Electrocochleography with Postural Changes in Perilymphatic Fistula and Meniere’s Disease: Case Reports

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Abstract

Perilymphatic fistula can be difficult to diagnose differentially prior to exploratory surgery. In this study, we investigated a new test technique in four case studies of subjects with perilymphatic fistula and compared our findings to results obtained in 20 normally hearing subjects with no history of vertigo and 10 subjects with Meniere’s disease. Recordings from an eardrum electrode were obtained with the subject in an upright position, after the subject had been lying in a horizontal position for 30 minutes with the test ear up and after 15 minutes with the test ear down. Stimuli consisted of high level clicks and tonebursts. Neither the summating potential (SP) and action potential (AP) amplitudes nor the SP/AP amplitude ratio were significantly affected by postural change in either the normal or Meniere’s disease group. The perilymphatic fistula subjects, in general, showed greater changes in the SP/AP, particularly for the 6000 Hz tonebursts, than did the other two groups. More data will be needed to determine if these findings are consistent in a large population of perilymphatic fistula patients.

Key Words: Action potential (AP), electrocochleography, Meniere’s disease, perilymphatic fistula, postural changes, summating potential (SP)

Electrocochleography provides a measure of the bioelectric potentials generated in the cochlea and eighth nerve. Although recordings of cochlear potentials have been reported as early as 1930 (Wever and Bray, 1930), the clinical application of electrocochleography is still under investigation. Several investigators have suggested that electrocochleography may allow evaluation of the physiologic status of the inner ear (Thornton, 1976; Coats, 1981; Glattke, 1983).

In recent years, several studies have addressed the effect of induced perilymphatic fistula on electrocochleographic recordings in animals. Most studies reported that the summating potential (SP) and action potential (AP) amplitude ratio (SP/AP) increases after induction of acute or chronic perilymphatic fistula (Arenberg et al, 1988; Campbell et al, 1992b; Campbell and Parnes, 1992; Campbell and Savage, 1992; Campbell and Abbas, submitted for publication). Because an enlarged SP/AP may also occur in Meniere’s disease, however, (Coats, 1981; Ferraro et al, 1983, 1985) an enlarged SP/AP may not distinguish between the two disorders.

Altering body position may change auditory threshold in the presence of perilymphatic fistula in a subset of patients (Fraser and Flood, 1982; Flood et al, 1985). Electrocochleography may be even more sensitive to these changes. Because Meniere’s disease involves a closed fluid inner ear system and perilymphatic fistula does not, changes in body position could encourage or discourage leaking in the latter but not the former case. In guinea pigs, experimental induction of perilymphatic fistula not only increased the SP/AP but markedly increased the SP/AP variability over time, with and without changes in body position (Campbell and Abbas,
Theoretically, in the presence of perilymphatic fistula, perilymph could refill with the ear up. Leakage could be exacerbated with the ear down. In Meniere’s disease and in normal subjects, because the fluid system is sealed, body position should have little if any effect on the SP/AP.

METHOD

Subjects

Because we were primarily interested in determining if postural electrocochleography showed different results in the presence of perilymphatic fistula than in normal subjects or patients with Meniere’s disease, we used two groups for comparison to our perilymphatic fistula case studies. The first subject group, the control group, comprised 20 adults with normal hearing (<15 dB HL from 250 Hz through 8000 Hz in both ears) and no history of vertigo. The second subject group comprised 10 subjects with Meniere’s disease (Alford et al, 1972; Harker and McCabe, 1980), and symptoms on the date of assessment were as described in a previous article (Campbell et al, 1992a). All subjects, except one Meniere’s disease subject, were sedated with 1 g chloral hydrate to reduce myogenic activity. On the date of assessment, an audiogram was obtained for each subject prior to testing.

Measurement Techniques and Equipment

A Nicolet Pathfinder II was used to present the stimuli and record the averaged results. Stimuli consisted of 6000 Hz tonebursts and 100 μsec clicks transduced through an Etymotic insert earphone. The tonebursts had a 3-cycle rise/fall time (.5 msec) with a 4-msec plateau. A Blackman gating function was used to generate the toneburst stimuli. The selection of stimuli was based on earlier animal studies (Campbell and Savage, 1992; Campbell et al, 1992b; Campbell and Parnes, 1992).

Stimuli were presented at rates of 11.7/sec at 120 dB SPL for tonebursts and 125 dB peSPL for clicks. The electrical recording was digitized over a 5.3-msec epoch following stimulus onset. A 2.88-msec prestimulus baseline was also obtained to serve as reference in amplitude measures. Responses to 2000 stimuli (1000 rarefaction and 1000 condensation) constituted each average. A replication of each average was obtained. Rarefaction and condensation stimuli were recorded separately, then added, to reduce the cochlear microphonic and stimulus artifact.

Locally constructed ear drum electrode was employed. This electrode consisted of fine silver wire encased in thin (1 mm) silastic tubing. The silver wire was hooked through a gelled foam tip, approximately 3 mm wide. The hook was then retracted into the tubing, leaving only the foam tip exposed. This electrode is similar to ones reported by Stypulkowski and Staller (1987) and Ferraro and Ferguson (1989). The electrode was placed with the patient in a supine position with the test ear up. The electrode was gently advanced along the inferior portion of the external auditory meatus until contact with the tympanic membrane was achieved. Care was taken to avoid contact between the gelled foam tip and the ear canal. Once contact had been obtained, the crushed foam tip of the insert earphone was advanced along the superior portion of the ear canal without touching the electrode. The earphone tip was then held in place for 60 seconds, until fully expanded. Once it was fully expanded, it held the electrode in place and delivered the acoustic stimulus, regardless of the patient’s position. When the patient was placed in the ear-down position, a foam doughnut cushion was placed around the ear to support the head. A silver earclip electrode (Nicolet) was placed on the contralateral earlobe and served as the reference. A silver electroencephalic disk electrode was placed on the forehead and served as ground.

Responses were filtered from 5 Hz to 3000 Hz, employing analogue filters and amplified (x300,000). Responses were then passed through zero phase shift digital filters with a 5 Hz to 3000 Hz bandpass. It was anticipated that employing an AC amplifier and these filter characteristics could result in some distortion of the SP; however, analysis of the SP and AP recorded through similar filter settings had been widely reported and appeared to be sufficient for clinical measurement of the SP/AP amplitude relationship (Ferraro et al, 1983; Gibson et al, 1983). All testing was performed in a double-walled sound suite (Industrial Acoustics Company).

An audiogram was obtained for each subject prior to testing on the date of assessment. Subjects were tested in an upright sitting position (upright position). Then each subject was placed in a horizontal position with the test ear up for 30 minutes and then retested in that position (ear-up position). For the normal control group, 10 subjects were placed with the left ear up and 10 with the right ear up. The subjects were then placed in the horizontal position with...
the test ear down for 15 minutes and then retested in that position (ear-down position).

**Analysis of Waveforms**

For both stimuli, amplitude of the SP was measured from the prestimulus baseline to the peak of the negative deflection or the shoulder preceding the AP (Coats, 1981; Ferraro et al, 1983). In humans, the SP is measured before the AP, rather than during the plateau portion of the stimulus for the 6000 Hz stimulus, to avoid contamination of the response by auditory brainstem response (ABR) activity. The contralateral reference electrode did allow waves III and V to be present in the recordings. If the kneepoint, or change in slope, in the recorded voltage function was clearly defined, that point in the waveform was used to measure the amplitude of the SP. In cases where the kneepoint was not as clear, the derivative (a measure of slope), of the waveform was obtained. The local maximum of the derivative function during the epoch preceding the AP was then used to indicate the measurement point for SP amplitude. The AP amplitude was measured from the prestimulus baseline to the maximal peak of the Nt.

**RESULTS**

Three dependent variables were determined to be of interest in this study: SP amplitude, AP amplitude, and the SP/AP amplitude ratio. The proposed primary variable of interest was the SP/AP amplitude ratio. Three independent variables were considered in the analysis: subject group, body position, and stimulus. Data were analyzed using a multivariate repeated measures design. Because only four subjects with perilymphatic fistula were included in the study, the data from these subjects were not included in the statistical analysis. The results for each subject with perilymphatic fistula are discussed as individual case studies at the end of this section. The results of the statistical analysis represent only the findings for the normal control and the Meniere’s disease subject groups. A .05 significance level was used throughout the human and animal studies. The dominant SP was invariably of negative polarity for humans and is plotted accordingly.

In Figures 1 and 2, the average SP/AP amplitude ratio is plotted for the click and for the 6000 Hz stimulus, respectively. In order to elucidate the changes within subjects, in Figures 3 and 4 the average amount of change in the SP/AP amplitude ratio is plotted for the click and for the 6000 Hz stimulus, respectively. In Figures 3 and 4, the value of the SP/AP amplitude ratio in the second position listed is subtracted from the value obtained for the first position listed (i.e., for the upright to ear-up change: change = SP/AP upright - SP/AP ear up). These changes were computed for each subject individually. The means and standard deviations of these changes were then computed.

**Normal Control and Meniere’s Disease Groups**

Body position did not significantly affect the SP/AP amplitude ratio, SP amplitude, or AP amplitude, nor did a significant difference between the two groups exist for any of the three dependent variables. The latter finding has been addressed in detail in a separate publication (Campbell et al, 1992a).
For the normal control group, the SP was larger for the 6000 Hz stimulus than for the click stimulus. AP amplitude was larger for the click stimulus than for the 6000 Hz stimulus in the Meniere’s group but not the normal control group. The SP/AP amplitude ratio was significantly larger for the 6000 Hz stimulus than the click stimulus for both groups.

Perilymphatic Fistula Subjects

Case 1

A 17-year-old woman (subject RB) had an oval window fistula in her left ear, secondary to head trauma, repaired in January of 1988. In May, she returned, complaining of recent vertigo but with essentially normal hearing, except for decreased sensitivity at 250 Hz. Her audiogram is presented in Figure 5. An exploratory tympanotomy was performed on the left ear. Initially, a welling-up of fluid was noted in the anterior portion of the oval window. This fluid was suctioned repeatedly and each time reaccumulated. No fistula was observed at the round window. Both windows were patched. The vertigo was eliminated after surgery and had not recurred for 2 months after surgery.

Electrocochleography was performed prior to surgery. Results for the click stimulus (Fig. 1, subject RB) showed a gradual decrement in the SP/AP amplitude ratio for both horizontal positions. The amount of change was similar, however, to that observed among the normal and Meniere’s disease subjects showing the greatest changes (Fig. 3, subject RB). For the 6000 Hz stimulus (Fig. 2, subject RB), the SP/AP amplitude ratio showed greater changes for this subject than for any subject in the other two groups (Fig. 4, subject RB). The ratio diminished after the subject had lain for 30 minutes with the test ear up and then doubled after the subject had lain for 15 minutes with the test ear down. For this subject, both the SP and AP amplitudes diminished with each successive position for both stimulus conditions.

An interesting finding for this subject was the presence of a prominent, replicable, positive SP component (+SP) preceding the negative SP component.
(-SP) for both stimuli. A +SP component of this magnitude was not noted in any subject in the normal control or Ménière's disease groups. Only the negative component is represented in the graphics.

Case 2

A 33-year-old male (subject SK) with a previous history of head trauma with subsequent migraine headaches and panic attacks developed a sudden shift in high-frequency hearing loss in the left ear (see audiogram, Fig. 6), vertigo, and perception of aural fullness in the left ear after sneezing. He was admitted to the hospital and kept at bedrest. Within 2 weeks, he experienced some improvement in hearing and was otherwise asymptomatic. Surgical exploration was never performed on this subject.

For the click stimulus, in the upright position, no SP was observed, although the AP was clear and replicable. The SP was clear and replicable in each of the subsequent horizontal positions (Figs. 1 and 3, subject SK). This pattern of response did not occur in any of the normal or Ménière's disease subjects. For the 6000 Hz stimulus, the SP/AP amplitude ratio increased slightly for the ear-up body position and more markedly for the ear-down horizontal position (Figs. 2 and 4, subject SK). An increase of 0.07 in the SP/AP amplitude ratio for 6000 Hz from ear-up to ear-down positions was noted for this and the previous fistula subject, but in only 1 of 20 normal subjects and 2 of 10 Ménière's disease subjects. For the click stimulus, SP and AP amplitudes were relatively stable over position. For the 6000 Hz stimulus, both the SP and the AP amplitudes increased with each successive position.

We wish to emphasize that this patient was never surgically explored. Therefore, a fistula was never objectively confirmed but was the presumed diagnosis. This case is included only as an interesting case study.

Case 3

A 54-year-old man (subject EM) with a previous fistula repair of the oval window in the right ear in 1985, developed vertigo and diminished hearing in the right ear. His audiogram is shown in Figure 7. The operation report described rapid flow of perilymph near the anterior crus of the stapes. With repeated suctioning, the perilymph quickly reaccumulated. A stapedectomy with patching was performed.

Prior to surgery, responses to the click stimulus (Figs. 1 and 3, subject EM) showed only slight change in the SP/AP amplitude ratio from the ear-up to the ear-down position, but showed a 0.044 decrement in the ratio from the upright to ear-up position. The overall SP and AP amplitudes for the click stimulus were similar across position. Clear and replicable 6000 Hz data were
not obtained for this subject, presumably due to the degree of high-frequency hearing loss.

**Case 4**

A 56-year-old woman (subject NM) was admitted to the hospital as a result of head trauma. The neurologic examination was normal, except for disorientation and confusion, which resolved by the following day. She also had persistent headaches. The computed tomography (CT) scan revealed a small subarachnoid hemorrhage and a suspected small left temporal contusion.

After improvement, she was subsequently released. Two months later, she returned, complaining of daily vertigo, particularly in response to head turn. She reported aural fullness, but no tinnitus or change in hearing. Audiologic assessment, however, revealed a bilateral high-frequency sensorineural hearing loss with good word recognition (Fig. 8). The patient was retested audiologically 3 weeks later, at which time hearing in the left ear had improved by 10 dB at 500 Hz and 20 dB at 6 kHz. An exploratory tympanotomy for the left ear was performed. Perilymphatic fluid was clearly present at the oval window. The fluid reaccumulated after suctioning. Questionable fluid was present at the round window. Preoperative electrocochleography with the click stimulus revealed little change in the SP/AP for the upright to ear-up postural change, but greater changes than any other subject in any other group for the ear-up to ear-down and the upright to ear-down postural changes. The primary change in the ear-down condition, however, was decreased in overall amplitude of the response, possibly suggesting a shift in auditory threshold, although auditory threshold was not assessed directly. It is possible that no replicable SP activity was recorded in the ear-down condition because of the decrease in overall amplitude. This decrement, however, did not occur in any other subject in any group.

For the 6000 Hz stimulus condition, the SP/AP amplitude ratio progressively increased from the upright to ear-up to ear-down position. In the ear-up position, a replicable positive SP component preceded the negative SP, as in Case 1 (subject RB). Because the +SP amplitude in this position exceeded −SP amplitude, the −SP was measured from +SP peak to −SP trough. Only a slight +SP was noted for the upright position. In the ear-down position, overall response amplitude had markedly decreased, as for the click stimulus. Because of the small overall amplitude, measurement of SP and AP amplitudes are probably less reliable. But the decrease in amplitude for the ear-down position did not occur in any subject in any other group.

All technical parameters were carefully controlled and rechecked, including stimulus delivery and electrode impedance.

**Summary of Results for Perilymphatic Fistula Subjects**

In general, while the electrocochleograms for the Meniere's disease and normal subjects were stable across positions, the results for the fistula subjects tended to be more variable across positions, particularly for the 6000 Hz stimulus. Perhaps the response to the 6000 Hz stimulus emanates from a more restricted region of the basilar membrane close to the site of lesion. The click stimulus, being broader in frequency, may elicit a response from a broader region, which could render the response to the clicks less sensitive to changes at a given point. While no characteristic change tended to occur for all subjects, the amount of response variability within each perilymphatic fistula subject may in itself be of interest.

**DISCUSSION**

The findings for the Meniere's disease and normal subject groups were consistent with
the original hypothesis of the study: in the presence of a closed fluid system, body position should have minimal, if any, effect on the SP/AP. The lack of significant difference between these two groups for SP/AP, irrespective of postural changes, has been addressed in detail in an earlier paper (Campbell et al, 1992a).

The primary finding for three of four of the perilymphatic fistula subjects was the marked variability in the response with postural changes and the presence of a prominent +SP in two of four cases. A +SP has been previously reported in some cases of Meniere's disease (Eggermont and Odenthal, 1974; Eggermont, 1976; Kanzaki et al, 1982; Gibson et al, 1983), but only with transtympanic recording techniques (Mori et al, 1982). Aberrant findings, such as the pronounced biphase SP with a noninvasive electrode, or SP/AP intrasubject variability over time, may not occur in all perilymphatic fistula subjects, but may be pathognomonic of this disorder when observed.

Marked SP/AP variability over time, with and without postural changes, has been reported in guinea pigs with induced perilymphatic fistula (Campbell and Abbas, submitted for publication). Perhaps fluid leakage from a perilymphatic fistula is variable over time or position, causing variability in the SP/AP. SP amplitude is dependent on intracochlear pressures (Davis et al, 1958; Butler and Honrubia, 1963; Durrant and Gans, 1975). The guinea pig model of perilymphatic fistula, however, is limited, because the cochlear aqueduct is patent in the guinea pig (Lempert et al, 1952; Duvall and Sutherland, 1972; Moscovitch et al, 1973) but may not be in humans (Lempert et al, 1952; Matteo, 1986).

An enlarged SP/AP in the presence of a patent perilymphatic fistula has been reported in guinea pigs (Campbell and Savage, 1992) and in humans (Arenberg et al, 1988). In the perilymphatic fistula subjects in this study, the SP/AP in the upright position was not markedly different from the subjects in the other two groups.

Although the SP/AP observed in perilymphatic fistulas may not be consistently different than findings in other subject groups for a single recording, the intrasubject variability in the response over time or position may possibly distinguish the perilymphatic fistula patient from normal subjects or Meniere's disease patients. This type of result, however, cannot be considered diagnostic of perilymphatic fistula at this time. Whether or not a clinically useful tool can be developed cannot be determined from animal studies or a few human subjects. Large-scale testing in a number of human subjects with perilymphatic fistulas will be required.

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Postural Electrocochleography and Perilymphatic Fistula/Campbell and Abbas


