

## SPONTANEOUS OTOGENIC PNEUMOCEPHALUS AND PNEUMATOCOELE

O. Soenen<sup>1</sup>, J.W. Casselman<sup>1</sup>, B. Lerut<sup>2</sup>

**We report the case of a 33-year old woman with an otogenic pneumocephalus associated with pneumatocoele. To our knowledge only 2 other cases have been documented. We discuss the imaging, treatment and causes of otogenic pneumocephalus.**

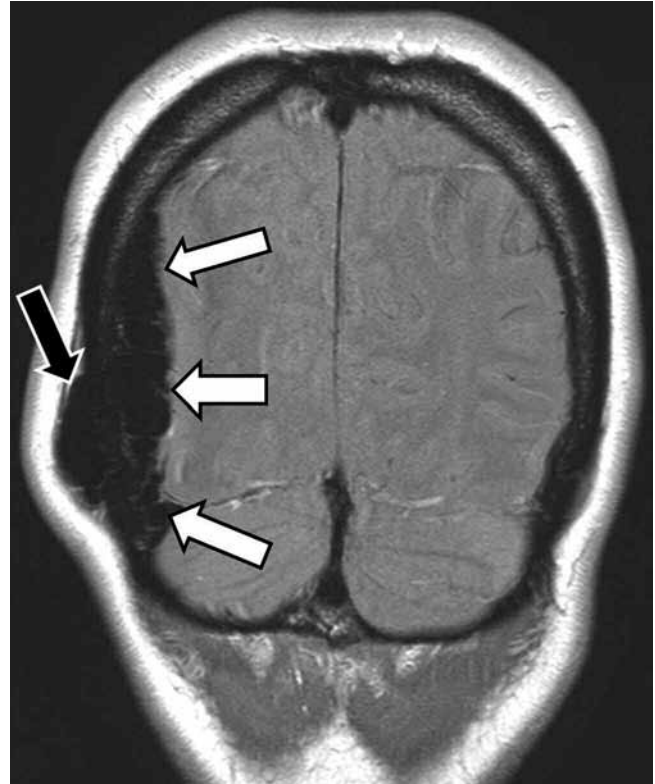
**Key-word:** Ear, diseases.

### Case report

A 33-year old woman, with no relevant medical history, presented with a retroauricular mass on the right side. The surgeon decided to puncture the mass and air was evacuated. One week later she was referred to an ENT surgeon for complaints of headache starting after puncture. Clinically there was polyposis nasi grade III. She had allergies (house dust mite) and had been sneezing for over 10 times a day, whereby she pinched her nose and thus induced a repetitive Valsalva manoeuvre. Lastly she also complained of a balance disorder she had been having for over a year. Subsequently an MR and CT study were performed to find the cause of this air-containing retroauricular mass.

MR study is best suited for soft tissue examination due to its high contrast resolution. In this case MR was performed first to exclude underlying infection or tumor. Due to lack of signal, air and -though less likely-calcified bone were considered possible options (Fig. 1). CT is the method of choice when it comes to the bone or air visualisation. CT indeed visualized an epidural air collection with associated pneumatocoele (Fig. 2). Following radiological studies, the ENT surgeon placed a transtympanic drain inducing air evacuation and a shift of the brain. Because of the risk of recurrence and of otogenic meningitis an obliterative mastoidectomy with cartilage in the additus ad antrum and bone paté filling up the mastoid (variation of Mercke procedure) was performed. During surgery a large bony defect was seen at the level of the tegmen

and the sigmoid sinus. Follow up imaging was performed with a cone beam CT. Cone beam CT is ideal in situations where a young patient is repeatedly exposed to radiation, because it provides a higher resolution at a lower radiation dose. A clas-

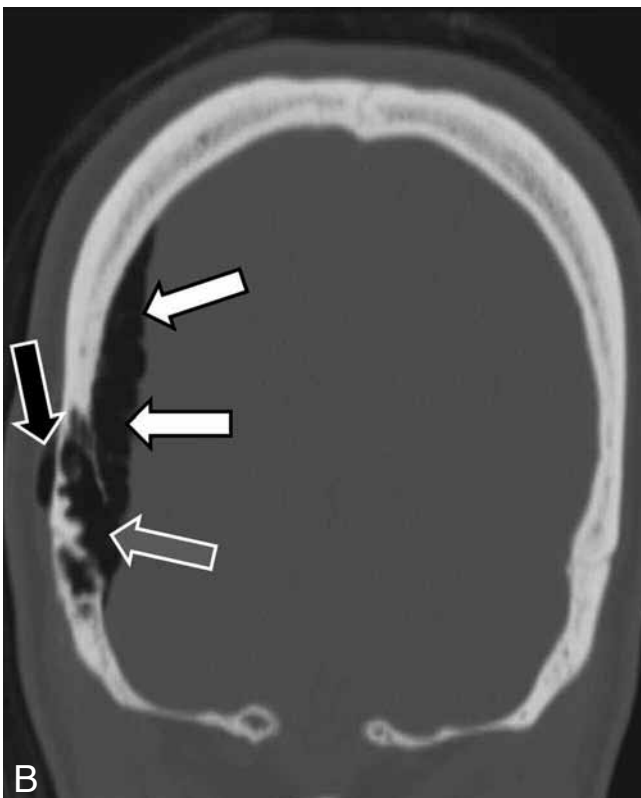
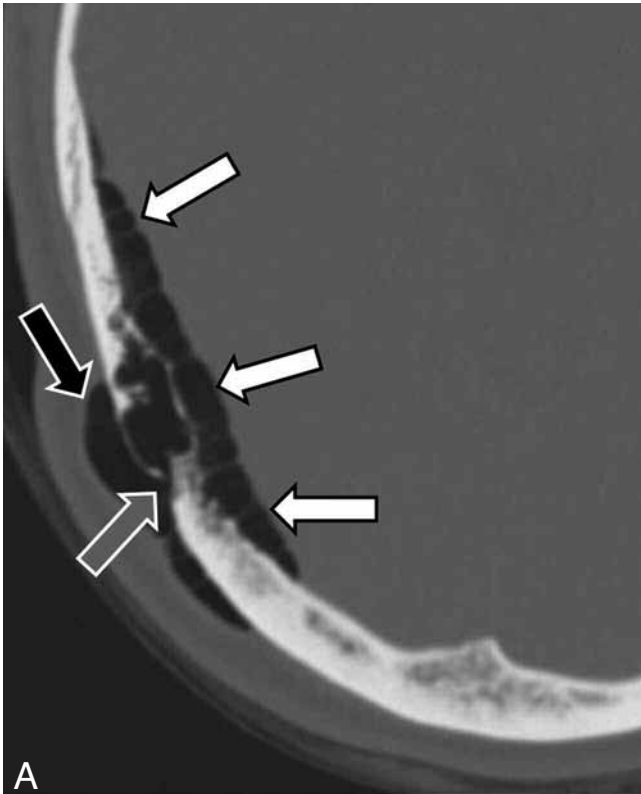


*Fig. 1.* — Coronal T2-weighted image. Initially MR was performed to exclude underlying infection or tumor. Hypointense epidural air collection (pneumocephalus) with mass effect on the brain (white arrows). Hypointense air filled subcutaneous pneumatocoele (black arrow).

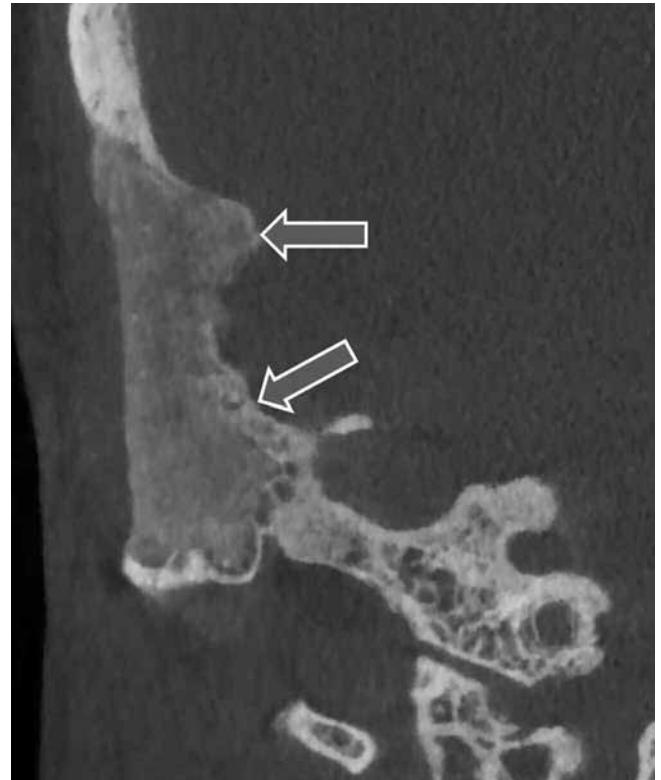
sic CT study with a bigger Field of View (FOV) was needed at the beginning to examine the origin of the air and the amount of air that went intracranially etc. Cone beam CT with its smaller FOV was a better option after surgery because only the local post-operative status of the petrous bone and the bone paté filling had to be evaluated (Fig. 3). To date the patient is relieved from her symptoms, has normal middle ear pressures on a tympanogram and is still followed by her ENT surgeon in the context of her polyposis nasi.

*From:* 1. Department of Radiology and Medical Imaging, 2. Department of ENT, AZ Sint Jan Brugge-Oostende AV, Brugge, Belgium.

*Address for correspondence:* Dr J.W. Casselman, M.D., Ph.D., Department of Radiology and Medical imaging, AZ Sint Jan Brugge-Oostende AV, Ruddershove 10, B-8000 Brugge, Belgium. E-mail: jan.casselmann@azsintjan.be



*Fig. 2.* — Axial (A) and coronal (B) CT image with bone window setting through the upper and most posterior part of the mastoid. The hyperpneumatisation of the bone resulted in a defect of the inner and outer cortex of the bone (grey arrows) and an epidural pneumocephalus (white arrows) and subcutaneous pneumatocoele (black arrows).



*Fig. 3.* — Coronal Cone Beam CT image through the posterior mastoid. The surgeon used bone to obliterate the hyperpneumatized bone and to close the cortical defects (grey arrows).

## Discussion

Otogenic pneumocephalus is defined as the presence of air in the intracranial cavity entering through the temporal bone. Pneumocephalus was first described in 1714 by Lecat. The most common causes of pneumocephalus are trauma to the head, ear infection, tumors of the skull base and iatrogenic (e.g. skull base surgery) and in these cases the air is most frequently found in the subdural and subarachnoid space or in the brain parenchyma or ventricles. However, spontaneous otogenic pneumocephalus and epidural air collections are both very rare. A literature research revealed 18 cases of spontaneous pneumocephalus until today. Of these 18 cases 8 were reported in the last 10 years, probably due to better and easier access to imaging. These patients present with a variety of symptoms including headache, dizziness, aphasia, hemiparesis or hemiplegia, visual symptoms etc, some of which can be attributed to the presence of epidural air. Less frequent symptoms are tinnitus and otalgia (1). The presence of a subcutaneous pneumatocoele was only reported in

two other cases and was the first and only complaint in these patients, which seems logic because the air is released which reduces the intracranial pressure. In this new case the air probably also escaped subcutaneously through a defect in the outer cortex of the upper and posterior part of the mastoid. The pathogenesis of spontaneous otogenic pneumocephalus remains uncertain. The hypothesis is that a defect in the temporal bone and/or a pressure difference between the temporal bone or middle ear and the intracranial space are at the origin. Moreover pressure changes or repetitive Valsalva manoeuvres can

lead to hyperpneumatization of the mastoid. Pneumocephalus can also very rarely be caused by a barotrauma (2), however sometimes no underlying mechanism is detected. Furthermore, predisposing defects in the petrous bone are not uncommon in the general population. In this case, the repetitive Valsalva manoeuvres probably led to the development of a pneumocephalus and pneumatocoele because of a pre-existing strong pneumatisation of the petrous bone with thinned over and underlying cortical bone. Treatment of a spontaneous otogenic pneumocephalus is usually managed surgically with an attempt

to relieve the intracranial pressure and a procedure to close the bone defect.

#### References

1. Kraysenbühl N., Alkadhi H., Junh H.-H., et al.: Spontaneous otogenic intracerebral pneumocephalus: case report and review of the literature. *Eur Arch Otorhinolaryngol*, 2005, 262: 135-138.
2. Tucker A., Miyake H., Tsuji M., et al.: Spontaneous epidural pneumocephalus. *Neurol Med Chir*, 2008, 48: 474-478.
3. Pennings R.J.E., Liauw L., Cremers C.W.R.J.: A spontaneous otogenic extradural pneumocephalus. *Otology Neurotol*, 2009, 30: 864.