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ORIGINAL ARTICLE

Diagnostic criteria of barotraumatic perilymph fistula based on clinical manifestations

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ABSTRACT

Conclusions: Patients with sudden hearing loss and dizziness after barotrauma should be suspected of barotraumatic perilymph fistula (PLF). Early surgical repair of PLF showed better hearing outcomes. Therefore, diagnostic criteria should help surgical indications of barotraumatic PLF.

Objectives: The aim of this study was to establish diagnostic criteria for barotraumatic PLF.

Methods: Twenty-four patients (26 ears) underwent surgery on suspicion of barotraumatic PLF. The causes of barotrauma and clinical symptoms were analyzed by surgical findings. Diagnostic criteria of PLF were proposed according to its clinical manifestations.

Results: Definite PLF (17 subjects) was confirmed by any evidence of perilymph leak through oval and round windows. For the other seven subjects (probable PLF), even though there was no evidence of perilymph leak, their clinical manifestations were similar to definite PLF. High frequency hearing loss occurred as an early symptom after barotrauma. Positional dizziness occurred a few hours after auditory symptoms (67%). Positional nystagmus was observed in 10 cases. The characteristics of positional nystagmus were multi-directional, longer in duration, smaller in amplitude, no reversibility, and no response to repositioning maneuver. Regarding the surgical outcomes, hearing was significantly improved in the early repaired PFL group, and dizziness was improved in 96% of patients.

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Introduction

Barotraumatic perilymphatic fistula (PLF) is an abnormal communication between the middle ear cavity and perilymphatic space of the inner ear due to pressure change. Internal barotrauma such as cough or nasal blowing could induce sudden pressure differences between the inner and middle ear space, thus causing rupture of the labyrinthine windows [1]. In addition, minor head trauma could act as external barotrauma into the inner ear. The location of PLF could be either round window (RW) or oval window (OW), depending on the mechanism of barotrauma, resulting in different clinical manifestations.

The diagnostic criteria of barotraumatic PLF are still controversial. Currently, there is no specific pre-operative diagnostic test. The most important clue to suspect PLF is clinical judgment of an otologist based on the patient's subjective symptoms with preceding barotrauma [2]. Most common symptoms of PLF are hearing loss and dizziness. Hearing loss is usually sudden after trauma. It can be progressive or fluctuating. It is usually accompanied by tinnitus and ear fullness. In addition, the complaint of dizziness is usually characterized by disequilibrium and motion intolerance. Dizziness can be provoked by position change with the affected side down. Treatment of barotraumatic PLF is either conservative or surgical. Previously, we have reported that early surgical exploration in suspected barotraumatic PLF is related to better hearing outcome [3]. Therefore, we retrospectively reviewed the clinical symptoms of surgically confirmed PLF in this study and proposed presumptive diagnostic criteria for surgical indications of suspected barotraumatic PLF.

Subjects and methods

Clinical data

From January 2005 through February 2016, 24 patients (26 ears) who underwent exploratory tympanotomy on suspicion of barotraumatic PLF were retrospectively reviewed. Barotraumatic PLF was suspected when patients developed symptoms of hearing loss and dizziness following known history of barotrauma. The Institutional Review Board of Samsung Medical Center approved this study (IRB No. 2015-12-101).

The causes of barotrauma were divided into two types (internal and external) depending on its origin. Barotrauma of internal origin included implosive and explosive types. Implosive forces such as nose blowing, flying, and diving could cause positive pressure into the middle ear, which is then transmitted to the ear via the oval or round window. Explosive barotrauma may be caused by Valsalva maneuver

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and increased cerebrospinal pressure that is then transmitted to the perilymph from the internal auditory meatus or by the cochlear aqueduct [1]. Barotrauma of external origin included minor blunt head trauma such as slap injury. However, severe head trauma causing temporal bone fracture and pneumolabyrinth were excluded. Pre-operatively, audiogram and video nystagmography were checked. Caloric test was done in nine patients to assess vestibular function. Temporal bone computed tomography (TBCT) was checked in every patient. Once PLF was suspected, surgical exploration was performed as soon as possible. Post-operatively, subjective symptoms were assessed and audiograms were obtained at least 1 month after the operation.

Exploratory tympanotomy and surgical findings

Exploratory tympanotomy was performed as follows. Under local anesthesia, the tympanomeatal flap was elevated via endaural approach. Meticulous bleeding control was done to prevent blood contamination in the middle ear cavity. After elevating the tympanomeatal flap, both round window (RW) and oval window (OW) were carefully evaluated for perilymph leak, membrane tearing, or other abnormality such as fibrous mesh and fistula hole. In some cases, an endoscope was used for better visualization of the RW and OW. After investigating in both windows, they were patched with soft tissue and fibrin glue to repair the fistula, regardless of whether PLF was clearly confirmed.

Definite PLF was defined as the presence of any evidence of PLF such as clear fluid collection, fibrous mesh, and fistula hole either in RW or OW. According to the location, definite PLF was divided into RW type, OW type, and undetermined type. Cases were included into undetermined type when the site was unclear, even though the evidence was present. If there was no evidence of definite PLF, it was classified as probable PLF.

Surgical outcomes

Surgical outcomes were analyzed based on hearing change and post-operative dizziness. The pure tone average of four frequencies (0.5 Hz, 1 kHz, 2 kHz, and 4 kHz) was calculated to analyze hearing outcome. Hearing improvement was defined as 'final hearing level less than 25 dB' or 'hearing gain greater than 15 dB' based on the last pure tone average. The post-operative dizziness was classified into four scales as follows: no improvement, partial improvement, complete improvement, and recurred after improvement. Improvement of dizziness was defined as partial and complete improvement.

To analyze the pre-operative hearing threshold according to origin, Kruskal–Wallis test was used. In addition, Mann–Whitney test, Kruskal–Wallis test, and Spearman analysis were performed to compare hearing changes.

Results

Subjects

A total of 24 patients (26 ears) underwent exploratory tympanotomy on suspicion of barotraumatic PLF. Among the 24 patients, two had exploratory tympanotomy on both ears simultaneously. The demographics and clinical characteristics of barotraumatic PLF patients are shown in Table 1. The mean age of these patients was 42 years old

Table 1. Demographics and clinical characteristics of suspected barotrauma perilymph fistula.

Case	Sex/age	Type	Initiating traumatic event	Time sequence of symptoms	TBCT	Symptom duration (days)	Operative findings	Origin
1	M/12	external trauma	trauma	HL + Dz	NL	6	fluid collection	RW
2-R	F/60	implosive	nose blowing	HL + Dz	air-fluid	2	fluid collection	RW
2-L	–	–	–	–	–	2	fluid collection	RW
3	M/17	external trauma	trauma	HL + Dz	NL	48	fibrous web	OW
4	M/44	explosive	straining	Dz → HL	NL	12	no evidence	unknown
5	M/12	external trauma	trauma	Dz → HL	NL	10	fibrous web	OW
6	F/32	explosive	straining	Dz → HL	NL	9	no evidence	unknown
7	F/52	explosive	straining	HL → Dz	NL	9	fluid collection	RW
8	F/40	implosive	nose blowing	Dz → HL	NL	7	fluid collection	unknown
9-R	M/34	explosive	straining	HL → Dz	NL	224	no evidence	unknown
9-L	–	–	–	–	–	224	no evidence	unknown
10	F/15	external trauma	trauma	HL → Dz	not done	45	fibrous web	unknown
11	M/16	external trauma	trauma	HL → Dz	NL	217	no evidence	unknown
12	F/64	implosive	altitude change	HL → Dz	NL	671	fistula (hole)	OW
13	F/54	implosive	altitude change	HL → Dz	NL	3	fistula (hole)	RW
14	F/45	explosive	straining	HL → Dz	NL	6	fluid collection	RW
15	F/43	explosive	straining	HL → Dz	NL	1	fluid collection	RW
16	M/56	implosive	altitude change	HL → Dz	NL	11	fluid collection	RW
17	F/58	implosive	altitude change	Dz → HL	not done	17	fluid collection	OW
18	M/57	explosive	cough	HL → Dz	NL	3	no evidence	unknown
19	M/59	external trauma	trauma	HL → Dz	NL	10	fluid collection	OW
20	M/50	implosive	nose blowing	HL → Dz	NL	7	no evidence	unknown
21	M/59	explosive	straining	HL → Dz	NL	4	no evidence	unknown
22	M/55	explosive	altitude change	HL → Dz	NL	7	fluid collection	RW
23	M/21	explosive	straining	HL → Dz	NL	6	fluid collection	unknown
24	F/49	implosive	ear probing	HL → Dz	not done	12	fluid collection	RW

Symptom duration was defined as interval time from onset of symptom to surgical repair. 'HL + Dz' stands for symptoms of hearing loss and dizziness that were presented simultaneously; 'Dz → HL' stands for dizziness followed by hearing loss several hours later; 'HL → Dz' stands for hearing loss followed by dizziness several hours later; HL: hearing loss; Dz: dizziness; TBCT: temporal bone computed tomography; OME: NL: normal; RW: round window; OW: oval window.

(range = 15–64 years). There were 13 males (14 ears) and 11 females (12 ears). The main symptoms of PLF were hearing loss and dizziness. The types of trauma included external barotrauma such as mild head blowing and slap injury (six ears) and internal barotrauma (20 ears). Internal barotrauma was sub-divided into implosive type (such as nose blowing, flying, diving, nine ears) and explosive type (such as Valsalva and straining, 11 ears). A high-resolution temporal bone CT was pre-operatively obtained with a 0.625-mm slice thickness in 21 of 24 patients. Among these patients, none showed congenital anomaly. Only one patient showed an air–fluid level in the middle ear cavity.

Surgical findings

During exploration, definite PLF was confirmed in 17 subjects. In the definite PLF group, RW fistula was observed in nine subjects, OW fistula was found in five subjects. The origin was undetermined in three subjects. In RW fistula, PLF was confirmed by identifying clear fluid (eight subjects) or tear of RW (one subject). In the OW type, PLF was confirmed by identifying clear fluid leak surrounding stapes (two subjects), fibrous mesh around stapes (two subjects) and slit-like fistula (one subject). In the group with undetermined origin, clear fluid leak (two subjects) and fibrous mesh (one subject) were found in the middle ear cavity. The remaining seven subjects showed no evidence of PLF such as perilymph leak, fibrous mesh, or fistula. Therefore, they were classified as probable PLF.

Clear fluid appeared to be leaked directly through the fistula, while fibrous mesh appeared to be formed after a period of time following fistula development. Surgery was carried out earlier (7.8 ± 4.4 days, $p = 0.031$) when clear fluid was found than that for fibrous mesh (34.3 ± 21.1 days) in Mann–Whitney test. Regarding the location of PLF according to the type of trauma, OW fistula was predominantly caused by external blunt trauma (3/5 subjects). However, RW fistula (8/9 subjects) and probable PLF (6/7 subjects) were caused by internal barotrauma such as straining, cough, nose blowing, and altitude.

Clinical manifestation

The main symptoms of PLF were always hearing loss and dizziness, regardless of the fistula site. Hearing loss was developed suddenly and immediately after trauma. It was usually accompanied by other auditory symptoms such as ear fullness and tinnitus. On pure tone audiograms, the descending type of sensorineural hearing loss was the most common one (Figure 1). Dizziness was developed several hours later than auditory symptoms in most cases (16/24 subjects, 66.7%), especially in RW fistula (7/9 subjects, 78%) and probable PLF (5/7 subjects, 71%) (Figure 2).

Dizziness was usually aggravated by positional change. On physical findings, nystagmus was found in 14 patients

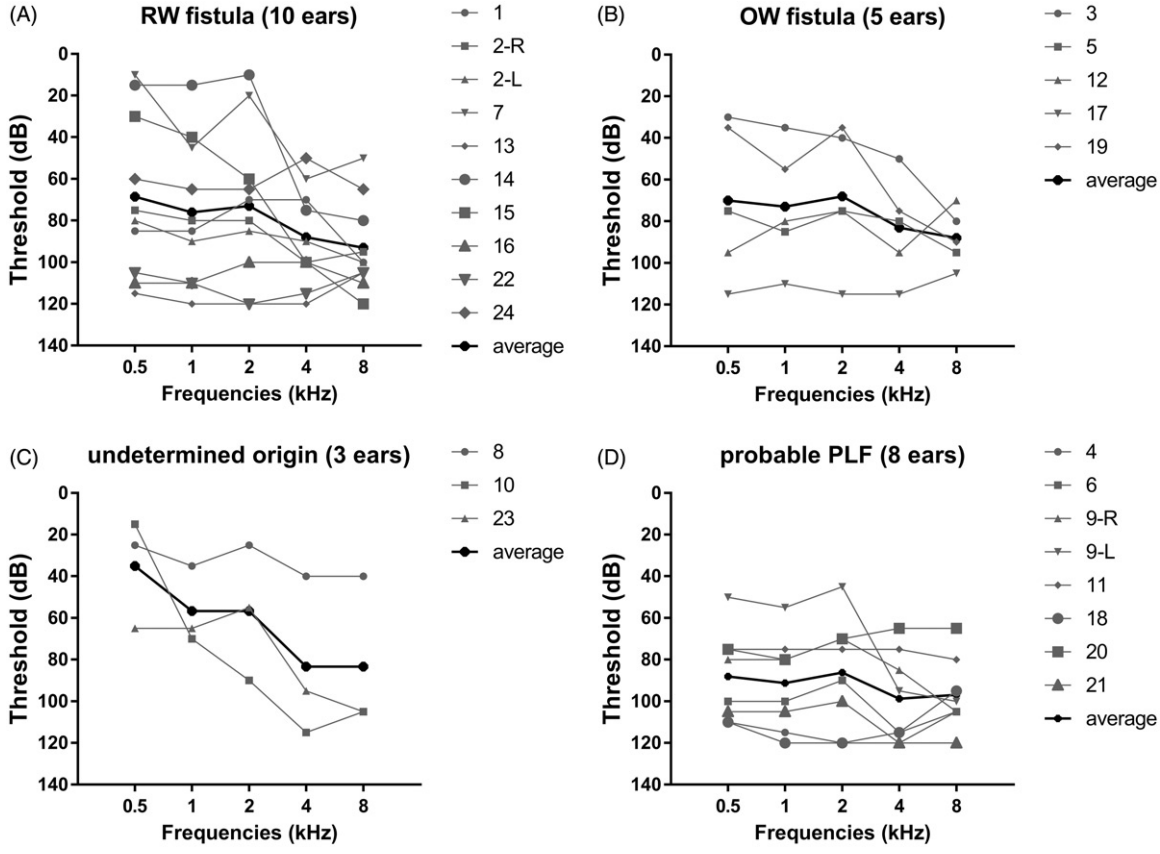

Figure 1. Pre-operative audiograms according to fistula site. On pure tone audiograms, the descending type of sensorineural hearing loss was the most common one. There was no difference in pure tone average at four frequencies (0.5, 1, 2, and 4 kHz) among RW (A), OW (B), undetermined (C), and probable PLF (D) in Kruskal–Wallis test ($p = 0.367$).

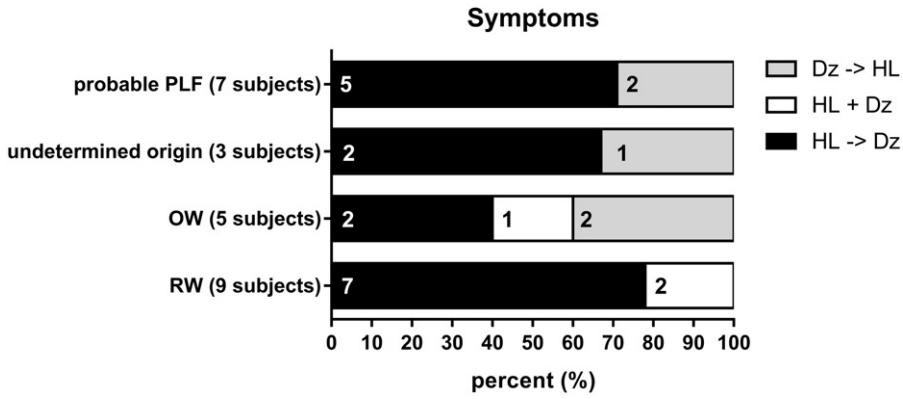

Figure 2. Time difference between symptom onsets. The main symptoms of PLF were hearing loss and dizziness. There was a time interval between symptom manifestation. Dizziness developed a few hours later than auditory symptoms in most cases (16/24, 67%), especially in RW fistula (7/8, 78%) and probable PLF (5/7, 71%). 'HL + Dz' stands for symptoms of hearing loss and dizziness that were presented simultaneously; 'Dz → HL' stands for dizziness followed by hearing loss several hours later; 'HL → Dz' stands for hearing loss followed by dizziness several hours later.

Table 2. Characteristics of nystagmus.

Case	Symptoms	PLF type	Caloric test (CP, %)	SN	Positional test	Dix-Hallpike test
4	Dz → HL	probable	vestibulopathy (100%)	present	unknown	
8	Dz → HL	undetermined	not done	present	unknown	
9-R	HL → Dz	probable	vestibulopathy (56%)	present	R) LB	R) LB
9-L	–	–	–	–	–	–
10	HL → Dz	undetermined	vestibulopathy (49%)	present	R) *UB*, L) *DB*	R) RB → B) *UB* (R > L)
14	HL → Dz	RW	not done	absent	R) LB, L) RB (R > L) → R) *UB*, L) *DB*	–
15	HL → Dz	RW	normal	absent	B) *UB* (R > L)	B) *UB* (R > L)
16	HL → Dz	RW	not done	present	B) *UB*, L) LB	
18	HL → Dz	undetermined	not done	present	R) *UB*	–
19	HL → Dz	OW	normal	present	B) *UB* → R) RB, L) LB	B) *DB*
20	HL → Dz	probable	vestibulopathy (35%)	present	R) RB, L) *UB* → R) RB	–
21	HL → Dz	probable	normal	present	L) LB	B) LB
22	HL → Dz	RW	normal	absent	R) RB, *UB*, L) LB	R) RB, *UB*, L) LB, *UB*
23	HL → Dz	undetermined	normal	present	'R) RB, *DB*, L) RB, *DB*	R) RB, *DB*, L) *UB*
24	HL → Dz	RW	normal	absent	R) RB, *UB*, L) LB, *UB*	R) RB, L) LB

R: right; L: left; B: bilateral; HL: hearing loss; Dz: dizziness; CP: canal paresis; SN: spontaneous nystagmus; RB: right beating; LB: left beating; UB: up-beating; DB: down-beating; RW: round window; OW: oval window.
RW fistula (five subjects) was more common than OW fistula (one subject). Parenthesis indicates intensity of nystagmus and ampullofugal direction of nystagmus presented in italic text.

(Table 2). Among these patients, positional nystagmus with or without spontaneous nystagmus was found in 10 (71%) patients. The pattern of positional nystagmus was similar to that of benign paroxysmal positional vertigo (BPPV). By positional test (Dix-Hallpike test and head roll test), upbeating torsional, downbeat torsional, and horizontal nystagmus were provoked, indicating that posterior, anterior, and lateral canal were involved, respectively. However, unlike typical BPPV, multi-directional and directional changing of positional nystagmus were evoked. Nystagmus was longer in duration, smaller in amplitude, and no reversibility in Dix-Hallpike test. In addition, positional nystagmus was not improved by otolith repositioning maneuver.

Surgical outcomes

After surgical repair of PLF, hearing was improved in 15 out of 24 subjects. The hearing outcomes were associated with surgical timing after symptom onset. The time period of operation after symptom onset was significantly ($p < 0.0001$) shorter in the hearing improvement group (mean = 7 days; range = 1–12 days) than that in the no improvement group (mean = 139 days; range = 6–671 days) in Mann–Whitney test. Hearing gain showed significant correlation with the timing of surgery ($rs = -0.639$, $p < 0.0001$) (Figure 3A), but not with pre-operative hearing threshold ($rs = 0.281$, $p = 0.219$). In addition, there was no significant difference in the degree of improvement of hearing with respect to frequency ($p = 0.126$), confirmation of perilymph leak ($p = 0.644$), or fistula sites (df = 3, $p = 0.428$) (Figure 3B).

Regarding dizziness, the overall improvement rate was ~96% (23/24 subjects). Dizziness was completely improved in 21 subjects after operation. However, two subjects underwent revision PLF repair. One patient had recurred dizziness with up-beating nystagmus on both positional tests. The patient showed web formation around stapes in exploratory tympanotomy. The patient's symptom was improved after sealing OW and RW. The other patients complained of persistent dizziness, even after the surgery, with persistent up-beating nystagmus on Dix-Hallpike test. The patient showed perilymph leak around RW in revision surgery. The patient's dizziness was improved after sealing the perichondrium on OW and RW. However, the symptom remained partially.

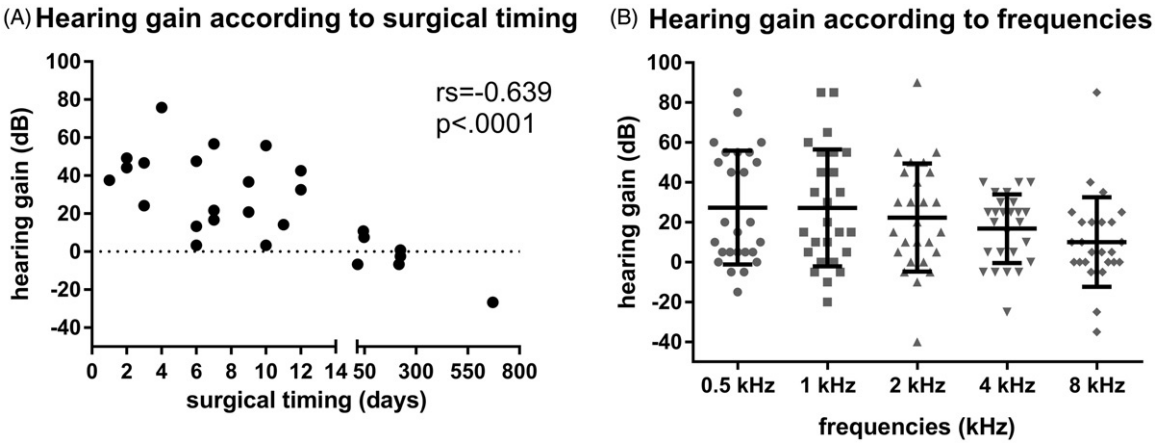

Figure 3. Surgical outcomes regarding hearing improvement. (A) The average of hearing gain at four frequencies (0.5, 1, 2, and 4 kHz) was strongly correlated with surgical timing in Spearman analysis ($rs = -0.639$, $p < 0.0001$). The mean follow-up period was 7 months (range = 1–58 months). (B) Hearing gain was not correlated with frequencies in Kruskal–Wallis test (df = 4, $p = 0.126$).

Discussion

In this study, we confirmed that early surgical repair of PLF was associated with better prognosis of patients with suspected barotraumatic PLF. Therefore, setting the criteria for determining suspected barotraumatic PLF with early surgical exploration will help these patients. By analyzing the clinical symptoms and signs of definite and probable PLF, we proposed the diagnostic criteria of barotraumatic PLF in Table 3.

Among various causes of PLF, traumatic PLF is the most common one [4–7]. Unlike severe blunt head trauma, barotraumatic PLF could cause perilymph leakage without damaging the endolymphatic membrane. Therefore, early surgical repair of PLF can offer the opportunity to recover hearing loss before the inner ear changes may settle and become irreversible [8]. Because inner ear function of these cases are highly likely to have already been changed irreversibly, traumatic PLFs caused by severe head trauma such as temporal bone fracture and pneumolabyrinth were excluded in this study.

There are two types of barotrauma: internal and external. History of internal barotrauma such as nose blowing, straining, and diving can be forgotten or even concealed by the patient [9]. A detailed history of barotrauma is very important for examining patients with sudden hearing loss and dizziness. The types of barotrauma appeared to be associated with the location of fistula. OW fistulas were predominantly caused by external minor trauma, as reported previously [10]. External trauma such as slap injury appeared to cause excessive ossicular movement and break annular ligament around stapes. RW fistulas and probable PLF were mostly caused by internal barotrauma.

If fistula occurred in the RW or OW, the basal turn of the cochlea would be firstly damaged. Therefore, hearing loss in high frequency and tinnitus will occur first. However, if further perilymph leak is prevented through early surgical repair, hearing recovery can be expected. In this study, improvement in hearing was associated with the time of surgery, but not with confirmation of perilymph leak, fistula sites, or pre-operative hearing thresholds. Hearing was significantly ($p < 0.0001$) improved in the early exploration group (mean = 7 days; range = 1–12 days). The degree of hearing improvement was also significantly correlated with surgical timing ($rs = -0.639$, $p < 0.0001$) (Figure 3A). Other studies have reported that only 17% and 20% of patients have improvement in hearing after surgeries [11,12]. Compared to previous results, the hearing outcome was better in this study because of the surgical timing. In previous studies, surgical intervention was performed after 6 weeks of conservative management or more than a month after symptom onset.

Table 3. Diagnostic criteria of barotraumatic perilymph fistulas.

Definite perilymph fistula
Probable barotraumatic perilymph fistula plus visual confirmation of perilymph leak through OW or RW via surgical exploration
Probable perilymph fistula
1. Preceding history of barotrauma
2. Sudden auditory symptoms (sensorineural hearing loss, tinnitus, and/or ear fullness)
3. One or more features of dizziness accompanying with auditory symptoms
A. Time difference between the onset of auditory symptoms and dizziness
B. Dizziness aggravated by positional change
C. Positional nystagmus (by Dix-Hallpike test and Supine Head Roll test) having at least one of the following characteristics
i. Multi-directional positional nystagmus
ii. Small amplitude, long duration, no reversibility
iii. No response to otolith repositioning maneuver

OW, oval window; RW, round window.

Dizziness occurred with auditory symptoms simultaneously or with time intervals. In particular, 67% of dizziness developed a few hours later than auditory symptoms, especially in RW fistula (78%) and probable PLF (71%). This could be a characteristic symptom of barotraumatic PLF. This indicates that it might take some time to affect the vestibular system after cochlear damage.

In addition, dizziness usually had a positional effect. In 10 patients, positional nystagmus was observed. Positional nystagmus had the following characteristics. The direction of nystagmus was mainly upbeating torsional, downbeating torsional, or ageotrophic horizontal direction, in which deflection of the cupula might occur in the direction from the excitatory stimulus (ampullofugal) to posterior canal, anterior canal, or horizontal canal. Pattern of nystagmus was multi-directional or directional changing as if multiple

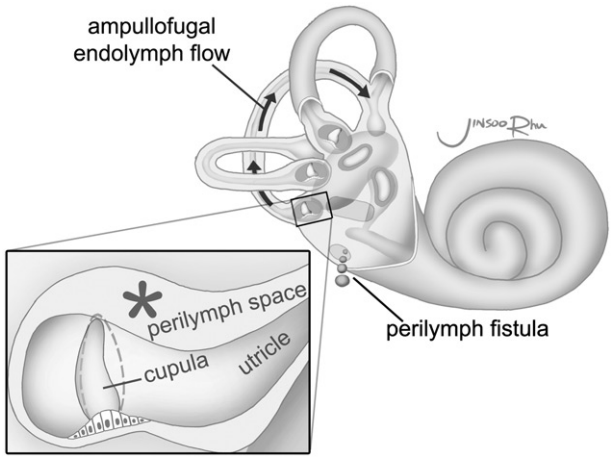

Figure 4. The mechanism of positional nystagmus in barotraumatic perilymph fistula. Perilymph leak through fistula by positional change induces the collapse of membranous labyrinth (asterisk). The collapsed membrane deflects the cupula of crista commonly in ampullofugal direction. The deflection of cupula in three semicircular canals induces excitatory signal to the canal simultaneously. Multi-directional and direction changing positional nystagmus could be evoked.

canals were influenced together. Nystagmus sustained longer in duration with slower velocity without reversibility. There was a tendency of no improvement by the otolith repositioning maneuver. These can be explained by the following reasons. In the animal model of PLF, leakage of perilymph can induce the collapse of endolymphatic membrane [13–15]. Collapse of the membranous labyrinth can also be formed by the outflow of perilymph through fistula when the movement of the perilymph is generated by the positioning. If the collapsed membranous labyrinth induces cupular deflection, positional nystagmus can occur. This may happen in all three semicircular canals. Therefore, multi-directional or directional changing nystagmus was evoked by positional tests (Figure 4).

Surgical intervention was very effective for dizziness control, despite the long duration of symptoms. Four patients whose symptom developed more than a month before surgery were completely improved ($n = 3$) or abated ($n = 1$) after surgical repair. The overall improvement rate for dizziness was ~96%. This result was comparable to that of previous studies [8,11].

Treatment principles of PLF can be divided into conservative and surgical treatment. Conservative therapy includes bed rest, with high dose steroid-like idiopathic sudden sensorineural hearing loss to wait until spontaneous closure of fistula. Conservative therapy can be applied when there is no dizziness at all, even if PLF is suspected and a considerable amount of time has passed after the symptom onset without change in hearing for a long duration. However, surgical repair is better when hearing loss occurs after barotrauma accompanied by delayed onset of dizziness. Dizziness is influenced by positional change with positional nystagmus at the time of examination. Because surgical technique is effective in improving hearing and dizziness, easy, and well tolerable to the patients without side-effects, we have no hesitation in deciding surgical exploration if unstable perilymph dynamics by PLF is suspected. Some studies have recommended exploratory tympanotomy in SSNHL patients who have no response to systemic treatment because PLF can be a possible cause of hearing loss [16,17]. However, hearing outcomes were not favorable in those studies. Based on our data, surgical repair for persistent hearing loss without dizziness after barotrauma is not recommended.

Conclusion

Early surgical repair of PLF is recommended to improve hearing and dizziness in clinically suspected barotraumatic PLF. To make a guideline for surgical indication in PLF, we proposed the diagnostic criteria of barotraumatic PLF. Sudden sensorineural hearing loss with dizziness immediately after known barotrauma should be suspected as PLF. Time delay of dizziness following auditory symptoms was evident in most fistulas. Positional nystagmus was observed. However, unlike typical BPPV, positional nystagmus in PLF showed multi-directional, longer in duration, and smaller in amplitude without reversibility. In addition, positional nystagmus was not improved by otolith repositioning maneuver. The identification of perilymph leakage is impossible in every case. However, early exploration of suspected PLF can increase the likelihood of PLF detection and improvement of their symptoms.

Disclosure statement

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

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