of nystagmus would also be present. In the early stages of PAN, shortly after alcohol intake, it is possible to systematically change the spontaneous rate of vertical cupulas [10]. However, this hypothesis has several issues that need to be addressed. First, although it is known that alcohol can cross the blood-brain barrier, there is no evidence that alcohol can also cross the blood-perilymph barrier. Second, the endolymphatic space is strictly separated from the perilymphatic space by the epithelial barrier function of tight junctions in the endolymphatic membrane, and the reticular lamina and supporting cells on the apical surface of hair cells provide a firm seal between the endolymph and perilymph. Most importantly, alcohol enters the endolymph before entering the cupula, as the cupula separates from the apical surface of the hair cells and supports the cells with the endolymph-filled subcupular space [10].

The "light debris (particle)" hypothesis

Light debris adhering to the cupula was thought to cause a light cupula due to buoyancy. Although no physical evidence has emerged, chemically produced substances from degenerative, swollen, and inflammatory cells and otoconial particles in the endolymph have been accepted as potential candidates for light debris [6].

Hypotheses of change in the density of endolymph and cupula were rejected because they were insufficient to explain the sudden emergence of changes in intensities, the information that it resolved within a few days and then recurred, and because it is known that the balance in the body is maintained by homeostasis [6, 11–13]. The light debris hypothesis was thought to explain the clinical features of persistent geotropic DCPN very well. The nystagmus resolves immediately when particles leave the cupula and recurs when attached to the cupula [6, 12]. Two situations have been found to be possible in the light debris theory: one is degenerated otoconia and the other is cells floating in the endolymph such as monocytes or lymphocytes [6]. The fact that there is generally unilateral influence also supports the light debris theory [14]. There is a case study in the literature that reported bilateral exposure, but this was not attributed to the light debris theory, but to the change of specific gravity in the endolymph or cupula due to systemic effects such as hormonal imbalance or being affected by ischemia of the inner ear [15]. In addition, studies have shown that the horizontal canal is frequently affected in case of light cupula. The heavy endolymph theory is insufficient to explain why the horizontal canal is predominantly affected and the anterior and posterior canals are rarely affected [11–13, 16, 17].

The rare occurrence of posterior canal involvement may be attributed to the fact that it is more difficult for light particles to adhere to the posterior SCC cupula than to cupulas in other canals [18].

When nystagmus characteristics were compared in patients with canalolithiasis and light cupula, it was found that the peak intensity, duration, latency, and slow phase rates of nystagmus were similar [16]. Nystagmus slow phase velocities and head rotation angle were also found to be symmetrical in patients with light cupula and cupulolithiasis at the null plane, which was determined as the point where nystagmus disappeared when turning the head from side to side [14]. Therefore, these pathologies were thought to be different but similar conditions, and they argued that the underlying cause of light cupula was the light debris hypothesis [14, 16]. In light cupula, the nystagmus ceases when the head is rotated 20–40° to the affected side (null plane-neutral point) and the geotropic nystagmus resumes and persists with more head rotation which shows that the cupula is deflected by the buoyant force of light particles less dense than the endolymph. Floating light residues herein mean monocytes or lymphocytes floating in the endolymphatic sac, and another possibility was thought to be a decrease in the density of cupula-associated otoconias due to the conversion of apogeotropic nystagmus due to cupulolithiasis to permanent geotropic [19].

Also, hearing impairment often occurs when the endolymph content changes rapidly [11]. If the light cupula phenomenon occurs with an increase in the specific

gravity of the endolymph, it was thought that the tectorial membrane, known as the cochlear counterpart of the cupula, would also be affected by head positions relative to the gravitational plane, because endolymphatic fluids circulate between the cochlea and vestibular organs within the endolymphatic membranes [11, 20]. However, the positional audiometry test performed on the patients showed that there was no change in the hearing threshold with the change of position. This result indicates that the light cupula is due to the light debris mechanism rather than the change in densities. It has been interpreted that the heavy endolymph theory occurs only in the vestibular organ without including the cochlea or that the effects of the light cupula can be altered by outer hair cells working to adapt to the cochlea despite the formation of a light tectorial membrane or heavy endolymph [20].

The "heavier endolymph" hypothesis

It has been hypothesized that endolymph specific gravity may be elevated due to an acute attack such as labyrinth hemorrhage, inadequate inner ear perfusion, or inflammation in the inner ear, resulting in a light cupula condition [6]. Despite all the results supporting the light debris theory, researchers using canalith repositioning maneuvers (CRM) to treat light cupula condition by moving the particles thought that this method was not effective and that the light cupula was due to density theories rather than light particles adhering to the cupula [21–25]. The good prognosis without any treatment suggested that the pathophysiology may not be otolithic [26, 27].

Researchers who think that intratympanic steroid injection (ITS) will affect inner ear inflammation, change the specific gravity of the endolymph or cupula, and will be effective in the treatment if the light cupula is due to the heavy endolymph theory, which is observed that after the ITS application, there was a resolution of nystagmus and that the strong side changed direction. This made the authors think that the heavy endolymph theory was valid [21].

When the nystagmus characteristics of the patients are examined, the observation of spontaneous nystagmus in the sitting position towards the opposite direction of the lesion can be explained by the upward deviation (ampullofugal deviation) of the cupula inclined in the sagittal plane and the base facing the upper and medial in the sitting position, and if the density of the endolymph is greater than the density of the cupula, a slight head movement would be sufficient to induce pseudo-spontaneous nystagmus [28].

The "altered endolymph/perilymph density ratio" hypothesis

Kim and Pham (2019) suggested that the density difference between the perilymph and endolymph causes the light cupula. For some reason, when the density of the perilymph is higher than that of the endolymph, the membranous canal containing the endolymph becomes afloat within the perilymphatic space under the constant influence of gravity, so the head-roll test would reveal the characteristic persistent geotropic DCPN [29].

The pathophysiology of the light cupula phenomenon is still unclear, but it has been demonstrated to arise from a different cause than LSCC BPPV. It is also conceivable that all the different hypotheses may be effective in light cupula; therefore, nystagmus characteristics and clinical course in patients should be considered for a more effective diagnosis and treatment process.

Comorbid disorders

It has been suggested that the light cupula detected in patients with sudden sensorineural hearing loss (SNHL) with positional vertigo is based on density difference rather than light debris theory because the time difference between hearing loss and onset of vertigo is not sufficient for otolithic displacement [30]. In these cases, the intensity of the nystagmus is stronger in turning the head towards the affected side and the null plane can be determined on the same side as the hearing loss. Light cupula may be associated with a change in the specific gravity of the endolymph as a result of various factors, such as disruption of the blood-labyrinth barrier, microcirculation disorder, inflammation, and acoustic trauma that can lead to an increase in protein concentration within the endolymphatic space [30, 31]. In a study conducted with light cupula patients, it was observed that geotropic nystagmus turned into apogeotropic. Two possible explanations have been considered for this result: light cupula formation with an increased specific gravity of the endolymph on the side of hearing loss, followed by an overcompensation of endolymphatic homeostasis and aggravation of light debris adhering to the cupula, and a heavy cupula due to the increased specific gravity of the cupula with an increase in the density of light debris attached to the cupula [32]. It has also been suggested that this pattern, which is also seen in patients with vestibular neuritis, arises from the same mechanism [33, 34].

In case studies, it has been suggested that cupula changes may occur in conditions such as Migraine, Sudden SNHL, and Meniere Disease [35–37]. An acute change in labyrinth function in migraine patients may cause a disorder in the dynamics of the cupula in the lateral or posterior semicircular canals and acute positional vertigo may develop in these patients. The temporal and morphological characters and features of nystagmus allow us to hypothesize a peripheral origin of these disorders and diagnose the associated light cupula [35]. In a case study, it was suggested that the cause of light

cupula-related positional nystagmus lasting 6 months was the morphological change in the cupula due to sudden SNHL [36]. Another case study presenting light cupula in a patient with Meniere also suggested a morphological and functional change in the cupula, but the relationship between endolymphatic hydrops and this change could not be explained [37].

A patient with acute otitis media on the right, permanent geotropic DCPN on the right, and hearing loss with null plane on the left was diagnosed with light cupula, and after intravenous antibiotic and myringotomy administration, constant irritative nystagmus was observed towards the contralesional side. It has been stated that toxic or inflammatory substances enter the inner ear, causing changes in the specific gravity between the cupula and endolymph, and different effects on inner ear function may be responsible for various manifestations of nystagmus [38]. In another case study, supporting the density change between endolymph and cupula was stated that changes in inner ear fluids such as penetration of toxic substances such as antibiotics or bleeding in the inner ear during middle ear surgery in a patient with cholesteatoma may cause light cupula. In addition, considering that stapes mobilization during surgery will damage the inner eardrum and interrupt the inner ear homeostasis; stapes mobilization may be the cause of light ipsilateral cupula [39].

In a case study of a patient who developed permanent geotropic DCPN after meningitis, the heavy endolymph theory is supported. The protein ratio of the cerebrospinal fluid increases due to meningitis and since the cerebrospinal fluid communicates with the perilymph via the cochlear aqueduct, the perilymph is also affected. This increases the osmotic gradient between the perilymph and endolymph, resulting in heavy endolymph and therefore a light cupula. The observation of bilateral involvement in the patient also supports this situation, considering that light cupula should affect the SSCs of both sides [40]. In another case study, it has been argued that the increase in perilymph density that occurs in cases such as hormonal imbalance, diabetes, and neurological problems makes the endolymph float and causes permanent geotropic DCPN [29].

In a study investigating positional nystagmus in patients with herpes zoster oticus, Ramsay Hunt Syndrome, accompanied by severe otalgia and characterized by vesicular eruptions in the external ear canal or auricle, was suggested that detected persistent geotropic DCPN may be due to a minor hemorrhage or altered specific gravity of the LSCC cupula or endolymph due to increased protein concentration in the inner ear [41].

In studies of patients with light cupula accompanied by comorbid disorders such as cancer, inner ear disease [6, 11, 42], sudden SNHL [28–31, 34], and Meniere's disease [42], the null plane at which nystagmus disappears, identified as the pathological side. This supports the knowledge that light cupula phenomenon is caused by changes in inner ear metabolism and should be considered in patients presenting with comorbid disorders. Diseases associated with the light cupula phenomenon have mostly been studied in case studies, so further studies are needed to learn more about possible disorders and to understand the underlying pathophysiological mechanism.

Incidence prevalence

In a study investigating the incidence of light cupula in different patient groups was found that the light cupula diagnosis rate was 4.9% (19 out of 388) among all BPPV types, 9.4% (19 out of 202) among patients with DCPN, and 14.2% (19 out of 134) among patients with geotropic DCPN [43].

When the direction of the lesion was examined, the studies showed predominantly right-sided involvement. In one study, 10 of 27 patients showed left-sided involvement, and 17 had right-sided involvement [6], whereas in another study, 5 left-sided and 13 right-sided light cupula were diagnosed in 18 patients [43]. In another study, the right-left ratio was given as 2:5 [27]. There is also a case study presenting bilateral involvement [40].

When the gender difference was examined, in a study, 65% of the 20 patients were female and 35% were male [24]; in another study, 62.5% of 16 patients were female and 37.5% were male [27]; and in another study, 73.6% of 19 patients were female and 26.3% were male [43]. The common result obtained from the studies is that the incidence is higher in women.

These findings suggest that light cupula may not be a rare disease and that some patients with geotropic DCPN may often be misdiagnosed as canalolithiasis LSCC-BPPV. Since the light cupula phenomenon is not considered clinically much, it should be considered that the prevalence may be higher and should be investigated in larger patient groups.

Prognosis

The duration of positional vertigo occurring with light cupula is longer than in patients with canalolithiasis [44]. In most cases, vertigo and positional nystagmus disappear within 2 weeks [40, 45]. In one study, nystagmus improved in 70.4% of patients within a week and in 88.8% within 30 days [6]. Severe cases are treated for a maximum of 2 months. This process is mainly due to the improvement of peripheral function. When the light debris hypothesis is accepted, the light particle is

completely absorbed as there is no foreign material and LSCC dysfunction is not permanent [12, 45]. Although in some cases patients receive vestibular suppressants [15, 17], the prognosis is generally good without any treatment [26, 39]. Patients usually do not experience recurrent episodes of nausea and vomiting by tilting their head to neutral point [27].

In one study, it was observed that the frequency of attacks varied from 1 to 3 times a year to once a week, and their duration varied from 2 min to 1 day. Approximately, 38% of patients who experienced both short- and long-term attacks reported full-day episodes of vertigo [24].

It was found that the rate of recurrence was higher in light cupula than canalolithiasis and cupulolithiasis, and it relapsed in 34 patients with a rate of 72.3%; this difference is because their mechanisms are different [12]. However, in another study, recurrence was observed in 33.3% of 27 patients, and since this rate is similar to cupulolithiasis, it was concluded that their mechanisms may be similar [6]. More than one recurrence has occurred in some patients [12]. In addition, the recurrence rate was found higher in women [43].

Diagnosis

Head roll test

In studies examining light cupula, the diagnostic criteria were determined according to the head roll test (HRT). In studies, light cupula was diagnosed by persistent geotropic DCPN lasting more than 1 min, the presence of null plane (detection of the affected side) at which nystagmus disappears with a slight head turn to the right or left [12, 20, 26, 46], and absence of nystagmus in the sitting position [19, 21]. However, differential diagnosis with central disorders is important because long-lasting geotropic DCPN can be also observed in central disorders [47]. It is also important to distinguish between LSCC BPPV and transient and persistent nystagmus occurring in light cupula; therefore, at least 1 min of recording should be taken. However, patients suffer from dizziness during nystagmus examination; therefore, it is preferable to distinguish transient nystagmus from permanent nystagmus in less than 1 min. Researchers investigating whether a 30-s observation period would be sufficient have observed a sensitivity of 82–100% (median 100%) and a specificity of 81–100% (median 97%) for distinguishing permanent geotropic DCPN after 30-s observation. In conclusion, they found that transient nystagmus can be distinguished from permanent nystagmus by observing nystagmus for 30 s. Patients should be instructed not to blink during the examination and to maintain their central ocular position [46].

In a study conducted with 12 patients with light cupula, the nystagmus characteristic in HRT was examined and the mean onset latency of nystagmus was observed 2.8±3.5 s while the mean recorded nystagmus duration was observed 86.5±23.8 s. In the affected ear down position, nystagmus peaked at 24.4±9.9 s with a peak intensity of 21.4±12.7°/s. A less intense geotropic nystagmus was observed in the unaffected side turn. The time constant was 11.9±5.6 s when the response was defined as the time that took to reach approximately 63% of the peak SPV (slow phase velocity of nystagmus). It was observed that the time to half-peak of SPV was 27.1±13.4 s [16].

While geotropic nystagmus occurred in the right and left positions, the nystagmus direction in the supine position was found to be towards the healthy side, that is, opposite the null plane [48]. In 2 patients without nystagmus in the supine position, the null plane was 0° and bilateral involvement was considered [43]. In addition, in a patient with posterior canal involvement, no nystagmus was observed since the cupula would be positioned almost perpendicular to gravity and was accepted as the null plane of the posterior canal and a persistent nystagmus with a torsional component occurred towards the unaffected side during right-left head rotation [18]. In another study, apart from geotropic nystagmus, 72% of patients had pitch-dependent nystagmus, and the direction of the nystagmus in the supine position (supination) is opposite to that in the prone position (pronation). The remarkable observation here is that there is a close correlation between the nystagmus intensity in the supine position and the difference in the slow phase velocities (SPVD) of the right and left nystagmus. Median SPVD was 2°/s (1–22°/s) in 20 patients, with >10°/s in 3 patients and ≥1°/s in 17 patients. Theoretically, the intensity of nystagmus in supination may reflect a deviation from the resting position of the cupula, that is, the magnitude of the difference between the density of the endolymph and the cupula. This difference influences the magnitude of the cupula deflection, as the cupula is more affected by gravity, as when the head is in a lateral position. Consequently, the lighter the cupula, the more it deviates in lateral positions and the greater the difference between ampullofugal and ampullopetal deviation as measured by SPV. However, the direction of nystagmus during supination and pronation is not a reliable indicator of the light cupula side, as anatomical and morphological changes in the cupula may shift the axis of the cupula [24].

Null plane (neutral point) Lateralization is based on identifying the stronger side of nystagmus during HRT in patients with transient DCPN but based on identifying null plane which is the nystagmus and vertigo disappeared in patients with persistent DCPN [28].

If the cupula is lighter than the endolymph for any probable cause, it becomes gravity sensitive and the LSCC cupula tilts sideways in the sagittal plane. Given the orientation of the LSCC cupula, a slight head rotation to one side in the supine position can align the ipsilateral cupula axis with the plane of gravity (Fig. 2). Thus, the side of the null plane always corresponds to the side of the light cupula. If only one side is affected in this empty plane, the nystagmus disappears and the direction of the nystagmus is changed with more head rotation [14, 20, 43, 49].

In studies, angle of null plane was found in the range of 0–30° [24], 15–58° (mean 25.4) [26], and 20–60° [45] while the patient's head was turned in the same direction as the slow phase speed. In one study, the angle from the cessation of nystagmus to its reappearance was accepted as the null plane and found to be 2° [48].

The null plane varies greatly. Some patients exhibit a large neutral position angle (more than 40°) [11]; therefore clinicians, should ensure that patients take a completely lateral position in the supine head rotation test. Anatomically, the angle between the supine position and the neutral point is considered as the angle between the cupula and the gravity vector in the affected SCC plane [27]. This differences in the null plane were thought to be due to anatomical variations, but this difference is too large to be explained by anatomical variation alone [11, 26]. Therefore, it may also depend on the position of the light particles [26] and in this case the diversity of the null plane; confusion of positional nystagmus with spontaneous nystagmus may also be the cause when position is insufficient on HRT [11].

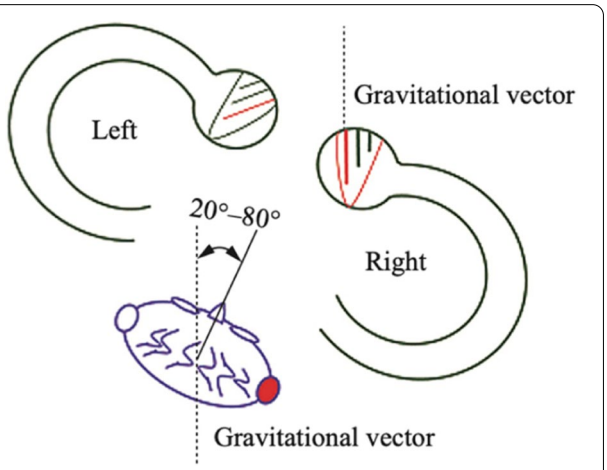

Fig. 2 Null plane [50]

In a study was observed that the severity of geotropic nystagmus on HRT in patients with light cupula is most severe when the head is turned to the affected side, that is, in the direction of the null plane. When the affected ear in the down position, it deviates in the ampullopetal direction, and the intact ear in the down position deviates in the ampullofugal direction. The intensity of the resulting nystagmus should be greater in the first case than in the second according to Ewald's second law [49]. In a study, while in 18 of 22 patients, the dominant side was on the lesion side, it is contralateral in 4 patients, and there is no laterality in 5 patients [6]. However, other studies do not support this finding, while no significant difference was found between the affected and unaffected side in one study [19]; in another study, only 4 of 25 patients had the same null plane and strong nystagmus direction [42]. This may be explained by the incompatibility of the cupula position with respect to the vertigo angle in HRT. The position of the cupula during HRT in permanent DCPN is different from that in transient DCPN. When the head is rotated 20–30° from the supine position to the lesion side, the cupula is parallel to the direction of gravity (null plane) and the nystagmus disappears. An additional 40–50° rotation is made to the lesion side; the cupula is about 40–50° downwards from the vertical axis. However, when performed on the opposite side of the lesion, the cupula is approximately 90–100° relative to the vertical axis. Therefore, stronger nystagmus may occur when turning to the opposite side of the lesion [42]. However, while maintaining a head position, afferent activity in SCCs weakens with adaptation, even if the cupula is permanently deviated. This adaptation makes it difficult to decide whether the nystagmus is lost due to attenuation or an eventual neutral position [10].

Considering these results, the null plane evaluation should be performed in patients with persistent geotropic DCPN; this is an important dividing point between canalolithiasis and light cupula. In addition, the search for the affected side in light cupula phenomenon should be done according to the direction of the null plane, not the strength of the nystagmus.

Vertical pattern Although the horizontal component is dominant, it is observed in the studies that the persistent geotropic DCPN includes vertical and horizontal components [19, 43]. Given that the change in endolymph density will not be limited to LSCC, it is reasonable to assume that positional nystagmus will have vertical and torsional components. However, vertical, and torsional components may be mixed due to the combination of activation or inhibition of vertical SCCs. Moreover, the exact alignment of the cupula in vertical SCCs with respect to the

gravity vector is not clearly defined, although the position of the cupula in the LSCC has been reported. The possibility that the vertical and torsional components originate from the horizontal canal has also been considered [43].. When unilateral LSCC is stimulated, it is natural for the eye movement to have a vertical component because the evoked canal causes a compensatory eye movement parallel to the canal. The lateral canal forms an angle of 18.8° with the Reid horizontal plane (typically about 7° nose up from the horizontal with the head upright). If the LSCC is stimulated, the eye rotation axis is not perpendicular to the Reid horizontal plane. Therefore, it is natural for nystagmus to contain a torsional component [19].

Bow and lean test

In the studies, the benefits of bow and lean tests (BLT) in diagnosis and lateralization were evaluated in patients with light cupula was defined by geotropic DCPN and the null plane in the head roll test. The general finding obtained in the tests performed by tilting the head 90° forward (bow) [20, 49, 51], 45° [20], or 60° backward (lean) [49, 51] while the patients are sitting upright in the chair is the observed permanent geotropic DCPN that direction of bow nystagmus (BN) corresponds to the affected side, while the direction of lean nystagmus (LN) corresponds to the opposite of affected side [19, 42, 48, 49, 52].

In a study trying to determine the effectiveness of the bow and lean tests in the diagnosis and lateralization of cupulopathy, first the head was tilted forward 90° and back 60° and then slightly tilted forward and backward to find the first null plane. Then, it was rotated 90° to the right and left with the head tilted back and 20°–30° to the right–left while tilted back to find the second null plane. Finally, it was rotated 90° to the right and left with the head tilted forward and rotated to the right–left with the head tilted forward to find the 3rd null plane [49]. Side of the affected by light cupula could be effectively find by determining the null planes where BN towards the lesion side and the LN towards the opposite side of the lesion [42, 49, 52]. Other studies have tried to find the null plane with a similar method, but it has been seen that it is more difficult to accurately measure the forward-backward tilt angle of the head and find the null plane in BLT compared to the roll test [35, 48]. However, in light cupula, continued rotation of the head 10–38° to the lesion side has been reported to reduce nystagmus [48].

When nystagmus intensities were compared, BN was found to be stronger than LN in light cupula while LN is stronger than BN in cupulolithiasis [52]. Considering this situation, although it is difficult to differentiate depending on the direction of nystagmus in diagnosis, comparison of nystagmus intensity between BN and LN may provide convenience.

The study investigating the correlation between BLT and roll test found that the results of the two tests were in agreement. In addition, nystagmus was observed in 100% of patients with light cupula on BLT, compared to 55% of patients with canalolithiasis [51]. This is a point to be considered in the differential diagnosis of the two conditions. However, in another study investigating the agreement between HRT and BLT, the agreement was only 33.3% [42]. This may limit the diagnostic value of BLT in light cupula, so more work is needed in this topic.

Dix Hallpike test

Although the number of studies examining the Dix-Hallpike test (DHT) in light cupula is limited, in a study examining patients with sudden SNHL and positional vertigo, while 11% patients with permanent geotropic DCPN have positive DHT and 52% of patients showed negative DHT [30]. In a case study, presenting the right posterior canal light cupula, right DHT revealed permanent torsional left-beating nystagmus. No nystagmus was observed in the left DHT and in the sitting position [18]. Considering the mechanisms underlying the light cupula phenomenon, vertical canal involvement is also possible; therefore, DHT should also be examined, and nystagmus patterns should be clearly identified.

Pseudo-spontaneous nystagmus

Although the term spontaneous nystagmus is used frequently, the term pseudo-nystagmus will be a more accurate definition considering the epidemiology of the pathology. Some studies have observed pseudo-spontaneous nystagmus in patients with light cupula [15, 18, 24, 49]. When interpreting nystagmus in patients with light cupula, it is important to understand the orientation of the LSCC plane within the temporal bone and the cupula within the LSCC, as nystagmus occurs relative to the alignment between the cupula axis and the direction of gravity. Since the anterior portion of the LSCC is positioned approximately 30° above the horizontal plane, the light cupula of the LSCC may deviate under the influence of gravity, resulting in spontaneous nystagmus. When the head is slightly bent forward, the LSCC becomes parallel to the horizontal plane and perpendicular to gravity [24, 49]. Pseudo-spontaneous nystagmus flashing against the lesion side may occur as the cupula tilts towards the canal side in the sitting position and induces ampullofugal flow. Pseudo-spontaneous nystagmus diagnosed in studies is directed towards the unaffected side and is permanent [24, 28, 49]. In a study, pseudo-spontaneous nystagmus was observed in 9 of 20 patients and was consistent with the aspect of nystagmus observed in the roll

test in 7 [28]. A very weak left-beating spontaneous nystagmus was also observed in a case study in which bilateral light cupula was diagnosed [15].

Caloric test

Studies examining caloric test results in patients with light cupula have obtained different results. In a study in which the rate of asymmetry was accepted as 25%, asymmetry was not observed in 11 (79%) of 14 patients; in 3 patients, the affected side showed a stronger response than the healthy side [45]. In another study, 2 out of 10 patients (20%) were found to have canal paresis on the lesion side, and caloric testing was seen as another way of identifying the lesion side of the light cupula [48].

In another study, in which 20 patients were examined, pathological response was obtained in 13, a discrepancy was found between the direction of nystagmus in the supine position and the vestibular disorder in the caloric response, and it was thought that this might be due to the topographic aspect of the long axis of the cupula or endolymphatic changes that changed the direction or morphology of the cupula. When looking whether the SPVD of right and left nystagmus would be an indicator of the affected side, its correlation with the side with a pathological response in the caloric test was evaluated; although no significant correlation was found, for 3 patients with SPVD >10°/s, the side with canal paresis and the side with higher SPV was found correlated [24].

Although there have been cases in which no canal paresis was observed [25], canal paresis was generally obtained on the affected side [13, 36]. In a study, bilateral paresis was observed [37]. The cause of canal paresis observed in caloric response may be central inhibition in the process of vestibular compensation, in which the cerebellum suppresses the vestibular nucleus, rather than damage to a sensory organ [13]. Abnormal results in the caloric response may result from morphological changes in the cupula as well as from endolymphatic disorders that change the morphology of the cupula.

VEMP

Vestibular-evoked myogenic potential (VEMP) outcomes in patients with light cupula have been studied in only two studies [24, 30]. While cervical VEMP (cVEMP) test showed pathological results in only 2 of 13 patients and ocular VEMP (oVEMP) test in 5 patients. The low percentage of pathological results in cVEMP indicates that the potential lesion site is not localized in the lower branch of the vestibular nerve and/or sacculus.

In the study examining the results of VEMP in patients who developed light cupula with sudden SNHL, no response was obtained, and this was assumed to be due to the effects of otolith organ dysfunction [30].

Pure tone audiometry and positional audiometry

Considering that endolymphatic fluids circulate between the cochlea and vestibular organs within the endolymphatic membranes; pure tone thresholds were assumed to be affected if the tectorial membrane density was relatively lower than that of the surrounding endolymph in patients with light cupula, and audiometry with positional changes was performed on patients (Fig. 3). Positional audiometry was performed as follows:

- First, the cochlear apex of the affected ear is directed anterolaterally and inferiorly, in an upright sitting position,
- By directing the cochlear apex of the affected ear to the top, the patient's head is turned 45° to the unaffected side and tilted backwards.
- By directing the cochlear apex of the affected ear downwards, turning the patient's head 45° to the unaffected side and leaning forward; air and bone audiometry were performed with appropriate masking methods.

It was assumed that the subtectorial space would expand in the cochlear apex-up position and narrow in the cochlear apex-down position, but test results found that a change in head position had little effect on the hearing threshold [20]. In case studies, no hearing loss was detected in patients who underwent audiometry [15, 29].

Unaffected hearing in case of light cupula, light cupula arose due to the light particle mechanism rather than the

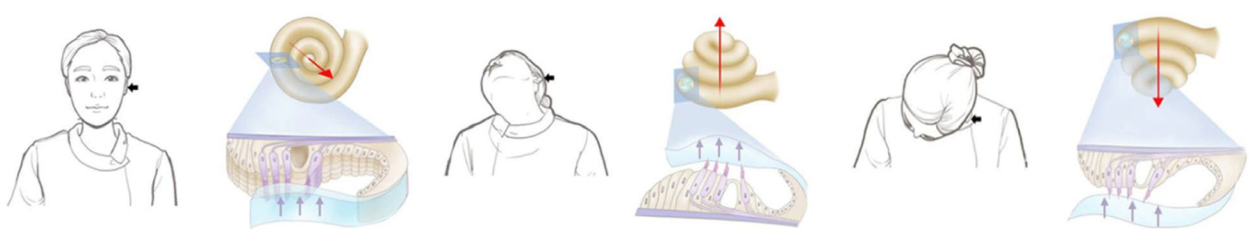

Fig. 3 Different head positions in positional audiometry [20]

change in densities, and it has been interpreted that the heavy endolymph theory occurs only in the vestibular organ without including the cochlea or that the effects of the light cupula can be altered by outer hair cells working to adapt to the cochlea despite the formation of a light tectorial membrane or heavy endolymph [20].

Treatment

Considering the light debris hypothesis, canalith reposition maneuvers are expected to be effective in removing particles from the cupula and are effective in healing, but these maneuvers are not successful in improving the light cupula [24, 40, 43, 47]. Although canal conversion was observed in some patients who underwent CRM (barbecue maneuver), this occurred in those with short-term geotropic DCPN and conversion to apogeotropic DCPN was observed. No effect was seen in patients with long-term DCPN [23].

In another study, the modified CRM was applied as follows, holding the patient in each position for at least 1 min until the nystagmus disappeared or if there was no nystagmus (Fig. 4).

- In the supine position (position 1), the patient's head is turned 60° to the healthy side
- With a hand-held vibrator, a 60-Hz vibration is applied for 30 s to the suprameatal triangle in the posterior superior area of the auricle on the lesion side (position 2)
- Next, the patient's head is rotated another 90° to the healthy side, thus turning it 150° from the starting position (position 3). This posture allows light particles from the cupula to move against gravity towards the utricle.
- The patient's head is then rotated another 90° from the starting position to 240° and oscillated to separate the particles from the utricle side of the cupula (position 4).
- Finally, the patient's head is held higher by another 90° (330° from the starting position) to drop the particles onto the common crus (position 5).

Geotropic DCPN was observed in 28 (80%) of 35 patients with light cupula who underwent modified CRM and in 28 (93.3%) of 30 patients who did not, and it was concluded that modified CRM was not effective in treatment [22]. Considering these results, the differential diagnosis between light cupula and canalolithiasis is important.

The use of ITS, CRM, and vestibular suppressants in the treatment of light cupula was compared, and no significant difference was found between the three methods in Dizzines Handicap Inventory and Visual Analogue Scale during follow-up, but better results were obtained in the ITS group after application [21].

Different methods of treatment have been tried in case studies. Visual suppression was tested by focusing a small light in the supine position in a 7-year-old boy diagnosed with light cupula, and nystagmus was markedly suppressed [13]. In another study, the patient, who was diagnosed with light cupula due to the disruption of the membranous labyrinth and enlargement of the cupula with idiopathic sudden SNHL, continued to experience positional vertigo and nystagmus, despite the administration of vestibular suppressants, and right LSCC plugging

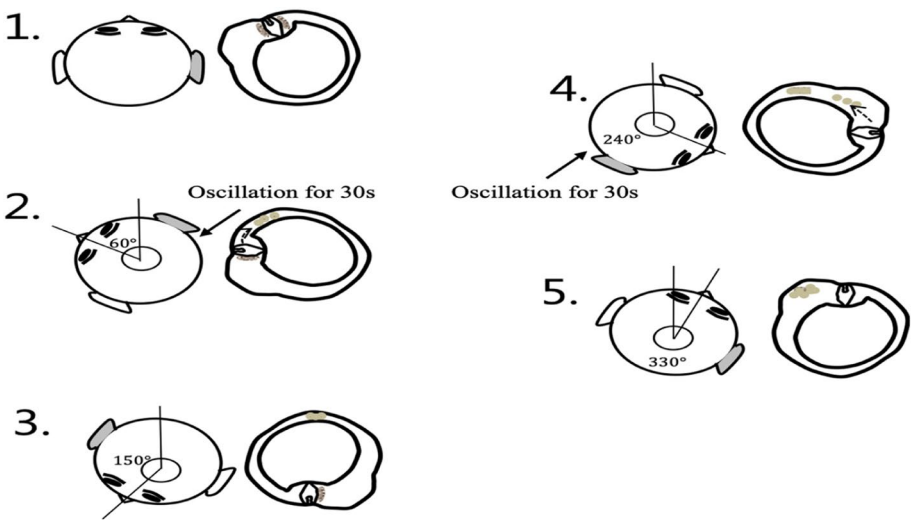

Fig. 4 Schematic of the modified cupulolith repositioning maneuver's 1–5 positions in the right horizontal canal light cupula

surgery was performed. Since the top of the cupula is attached to the ampullary wall, an occluded SCC creates a closed space between the cupula and the occluded area in surgery. Although the cavity is surrounded by soft tissue such as the membranous labyrinth and cupula, it acts as a solid component and prevents compression or expansion of the endolymph; therefore, the cupula is fixed. After surgery, no response was obtained on the caloric test, and the patient did not complain of any dizziness and was able to return to daily activities [35]. In another study, it was assumed that transcutaneous vagus nerve stimulation would affect autonomic control in the activation of SCC and/or otolith organs in the vestibular system. When administered to the patient, the nystagmus resolved, and the patient reported decreased dizziness [53].

The unclear pathophysiology of the light cupula phenomenon causes difficulties in guiding the treatment plan. First of all, light cupula phenomenon should be distinguished from LSCC BPPV and the underlying mechanism should be clearly understood by paying attention to the nystagmus characteristics and the treatment plan should be guided accordingly. Light cupula phenomenon should be considered in cases resistant to CRM.

Conclusion

Light cupula mechanism has been described for patients with positional vertigo who exhibit a persistent geotropic DCPN without delay or fatigue. Normally, the density of the cupula is equal to the endolymph surrounding it, and gravity does not affect it. However, deflection of the cupula under the influence of gravity can occur when the cupula becomes lighter than the surrounding endolymph. Under these conditions, persistent positional nystagmus can be observed under the influence of gravity, and the person will experience dizziness as long as they stay in that position. The pathophysiology of the light cupula phenomenon is still unclear, but it has been demonstrated to arise from a different cause than LSCC BPPV. It can be considered that all the different hypotheses may be effective in light cupula. Therefore, nystagmus characteristics and clinical course should be considered in patients for a more effective diagnosis and treatment process.

In addition, the results obtained from the studies show that light cupula may not be a rare disease and that in some patients with geotropic DCPN may be frequently misdiagnosed as canalolithiasis LSCC-BPPV. The unclear pathophysiology of the light cupula phenomenon causes difficulties in guiding the treatment plan. It should be considered in cases resistant to CRM, and a treatment plan should be established according to the underlying mechanism by paying attention to the nystagmus characteristics. Further studies are needed to elucidate the clinical aspect of the light cupula phenomenon and facilitate its diagnosis.

Abbreviations

BN: Bow nystagmus; BLT: Bow and lean test; BPPV: Benign paroxysmal positional vertigo; cVEMP: Cervical vestibular-evoked myogenic potential; DCPN: Direction-changing positional nistagmus; DHT: Dix-Hallpike test; HRT: Head roll test; ITS: Intratympanic steroid enfection; CRM: Canalit reposition maneuver; LN: Lean nistagmus; LSCC: Lateral semicircular canal; oVEMP: Ocular vestibular evoked myogenic potential; PAN: Positional alcohol nistagmus; SNHL: Sensoryneural hearing loss; SCC: Semicircular canal; VEMP: Vestibular-evoked myogenic potential; SPV: Slow phase velocity; SPVD: Slow phase velocity difference.

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