



Original Article

Malignant Otitis Externa – A Retrospective Study of 15 Patients Treated in a Tertiary Healthcare Center

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OBJECTIVE: Malignant otitis externa (MOE) is an uncommon but potentially fatal disease of the external auditory canal. The study aimed at evaluating the demographic profile, coexisting disabilities, clinical presentations, and management of MOE

MATERIALS and METHODS: This is a retrospective study of patients with MOE who were treated at the Otorhinolaryngology Department of our institution. The case records of patients treated between 2006 and 2013 for MOE were reviewed from the Medical Records Department of the hospital. The details were tabulated in a master chart, and the data were analyzed.

RESULTS: Fifteen patients with MOE were treated as inpatients at the Otorhinolaryngology Department of our hospital during the study period. Of these, 12 were males and three were females. Among the 15 patients, 14 were diabetic and one was non-diabetic. Earache was the most common symptom observed in all patients; edema and granulations in the ear canal was the most common sign observed in 12 patients. *Pseudomonas aeruginosa* was isolated in 11 patients. All patients were managed medically with intravenous antibiotics.

CONCLUSION: MOE is a rare but aggressive condition affecting the external ear, which is commonly observed in elderly diabetic individuals. Immune senescence may be the cause of MOE in elderly people. *Pseudomonas* is the most common causative organism isolated in this condition. Most of these patients can be managed with medical treatment; reserving surgery only for the removal of granulation tissue and for histopathological examination.

KEYWORDS: Malignant otitis externa, *Pseudomonas aeruginosa*, diabetes mellitus, biopsy

INTRODUCTION

Malignant otitis externa (MOE) is an aggressive form of inflammation of the external auditory canal and is commonly caused by *Pseudomonas aeruginosa*. It affects elderly diabetics and immunocompromised people. The infection begins as an inflammation of the external auditory canal that later spreads to the deeper structures, such as cartilage and the bone of the skull base.^[1] The condition was first reported by Toulmouche in 1838^[2]. It is also called as necrotizing otitis externa or skull base osteomyelitis because of the aggressive nature and involvement of the skull base in some patients. Chandler published a report on this condition in 1968, and he named the condition as MOE^[3]. Once considered as a fatal condition, this disease can now be controlled with the advantage of better diagnostic and therapeutic modalities. The treatment of this aggressive disease is primarily medical, the antibacterial agents to be decided after culture and sensitivity of the ear discharge. Quinolones proved to be an excellent therapeutic agent in the beginning, but later, the infection became resistant to quinolones. Antipseudomonal cephalosporins or other antimicrobial agents proved to be more effective. In some cases, fungal infections may be present after some weeks with antimicrobial treatment over the course of the disease. This may result in the deterioration of a patient's condition even after having shown a good initial response to treatment. Surgery has a limited role in the treatment of MOE. Most of the times, surgery is limited only to the biopsy of the granulation tissue. A retrospective study of 15 cases of MOE is presented here.

MATERIALS and METHODS

This is a retrospective study of patients with MOE who were treated in the Otorhinolaryngology department of our institution. This study was approved by the Ethics committee of our institution, "KSHEMA Ethical Committee." The study aimed at evaluating the demographic profile, coexisting disabilities, clinical presentations, and management of MOE. The case records of patients treated between 2006 and 2013 for MOE were reviewed from the Medical Records Department of the hospital. The details were tabulated in a master chart, and the data were analyzed.

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Table 1. Clinical presentations of malignant otitis externa

Clinical features	Number of patients (n=15)	Percentage
Earache	15	100
Ear discharge	13	87
Temporomandibular joint pain	3	20
Edema and granulations in EAC	12	80
Edema of EAC	3	20
Co-existing CSOM	3	20
Polyp in EAC	2	14
Swelling in preauricular region	2	14
Facial palsy	1	7

EAC: external auditory canal; CSOM: chronic suppurative otitis media

Table 2. Organisms isolated and their sensitivity to antibacterial agents (n=15)

Number of patients	Organisms	Antibacterial Sensitivity
7	<i>Pseudomonas</i>	Ceftazidime
4	<i>Pseudomonas</i>	Ciprofloxacin
1	<i>Klebsiella</i>	Ceftriaxone
1	Diphtheroids	Ceftriaxone
2	No growth	-

RESULTS

Fifteen patients of MOE were treated as inpatients in the Otorhinolaryngology department of our hospital during the study period. Of these, 12 were males and three were females. Among the 15 patients, 14 were diabetics and one was non-diabetic. The age of the patients ranged between 25 and 82 years. The youngest patient in the study was a 25-year-old male with type 1 diabetes mellitus. The eldest patient was an 82-year-old male who was non diabetic.

The diagnosis of MOE was made on the basis of the patient's history, clinical examination, microbiology of ear discharge, and radiological examination. The clinical presentations of these patients are shown in the Table 1. Earache was the most common symptom seen in all patients, followed by ear discharge, which was blood stained. Noticeably, none of the patients had fever at presentation. The examination of the ear revealed edema and granulations in the ear canal in 12 patients (80%) and canal wall edema in three. Three patients had pain related to the temporal joint of the corresponding side. Coexisting mucosal type of chronic suppurative otitis media was seen in three patients. Two patients had a polyp in the ear canal. Two patients had a swelling in the preauricular region. One of the patients had facial palsy on presentation.

The total leukocyte count was within normal limits in 14 patients. Only one patient had an elevated total leukocyte count of 12,700/cmm. Elevated erythrocyte sedimentation rate (ESR) was observed in all the patients. Three patients had ESR above 100, the highest being 117 mm/h. Four had ESR between 51 and 100. Eight patients had ESR between 20 and 50; the lowest ESR was 20mm/h observed in one patient. Blood sugar levels were raised in the 14 diabetics. Blood sug-

**Figure 1.** High resolution computed tomography scan of the temporal bone scan of a patient with malignant otitis externa showing erosion of the mastoid segment of the facial nerve canal

ar levels were within normal limits in one patient. Biochemically, all patients had a good renal function.

Culture of the ear discharge yielded *P. aeruginosa* in 11 patients, *Klebsiella* species in one, and diphtheroids in one. The ear discharge of two patients did not yield any growth. In one of the patients with no growth, the histopathological examination of granulation tissue from the ear canal showed the presence of *Klebsiella*.

Computed tomography (CT) scan was performed in four patients; one with facial nerve palsy and three with temporomandibular joint pain. High resolution CT of temporal bone showed erosion of the mastoid segment of the facial nerve canal (Figure 1). The CT scan showed erosion of the temporomandibular joint in one of the patients with preauricular swelling (Figure 2, 3). The other two patients had only soft tissue thickening with rarefaction of bone without gross destruction.

The patients were treated with intravenous antibacterial medications according to the sensitivity test report. Nine patients were treated with ceftazidime, four with ciprofloxacin, and two with ceftriaxone (Table 2). Two patients in whom there was no growth in the ear discharge were empirically treated with ceftazidime, considering the fact that the clinical features were consistent with MOE. Topical acetic acid drops were instilled for all patients. Antibiotic ear drops were not instilled in these cases as protocol. Microscopic examination and cleaning of the ear canal was conducted in all patients. In patients with granulation in the ear canal, a biopsy was taken. Histopathological examination of the biopsy showed features of inflammatory granulation tissue.



Figure 2. Computed tomography scan (axial) of a patient with malignant otitis externa showing erosion of the temporomandibular joint

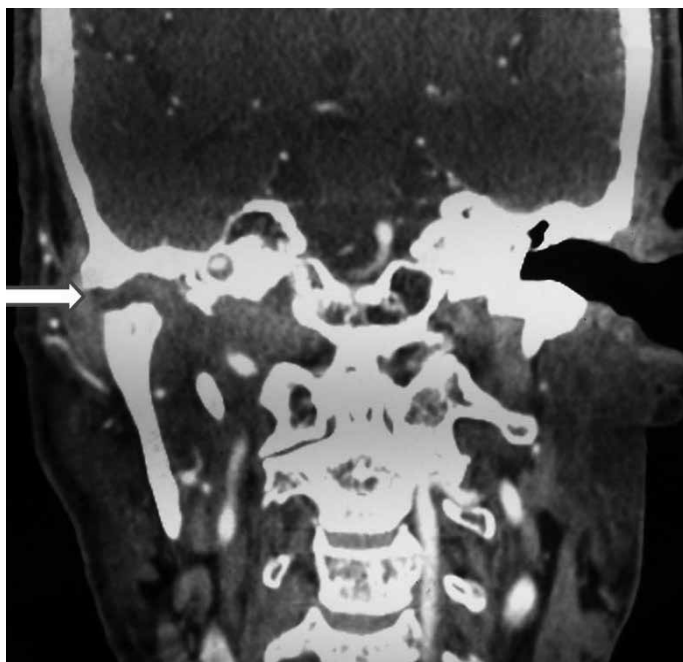


Figure 3. Computed tomography scan (coronal) of a patient with malignant otitis externa showing erosion of the temporomandibular joint and soft tissue opacity in the tympanic cavity

The average duration of treatment was 22 days. Facial nerve palsy did not improve in spite of physiotherapy during the study period. Two patients had a recurrence of the disease. One patient with temporomandibular joint erosion had longstanding pain even after the completion of treatment, and the same patient had a recurrence of the disease twice—once after three months and the other after one

year. The other patient had a recurrence of the disease once—four months after completing treatment.

DISCUSSION

MOE is an aggressive form of external ear infection. The condition, although rare, is occasionally fatal. It was described as pyocutaneous osteomyelitis of the temporal bone by Meltzer in 1959 [4]. Some authors mentioned that Toulmouche described this condition as early as 1838 [2]. It was Chandler who published the first series of this condition, and named this condition MOE [3]. This aggressive infection is commonly seen in immunocompromised people, such as those with diabetes mellitus, human immunodeficiency virus/acquired immune deficient syndrome (HIV/AIDS), chemotherapy induced aplasia, refractory anemia, chronic leukemia, lymphoma, splenectomy, and renal transplantation [5].

We have treated 15 patients over last eight years in the Otorhinolaryngology department of our tertiary care hospital. This shows the rarity of this condition. Out of the 15 patients, 14 were diabetics (94%). Only one patient, who was a male aged 82 years, was a non-diabetic. Most of the authors have noted that people with diabetes are more prone to this infection [5, 6]. The vulnerability of diabetics to MOE is because of the endarteritis, microangiopathy, and small vessel obliteration due to the disease [5]. Immunosenescence can be explained as the cause of MOE in elderly people because in our study, the only diabetic was an elderly male aged 82 years [7]. MOE is also reported in immunocompromised children or children with diabetes; however, the incidence is not as common as that seen in elderly diabetics [5].

Clinical features of MOE include severe otalgia, purulent otorrhea, aural fullness, and hearing loss. Otalgia is more severe and out of proportion to the clinical findings and persists after the usual treatment of otitis externa. Temporomandibular joint pain, hemifacial pain, headache, and trismus are the other common features when there is anterior extension of the disease. Facial nerve palsy can be the presenting feature in some cases [5].

Elevated ESR has been reported by many authors, though it is not diagnostic. It has been found to decrease with treatment and can be considered as a prognostic marker [8]. In our study, all patients except one had elevated ESR. However, we could not retrieve follow-up ESR in these patients. Granulations in the external auditory canal are the most common finding reported in literature.

Levenson's criteria are useful for the diagnosis of MOE. The criteria include refractory otitis externa, severe nocturnal otalgia, purulent otorrhea, presence of granulation tissue in the external auditory canal, growth of *Pseudomonas* in the culture from ear discharge, and presence of diabetes or immunocompromised state [9].

The facial nerve is the most commonly affected cranial nerve. This is because of the proximity of the stylomastoid foramen. In our series, only one patient had facial nerve palsy. The facial nerve palsy did not recover even after the completion of treatment of MOE. Rajput et al, in their series of 21 cases, reported eight patients with cranial nerve palsy, five with facial palsy, two with facial and CN X palsy, and one with facial and CN V palsy [6]. The palsy did not recover during the treatment and follow-up period, which also occurred in our study.

Corey et al in 1985, in their series of 83 patients, reported facial palsy in 58 (70%) patients^[10]. Mani et al.^[11] concluded that the prognosis of MOE with facial palsy is not different from those without palsy.

P. aeruginosa is the commonest organism isolated in most of the reports^[6]. Other bacteria such as *Staphylococcus aureus*, *S. epidermidis*, *Proteus mirabilis*, *Klebsiella oxytoca*, and *P. cepacia* have been isolated in MOE^[5]. In our study, out of the 15 patients, 11 (73%) had *P. aeruginosa*. *Klebsiella* and diphtheroids grew in one patient each. The culture did not grow any organism in the two patients, which is possibly due to an error in sampling. Interestingly, in one of these two patients, the histopathological examination suggested the presence of *Klebsiella*. The condition of these patients improved when they were empirically treated with ceftazidime.

Fungi are occasionally isolated from MOE patients. Fungal otitis externa is considered when a patient with symptoms and signs of MOE does not respond to the appropriate treatment for MOE. The bacterial culture in these patients will be negative^[12]. Fungal MOE is found more in AIDS patients than in diabetics^[5]. Fungal culture was not performed in any of our patients. However, the authors feel that fungal culture was indicated in at least one of the patients where the bacterial culture was negative.

An imaging study is mandatory when a patient presents with complications, such as facial palsy or temporomandibular joint pain. CT scan is a better tool to evaluate the presence of bony erosions. Mani et al reported that the clinical course does not closely correlate with the CT scan findings^[11].

A technetium-99m bone scan is a more sensitive investigation because it is positive in all cases of MOE. As the test is based on binding to osteoblasts, which are also seen in neoplasms and during trauma, it is not specific to MOE. A gallium 67 scan is positive in soft tissue and bone infections, where it binds to lactoferrin. The uptake returns to normal after the infection is cleared. Hence, a gallium 67 scan is more useful than Technetium-99m in diagnosis and also in monitoring the response to treatment and detecting recurrence.^[5] None of the patients were evaluated with a radionuclide scan because of the non-availability of this instrument.

Treatment of MOE is mainly medical. Strict control of diabetes mellitus is extremely important in controlling the disease progression. Oral ciprofloxacin has been the treatment of choice in the past.^[13] However, with the widespread and irrational use of ciprofloxacin, the number of resistant cases is increasing. Parenteral antibiotics depending on the culture and sensitivity are administered. Currently, antipseudomonal penicillin-like carbenicillin and cephalosporins like ceftazidime and amino glycosides are the commonly used drugs. However, nephrotoxicity and ototoxicity need to be monitored when administering aminoglycosides as diabetics are at risk of compromised renal function. The duration of treatment is more prolonged in cases of otitis externa. The response to treatment can be determined by the improvement of symptoms, imaging studies, and radionuclide bone scans. Six to eight weeks of therapy is required for complete remission of the disease. In exceptional cases, the duration of treatment may exceed several months. In our study, the average duration of treatment was only 22 days, and during follow up, two patients

had a recurrence of the disease. The authors feel that the recurrence could be actually residual disease because of inadequate therapy.

Surgery has a limited role in the management of MOE. The role of surgery is the debridement of necrotic material in cases of extensive disease. Mastoidectomy, as a part of debridement, may be needed to clear the disease in serious cases. In our study, surgical management in the form of granulation removal and biopsy was conducted in 12 patients. Histopathological examination revealed inflammatory granulations in all these patients.

The recurrence of MOE after complete treatment is not uncommon. Recurrence rates of 15-20% have been reported in literature.^[14] In our study, two patients had recurrence of the disease (14%). One patient who had temporomandibular joint involvement in the first presentation had a recurrence after three months of completion of treatment. He had a recurrence again after one year. The other patient had recurrence once after four months, which was treated successfully. Mortality because of MOE has dropped from 50% in an earlier report of Chandler to 0-15% reported by Naroznyet al.^[3, 15] In a recent study, Loh et al reported a mortality rate of 21%. All 15 patients were free from the disease during the study period^[16]. No deaths occurred due to MOE.

Synchronous MOE and squamous cell carcinoma of external auditory canal have been reported by Chin et al.^[17] Both conditions may present with cranial nerve palsies. Imaging studies in both these conditions may show similar features as both have a tendency of bone erosion and destruction of soft tissues. This emphasizes the need for biopsy of granulation tissue in patients with MOE, especially when the disease is not responding to treatment. Early diagnosis of malignancy is essential to early initiation of treatment to prevent the spread of the disease.

We conclude that MOE, which is commonly seen in elderly diabetics, is a rare but aggressive condition affecting the external ear. Immunosenescence may be the cause of MOE in elderly people. *Pseudomonas* is the most common causative organism isolated in this condition. MOE has to be considered when the symptoms are out of proportion with the clinical findings of otitis externa, thus warranting further evaluation. Most of these patients can be managed with a medical line of treatment with surgery only for the removal of granulation tissue and for histopathological examination. Appropriate antibacterial medication for an appropriate duration is necessary for complete cure and for preventing recurrence, which could actually be a residual disease. Considering the uncommon nature of the disease, a high index of suspicion is required for diagnosis.

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