



A retrospective review of 14 cases of malignant otitis externa

Saldanha Marina^{*}, M.K. Goutham, A. Rajeshwary, Bhat Vadisha, T. Devika

Department of Otorhinolaryngology, K.S.Hegde Medical Academy, Nitte(deemed to be University), Mangalore, Karnataka, India

ARTICLE INFO

Article history:

Received 21 September 2018

Received in revised form

28 December 2018

Accepted 8 January 2019

Keywords:

Malignant otitis media

Granulation tissue

Pseudomonas aeruginosa

Diabetes

ABSTRACT

Background: Malignant otitis externa is an inflammatory condition of the external ear which has the propensity to spread to the skull base. It can be a difficult entity to treat as clinical presentation varies and response to treatment differs between patients. We reviewed cases of malignant otitis externa in our setup to document the epidemiology and outcome of management.

Methods: This is a retrospective case review observational study from January 2013–December 2017. Fourteen patients diagnosed with malignant otitis externa in our tertiary referral centre were included in the study. Based on hospital protocol, empiric treatment was started. After discharge, the patients follow up visits to the hospital were also documented.

Results: Otalgia was the most common symptom. Edema and congestion of the external auditory canal were observed in most cases. Diabetes was present in all patients. Three cases had associated facial palsy, and one patient had involvement of 7th, 9th, 10th, 11th and 12th cranial nerve. Two patients with facial palsy recovered. *Pseudomonas aeruginosa* was the most common organism isolated (50%).

Conclusions: In our series, malignant otitis externa invariably presented with severe otalgia. Lower cranial palsies were also seen. Methods to evaluate complete eradication of disease should be centered on clinical symptoms and signs, but the measurement of erythrocyte sedimentation rate or radiological imaging may be used as a useful adjunct when there is uncertainty.

© 2019 PLA General Hospital Department of Otolaryngology Head and Neck Surgery. Production and hosting by Elsevier (Singapore) Pte Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

Malignant otitis externa (MOE) is an infection of the external auditory canal which can spread to the mastoid process and skull base (Singh et al., 2005). Severe otalgia, purulent ear discharge are the common symptoms at presentation. On otoscopic examination, this condition has clinical signs similar to otitis externa; however, the presence of edema and granulations at the osseo-cartilaginous junction in the external auditory canal are more in favor of MOE (Ali et al., 2010). The ability of this disease to spread through the skull base increases its propensity to present with nerve palsies, the facial nerve being the most common nerve involved, followed by other cranial nerve palsies (IX, X, XII). This disease is mostly seen in patients with diabetes and immunocompromised status. The most

common organism isolated is *Pseudomonas aeruginosa* and the widely accepted empiric treatment based on studies is third-generation cephalosporins and fluoroquinolones (Singh et al., 2005; Franco-Vidal et al., 2007; Vadish et al., 2015). It has been noted that some patients do not respond to this treatment and in those cases, fungal MOE due to *Aspergillus* should be kept in mind and biopsy should be sent to rule out the same (Hasibi et al., 2017). Once the patient is symptomatically better, the otolaryngologist faces the dilemma as to when to stop the medications and prevent the chances of recurrence. In this aspect radionuclide scans, i.e., Gallium- 67- citrate (⁶⁷Ga) or 111Indium (¹¹¹In) play a significant role as they identify areas of residual infection and these scan return to normal when the infection subsides (Courson et al., 2014).

We aimed to evaluate the clinical presentation and response to treatment in patients who presented to our tertiary referral hospital with malignant otitis externa.

2. Materials and methods

A retrospective observational study from 2013 to 2017 conducted in a tertiary referral hospital. Ethical clearance was obtained

^{*} Corresponding author.

E-mail addresses: saldanhamarina@gmail.com (S. Marina), mkgoutham565@gmail.com (M.K. Goutham), rajeshwarisomayaji@gmail.com (A. Rajeshwary), bvadish@yahoo.co.in (B. Vadisha).

Peer review under responsibility of PLA General Hospital Department of Otolaryngology Head and Neck Surgery.

from the K.S.Hegde Institutional Ethics committee, Mangalore, India. Identifying information was not documented, and all the data recorded was kept confidential. The case records were reviewed, and 14 patients with malignant otitis externa were identified.

Data recorded was.

1. Time of presentation after the onset of symptoms
2. Presenting symptoms
3. Presenting signs
 - a. External auditory canal edema and congestion
 - b. Granulations
 - c. Tympanic membrane
4. Organism isolated
5. Treatment started according to the protocol
6. Imaging studies- High resolution Computed Tomography (HRCT) Temporal Bone
7. Erythrocyte sedimentation rate (ESR), Haemoglobin and HbA1c pre-treatment and ESR post-treatment.
8. Response to treatment was based on the reduction of pain and otoscopy/examination under microscope to examine for a decrease in canal wall edema, congestion, and granulations.

The treatment protocol in our hospital is to start with Injection Ceftazidime 1 gm thrice a day as the monotherapy and then depending on the culture sensitivity report we continue or add other antibiotics. After commencement of treatment, patients who showed improvement regarding the reduction of pain and decrease in ear discharge within two weeks were discharged. These patients were advised acetic acid ear drops and oral/intravenous antibiotics depending on the sensitivity report. They were called for follow up one week after discharge. Patients whose symptoms persisted, and depending on the extent of involvement, local debridement was done.

2.1. Statistical analysis

The data collected was entered into Microsoft Excel spreadsheet and analyzed using IBM SPSS Statistics Version 22 (Armonk, NY: IBM Corp). Frequency and percentage were calculated for the demographic profile, clinical and laboratory characteristics of the patients.

3. Results

Fourteen patients, thirteen males and one female with malignant otitis externa were included in the study. The ages ranged from 43 to 81 years (mean, 60 years).

3.1. Clinical presentation

Otalgia was the most common symptom at presentation. Patients presented within 20 days to 4 months after the onset of otalgia. Further details of the clinical symptoms and signs are shown in Table 1. Facial palsy was observed in three patients and one patient who was 81 years old presented with 7th, 9th, 10th, 11th and 12th cranial nerve palsy.

3.2. Associated co-morbidities

All patients had diabetes, and all presented with elevated blood sugar levels. Five patients had associated hypertension. One patient had hypertension, chronic kidney failure, and ischemic heart disease.

Table 1
Clinical presentation and investigations among patients with MOE.

Patient characteristics	Number (Frequency%)
Otalgia	14 (100)
Ear discharge	10 (71)
External ear granulations	8 (57)
External ear edema	12 (85)
Co-existing CSOM	3 (21)
Cranial nerve palsy	
7th nerve	3 (21)
7, 9, 10, 11, 12th nerve	1 (7)
Co-morbidities	
Diabetes mellitus	14 (100)
Hypertension	5 (35)
Ischemic heart disease	2 (14)
Chronic kidney disease	1 (7)
Laboratory investigations	
ESR (mm/hr)	
<20	4 (29)
20–40	6 (42)
>40	4 (29)
HbA1c (%)	
<7 (good control)	2 (14)
7–10 (poor control)	9 (64)
>10 (very poor)	3 (21)
Haemoglobin (g/dl)	
<10	1 (7)
>10	13 (93)

CSOM- Chronic suppurative otitis media, ESR- Erythrocyte Sedimentation Rate.

3.3. Microbiological report and laboratory investigations

Pseudomonas aeruginosa was the most common organism isolated (50%). No growth was seen in 29% cases (Fig. 1.)

Details of Erythrocyte sedimentation rate (ESR), Hb A1c and hemoglobin (Hb) at presentation has been shown in Table 1. The mean ESR was 55 mm/h, with a range of 12–88mm/hr.

Thirteen patients were diabetic with high sugar levels at presentation. Based on HbA1c, majority (64%) had poor diabetic control. Only one patient had low hemoglobin due to his chronic kidney disease.

3.4. Treatment protocol

On review of the antibiotic therapy based on the antimicrobial sensitivity report, 12 patients received monotherapy only (ceftazidime for 11, linezolid for one patient with methicillin-resistant *Staphylococcus aureus* (MRSA)). One patient with E-coli isolates, based on sensitivity report received a combination of Ciprofloxacin plus ceftazidime, with *Klebsiella* isolates received meropenem plus Ciprofloxacin. Gentamicin aural drops were used in four cases. Acetic acid ear drops (for two to three weeks) were

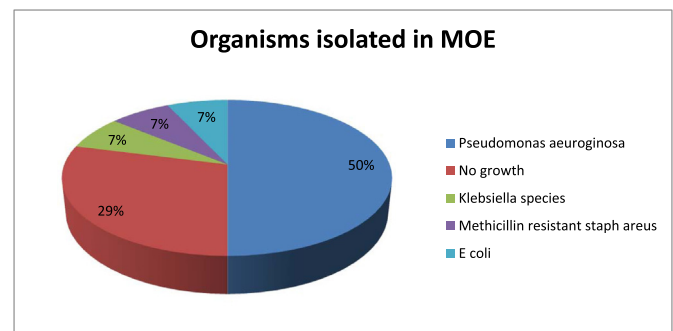


Fig. 1. Pie-chart depicting the organisms isolated from discharge in MOE.

given in all cases.

The treatment of duration varied but the mean duration observed was three weeks.

3.5. Response to treatment and follow up

After receiving treatment; eight (57%) patients had reduction in symptoms of otalgia and decrease in ear discharge within 10–14 days. They were discharged with the advice to continue acetic acid ear drops and antibiotics for one more week. They were called for follow up on week after discharge. One patient with multiple cranial nerve palsies had a hospital stay for two weeks and wanted discharge on request. He was lost to follow up. Four (29%) patients the response to treatment was poor. Hence, we did local debridement by removing the granulation tissue, necrotic tissue from the external auditory canal. Two patients improved after debridement. The pain persisted in two patients. Three patients had facial palsy with MOE, post-treatment the facial palsy completely recovered and their symptoms also improved within two weeks. One patient had persistent facial palsy after treatment.

Two patients came with other ear involvement after three months, and the affected side ear pain had subsided. Three patients came after nine months with repeat symptoms of MOE.

We could follow up six patients for 12 months. All six patient's symptoms subsided after four weeks of treatment, and for 12 months they were symptom free. ESR was done in six patients three weeks after treatment, and all showed a reduction in ESR.

4. Discussion

Malignant otitis externa is a difficult entity to treat as not only is the treatment required for a prolonged period but even the response to treatment has to be regularly monitored. Undiagnosed or partially treated MOE can progressively spread to the skull base and cause major complications such as thrombosis of lateral sinus or internal jugular vein, meningitis, Bezold's abscess and paralysis of cranial nerves (Singh et al., 2005; Nawas et al., 2013).

Similar to other studies (Courson et al., 2014; Hasibi et al., 2017), the most common presenting symptom was otalgia, which was associated with temporal and occipital headache. The presence of external auditory canal discharge, edema and granulations are all features of MOE which were present in the cases.

In our study, thirteen patients had uncontrolled diabetes. The risk of developing MOE in diabetes is more due to endarteritis, microangiopathy and small vessel obliteration (Carfrae et al., 2008). We reviewed the HbA1c status and observed that nine patients had poor control (67%).

ESR was more than 20 mm/h in 14 patients. ESR of 6 patients was repeated after three weeks, and it was noted that their ESR had come to normal. This co-related with the reduction in their symptoms and signs. This shows that ESR can be used as an adjunct to monitoring response to treatment especially in cases where repeat scan is not feasible (Rubin and Yu, 1988; Loh and Loh, 2013). HRCT temporal bone was done in all patients. Repeat HRCT was done in 4 patients after three weeks of therapy, and it showed reduced soft tissue intensity in the mastoid region. Due to non-availability of radioactive scans in our institute and the cost factor we did not do Gallium scan in our patients. Continuation of treatment was based on the response of patients to treatment and reduction of ear discharge, edema, and granulations.

Our relapse rate was 21% which is similar to other studies (Singh et al., 2005). In comparison, 2.6% was the relapse rate in another study (Hasibi et al., 2017) and in which patients who did not respond with antibacterials were switched to antifungals and they showed a favourable response.

In our series three patients (21%) presented with facial nerve palsy and two showed complete resolution of the palsy, one had persistent facial palsy. Studies have shown that facial nerve palsy is significantly less likely to improve after treatment, but the presence of cranial nerve involvement does not affect the overall prognosis (Mani et al., 2007).

Medical line of management forms the mainstay of treatment (Ridder et al., 2015). Third generation cephalosporins, intravenous ceftazidime, fluoroquinolone, carbapenems are commonly used drugs (Loh and Loh, 2013). In cases of negative culture report, studies have shown that empiric treatment with the above medication showed positive results. In our institute, we also follow the same protocol and 57% of the patients showed a favourable response within two weeks. The clinical challenge for the clinician is to decide when to stop treatment. Due to cost factor of radio-nuclide scan to monitor follow up, the protocol in our institution is to continue the antibiotics for a further two weeks and depending on the response of the patient further management is decided.

The limitation of our study was that the sample size was small and in patients who did not have a favorable response to treatment we did not send tissue biopsy specimens for fungal culture.

5. Conclusions

In our series, malignant otitis externa invariably presented with severe otalgia. Edema with congestion of the external auditory canal and lower cranial palsies were also seen. Diabetes as a predisposing factor was observed in all cases. Methods to evaluate complete eradication of disease should be centered on clinical symptoms and signs, but the measurement of erythrocyte sedimentation rate or radiological imaging may be used as a useful adjunct when there is uncertainty.

Declarations

Funding

No funding

Conflict of interest

No conflict of interest.

Ethical approval

Yes.

Acknowledgements

Nil.

References

- Ali, T., Meadi, K., Anari, S., ElBadawey, M.R., Zammit-Maempel, I., 2010. Malignant otitis externa: case series. *J. Laryngol. Otol.* 124 (8), 846–851.
- Carfrae, M.J., Kesser, B.W., 2008. Malignant otitis externa. *Otolaryngol. Clin. North Am.* 41, 537–549.
- Courson, A.M., Vikram, H.R., Barrs, D.M., 2014. What are the criteria for terminating treatment for necrotizing (malignant) otitis externa? *Laryngoscope* 124 (2), 361–362.
- Franco-Vidal, V., Blanchet, H., Bebear, C., Dutronc, H., Darrouzet, V., 2007. Necrotizing external otitis: a report of 46 cases. *Otol. Neurotol.* 28 (6), 771–773.
- Hasibi, M., Ashtiani, M.K., Motassadi, M.Z., Yazdani, N., Borghei, P., Kuhi, A., et al., 2017. A treatment protocol for management of bacterial and fungal malignant external otitis: a large cohort in Tehran, Iran. *Ann. Otol. Rhinol. Laryngol.* 126 (7), 561–567.
- Loh, S., Loh, W.S., 2013. Malignant otitis externa an Asian perspective on treatment outcomes and prognostic factors. *Otolaryngol. Head Neck Surg.* 148 (6),

- 991–996.
- Mani, N., Sudhoff, H., Rajagopal, S., Moffar, D., Axon, P.R., 2007. Cranial nerve involvement in malignant external otitis: implications for clinical outcome. *Laryngoscope* 117 (5), 907–910.
- Nawas, M.T., Daruwalla, V.J., Spierer, D., Micco, A.G., Nemeth, A.J., 2013. Complicated necrotizing otitis externa. *Am. J. Otolaryngol.* 34 (6), 706–709.
- Ridder, G.J., Breunig, C., Kaminsky, J., Pfeiffer, J., 2015. Central skull base osteomyelitis: new insights and implications for diagnosis and treatment. *Eur. Arch. Oto-Rhino-Laryngol.* 272 (5), 1269–1276.
- Rubin, J., Yu, V.L., 1988. Malignant external otitis: insights into pathogenesis, clinical manifestations, diagnosis and therapy. *Am. J. Med.* 85 (3), 391–398.
- Singh, A., Al Khabori, M., Hyder, M.J., 2005. Skull base osteomyelitis: diagnostic and therapeutic challenges in atypical presentation. *Otolaryngol. Head Neck Surg.* 133 (1), 121–125.
- Vadish, B., Ajaz, A., Satheesh, K.B., Rajeshwary, A., Srinath, D.P.K., Marina, S., 2015. Malignant otitis externa—a retrospective study of 15 patients treated in a tertiary healthcare center. *J. Int. Adv. Otol.* 11 (1), 72–76.