

## Short Communication

# Otitis externa due to *Stenotrophomonas maltophilia* in an immunocompetent patient: Case report

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***Stenotrophomonas maltophilia* is a gram negative bacterium that primarily affects immunocompromised and clinically debilitated patients. It is known to affect many tissues and organ systems of the body, but it rarely causes otitis externa. We reported a case of otitis externa due to *S. maltophilia* in a 22-year-old immunocompromised male who had bilateral otalgia and ear discharge, as well as left facial nerve palsy in combination with hearing loss. Further investigations revealed *S. maltophilia* in ear drainage cultures, including a deep swab taken during surgical debridement. Systemic antibiotics in combination with local treatment were effective in controlling the infection and resulted in the resolution of his signs and symptoms, except for the hearing loss. We believe that early identification of the pathogen and treatment of the infection might lead to a favorable outcome.**

**Key words:** *Stenotrophomonas maltophilia*, opportunistic infection, otitis externa.

## INTRODUCTION

*Stenotrophomonas maltophilia* (previously *Pseudomonas* and *Xanthomonas maltophilia*) is a ubiquitous, aerobic, non-fermentative, gram-negative bacillus (Denton and Kerr, 1998) that is known to colonize the respiratory tract, wound, urinary tract, catheter tips, blood, and intra-abdominal fluids in hospitalized patients (del Toro et al., 2002). Even though the bacterium was formerly thought to have low pathogenicity, recent reports show that it can cause bacteremia and other serious infections (Nseir et al., 2006), especially in immunocompromised hosts (Senol et al., 2002). More so, infection with this pathogen is associated with considerable mortality in severely immunocompromised and clinically debilitated patients (Senol, 2004).

Several cases of *S. maltophilia* infection have also

been described in immunocompetent patients (Nseir et al., 2006; Thomas et al., 2010). While the bacterium is known to cause a wide range of infections in both immunocompetent and immunocompromised individuals, there are seldom any reports on cases of otitis externa (OE) caused by this pathogen. A single report describes a case of OE due to *S. maltophilia* in a patient, who had chronic lymphocytic leukemia (Börner et al., 2003). We present a case of OE caused by *S. maltophilia* in an immunocompetent patient, which to the best of our knowledge is the first report of OE due to this pathogen in an immunocompetent individual.

## CASE PRESENTATION

A 22-year-old male presented to the Emergency Department at King Abdul-Aziz University Hospital with a two-week history of bilateral ear discharge that was purulent, foul smelling and bloody. It was associated with bilateral otalgia, decreased hearing, left facial weakness,

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and mild vertigo. There was no history of fever, tinnitus, nausea or vomiting.

The patient had no history of recent antibiotic intake or trauma. There was no history of diabetes or known medical illness, and his family history was unremarkable.

On examination the patient had a normal temperature, pulse, respiratory rate and blood pressure. He was conscious, oriented, and he had a pain score of 4 out of 10. There was facial asymmetry with grade V weakness of the left facial nerve. Examination of both external auditory canals showed necrosis and sloughing of the mucosa with yellowish discharge; there was no mastoid tenderness. On otoscopy, both external auditory canals were hyperemic and there was bilateral tympanic membrane perforation. Examination of the nose, nasopharynx and throat was unremarkable. Fiber optic examination of the larynx showed bilateral mobile vocal cords. Pure tone audiogram showed mild to moderate mixed type hearing loss.

A provisional diagnosis was made for bacterial OE. Differential diagnoses included viral and fungal OE. Laboratory investigations revealed the following: white blood cell count 19.1 K/uL (reference range 4.5 to 11.5 K/ $\mu$ L) with 87% neutrophils (reference range 50 to 70%); C-reactive protein 12.30 mg/L (reference range 0 to 3 mg/L); random blood glucose 7.0 mmol/L (reference range 3.9 to 6.7). Urinalysis, serum electrolytes, liver function tests, and peripheral blood film were normal. Human immunodeficiency virus (HIV) serology, hepatitis serology, and varicella zoster virus IgG were all negative. Serological assays for herpes simplex virus 1 and 2 (HSV-1 and HSV-2) IgM were negative; the results were weakly positive for HSV-1 and HSV-2 IgG. Anti-nuclear antibodies yielded a titer of 1:80 (mildly positive). Cytoplasmic anti-neutrophil cytoplasmic antibodies were negative. Complement and immunoglobulin levels were both normal. Computed tomography (CT) scan of the temporal bone showed bilateral soft tissue filling of the anterolateral parts of the middle ear with normal facial nerve course and appearance. Brain CT and magnetic resonance imaging (MRI) were unremarkable with no intracranial extension of the infection.

Ear drainage cultures were done, and the patient was admitted for surgical debridement. Deep swabs were taken during surgery and sent for culture and histopathology. Treatment was initiated empirically with per oral ciprofloxacin and metronidazole. Per oral acyclovir and prednisolone were also included in his treatment.

*S. maltophilia* was isolated from the specimens, and susceptibility testing showed the species was susceptible to trimethoprim/sulfamethoxazole and piperacillin/tazobactam. Acid fast bacilli (AFB), fungal and viral cultures yielded negative results, and histopathology revealed only necrotic tissue. A definitive diagnosis of OE due to *S. maltophilia* was made. The patient's antibiotics

were changed to intravenous piperacillin/tazobactam based on susceptibility tests, and acyclovir was discontinued. Daily cleansing of both ear canals was performed followed by application of topical dexamethasone and gentamicin. There was a remarkable improvement in his signs and symptoms within three weeks of initiating treatment, but he, unfortunately, developed drug-induced hepatitis, warranting the cessation of piperacillin/tazobactam. He was started on intravenous tigecycline, which was prescribed for a period of three weeks. He also developed adrenal insufficiency during the course of treatment, necessitating the tapering of prednisolone. By the end of the third week, the ear discharge and facial paralysis had resolved; however, he had sequelae in the form of hearing loss.

## DISCUSSION

*S. maltophilia* is emerging as a known cause of infection in the nosocomial setting (Metan and Uzun, 2005). The risk factors that have been reportedly associated with this pathogen include intensive care admission, prior use of broad-spectrum antibiotics, mechanical ventilation, malignancy, cystic fibrosis, neutropenia, presence of central venous catheters, prolonged hospitalisation, debilitation and immunodeficiency (Denton and Kerr, 1998). Community-acquired infections due to *S. maltophilia* have also been reported in the literature (Falagas et al., 2009). In the study conducted by Falagas et al. (2009) it was found that malignancy, HIV infection and prior hospitalization were the most common comorbidities in immunocompetent patients who developed *S. maltophilia* infections. The patient in our case did not have any of these risk factors or a serious comorbid condition.

*S. maltophilia* is a frequent colonizer of human tissues and hospital equipment (del Toro et al., 2002; Di Bonaventura et al., 2004). Clinicians may be faced with a difficulty when they encounter positive cultures of *S. maltophilia* because it is hard to differentiate colonizers from pathogens. While there is evidence that delayed effective treatment for *S. maltophilia* is a risk factor of infection-attributed mortality in patients with positive cultures of the pathogen (Kwa et al., 2008), clinicians have to bear in mind that indiscriminate treatment of all positive cultures may result in overuse of antibiotics and the emergence of resistant strains. In our case, the decision to treat was based on his clinical presentation, repeated negative cultures for AFB, viruses and fungi and the fact that *S. maltophilia* was isolated after multiple ear drainage cultures, including a deep one. Given that our patient was weakly positive for HSV-1 and HSV-2 IgG and the fact that viruses are a frequent cause of OE, we also started him on acyclovir. However, we interrupted

the treatment when viral cultures of the ear discharge were negative.

In general, the antibiotic options for the treatment of *S. maltophilia* infections are limited because the bacterium exhibits resistance to many commonly prescribed antibiotics, including beta-lactam agents, carbapenems and aminoglycosides (Nicodemo and Paez, 2007). Selection of the appropriate antimicrobial agent is crucial in improving the outcome of *S. maltophilia* infections, especially in patients who develop bacteremia (Metan and Uzun, 2005). According to recent reports, trimethoprim/sulfamethoxazole is the most effective antimicrobial agent in the treatment of *S. maltophilia* infections. Ciprofloxacin, ceftazidime or ceftriaxone, and ticarcillin/clavulanate, alone or in combination with other antibiotics, may be considered as alternative options when co-trimoxazole cannot be used (Di Bonaventura et al., 2004; Falagas et al., 2008). Our choice of antibiotics in this case was guided by the results of susceptibility studies. The administration of piperacillin/tazobactam in our patient produced a marked improvement in his condition, but the medication was discontinued when he developed adverse effects. Tigecycline, which was prescribed as a substitute for piperacillin/tazobactam, proved effective in controlling the infection in our case. Tigecycline has been shown to have a high activity against *S. maltophilia* in *in vitro* studies conducted by Betriu et al. (2002), and it was proposed as an additional agent in the treatment of infections caused by this pathogen.

The mechanism underlying the development of hearing loss in our patient is unclear. In addition to the fact that there are no reports in the literature that cite hearing loss as a result of the direct effect of *S. maltophilia*, the antibiotics that we used in the treatment of the infection in this patient are not known to have significant adverse effects on hearing. We cannot, however, exclude the possibility that other factors might have contributed to the development of impaired hearing in our patient. Hence, it is possible that there is still more to learn about the natural history of this pathogen and its impact on hearing.

## Conclusion

Otitis externa due to *S. maltophilia* is a rare finding in immunocompetent individuals. Multiple ear drainage cultures and susceptibility tests can be of invaluable help in establishing the diagnosis and in choosing the most appropriate antibiotic treatment for patients. In addition, early identification of the pathogen and treatment of the infection might lead to a favorable outcome.

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