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NECROTIZING OTITIS EXTERNA: A DEADLY MISFORTUNE

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ABSTRACT

Necrotizing otitis externa or malignant otitis externa (MOE) is an emerging problem in developing countries. It is an invasive, potentially life-threatening condition affecting immunocompromised people. The aim of this study is to analyze demographic data, clinical findings, diagnosis and prognostic markers in the management of this condition. This is a retrospective study including 11 patients diagnosed and treated for MOE at a tertiary care center. Pseudomonas aeruginosa was the most common causative organism. All the patients were managed medically and symptomatic improvement was seen with meticulous aural toileting, intravenous antimicrobials and control of the comorbid condition. This study signifies that conservative management has a better outcome than surgery.

KEYWORDS

Malignant Otitis Externa, Diabetes, Immunocompromised, Pseudomonas aeruginosa, granulation tissue.

INTRODUCTION

Malignant otitis externa (MOE) is a rare, rapidly spreading, life-threatening invasive infection of the external auditory canal and the lateral skull base. Toulmouche first described this condition.[1] The term Malignant Otitis externa was coined by Chandler.[2] Necrotizing otitis externa and skull base osteomyelitis are the other terms used to describe this condition. In recent years, there has been a significant rise in the incidence of MOE, especially in the elderly and immunocompromised patients. This may be attributed to an increase in awareness of the disease, better diagnostic modalities, and an increase in the geriatric and diabetic population in India.(1) The term 'malignant' is a misnomer as it is not a neoplasm but is labelled so due to its aggressive clinical behaviour, poor treatment outcome, and high mortality rate. The term 'necrotizing' emphasizes the destructive nature of the infection.[4]

MOE is described as a disease of an elderly patient with diabetes.[2] Diabetic patients are more prone for MOE due to microangiopathy, hypoperfusion, and diminished host immune response. It begins in the external auditory canal, and then spreads to the skull base and jugular foramen via the fissures of Santorini and the stylomastoid foramen, eventually resulting in local extension to the apex of the petrous bone. Venous channels and fascial planes facilitate the tracking of infection along the dural sinuses, which may result in spread to distant tissue.[5] The facial Nerve is the most affected Cranial Nerve, but glossopharyngeal, vagus, accessory, or hypoglossal nerve involvements can also occur.[6]

The bacterial origin is almost always Pseudomonas aeruginosa, but cases of staphylococcus aureus, Proteus mirabilis and some fungal species such as Candida albicans and aspergillus have been reported. Common presenting symptoms of MOE include otalgia, otorrhea, aural fullness, and hearing loss.[7]

The awareness and understanding of this disease is necessary to formulate a treatment plan. Hence, in this study, we retrospectively audit the case records of 11 patients of MOE and our experience in managing them.

MATERIALS AND METHODS

This is a retrospective study that includes 11 patients diagnosed and treated for MOE at the department of ENT in a tertiary care center during the period of July 2020 to December 2022. All the patients were treated as in-patients during the study period. The diagnosis was made based on Levenson's Criteria shown in table 1. Data collected on gender, age, comorbid condition, clinical presentation, microbiological investigations in the form of bacterial culture, routine blood tests along with inflammatory markers such as Erythrocyte sedimentation rate (ESR) and C reactive protein (CRP), treatment given (antibiotics given, route, and duration) adverse events, complications, and the outcome was analysed. Computed tomography (CT) was performed to identify the spread of the disease. A biopsy was taken from all patients with granulations in the external auditory canal to exclude malignancy. Microscopic examination and cleaning of the external auditory canal were performed for all patients. House Brackmann (HB) scores were used for facial nerve grading. [Table 2] Poor glycaemic control was defined as HbA1c >7.5 at the time the condition was first diagnosed. An adverse outcome was defined as one or more of the following: i)greater than 4 months to the resolution of symptoms, ii)deceased within 6 months, iii)permanent facial nerve palsy (partial or complete). These three outcomes were selected from the data as having the most severe impact on the patients. There is no current validated model within the literature which categorises patient adverse outcomes.[8]

Table 1. Levenson's Criteria

• Refractory Otitis externa for more than 2 weeks.
• Severe nocturnal otalgia.
• Purulent otorrhea.
• The presence of granulation tissue in the bony cartilaginous junction of external auditory canal.
• Growth of Pseudomonas aerugenosa in the culture from ear discharge.
• Immunocompromised state.

Table 2. House Brackmann (HB) Scores

Grade	impairment
I	Normal
II	Mild dysfunction (slight weakness, normal symmetry at rest)
III	Moderate dysfunction (obvious but not disfiguring weakness with synkinesis, normal symmetry at rest) Complete eye closure w/o maximal effort, good forehead movement
IV	Moderately severe dysfunction (obvious and disfiguring asymmetry, significant synkinesis) Incomplete eye closure, moderate forehead movement
V	Severe dysfunction (barely perceptible motion)
VI	Total paralysis (no movement)

RESULTS

Among the 11 patients treated for MOE, nine (82%) were male and two (18%) were female. All of them were diabetic. The age of patients ranged between 44 and 78 years. The youngest in the study was a 44-year-old male and the eldest was a 78-year-old female. The clinical presentations of these patients are shown in Table 3.

Table 3. Clinical Presentations Of Malignant Otitis Externa

Clinical features	Number of patients (n=11)	Percentage
Earache	11	100
Ear discharge	11	100
Temporomandibular joint pain	1	9
Granulations in EAC	10	91
Pre-existing CSOM	2	18
Polyp in EAC	1	9
Facial palsy	7	64
EAC: external auditory canal; CSOM: chronic suppurative otitis media		

All eleven patients (100%) had ear earache which was excruciating, nocturnal, dull, nagging type of pain affecting the external auditory canal, mastoid and the surrounding region. Associated ear discharge was present in all eleven patients (100%) which was blood stained in ten patients. Pre-existing CSOM was seen in two patients (18%). Seven patients (64%) had facial nerve palsy ranging from grade 3 to 4 according to the House Brackmann scoring system. There was single patient(9%) who presented with pain and swelling around temporomandibular joint (TMJ). The examination of ear revealed edema and granulation tissue in ear canal in ten patients (91%) and one patient (9%) had aural polyp.

The total leukocyte count was within normal limits in nine patients(82%). Only two patients(18%) had an elevated total leukocyte count of 12,700/ cmm and 13,000/cmm. Elevated erythrocyte sedimentation rate (ESR) was observed in all the patients. Three patients(27%) had ESR above 100, the highest being 117 mm/h. Four(36%) had ESR between 51 and 100. Four patients(36%) had ESR between 20 and 50; the lowest ESR was 35mm/h observed in one patient. The changes in CRP levels in association with disease progression was also assessed. It was found that significant reduction in inflammatory markers was associated with quicker disease resolution. Blood sugar levels were raised with poor glycemic control in 8 patients(73%) and were normal in three patients(27%). Biochemically, all patients had good renal function.

Culture of the ear discharge yielded Pseudomonas aeruginosa in eight patients(73%), and Methicillin-resistant staphylococcus aureus (MRSA) in one(9%). The ear discharge of one patient(9%) did not yield any growth. The organisms isolated from ear discharge and their sensitivity to antibacterial agents is shown in table 4. HRCT bilateral temporal bone was performed in eight patients(73%), seven(64%) with facial palsy and one(9%) with temporomandibular joint pain. It showed erosion of mastoid segment of facial nerve in all seven(64%) patients and soft tissue thickening with rarefaction of bone without gross destruction for the patient with TMJ swelling.

Table 4. Organisms Isolated And Their Sensitivity To Antibacterial Agents.

Number of patients	Organisms isolated	Antibacterial Sensitivity
8	Pseudomonas	Ciprofloxacin
2	MRSA	Piperacillin and tazobactam
1	No growth	-

The patients were treated with intravenous antibacterial medications according to the sensitivity test report. Eight patients(73%) were treated with ciprofloxacin iv bd hourly for at least 6 weeks and two(18%) were given Piperacillin and tazobactam 4.5 gm tds for 6 weeks and in patient with no growth on culture of ear discharge, he was empirically started with ceftazidime, as the clinical features were consistent with MOE. Topical steroidal eardrops were given to patients with granulation tissue.

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 chronic inflammation.

Facial nerve palsy did not improve despite physiotherapy for 4 patients (47%) during the study period. Improvement in facial palsy was seen in three patients (43%) without complete resolution. One patient had recurrence of disease 4 months after completing treatment. one patient with temporomandibular joint involvement had long standing pain even after completion of treatment.

DISCUSSION

MOE is a rapidly spreading infection, which originates at bony cartilaginous junction of external auditory canal. Its etiology is attributed to infection after trivial trauma like aural irrigation or ear pricking. It starts as cellulites of the deep periauricular tissue that subsequently spreads through the fissures of Santorini to the adjoining skull base, causing periostitis, osteitis, chondritis, osteomyelitis and, eventually, multiple cranial nerve palsies.[3] It is referred to as “skull base osteomyelitis” once the bone infection is confirmed.[9]

In our study, the average age at diagnosis of MOE was 60 + 5.5 years. All the patients had type 2 diabetes mellitus with poor glycemic control in 73% cases. Other comorbidities associated with MOE are hepatitis B infection, HIV, immunosuppression after organ transplant. MOE is also reported in immunocompromised children or children with diabetes; however, the incidence is not as common as that seen in elderly diabetics.[10]

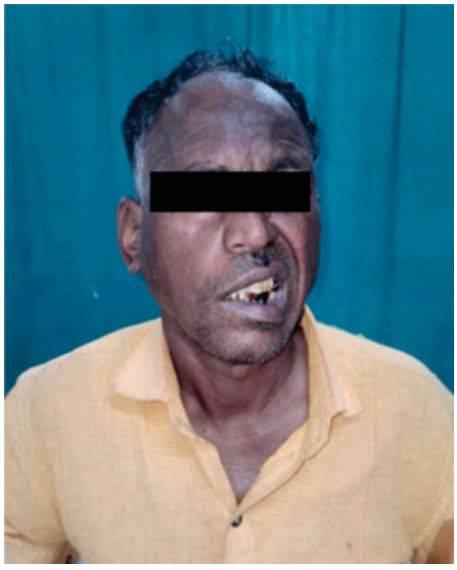

Fig 1. Patient With Grade 5 Left Facial Nerve Palsy.

The most common presenting symptom in our study was severe excruciating earache with discharge, which was out of proportion to clinical features, followed by granulation tissue in the EAC, facial asymmetry, TM joint pain and polyp in EAC. MOE spreads rapidly around the skin and soft tissues of external auditory canal, leading to the involvement of periosteum which could be the reason for severity of earache in all patients. It later spreads through the haversian canals of the compact bone producing multiple abscesses and sequestrum.

Levenson's criteria is useful in diagnosis of MOE. Facial nerve is the most commonly affected nerve. In our series, the facial palsy did not recover completely even after completion of treatment. 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.[11] The facial palsy did not recover even after completion of treatment, as in our study also.

The most common organism isolated on culture of ear discharge in our study was Pseudomonas aeruginosa (73%) and in one patient no growth was seen on bacterial culture which could possibly be due to sampling error. 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.[10]

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.[6]

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.[10] Due to its unavailability, none of our patients were subjected to radionuclide scans.

MOE can be managed with medical line of treatment. Strict control of blood sugar levels in diabetic patients play crucial role in arresting 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.[14]

Hyperbaric oxygen is gradually gaining recognition as a beneficial adjuvant therapy. Phillips and Jones in their Cochrane review, however, concluded that there is no clear evidence that hyperbaric oxygen is more beneficial than antibiotic therapy.[15]

Response to treatment is assessed by improvement of symptoms, imaging modalities like radionuclide bone scan. Inflammatory markers like ESR and CRP can be used to monitor disease progression. On an average, 6-8 weeks of medical therapy is required for complete remission of disease, but in some cases duration of treatment may exceed several months. In our study, average duration of treatment was 45 days.

The role of surgery in the treatment of MOE is restricted to biopsy to rule out malignancy, draining of microabscess, debridement of necrotic tissue and removal of sequestrum. Mastoidectomy can be done as part of debridement in extensive involvement.

Recurrence rate of MOE reported in literature is 15-20%.[16] In our study, one patient (9%) had recurrence four months after completion of treatment. No deaths occurred due to MOE.

CONCLUSION

To conclude, we audited case records of patients included in this study and the results corroborate with prior literature confirming again that MOE is disease affecting elderly males with Diabetes. During earlier years, radical surgical debridement was preferred due to extensive necrosis and uncontrolled spread of infection around temporal bone. The outcome of such surgical procedures was limited and some authors even reported that extensive surgical debridement may help in the spread of infections by exposing the healthy bone.[17] However the trend in management of MOE has shifted from surgical to medical management owing to the availability of parenteral antibiotics with good penetration, meticulous aural toilet, analgesics, topical steroids along with aggressive control of comorbid condition. Our study point out the significance of this changing trend towards conservative management with better outcome and less morbidity.

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