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Cochlear implant challenges encountered in tuberculous otitis media

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ABSTRACT

Tuberculous otitis media (TOM) is rare in ENT department, and is frequently misdiagnosed as otitis media. Thus early systemic treatment is very important for TOM. We reported a case report with TOM to highlight development of the disease and difficulties in clinical treatment in late stage of TOM. Implantation of ossified and eroded cochlea poses many unique challenges to both the surgeon and programming team. With thorough preparation and complete knowledge about characters of specific issues, implantation would be performed successfully, and patients with ossified cochlear could benefit from cochlear implantation.

1. Introduction

Tuberculous otitis media (TOM) is a rare form of chronic otitis media and extrapulmonary tuberculosis, which is difficult in diagnosis. It only accounts for 0.04%-0.90% of suppurative otitis media cases in developed countries but incidence has been in a steady rise in areas where tuberculosis is still endemic[1-3].

Mycobacterium tuberculosis (TBC) causes TOM in three ways: (1) haematogenous spread of the infection in patients with other forms of TBC through blood circulation system or lymphatic circulatory system; (2) infection imported through perforated tympanic membrane; (3) aspiration of infected food through the eustachian tube during drinking or nursing in patients with eustachian tube dysfuction[4].

TOM may occur at any age. The classic symptoms include multiple perforations of tympanic membrane, painless otorrhoea, and frequent complications include sensorineural hearing loss, fistulae, exuberant granulations, paralysis of the facial nerve, labyrinthitis, tuberculous osteomyelitis, mastoiditis, and spread of infection to the central nervous system. TOM may have association with pulmonary TBC or not[4]. The diagnosis should be taken into account for patients who have no response to standard antibiotic therapy of fungal external otitis or bacterial otitis media. Therefore, in case of serious complications early diagnosis and treatment are imperative[5].

High-resolution CT of temporal bone is highly sensitive in detecting soft tissue disease and bony erosion. Preservation of mastoid air cells without sclerotic change, mucosal thickening of the bony Ehrlich ascites carcinoma (EAC), or a soft tissue extension to the EAC without erosion of scutum are characteristic CT findings and would be helpful for the early diagnosis of TOM[6]. Besides, CT scans of temporal bone also indicate bone destruction, labyrinth ossification, and surrounding tissue erosion in the late stages of TOM[4].

The standard and effective treatment of TOM is antituberculous medication, and four drugs are usually used daily during the intensive phase: isoniazid, rifampicin, pyrazinamide and ethambutol. Patients should be treated for at least 6 months, and even longer in cases of disseminated tuberculosis and tuberculous meningitis[7].

For severe and profoundly hearing impaired patients, cochlear implantation is the establishment method of auditory rehabilitation. Complications after cochlear

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implant include improper placement of the electrode, skin flap-related complications, device dysfunction as a result of a manufacturing defect, or trauma to the electrode array during insertion[2-4].

Most reports of TOM focus on its early diagnosis and treatment aspects, but rarely discuss the complex complications in TOM, and provide little information on how to handle the difficult situation in cochlear implant. We report a rather complicated case of tuberculous otitis media rarely encountered in cochlear implant surgery.

2. Case report

A 31-year-old Asian woman was admitted to the ENT department with a ten-year history of bilateral painless otorrhoea and progressive hearing losss accompanied by tinnitus and dizzines. Six years ago she suffered from moderate to profound bilateral hearing loss which couldn't be treated by amplification or multiple local or systemic antibiotics. She underwent plastic surgery of the external auditory meatus due to external auditory canal stenosis in other hospital 5 years ago. This patient underwent treatment in our department 3 years ago, and has had a sequence of operations up to now.

Otoscopic examination revealed subtotal tympanic membrane perforation and granulation tissue in the middle-ear cleft, no mucopurulent discharge in the bilateral middle ear and meatus. The lungs were normal which was confirmed by chest X-ray.

Pure tone audiography revealed a profound sensorineural hearing loss in the bilateral ear. A MRI scan of the temporal bones showed soft tissue material filling the mastoid process (Figure 1), and CT provided similar information.

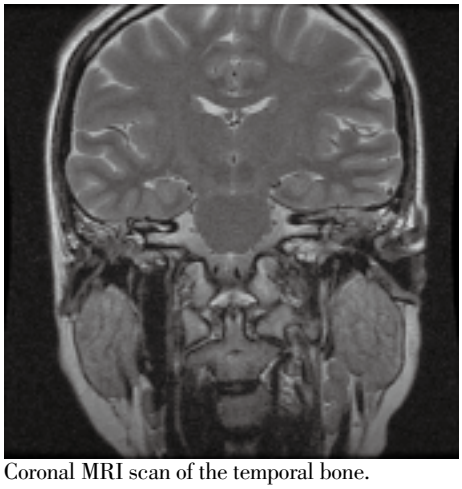

Figure 1. Coronal MRI scan of the temporal bone.

Then the patient underwent left modified radical mastoidectomy. Intraoperatively, the cortex of the mastoid bone was found to be eroded and the antrum filled with granulation tissue. Pale yellow precipitate was found in tympanum. The malleus, incus and stapes were found to be necrotic. Part of superior semicircular canal, posterior semicircular canal and facial nerve canal were eroded, and joint capsule exposed about 0.3 cm $\times$0.5 cm. Pathological changes were completely removed, and defects were covered by temporal fascia. Rapid pathological changes of granulation tissue and pale yellow precipitate confirmed inflammation. Subsequent culture of tissue revealed *Mycobacterium* species, identified as *Mycobacterium*.

After surgery, this patient revealed a left House-Brackmann grade Ⅱ facial nerve palsy. However, facial electromyogram showed that bilateral facial nerve had neurogenic damage, and reductions in the amplitude ratio of the action potential were 84% in the left side and 2% in the right side. The patient was treated with antituberculous medication and started quadruple therapy (*i.e.* rifampicin, pyrazinamide, pyridoxine and ethambutol) for at least 6 months until contralateral surgery.

Three months later, this patient had a progressive right-sided facial paralysis, but failed to seek timely medical treatment. Six months after the first surgery, clinical examination revealed a right-sided House-Brackmann grade Ⅳ facial nerve palsy. Facial electromyogram indicated severe neurogenic damage of bilateral facial nerve, and reduction in the amplitude ratio of the action potential were 60% in the left side and 97% in the right side. CT scan different from a previous radiograph were shown in Figure 2.

Then subtotal resection of temporal bone and facial nerve decompression of right ear was performed under general anaesthesia. During this procedure, plenty of cheese-like substance, edema granulation and mucopurulent discharge were seen in the external auditory canal and mastoid air room, the tympanic membrane was found to be absent, the anterior wall of the external auditory canal and temporomandibular joint were eroded, the mastoid segment and the tympanic segment of facial nerve were surrounded by granulation tissue, and fistula was found in lateral semicircular canal. During the surgery, we selectively resected the lateral semicircular canal, decompressed the mastoid segment and the tympanic segment of facial nerve, and defects were covered by temporal fascia. The pathological changes of granulation tissue and pale yellow precipitate confirmed the diagnosis as *Mycobacterium*.

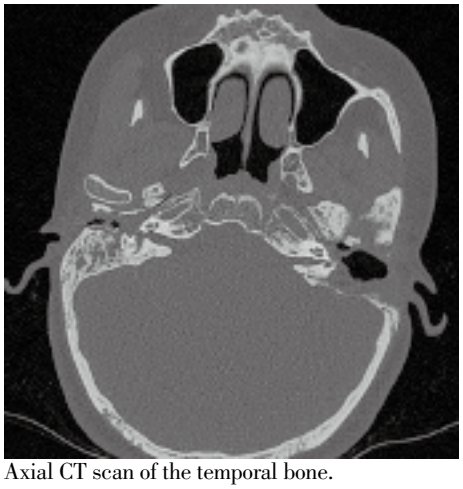

Figure 2. Axial CT scan of the temporal bone.

One year later after the second surgery and systematic antituberculous medication, the patient had a strong desire to go back to the hearing world, so she came back and asked for cochlear implantation. Based on adequate preoperative preparation and detailed radiography (Figure 3), we planed to implant a multi-channel cochlear implant (Combi 40+S, Med-El) on the right side. Unfortunately, anatomical structures such as round window and oval window could

not be clearly identified, even if we drilled the promontory gently during the surgery. Besides, due to some granulation in the tympanic cavity, the surgery had to be stopped.

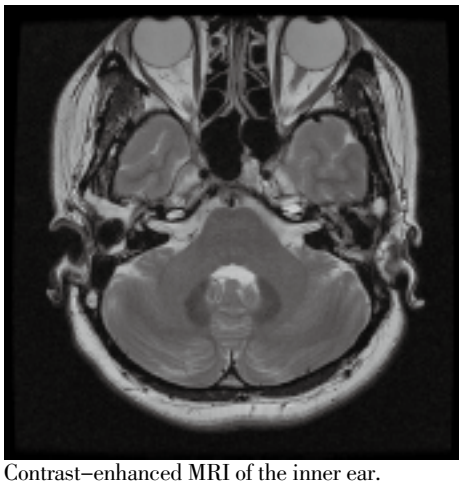

Figure 3. Contrast-enhanced MRI of the inner ear.

Five months later after the third surgery, the cochlear implantation was performed on the same side, this time we considered tympanic ostium of eustachian tube as the anatomical landmark, and drilled along the proximal tip of the basal turn of the cochlea carefully. There was a narrow space for cochlear implant due to cochlear ossification, then electrode array was inserted gently about 1 cm during the process. In the telemetric revision all the electrodes had good impedance. Postoperative CT examination proved that electrode lay in the right position (Figure 4).

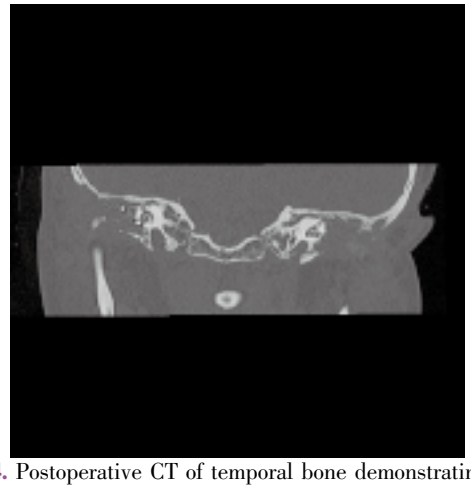

Figure 4. Postoperative CT of temporal bone demonstrating correct positioning of electrode array.

However, one month later after the fourth surgery, when her right implant turned on for the first time, she still couldn´t hear the sound. CT revealed that electrode array position had been changed compare to previous X-ray examination (Figure 5). So we deduced that electrode position might have changed after the implantation, and an optimal choice for the sake of the patient was to adjust the electrode position as soon as possible. Intraoperatively, we created a perimodiolar canal to hold the electrode array and cleared calcifications of soft tissue in the labyrinth (Figure 6a, 6b). All the electrodes had good impedance in the telemetric revision.

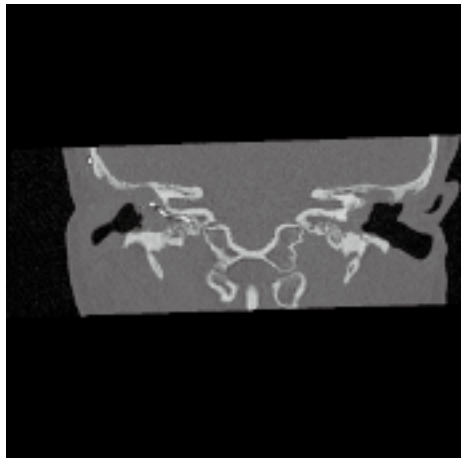

Figure 5. CT scan showing electrode array position changes.

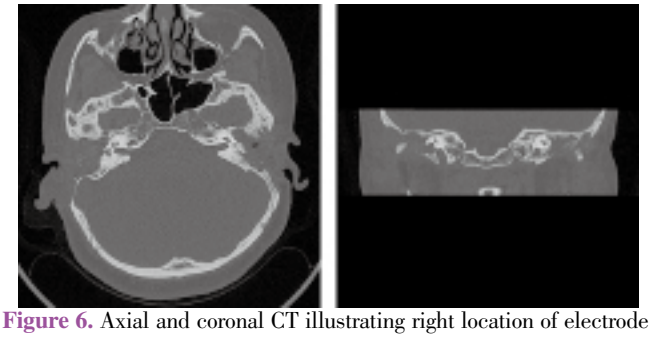

Figure 6. Axial and coronal CT illustrating right location of electrode array.

After the sixth surgery, she received ongoing management by an audiologist including programming the implant parameters and monitoring device performance from electrical threshold and dynamic range data[8]. Follow-up audiological evaluations revealed that sound and speech detection and auditory reception of speech were improved.

The patient´s bilateral otorrhoea and otalgia had settled, and through acupuncture and physiotherapy, her facial palsy had partially recovered (to House-Brackmann grade Ⅲ in the right and grade Ⅱ in the left side) after a year of follow up.

3. Discussion

It is an interesting case, because the lower tuberculous otitis media was observed in the ENT department. It remains a diagnostic challenge for many clinicians, because of its unspecific clinical features and inability to confirm the infection by microbiological and histopathological examinations. A high level of clinical suspicion is needed for early diagnosis and antitubercular therapy should be started as soon as possible to prevent the possible complication[9].

However, unlike previously reported in the literature, this patient had serious complications including profound hearing loss, labyrinth ossification and facial paralysis when she was admitted to our department. The patient´s condition posed a great challenge to clinicians because it was very difficult to control, despite anti-tuberculosis chemotherapy and radical mastoidectomy. On one hand, the physical structures were already destroyed, on the other hand it was very difficult to insert electrode implantation into the appropriate location because of labyrinth ossification[10].

A high resolution computed tomography scan is necessary

to assess the preoperative bony anatomy of the temporal bone. However, its use alone may not provide sufficient details regarding the degree of ossification, as a wide variance of sensitivities for identifying ossified cochlea has been reported[11,12]. Attenuation of intracochlear fluid signal in T2-weighted magnetic resonance imaging has proved to be a more sensitive method of assessing blockage[13]. Similarly, contrast-enhanced T1-weighted images can detect early fibrosis and may aid the early implantation[14].

The greatest surgical difficulty in the cochlear implant is the identification of the area of the round window. Facial nerve, chorda tympani nerve and round window offer special reference to the approach route for cochlear implant surgery[15]. In this case, tuberculous enroded the regular landmarks, and few anatomical structure provided the approach route for implant. So we considered tympanic ostium of eustachian tube as the anatomical landmark, and postoperative radiology showed electrode array was inserted in the cochlear region. As to the deviation of the electrode array from its intended location, we attributed it to a shallow depth insertion in the scala tympani.

It is difficult but not impossible to do a cochlear implant in such ossified cochlea, Smullen[16] has classified cochlear ossification in three degrees of obliteration: (1) limited to the niche of the round window; (2) inferior segment of basalturn up to 180°, and (3) the obliteration goes beyond basalturn. For patients with stage Ⅰ or Ⅱ ossification, the electrode can then be inserted normally through scala tympani or vestibular scala after clearing calcifications of soft tissue in the labyrinth. While for stage Ⅲ Gibson[17] recommend drill a cleft so that the electrode array can be inserted along the position of the basal coil. In some stage Ⅲ cases, we can also implant a split electrode array. Two cochleostomies are made through the facial recess. The first is the tunnel into the inferior basal turn, and the other is into the second turn of the cochlea[16].

Various types of electrodes including short arrays and split arrays have been designed for the situation of narrow canal as in this case. We considered C40+S to be appropriate for this patient, since its array (12.1 mm) is shorter than regular ones[8], and the operative cavity was rather complicated in this patient. This severe degree of cochlear ossification poses a challenge to the cochlear implant surgeon.

It is well known that patients with ossified cochleas may benefit from implantation, and yet few has received cochlear implant in cases of TOM[10]. Although the process is tortuous, our patient still benefit from cochlear implant. Moreover, we have accumulated valuable experience in this process.

This case highlights the cochlear implant challenge posed by tuberculous otitis media with complications. Early diagnosis and treatment is very important in TOM. A high index of clinical suspicion of tuberculousotitis media is required in patients who have no response to standard antibiotic therapy for chronic middle-ear infection. Antituberculous medication must be available throughout the course of treatment. CT and MRI imaging of the temporal bone provide useful information for diagnosis and surgery. Furthmore, application of different surgical techniques for different situations in ossified cochlea, and tympanic ostium of eustachian tube can be used as reliable signs of cochlear implant in such patients. Patients with ossified cochlear could benefit from cochlear implantation.

Conflict of interest statement

We declare that we have no conflict of interest.

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