CASE STUDY

Pneumolabyrinth: An Atypical Complication in a Cochlear Implant

Neumolaberinto: una complicación atípica en un implante coclear

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A 69 year old patient with a history of extra and intracranial vestibular schwannoma of 3 cm in diameter in the left ear, treated with external hypofractionated radiotherapy (30 Gy/12 Fr). Five years later, the patient presented with sudden hearing loss of unknown origin and as a result developed right-ear cophosis. Radiological study showed no malformations of the said ear nor any other causes to explain the hearing loss.

Due to the presence of cophosis in both ears, we proceeded to insert AB HR90K/HiFocus® cochlear implants in the right ear performed by cochleostomy. There were no notable postoperative complications and the transorbital X-ray showed that the implant electrodes had been correctly positioned.

Two years after the implantation, the patient sought consultation as a result of spontaneous presentation of a sensation of occupation of the middle right ear and swelling in the temporal cranial cavity, which coincided with the area where the receiver-stimulator of the cochlear implant was located.

On examination, a crepitant swelling was observed in the right temporal region with no presence of liquid. The otoscopy showed a dull and completely bulging tympanic membrane, but with no hydro-aerial levels.

A CT scan of the temporal bone was performed which showed an image of pressurized air occupation in middle ear, inner ear and right mastoid structures. The cochlear implant was correctly positioned in the scala tympani (Fig. 1).

On suspicion of a perilymphatic fistula in the cochleoestomy area, a surgical review was performed where distension of the soft parts of the mastoid cavity were observed. These were compatible with mastoid air entrapment (Fig. 2). The cochleostomy was reviewed and no evidence of perilymphatic fistula was observed; however, the interphase of the electrode guide with the cochleostomy was reinforced with the application of muscular tissue and fibrin glue.

No improvement was observed post operatively in X-rays, nor did clinical symptoms improve. The cause was therefore believed to be the existence of a tubal valve mechanism, and the patient was therefore referred for transtympanic drainage as an outpatient procedure.

After drainage a progressive improvement of symptoms was observed and the disappearance of emphysematous inflammation in the temporal bone area.

A control TC scan was performed again, which showed the persistence of a small amount of air around the electrodes located inside the cochlear, the disappearance of air which

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Figure 1  Occupation by air at middle, inner and right mastoid level. Electrode guide positioned in the scala tympani. Swelling of the external mastoid wall.

occupied the middle and inner ear, which at that time was filled with liquid. The air located in the mastoid cavity also disappeared (Fig. 3).

Discussion

Air in the inner ear or pneumolabyrinth is a radiological sign which was first reported by Mafee et al.1

The most frequent cause of pneumolabyrinth is perilymphatic fistula.1 There are several reasons for its appearance, the most frequent of which are fractures of the petrous portion of the temporal bone, labyrinth trauma caused by the displacement of the stapes during otosclerosis surgery, an oval or round window injury after major Valsalva manoeuvre or after the insertion of a cochlear implant in patients affected by inner ear malformations.2-4

The presence of air in the inner ear, or pneumolabyrinth, associated with entrapment of air in the mastoid cavity, is a rare occurrence in patients with cochlear implants. The few cases reported in literature are associated with patients who suffer from some type of malformation in the inner ear, such as, for example, large vestibular aqueduct syndrome,5-7 in cases of patients who have a peritoneal ventricle valve8 or in patients who have had a major Valsalva manoeuvre.

Figure 2  (A) Swelling of the external mastoid wall from pressurized air. (B) Opening of the external wall with observation of a normal cavity with an electrode guide in its interior.

Figure 3  Disappearance of the air in the inner ear structures and mastoid attachment.
There are different ways in which air can enter the cochlear vestibular system, particularly after the instalment of a cochlear implant. The most probable point of entry is through cochleostomy, at the entry level of the electrode guide.

It has been reported that the diameter of the cochleostomy may have an effect on the appearance of a perilymphatic fistula in the postoperative period. Robey et al. studied the effect different cochleostomy diameters (1 mm vs 1.5 mm) had on the appearance of perilymphatic fistula in the middle ear, although after the intracochlear pressure had increased there were no statistically significant outcomes. Despite this, it is important to underline the importance of the cochleostomy sealing following insertion of the cochlear implant, whether this be with muscle or fat and fibrin glue.

Perilymphatic fistula cannot be the cause in our case, since the presence of air was at the correct level in the inner ear, the middle ear, and the mastoid cavity. Furthermore, the radiological image did not improve after surgical review and resealing of the cochleostomy.

The only plausible explanation would be the existence of a valvular mechanism in the Eustachian tube which prevented normal evacuation of air of the middle ear to the nasopharynx and caused hyperpressure in all middle ear structures and from there penetrated the inner ear to the mastoids.

In these cases the problem is simply resolved by transtympanic drainage, as occurred in our case.

The reason behind how a valvular mechanism occurred in the Eustachian tube is a further issue for research.

No cause was discovered in our patient but it is possible that a history of radiotherapy could be related to the appearance of the valvular mechanism at Eustachian tube level.

To conclude, in the presence of a pneumolabyrinth in a patient with a cochlear implant, one not only has to rule out the presence of a perilymphatic fistula which would require surgical review and the sealing of both oval and round windows, but also rule out that the cause could be a tubular dysfunction, especially if in addition to the pneumolabyrinth there is a presence of entrapped mastoid air.

The most beneficial and less aggressive option in these cases would be transtympanic drainage.

Conflict of Interests

The authors have no conflict of interest to declare.

References