Malignant External Otitis with Facial Nerve Paralysis

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Case Presentation

A 62 years old man was referred to our department experiencing right otalgia, purulent ototomea, ear loss and a grade III right facial nerve palsy according to the House Brackmann scale.

He was affected by decompensated insulin-dependent type 2 Diabetes. The patient had a history of non cholesteatomatous ipsilateral chronic otitis treated with tympano-mastoid surgery 10 years before, without any sign of recurrence.

At the examination there was a purulent discharge from the right ear, the canal appeared red and swollen, the tympanic membrane was not visible and the retro auricular and preauricular areas showed redness and swelling. Meningeal signs were negative, without any cervical lymphadenopathy or any alteration of the blood exams.

CT scan showed opacification of the right middle ear and mastoid cavity, with osteolysis of the facial canal and of the tegmen tympani, while MRI with contrast showed a mild enhancement in the meninx.

The scintigraphic evaluation with 99-Technetium confirmed the osteomyelitis with increased uptake of radio-marked Difosfonate in the temporomandibular joint, mastoid cells, semicircular canals, cochlea, middle and inner ear and in the greater wing of the sphenoid bone.

The first antibiogram was positive for P. Aeruginosa and, according to the Infectivologist’s evaluation; a specific therapy with intravenous 400mg Ciprofloxacin every 12 hours was undertaken. We also started a corticosteroid therapy with the support of a diabetes specialist.

Because of the MRI results we asked for a neurological evaluation; a rachicentesis was performed excluding the hypothesis of meningitis. The EMG and ENG confirmed the right 7th nerve palsy with a decreased conduction velocity of all the branches of the nerve.

Hyperbaric oxygen therapy was not undertaken, because of the lack of a clear evidence demonstrating its efficacy (level of evidence Grade D) [1]. We took a biopsy of a polypoid formation of the ear canal that resulted to be hyperplastic tissue with dyskeratosis and granulation tissue.

After ten days of therapy, symptoms were reduced and the paralysis of superior branches of the facial nerve had mildly improved but the persisting swell of the ear canal did not allow a thorough evaluation of the tympanic membrane. Therefore an explorative tympanotomy was performed and few days after surgery the marginalis branch paralysis of the facial nerve had improved according to ENG results and the culture swab showed a polymicrobial flora.

Keywords: Malignant external otitis; Tympanotomy; Facial nerve paralysis; Otitis; External auditory canal

Case Report

Malignant External Otitis (MOE) is a severe infection of the external auditory canal and the skull base that is often seen in elderly diabetic patients with a high mortality and morbidity rate: in more than 98% of cases the causative pathogen is P. aeruginosa.

Here we describe a case of MOE associated with facial nerve paralysis in a patient hospitalized.

The 62 years old man was referred to our department experiencing right otalgia, purulent otomea, ear loss and a grade III right facial nerve palsy according to the House Brackmann scale. He was affected by uncompensated insulin-dependent type 2 Diabetes. The patient had a history of non cholesteatomatous ipsilateral chronic otitis treated with tympano-mastoid surgery 10 years before, without any sign of recurrence.

After ten days of specific antibiotic and corticosteroid therapy, symptoms were reduced and the paralysis of superior branches of the facial nerve had improved but the persisting swell of the ear canal did not allow a thorough evaluation of the tympanic membrane. Therefore an explorative tympanotomy was performed and few days after surgery the marginalis branch paralysis of the facial nerve had improved according to ENG results and the culture swab showed a polymicrobial flora.

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After ten days of therapy, symptoms were reduced and the paralysis of superior branches of the facial nerve had mildly improved but the persisting swell of the ear canal did not allow a thorough evaluation of the tympanic membrane. Therefore an explorative tympanotomy was performed, without any sign of otorrea, osteolisis or cholesteatoma but it was determined. Few days after surgery the marginalis branch paralysis of the facial nerve had improved according to ENG results and the culture swab showed a polymicrobial flora. Moreover the swelling of the auditory canal was reduced.

The antibiotic therapy was carried on after the dismissal with an association of oral Amoxi-clavulanic Acid 1g 3/die and Ciprofloxacin 500mg2/die for 4 weeks. The weekly follow-up showed an improvement of the oedema and after 1 month the symptomatology was disappeared and the facial nerve function had further ameliorated according to ENG and clinical signs.

After three months, at the scintigraphic control with marked 99-Tg...
leucocytes, the inflammation and the facial nerve palsy were solved.

**Discussion**

Toulmouche reported the first case of Malignant External Otitis (MEO) in 1838. In 1959, Meltzar reported a case of Pseudomonal osteomyelitis of the temporal bone, but the name was coined by Chandler that defined malignant external otitis (MEO) as an unresponsive infection of the external ear canal associated with granulation tissue at the junction of the osseous and cartilaginous portions that occurs almost exclusively in elderly and diabetic patients.

Most cases of MEO (86–90%) have been reported in diabetic patients. This is probably due to immunodeficiency and to abnormalities of small blood vessels.

There is also a group of non-diabetic immunocompromised patients that can develop the disease (e.g. malignancy, chemotherapy, malnutrition etc.). Even if the most commonly causative organism of MEO is Pseudomonas Aeruginosa, there are other organisms that have been isolated, such as Proteus mirabilis, Aspergillus fumigatus, Proteus spp, Klebsiella spp, and Staphylococci. The majority of fungal MOE occurs in immunosuppressed individuals with AIDS [2].

The symptoms of MOE are severe unremitting throbbing otalgia, purulent otorrhea, the sensation of a blocked ear, and hearing loss.

Cranial nerves can be affected by inflammation and the facial nerve (VII) is the most commonly affected.

In 1987 Cohen and Friedman listed a set of major and minor criteria for MEO and suggested that the diagnosis was established when all the major criteria were present.

The major criteria were: pain, edema, exudate, granulations, microabscess (when operated), positive bone scan or failure of local treatment often more than one week, and possibly *Pseudomonas* spp in culture. The minor criteria are diabetes, cranial nerve involvement, positive radiograph, debilitating condition and old age. All of the obligatory criteria must be present in order to establish the diagnosis.

Levenson’s criteria can also be used for diagnosis. Criteria include: refractory otitis externa, severe nocturnal otalgia and purulent otorrhea associated with *Pseudomonas* infection and granulation tissue in an immunocompromised or diabetic patient [3].

Both Levenson, Cohen and Friedman’s criteria were present in our patient.

Laboratory parameters are generally normal in malignant external otitis, with the exception of an elevated erythrocyte sedimentation rate (ESR) and/or C-reactive protein (CRP).

The leukocyte count in malignant external otitis (MOE) is usually normal or mildly elevated.

CT scanning is ideal for the assessment of bone erosion in MOE while MRI results are better than CT at demonstrating medial skull base disease due to its ability to delineate changes in the fat content of the marrow [4].

Gallium citrate scanning can be used to follow disease activity, since the radioisotope is incorporated into granulocytes and bacteria. Bone scanning with technetium (Tc 99m), where the radionuclide tracer accumulates at sites of osteoblastic activity, is a very sensitive exam but it is not specific, since there are reports of positive bone scans in simple external otitis, and bone scans are not suitable for following response to treatment since they do not normalize.

A biopsy is indicated in order to differentiate MOE from squamous cell carcinoma of the external auditory canal.

The treatment of MOE mainly consists of strict glycemic control, aural toilet, systemic and ototopic antimicrobial therapy, and hyperbaric oxygen therapy [2].

The use of fluoroquinolones (especially ciprofloxacin monotherapy) or ceftazidime monotherapy has been proved to be very effective.

Patients with ciprofloxacin-resistant *P. aeruginosa* require parenteral antibiotics with antipseudomonal beta-lactam antibiotics with or without an aminoglycoside [5].

The antibiotic treatment should be carried on until the resolution of osteomyelitis. The response to the treatment can be evaluated with a gallium citrate Ga 67 scan, which should be repeated every 4-6 weeks, until the normalization of the exam.

Hyperbaric oxygen therapy should be used only as an adjunct to antimicrobial therapy especially for those patients experiencing a poor response to therapy, or with recurrent cases [3,6].

Surgery is indicated for local debridement, removal of necrotic bone tissue, or abscess drainage, while facial nerve surgical decompression is not indicated.

MOE is characterized by a recurrence of 9-27% and this is usually related to the inadequate length of the therapy.

Its Complications include meningitis, abscess, sagittal, Dural and cavernous sinus thrombosis.

Most current studies report a mortality rate of less than 10%, but it remains high in patients with cranial neuropathies (other than VII), intracranial complications, or with irreversible systemic immunosuppression.

**Conclusion**

Malignant otitis externa is an aggressive infection that can represent a life-threatening condition.

In our patient, a prolonged steroid and antibiotic treatment and a strict control of glycemic levels permitted to control the osteomyelitis, but the surgical approach appeared essential for the resolution of the disease.

The aural toilet with the elimination of the necrotic and granulation tissue, lead in fact to the aeration of the tympanic cavity, helping the resolution of the paralysis, especially the function of the marginal branch.

In our patient we obtained the complete resolution of the infection without any recurrence over a year confirming, according to the literature, that the presence of the facial paralysis does not represent a negative prognostic factor.
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