

## Malignant otitis externa in an immunocompetent infant- A case report and review of literature

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### Abstract

In infants, malignant otitis externa presents extremely rarely with only nine cases reported till date in the world medical literature. This clinical record documents the first case of the malignant otitis externa in an immunocompetent 3 month old female infant, who was treated successfully with intravenous antibiotics and local debridement. In addition this case report also reviews the scant medical literature on the cited disease.

**Keywords:** Malignant otitis externa, Infant.

### Introduction

Malignant otitis externa (MOE) is a life threatening, progressive bacterial infection of the external auditory canal (EAC), mastoid and skull base with multiple cranial nerve palsies, with a mortality of 30–80%. It is most commonly caused by *Pseudomonas aeruginosa* and primarily affects elderly diabetics and HIV patients.<sup>(1)</sup> Children are rarely affected and a state of immunocompromised is generally associated in these cases. Only nine cases of malignant otitis externa in infants have been described in the literature till date.<sup>(1)</sup> The earliest reported age is one month. With this background we present a 3 month old female child who was diagnosed with malignant otitis externa and successfully treated.

### Case Report

A three month old female baby, resident of New Delhi, was referred to our hospital by a local doctor with chief complaints of fever and left side otorrhoea for 4 days and decreased urine output for 2 days. Ear discharge was profuse, purulent, and greenish and blood stained. On general physical examination she was well nourished (weight – 6 kg), the child had left sided facial weakness and deviation of angle of mouth to the right on crying and incomplete eye closure (Grade V facial palsy). On local ENT examination, pinna was normal; ulceration, necrosis and blackening were present in the left external auditory canal. The skin was macerated, bone partially exposed and granulation tissue was present with foul smelling discharge, tympanic membrane was not visible and post auricular and preauricular regions were normal. Right ear examination was normal. Her blood investigations showed anemia (Haemoglobin–8.1 gm/dl) with total leucocyte count- $6.8 \times 10^3$ ,<sup>(3)</sup> platelet count-68000, and ESR-14 mm/h. The blood sugar was normal. Aural swab was sent for culture sensitivity which showed no growth and empirically piperacillin was started. High

resolution CT scan of temporal bone revealed fluid/soft tissue thickening involving left external auditory canal, middle ear and ill defined focal bony destruction involving mastoid and petrous temporal bone. Ossicles were normal. The patient responded well to treatment with antibiotic, local debridement of lesion and regular dressings. However, facial paralysis improved slightly but persisted.

### Discussion

Malignant otitis externa is a necrotising form of otitis externa which gets initiated from the external auditory meatus and then spreads into the surrounding tissues leading to various cranial nerve palsies and even can lead on to death. Typical clinical presentation of malignant otitis externa is elderly diabetic patient with severe, unremitting otalgia, aural fullness, otorrhea, and hearing loss.<sup>(2)</sup>

The diagnosis of MOE is based upon typical clinical presentation and examination of the ear which shows oedema of external auditory canal with profuse discharge and granulations in the floor of external auditory canal and its bone erosion. Laboratory and imaging studies generally support the diagnosis. HRCT is sensitive in diagnosing and detecting involvement of the skull base. Most common organism involved in these patients is *pseudomonas aeruginosa*. Other bacteria, including *staphylococcus aureus*, *S epidermidis*, *proteus mirabilis*, *klebsiella* and fungus (*Aspergillus fumigates*), have also been reported.

Patients having HIV-associated MOE are younger than the average patient who has diabetes associated MOE. Fungal is the most common causative agent in patients who have HIV than in diabetic patients, particularly patients with severe AIDS.<sup>(1)</sup> Although rare, MOE has been reported in children who have immunocompromised states, including IgG subclass deficiency, IgA deficiency, acute monocytic leukemia, iatrogenic neutropenia secondary to induction

chemotherapy for acute lymphoblastic leukaemia, and bone marrow transplantation.<sup>(3,4)</sup> Compared with adults, diabetes is not as common a comorbidity in children (21% in one review) as are other immunocompromised states.

Among children, Infants are rarely affected with MOE. Till date only 9 cases of MOE have been reported in the English medical literature (Table I). The course of MOE in children has an acute onset, with more toxic initial symptoms of fever, malaise, and leucocytosis. One review reported a lower incidence of facial nerve paralysis in children. But another study cited a higher incidence because of the less-developed mastoid process and closer proximity of the facial nerve and stylomastoid foramen to the EAC. Facial paralysis has no prognostic significance to overall recovery, because both studies agree that children generally have a more favourable prognosis regarding mortality than adults. The prognosis for facial nerve recovery in children is poorer if the patient presents with the palsy.

MOE in infants is generally associated with some immunocompromise. The diagnosis of MOE is made on clinical basis in both adults as well as infants. Most of the cases of infants are also caused by pseudomonas aeruginosa as has been seen in adults but causative agent is different in AIDS patients (mostly fungal).

If we analyse all the cases of infantile MOE compared to the adults, this disease entity seems to be representing a different spectrum of the same disease as the point raised by Robert Mills in 1986.<sup>5</sup> As more and more number of cases is being diagnosed, we may get a

clear picture of this disease entity in infants. Association of MOE with high mortality and morbidity always necessitates a high index of clinical suspicion and early intervention for successful management of these cases whether in infants, children or adults.

The present case is only the second case of MOE in infants which was immunocompetent and youngest ever immunocompetent child with MOE with negative culture, hitherto unreported in medical literature.

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#### Author's contributions

Dr. Arunabha Chakravarty contributed towards clinical management of the patient; conceptualization and revision of the manuscript. Dr. Sunil Garg contributed towards the literature search and drafting the manuscript. Dr. Sunil Garg also helped in clinical management and revision of the manuscript.

#### Declaration of the conflicting interests

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#### Ethical Approval

Consent was taken from parents of the children for surgery and publication of the case report.

**Table 1: Synopsis: Malignant otitis externa cases in infants**

Sr no.	Author/Year	Age	Sex	Immune Status	Organism	Facial Paralysis
1	Coser/1974 <sup>6</sup>	2 months	Not known	Unknown	pseudomonas aeruginosa	Recovered/Permanent
2	Joachims/1976 <sup>3</sup>	7 months	M	Anemia	pseudomonas aeruginosa	Recovered/Permanent
3	Coser/1980 <sup>2</sup>	5 month	M	Anemia/malnourished	pseudomonas aeruginosa	Recovered/No
4	Coser/1980 <sup>2</sup>	6 months	M	Anemia/malnourished	Proteus mirabilis	Recovered/No
5	Rubinstein/1980 <sup>7</sup>	4 months	M	Neuroblastoma (on chemotherapy)	pseudomonas aeruginosa	Recovered/No
6	Nir/1990 <sup>8</sup>	3 month	F	Granulocytopenia	pseudomonas aeruginosa/proteus rettgeri	Recovered/No
7	Paul/2001 <sup>4</sup>	1 month	F	IgA deficiency	pseudomonas aeruginosa	Recovered/Permanent
8	Patigaroo/2009 <sup>9</sup>	5 month	Not known	Transient Neutropenia	pseudomonas aeruginosa	Recovered/Permanent
9	Bains/2010 <sup>10</sup>	10 month	F	Well nourished	Enterobacter	Recovered/No

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