Clinical Report

Sub-Galeal Pneumocele 20 Years after Cochlear Implantation

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This case report documents a rare case of a subgaleal pneumocele occurring more than 20 years after cochlear implantation. On presentation, the presence of air under the scalp followed vigorous nose blowing was observed. The patient was treated conservatively with a pressure dressing, which resulted in complete resolution of the surgical emphysema. Only a handful of such cases have been reported in the literature. Most of these cases occur early after cochlear implantation. Our case is even more unique as it occurred 24 years following implantation and presented with a painless swelling associated with deterioration in cochlear implant performance. A review of the current literature is included in the discussion.

KEYWORDS: Cochlear implantation, postoperative complications, subcutaneous emphysema

INTRODUCTION
In competent hands, cochlear implantation is a low-risk procedure. The most common complications are infection (wound infection and meningitis), facial nerve trauma, electrode migration/misplacement, vertigo, and tinnitus [1, 2]. Pneumocele following cochlear implantation is very rare [3-10]. Our case is even more unusual as the pneumocele developed more than two decades following cochlear implantation.

CASE PRESENTATION
An 80-year-old lady presented with a large swelling over her cochlear implant site. She had undergone right-sided cochlear implantation 24 years prior at another institution. Her implant had worked well until the development of the swelling when implant performance, and consequently her hearing, deteriorated. The swelling varied in size from day to day, and several times in a single day. It had been present for 6 weeks prior to presentation at the Otology & Neurotology Clinic at the University of Louisville. The patient denied dizziness, pain, or headache. She did report vision problems as well as past medical history of hypertension and hypercholesterolemia.

On examination, a fluctuant, crepitant, diffuse, non-inflamed, non-tender 3-cm swelling was noted. It was centered directly over the receiver–stimulator package. Sustained digital pressure over the swelling resulted in sudden decompression and emptying along with resolution of the swelling. The swelling refilled gradually after a variable period. Increase in swelling was associated with nose blowing. There was no evidence of cerebrospinal fluid rhinorrhea or otorrhea. The tympanic membrane appeared normal and facial strength and symmetry was also normal. A high resolution computerized tomography scan (HRCT) of the temporal bone was requested.

HRCT demonstrated bilateral advanced otosclerosis. An electrode was visualized within the right cochlea, and air was seen within the mastoidectomy cavity. A subgaleal air-containing pocket was also visualized around the receiver–stimulator package. A column of air around the electrode connected this subgaleal pocket with the air in the mastoid cavity (Figure 1, 2). There was no evidence of fluid collection or other bony dehiscence in the skull. Options including ventilation tube placement were presented to the patient and a pressure dressing was applied for 4 weeks. Antibiotics were not prescribed as there was no evidence of infection or inflammation. At follow-up examination a month later the swelling appeared to have resolved. She was advised against forceful nose blowing. The patient has been trouble-free for over a year.

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DISCUSSION

Pneumocele following cochlear implantation is a rare event \[3-10\]. It generally occurs in the early postoperative period. This case is extremely unusual as the pneumocele developed some two decades following cochlear implantation. The most likely mechanism in our case appears to have been the flow of air up the Eustachian tube due to a pressure difference generated between the nasopharynx and middle ear cleft by forceful nose blowing. The air column can be seen to track through the middle ear and mastoidectomy cavity around the electrode path and to collect around the receiver–stimulator package (Figure 1, 2).

Figure 1. Axial computed tomography scan reveals a pneumocele is visualized around the receiver–stimulator package.

Figure 2. Coronal computed tomography scan demonstrates air tracking along the electrode from the mastoid to the pneumocele.

In almost every case report, a Valsalva maneuver or a Toynbee maneuver is implicated in the causation of subcutaneous emphysema after cochlear implantation \[3-6, 8-10\]. Most studies do not report serious complications as a result of pneumocele but in a series of four cases, one case developed an infection, which was treated successfully with antibiotics \[6\]. Of these four cases, two others resulted in diminished performance of the cochlear implant. Only one case occurred 7 years following implantation, while the rest presented early. Typically, this complication occurs between 1 and 10 weeks following implantation. Our case is unique as the patient presented some two decades following implantation without evidence of infection. Unlike Khatwa et al. \[7\] and Backous et al. \[8\] antibiotics were not administered in our patient as she demonstrated no evidence of clinical infection or inflammation. However, we agree that it may be reasonable to administer antibiotics as the authors recommend \[7, 8\].

Various modalities of treatment have been suggested in the literature. Some physicians chose to treat with antibiotics and pressure equalization tubes \[8\]. Ventilation tubes prevent a pressure build-up as they act as relief valves. Aspiration of the air under strictly aseptic conditions has also been recommended \[3, 6, 9\]. In our case, a pressure dressing for 4 weeks without surgery was extremely effective. Close follow-up examinations were necessary to make certain that an infection did not develop.

All patients with cochlear implants should be counseled on avoiding the performance of forceful or excessive nose blowing particularly in the early postoperative period. However, as is evidenced in our patient, this complication can occur decades after implantation as well. Although rare, it is important to try and prevent this complication. Hagr and Bance \[9\] reported a rare case of an intraparenchymal pneumocephalus in addition to a subcutaneous pneumocele. This was thought to be the result of habitual nose blowing due to an untreated sinus disease. Their patient was treated with antibiotics and ventilation tube placement. They recommended that nasal disease such as nasal polyposis be treated prior to cochlear implantation. Another case of pneumocephalus, evidently as a result of performing Valsalva maneuver up to 30 times a day, was reported by Gillett et al. \[10\]. Their patient was treated conservatively with resultant spontaneous resolution, followed by insertion of a Shah mini grommet for the prevention of repeat occurrences.

CONCLUSION

An extremely rare complication of a pneumocele developing more than two decades following cochlear implantation is discussed. A method to prevent and alleviate the condition and a synopsis of the existing literature is presented.

Ethics Committee Approval: Ethics committee approval was received for this study from the ethics committee of the University of Louisville.

Informed Consent: Verbal informed consent was obtained from the patient who participated in this study.

Peer-review: Externally peer-reviewed.


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REFERENCES