

Perilymph fistula: the patients' experience

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Abstract

Objective: This study aimed to assess the experiences and outcomes of patients who underwent surgical repair of a perilymph fistula in Norfolk, UK.

Methods: The study involved a retrospective questionnaire-based patient survey and case note review of patients who had undergone tympanotomy and perilymph fistula repair between 1998 and 2012 in two district general hospitals.

Results: Fourteen patients underwent 20 procedures, of whom 7 completed the pre- and post-operative Vertigo Symptom Scale. In five patients, there was no obvious precipitating cause. Perilymph fistula was precipitated by noise in one patient, by a pressure-increasing event in six patients and by trauma in two patients. The Vertigo Symptom Scale scores showed a statistically significant improvement following surgical repair, from a median of 67 (out of 175) pre-operatively to 19 post-operatively.

Conclusion: In selected patients with vertigo, perilymph fistula should be considered; surgical repair can significantly improve symptoms.

Key words: Perilymphatic Duct; Fistula; Vertigo; Outcomes Assessment

Introduction

Perilymph fistula is defined as an abnormal connection between the perilymph compartment of the inner ear and the middle ear. Some cases are caused by trauma or erosive disease (e.g. cholesteatoma), or following ear surgery, particularly stapedectomy. Twenty per cent of cases have been reported to be spontaneous; however, many of these were associated with a preceding pressure-increasing event.¹

There is no agreed 'gold standard' diagnostic tool, and currently diagnosis of a perilymph fistula is based on a compatible history, examination and investigations. Direct observation at tympanotomy may be the only way to definitively identify a perilymph fistula, but even this is controversial, with a wide range of positive identification rates reported in the literature (24–100 per cent).²

Patient history features which may be suggestive of a perilymph fistula include a pressure-increasing precipitating event that causes episodic vertigo, and fluctuating hearing loss sometimes associated with tinnitus and aural fullness. These can be provoked by further pressure-increasing events such as coughing, straining or sneezing. Some patients report vertigo in the presence of loud sounds (Tullio phenomenon). It is

notable that this clinical picture may be similar to Ménière's disease and superior semicircular canal dehiscence. The former is not usually associated with pressure-increasing events, and has specific diagnostic criteria set out by the American Academy of Otolaryngology – Head and Neck Surgery.³ Superior semicircular canal dehiscence is diagnosed on computed tomography (CT) scans.

Clinical examination findings are often normal, particularly if the patient presents some time after the precipitating event; they may, however, have a positive 'fistula test' result. The Fraser positional test may yield positive results, wherein audiometric thresholds improve after lying with the affected ear uppermost for 30 minutes.

Imaging is often not sensitive enough to detect a small leak, but high resolution CT scanning may show abnormalities predisposing to a perilymph fistula and may show superior semicircular canal dehiscence. Magnetic resonance imaging (MRI) with intrathecal gadolinium has been used to detect leaks, but is not without complications.⁴

In light of this diagnostic uncertainty, management decisions can be difficult. The options include conservative measures, such as bed rest with head elevation to

30° and the avoidance of pressure-increasing activities.⁵ The alternative is surgical exploration via tympanotomy and repair of the leak with a graft.

The literature is very sparse regarding perilymph fistulas, and there appear to be no formal patient-reported outcome measures that have been used following tympanotomy and perilymph fistula repair. Outcomes are difficult to assess because of the lack of an accurate diagnostic test for pre- and post-intervention comparison. We therefore analysed a case series undergoing tympanotomy and fistula repair, to see whether this treatment improved patients' symptoms, using a validated patient-reported outcome tool.

Materials and methods

In order to test the null hypothesis that there is no difference in vertigo before and after perilymph fistula repair, we undertook a retrospective questionnaire-based survey and case note review. Patients who had undergone exploratory tympanotomy and perilymph fistula repair between 1998 and 2012, at two hospitals in Norfolk UK, were included.

A presumed diagnosis of perilymph fistula was made following a clinical assessment. The relevant clinical and operative investigations and findings were recorded, including intra-operative video evidence of a perilymph fistula.

Surgery was performed via an endaural approach and a tympanomeatal flap was raised to examine the middle ear. An anaesthetic Valsalva manoeuvre was performed to assess for a leak. If a leak was identified, this area was plugged with fat harvested from the subcutaneous tissue at the incision site. If no leak was identified, the area plugged was at the discretion of the surgeon. Post-operatively, patients were advised not to strain. They were discharged the same day or the following day. The first review occurred two weeks later and further follow-up appointments were arranged on an individual basis.

Yardley and colleagues' Vertigo Symptom Scale was used to assess patient outcome.⁶ In this validated questionnaire, 5-point Likert scales are used to assess patients' experience of 23 symptoms during an average month, wherein 0 equates to 'never' and 4 to 'very often, on average once a day'. A questionnaire pack, which included pre- and post-operative questionnaires, was sent to the patients, with a covering letter and reply envelope.

Data were analysed statistically using a Wilcoxon signed-rank test; a *p* value of less than 0.05 was considered statistically significant.

Results

Fourteen patients underwent 20 procedures between November 1998 and July 2012. Seven patients were female and seven were male. Mean age at surgery was 43 years (range, 15–73 years).

The main presenting symptom was vertigo. Nine patients also had tinnitus and 13 complained of

hearing loss. Precipitating events occurred in nine patients; these consisted of acoustic trauma, sneezing, straining, a self-performed fistula test, nose blowing, leaning down a manhole whilst reading water meters, assault and barotrauma from a football hitting the patient's ear. Five patients could not recall any precipitating event. The vertigo was provoked by the patient performing a fistula test on themselves in three patients.

All patients underwent tympanotomy. A definite leak was seen in eight patients during an anaesthetic Valsalva manoeuvre. A likely leak was seen in seven patients and no leak was seen in five patients. Table I shows the location of the leaks. In 10 cases, the round window niche was plugged. In nine cases, both the round window and the oval window and/or stapes footplate were plugged. In one case, just the footplate was plugged. In the majority of cases, fat was used, but in one case fascia was used.

The mean follow-up time was 29 months (range, 0.5–132 months). In 18 of the operations, there was a report of benefit. One patient reported no change and a further patient was awaiting follow up at the time of writing. The Vertigo Symptom Scale was returned in seven cases, giving a response rate of 37 per cent. The data were analysed graphically and did not follow a normal distribution, as confirmed by a Shapiro–Wilk test. Therefore, non-parametric statistics were used to analyse the data. The Vertigo Symptom Scale scores improved from a median of 67 (out of 175) pre-operatively to 19 post-operatively. The Wilcoxon signed-rank test statistical result was $W = 21$. Using a table of critical values of statistical results for small samples, the *p* value was less than 0.05 for a non-directional test. This indicates a statistically significant improvement in vertigo symptoms, from pre- to post-operation (Figure 1).

Six patients experienced recurrence of symptoms, between 4 months and 11 years after the surgery. In one case, symptoms recurred four months post-operatively – this was associated with heavy lifting. Three patients underwent revision surgery, one of whom had two revisions. One patient had bilateral sequential perilymph fistulas.

There was no difference in pre- and post-operative audiogram findings for the majority of patients with complete audiometric data. In three patients, pure tone averages (taken at 0.5, 1, 2 and 4 kHz) showed an elevation in thresholds of 14, 15 and 16 dB.

TABLE I
LOCATION OF PERILYMPHATIC LEAKS*

Location of leak	Definite leak	Likely leak	Total
Oval window	4	2	6
Round window	4	5	9
Total	8	7	15

Data represent numbers of leaks. There were five cases in which no leak was found. *Identified intra-operatively during tympanotomy, provoked by an anaesthetic Valsalva manoeuvre.

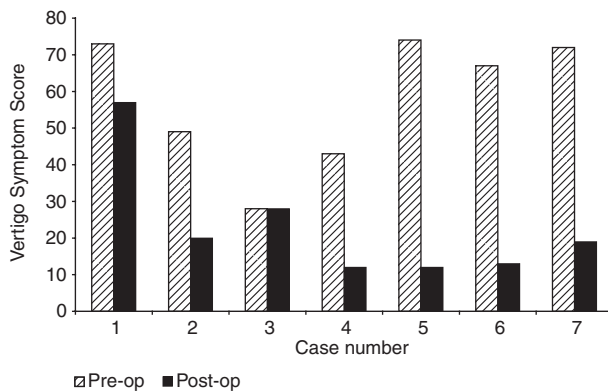


FIG. 1

Graph showing the Vertigo Symptom Scale scores (for the seven patients who completed the scale pre- and post-operatively) before and after exploratory tympanotomy and perilymph fistula leak repair. In all except one case, the median score improved (when pre-operative ('pre-op') scores were compared with post-operative ('post-op') scores; $p < 0.05$).

Illustrative cases

Case one. This was a 24-year-old, previously fit male storeman who was struck on the side of the head by a football. Two weeks later, he noticed muffled hearing, and began to experience attacks of dizziness lasting up to 2 hours and occurring up to 5 times a day. During an attack, movements would result in vomiting. He was unable to work.

Five months after the injury, he was seen by an ENT surgeon. A possible fluid meniscus was visible behind the ear drum. Audiogram findings were normal and a caloric test of vestibular function revealed symmetrical responses. A tympanotomy was performed, and despite the fact that no perilymph fistula was apparent, small fat plugs were placed in the oval window and over the footplate of the stapes under the arch.

The patient was very well for three months, but developed a recurrence of vertigo and vomiting after going back to work and lifting a heavy box. A repeat tympanotomy revealed fluid accumulating under the round window plug when an anaesthetic Valsalva manoeuvre was performed. The fat plug procedure was repeated and the patient reported immediate improvement.

This is a case of definite perilymph fistula, with a clear cause and a good response to surgery. No perilymph fistula was evident at the first operation, and the audiogram and vestibular function test results were normal. The diagnosis was suggested by the history of trauma and the suspicion of a fluid meniscus on otoscopy.

Case two. This was a 40-year-old water meter reader, who reported a sudden episode of vertigo whilst driving 1 month previously, with right-sided hearing loss. The vertigo was continuous and the patient was unable to stand unsupported. The vertigo was worse when the patient was lying down.

At two months, vestibular testing revealed a 52 per cent right canal paresis. At four months, tragal pressure caused vertigo and the diagnosis of perilymph fistula was first considered. Positional audiometry (Fraser test) was positive. The patient underwent exploratory right tympanotomy, where a likely leak was seen on the footplate when an anaesthetic Valsalva manoeuvre was performed. Fat plugs were placed in the round and oval windows. The patient reported post-operative improvement.

Six years later, the patient reported that some eight months earlier he had experienced sudden hearing loss on the (non-operated) left side and dizziness when lying on the left. Fraser testing revealed a positional change in hearing thresholds. He underwent a left tympanotomy; a likely leak was seen at the footplate, and fat was placed in the round and oval windows, with resolution of symptoms.

Unfortunately, the patient re-presented with continuous vertigo after another 5 years, and the fistula test result was positive on the right side (which had been operated on 11 years previously). He underwent a revision right tympanotomy, during which a likely leak was seen at the round window niche. This area alone was plugged with fat and the vertigo again resolved.

This is a case of bilateral sequential perilymph fistula. It is possible that the patient's profession of water meter reader may have been contributory, as he spends a significant amount of time with his head lowered below the horizontal level in a manhole. For each episode, his fistula test result was positive, suggesting the diagnosis.

Case three. This was a patient who worked as a commercial pilot and also ran an agricultural smallholding. He presented with a bubbling sensation in his left ear, which occurred daily on and off, and had experienced two episodes of sudden collapse, one early in the morning when lifting a sack of grain and one in the evening when lifting a fence post. He reported light-headedness prior to falling, but no rotation. For some days after the falls, he experienced a sensation of 'being on a ship'.

The findings of an audiogram at this time were normal. Unterberger testing showed peculiar stepping back and to the side, and then rotation. Caloric testing showed a 24 per cent left canal paresis. An MRI of the brain was normal.

The patient was referred for a programme of vestibular rehabilitation exercises, but he made no progress. He described persistent motion-provoked dizziness, and had been unable to return to work as a pilot.

At one year after presentation, an audiogram showed a left-sided hearing loss of 40 dB. Repeat MRI was normal. However, over the next three months, the patient's hearing deteriorated to 90 dB, and a decision was made to explore the ear for a suspected perilymph leak. At surgery, fluid was seen to accumulate in the round window niche when an anaesthetic Valsalva

manoeuvre was performed. This can be seen in supplementary movie 1 at <http://dx.doi.org/10.1017/S002221511600030X>. A small fat plug was placed over the round window. On follow up, his vertigo improved, with Vertigo Symptom Scale scores decreasing from 72 (out of 175) to 19.

In this case, there was a definite leak intra-operatively and a pressure-increasing event which precipitated symptom onset.

Discussion

In this study of patients with suspected perilymph fistulas who underwent tympanotomy and fistula repair, the majority (14 patients, 18 out of 19 operations) reported an improvement in their symptoms. Six of the seven patients who completed the Vertigo Symptom Scale reported symptom improvement, with a statistically significant reduction in scores, from a median of 67 (out of 175) pre-operatively to 19 post-operatively. No patient reported a deterioration in symptoms.

Perilymph fistula is an enigmatic condition with a rather heterogeneous presentation. An excellent review of the subject was written by Hornibrook from New Zealand in 2012.⁷ There is long-running controversy regarding the condition. Indeed, influential otologists have previously denied the existence of spontaneous perilymph fistula on the basis that they have never observed a case.⁸ It has been suggested that these patients may in fact have Ménière's disease or, since 1998, superior semicircular canal dehiscence.⁹

The intra-operative presence of a visible leak has been the mainstay of diagnosis; however, this has been postulated to be due to local anaesthetic accumulation or cerebrospinal fluid, rather than perilymph. Some groups have tested the fluid for β_2 -transferrin, with positive results in 5–66 per cent of cases.^{10,11} β_2 -transferrin is not specific to perilymph as it is also found in cerebrospinal fluid. Cochlin-tomoprotein, however, is specific to perilymph, and has been found in 'some cases' of perilymph fistula.¹² These tests are likely to have low sensitivity as the volume of the perilymph is only 75 μ l and the leak may be intermittent.¹³

The most useful audiology test is the Fraser positional test. This is simple to perform, and if it is positive the diagnosis is more secure. Caloric tests of vestibular function may be normal or abnormal, and do not seem to be especially useful. Other vestibular tests such as vestibular ocular reflexes and positional testing are not diagnostic of the condition.

Endoscopic assessment of a leak prior to tympanotomy has been performed to exclude local anaesthetic and open surgery as confounding factors. This has correlated poorly with tympanotomy findings (i.e. in the absence of a leak observed endoscopically, a 'leak' was found at tympanotomy).¹⁴

It is generally accepted that perilymph fistulas can occur following surgery and head injury.^{15–17} The

first paper describing 'spontaneous' perilymph fistula was written by Stroud and Calceterra in 1970.¹⁸ They described minor pressure-increasing events preceding the symptoms (e.g. laughing, singing in a choir, leaning over when washing a car and boxing). There are case series that support the existence of perilymph fistula, but none of those reports used a validated questionnaire to assess patient-reported outcome.

We acknowledge some pitfalls in the methodology; for instance, this was a retrospective study and patients can be unreliable when recalling earlier events. Being a case series, it lacks a control group and therefore any placebo effect cannot be accounted for. As discussed earlier, the diagnosis of perilymph fistula is fraught with difficulties, and it is conceivable that some patients earlier in this series actually had superior semicircular canal dehiscence as the time scale of this study covers the period when this condition was first reported. Intriguingly, surgery performed to occlude the round window may be rather effective in treating superior semicircular canal dehiscence. In this series, some patients only had short follow-up periods because appointments were not attended; however, most patients had a follow-up period of more than six months.

Our findings demonstrate the variable nature of perilymph fistula presentation. The clinical presentation was similar to that in other studies, with the majority of perilymph fistulas being preceded or provoked by a pressure-increasing event.¹⁸ Five out of 14 patients (36 per cent) did not recall any preceding pressure-increasing event. This is similar to other series (20–35 per cent).^{1,19–21} It has been suggested that those who do not volunteer a pressure-increasing event may not have been aware of it, as it was a minor event.²¹

Definite and likely intra-operative leaks were found in 15 out of 20 operations (75 per cent), with no leak being found during 5 operations (25 per cent). A summary of 18 studies published from 1968 to 1990 revealed a positive identification of a leak in an average of 71 per cent of cases (range, 24–100 per cent).² It is difficult to compare many of these studies as there is a variety of different thresholds for defining a leak. The inclusion criteria are also varied, with many of the studies including post-surgical perilymph fistulas. Nevertheless, our data are similar to the average leak rate. Direct observation of a tiny fluid leak is rare at surgery, but an anaesthetic Valsalva manoeuvre has in some cases resulted in the obvious accumulation of fluid in the round window or around the stapes footplate (see supplementary movie 1). We feel that this finding may help refute the argument that supposed leaks may be a result of local anaesthetic, as this would not suddenly appear on an anaesthetic Valsalva manoeuvre.

In our cohort, 9 out of 15 of the leaks (60 per cent) occurred at the round window and 6 (40 per cent) occurred at the oval window and/or stapes footplate. The case series summary mentioned above showed

an average of 58 per cent of leaks occurring at the oval window, 24 per cent at the round window and 18 per cent at both.² This difference may reflect the fact that many of these studies included post-stapedectomy patients, which will inevitably increase the proportion of leaks occurring at the oval window.

In our series, the round window alone was repaired in 10 cases, the oval window alone in 1 case, and both the oval and round windows in 9 cases. A study in which a questionnaire was completed by ENT surgeons in the USA revealed that 78 per cent of respondents repaired both windows, regardless of findings.²²

Eighteen out of 19 patients (95 per cent) reported vertigo improvement following tympanotomy and perilymph fistula repair. One patient did not notice any improvement, but she did not report deterioration either. The literature mostly reports slightly lower rates of improvement, at 71–100 per cent, with an average – based on six papers – of 77 per cent.^{1,2,19,21,23,24}

The only significant complication in our study was symptom recurrence. This occurred in 4 out of 14 patients (29 per cent), with a single recurrence in 3 patients and 1 patient requiring 3 procedures in total. In one of these patients, a leak was positively identified at the oval window during the original procedure. This area alone was plugged with fat, but the symptoms recurred after seven years. Again, at tympanotomy an oval window leak was observed, but this time the oval and round windows were plugged with fat. On follow up, the patient reported great improvement of symptoms (to ‘100 per cent’). On both occasions, the Fraser test result was positive. In another patient (case 2, described above), the recurrence occurred at 11 years and may have been due to the patient’s continued ‘head down’ position (associated with his occupation as a water meter reader). Another patient developed symptoms after an episode of barotrauma (a football hit his ear). At the first tympanotomy, no leak was observed, and both the round and oval windows were plugged. The recurrence occurred at four months, immediately after lifting some heavy boxes. At the revision tympanotomy, a leak was observed under the round window fat plug, which was removed and replaced. This resulted in symptom resolution. The remaining patient who suffered recurrence had undergone previous beneficial surgery elsewhere. At tympanotomy, a likely leak was identified at the round window. The symptoms recurred at one year. No leak was seen at surgery, but the round window plug was replaced. Initially, there was a good response, but there was a further recurrence after another two years. This time, a leak was observed at the round window and the fat plug replaced. The vertigo was much improved after each operation. Other studies have reported recurrence and re-exploration rates of 17, 21 and 8–47 per cent.^{2,19,25}

Our study showed a statistically significant improvement in Vertigo Symptom Scale scores when compared

before and after surgery. This questionnaire has been used previously in our hospitals to assess the efficacy of other interventions for vertigo.^{26,27}

Despite its limitations, this study provides evidence to suggest that correctly selected patients with symptoms suggestive of perilymph fistula can benefit from tympanotomy and perilymph leak repair. Many of the patients in this study had experienced symptoms for several years and were very appreciative of the benefit they received, although the symptoms recurred in some cases. None of our patients had any complications. Therefore, even if the diagnosis is uncertain, tympanotomy and plugging of the round and oval windows may still be justifiable. This simple operation is easily within the competence of a general otologist and is quite unlikely to make the situation worse.

- **Perilymph fistula often presents after a pressure-increasing event, with vertiginous symptoms, with or without aural symptoms**
- **There is no ‘gold standard’ diagnostic test for a perilymph fistula**
- **Clinical diagnosis is based on patient history, ideally with a precipitating event, and positive fistula or Fraser test results**
- **Treatment involves tympanotomy and leak repair; however, intra-operative identification of a leak is controversial**
- **This paper shows evidence of a leak captured intra-operatively on video**
- **Vertigo improved in most patients, as assessed using a validated questionnaire**

Until a gold standard test is identified, the mainstay of diagnosis is going to be based on clinical suspicion. The otologist should at least consider perilymph fistula as a differential diagnosis. To help clarify perilymph fistula management, it would be useful to perform a prospective cohort study, ideally comparing conservative versus surgical management. The surgical group could be subdivided into those with a positive leak who undergo repairs and those who do not undergo placement of a fat plug. If local anaesthetic is used, fluorescein can be used to mark it, to distinguish it from perilymph. Absolute exclusion criteria would include stapes surgery and superior semicircular canal dehiscence. The Vertigo Symptom Scale could be used to compare these three groups. This would help to answer some of the questions regarding this enigmatic condition.

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Appendix 1. Supplementary video material

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