**Malignant or Necrotizing Otitis Externa Complicated by Peripheral Facial Paralysis in an Immunocompromised Diabetic Patient : A Case Report**

 **Abstract**

Necrotizing otitis externa is a serious condition that poses increasing challenges in therapeutic management. We report a case of necrotizing otitis externa complicated by facial paralysis in an unbalanced diabetic patient.

Malignant otitis externa (MEO), also known as necrotizing otitis, is a rare but severe infectious disease of the external auditory canal (EAC) that can spread to the adjacent soft tissues and bone.

The predisposing condition is diabetes, with the onset occurring electively in elderly diabetic or immunocompromised patients.

Glycaemic control combined with antibiotic therapy adapted to the pathogen resulted in regression of clinical signs and cure of the infection.

Keywords: Diabetes, Otitis, external, necrotizing, treatment

INTRODUCTION

Malignant otitis externa (MOE), also known as progressive necrotising otitis externa (PNEO), is an invasive infectious pathology with a high morbidity, which occurs mainly in immunocompromised, elderly and diabetic subjects.

It is an osteitis of the temporal bone, generally due to Pseudomonas Aeruginosa, which starts in the EAC and then spreads to the base of the skull, causing damage to the cranial nerves.

It is therefore a diagnostic and therapeutic emergency, which can be life-threatening.

Facial paralysis is a rare complication of otitis externa. It is more common in cases of necrotising otitis externa (NEO) with skull base osteomyelitis, particularly in the presence of co-morbidities. (1).

Treatment is based on glycaemic control, local treatment, appropriate prolonged dual antibiotic therapy, and possibly surgery(2).

Case Présentation :

This was a 52-year-old insulin-dependent diabetic patient treated with mixtard®30 and actrapid®30 for 30 years, with amputated toes, who was admitted to hospital for management of malignant right otitis externa which had been evolving for 6 months.

He presented with otalgia resistant to the usual analgesics and antibiotics, hypoacusis and tinnitus.

Clinical examination was marked by swelling in the region of the temporomandibular joint followed by right peripheral facial paralysis on the side of the diseased ear. Otoscopy showed a narrowed EAC covered with whitish deposits with black spots. The diagnosis of OEM was suspected. The patient was admitted to hospital for management.

The biological work-up showed a CRP of 25mg/l, which increased progressively despite the initiation of treatment, hyperglycaemia of 28.01 mmol/l and glycosylated haemoglobin (HbA1c) of 13%. Bacteriological samples showed the presence of staphylococcus.

The tonal audiogram showed mixed hearing loss in the right ear and a sensorineural component in the left ear.

CT and MRI were consistent with acute malignant otitis involving the parapharyngeal and periauricular spaces and the right temporomandibular joint.

Bone scan of the rock showed active and extensive osteitis of the right rock.

Treatment consisted of parenteral dual antibiotic therapy with a 3rd generation cephalosporin (C3G) (ceftriaxone) 2g/24h and a fluoroquinolone (Ciprofloxacin 200 mg x2/24h). Local treatment consisted of calibrating the CAE with otowick pop instillation and 5 drops of antibiosynalar x2/day for 15 days. Symptoms persisted, prompting a therapeutic adjustment to Fortum 1g x3/d and ciprofloxacin 400 mg x2/d and mastoidectomy to decompress the nerve.

After this procedure, the outcome was favourable. Diabetes was managed by diabetologists with Mixtard®30 and Actrapid®30. Oral ciprofloxacin 1g/day was continued for 30 days.

   

FIG 1 : Otoscopy of the right ear showing whitish secretions and black spots.

FIG 3 : The MRI supports a diagnosis of malignant otitis externa infiltrating the peri-auricular space, temporomandibular region, and right parapharyngeal space.

FIG 2 : CRT SCAN ROCK in favor of malignant otitis externa with invasion of the temporomandibular region.

FIG 4 : The CRP curve declines from the third week onwards despite treatment.

**Discussion :**

The predisposing condition is essentially diabetes (75-95%). The occurrence or severity of OME correlates with diabetic imbalance. Bacteriological examination, antibiotic susceptibility testing and medical imaging should be performed systematically, and treatment is based on broad-spectrum antibiotic therapy combined with optimised insulin therapy(2)

OME is a rare and serious complication of otitis externa. We report a case with a complication of peripheral facial paralysis. This pathology corresponds to cellulitis of the EAC, originating at the osteo-cartilaginous junction, giving rise to perichondritis, osteitis of the tympanal bone and of the temporomandibular joint (3).

 In the absence of effective treatment, this infection spreads to the base of the skull, reaching the infra-temporal fossa, the parapharyngeal space, the nasopharynx and the intracranial compartment. The facial nerve is primarily affected in the stylomastoid foramen. Spread of the infection to the jugular foramen can lead to thrombophlebitis of the lateral sinus and paralysis of other cranial pairs (IX, X, XI, XII). In our patient, facial involvement was present, stage IV with Charles Bell's sign, for which an ophthalmic opinion was sought in favour of exposure keratitis and additional management (4).

Several authors have reported a predominance of males. Elderly diabetic, poorly balanced or immunocompromised subjects are the main predisposers to infection. Diabetes is the main factor predisposing to the development of OME, with a prevalence of 75-95%. Rubin et al report that hyperglycaemia is not a factor in the pathogenesis of the disease, but rather the vascular complications of diabetes (microangiopathy) (5).

Histopathological studies of the capillaries in the skin and subcutaneous tissue around the temporal bone have shown that diabetics have a thickening of the subendothelial basement membrane, resulting in hypoperfusion of the tissues and reduced resistance to infection, which spreads from person to person (5).

Pseudomonas aeruginosa is the germ responsible for over 95% of cases. Pseudomonas aeruginosa is a commensal germ of the skin, which colonises the EAC after swimming in a pool or washing an ear. It becomes pathogenic when the immune defences are impaired or after a break in the skin. Other germs may also be involved, such as staphylococcus epidermidis, which is positive on bacteriological study, and more rarely fungal agents such as Aspergillus fumigates (6).

The biological work-up shows CRP to be generally accelerated; it is a non-specific inflammatory marker for the diagnosis of the disease, but can be used in follow-up as an indicator of response to treatment (7).

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CT and MRI scans are used to confirm the diagnosis and assess the extent of lesions, particularly in the bone (temporal bone, base of skull, temporomandibular joint). An MRI was performed on the patient. It showed thickening of the soft parts of the EAC and filling of the mastoid cells in all cases and lysis of the mastoid. However, its usefulness in follow-up is limited given that the lesions of demineralisation and erosion of the bone cortex appear late and disappear slowly after healing. In addition, the lesions seen on CT are not specific to OENP and can also be seen in malignant tumour pathology of the EAC.

Scintigraphy or PET scans are more effective.

 These two examinations were performed on the patient. (8)(9)(10)

Treatment of OME is essentially medical, and should be initiated as early as possible in a specialised setting. There are 3 essential components to this treatment, combining correction of immunodepression or glycaemic control with diabetes control, daily local treatment and effective prolonged systemic antibiotic therapy (13).

Switching to insulin therapy is mandatory, regardless of diabetes control, and all oral antidiabetic agents must be discontinued. The management of our patient was in line with these guidelines, with intensified insulin therapy based on repeated complete glycaemic cycles (before meals and 2 hours after meals). Local treatment included cleaning, calibration, debridement of the external auditory canal and instillation of antibiotic drops (11).

Currently, the antibiotic therapy recommended by the majority of authors is the parenteral combination of a fluoroquinolone (ciprofloxacin or ofloxacin) with C3Gs (ceftazidime or ceftriaxone), followed by a quinolone as soon as clinical improvement is seen.

Anti-Pseudomonas molecules are prescribed as a first-line treatment, and treatment is adjusted according to the results of microbiological samples. In limited forms of PENO, some authors recommend a single course of oral ciprofloxacin at a dose of 1.5g/d in two doses for 6 to 8 weeks. However, given the increasing frequency of ciprofloxacin-resistant Pseudomonas, the combination of an aminoglycoside or a third-generation cephalosporin is essential. (12)

Surgery has a limited role in the treatment of skull base osteitis. According to some authors, it is indicated in cases where the disease has not progressed well under medical treatment, and should be limited to purely local procedures in order to avoid extending the lesions to the healthy bone.

It consists of removal of bone sequestration, debridement of infected tissue and drainage of purulent collections. For others, however, it involves mastoidectomy or even subtotal petrectomy (depending on the extent of the lesions) with decompression of the facial nerve in the event of facial paralysis.

We performed mastoidectomy when the symptoms persisted despite well-managed medical treatment. All patients in our study had a favourable outcome.

**CONCLUSION :**

ENT is a rare and serious infection of the EAC, which frequently occurs in elderly diabetic or immunocompromised patients.

Diagnosis and management are a challenge for ENT physicians.

Pseudomonas aeruginosa is responsible for over 95% of cases of OME.

Its prognosis has improved markedly since the advent of antibiotics active against this germ, and involves a number of aspects: correction of immunodepression or control of diabetes, local treatment of the EAC, prolonged parenteral dual antibiotic therapy and mastoidectomy in certain cases.

**Consent**

As per international standards or university standards, patient(s) written consent has been collected and preserved by the author(s).

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 References

1.Physician. 1er février 2021 ; 16(1) : 117-120. doi : 10.51866/cr1108. PMID : 33948150 ;

2.P L, C ANY, Np B, Ab MN, J M, L N. Malignant Otitis Externa in Libreville : A Report of Four Cases. HEALTH SCIENCES AND DISEASE [Internet]. 2024 [cité 3 sept 2024];25(1). Disponible sur: https://www.hsd-fmsb.org/index.php/hsd/article/view/5145

3.Habra B, El Mghari G, El Ansari N. Otite externe maligne ou nécrosante chez le diabétique : à propos de 10 cas. Annales d’Endocrinologie. sept 2015;76(4):550.

4.Schultz P, Riehm S, Carpentier AS, Boivin G, Schlienger JL. Otite externe nécrosante (otite maligne externe) et diabète. Médecine des Maladies Métaboliques. févr 2011;5(1):53‑4.

5.Rubin Grandis J, Branstetter BF, Yu VL The changing face of malignant (necrotizing) external otitis: clinical, radiological, and anatomic correlations Lancet Infect Dis, 4 (2004), pp. 34–39

6. Moata H, EL Mghari G, EL Ansari N, Ait el abdia R, Rochdi Y, Nouri H, Aderdour L and Raji A. les otites nécrosantes: lorsque l’hyperglycémie prend sa part: à propos de 32 cas.  International Journal of Advanced Research 2019; 7(1): 394-399

7. Stephen S, Subashini B, Thomas R, Philip A, Sundaresan R. Skull Base Osteomyelitis Caused by an Elegant Fungus. J Assoc Physicians India 2016 Feb;64(2):70-71

8.Peled C, El-Seid S, Bahat-Dinur A, Tzvi-Ran LR, Kraus M, Kaplan D. Necrotizing otitis externa-analysis of 83 cases: clinical findings and course of disease. Otol Neurotol 2019;40:56–62.

9.Altuna Mariezkurrena X, Gómez Suárez J, Luqui Albisua I, Vea Orte JC, Algaba Guimerá J. [Prevalence of exostoses among surfers of the Basque Coast]. Acta Otorrinolaringol Esp 2004 ; 55 : 364-8.

10. Ismail H, W.P. Hellier, V. Batty Use of magnetic resonance imaging as the primary imaging modality in the diagnosis and follow-up of malignant external otitis J Laryngol Otol, 118 (2004), pp. 576–579

11. Al-Noury K, Lotfy A. Computed tomography and magnetic resonance imaging findings before and after treatment of patients with malignant external otitis. Eur Arch Otorhinolaryngol 2011 Dec;268(12):1727-34

12. Hasibi M, Ashtiani MK, Motassadi Zarandi M, Yazdani N, Borghei P, Kuhi A, Dabiri S, Hosseini R, Sardashti S. A Treatment Protocol for Management of Bacterial and Fungal Malignant External Otitis: A Large Cohort in Tehran, Iran. Ann Otol Rhinol Laryngol. 2017 Jul;126(7):561-567

13. Stevens SM, Lambert PR, Baker AB, Meyer TA. Malignant Otitis Externa: A Novel Stratification Protocol for Predicting Treatment Outcomes. Otol Neurotol 2015 Sep;36(9):1492-8 12

14.Glikson E, Sagiv D, Wolf M, Shapira Y. Necrotizing otitis externa: diagnosis, treatment, and outcome in a case series. Diagn Microbiol Infect Dis 2017 Jan;87(1):74-78