# *Stenotrophomonas maltophilia* CSF infection in infants after neurosurgery

## Introduction

Two infants with recently treated hydrocephalus (by ventriculoperitoneal shunt) re-presented with meningitis. CSF culture confirmed *Stenotrophomonas maltophilia* infection in both cases. They were successfully treated with removal of the shunt, insertion of an external ventricular drain and completion of a course of trimethoprim and sulfamethoxazole antibiotic therapy (also known as co-trimoxazole).

## Discussion

*S. maltophilia* meningitis is extremely rare with only 30 cases previously reported. Predisposing risk factors include prematurity, intracranial haemorrhage, malignancy, neurosurgical intervention, and recent carbapenem treatment (Sanyal and Mokaddas, 1999; Caylan et al, 2002). This article describes the third and fourth paediatric cases to date of *S. maltophilia* meningitis, in whom previous ventriculoperitoneal shunt insertion and initial carbapenem therapy (both cases) and prematurity (one case) were risk factors.

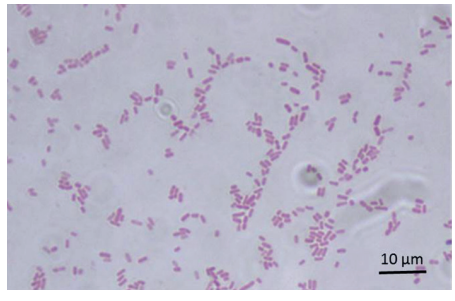
Unusually, these two cases did not manifest in the immediate postoperative period in hospital but rather in the community several weeks after surgery. In contrast, the two previously reported paediatric cases occurred within the first 3 weeks following external ventricular drain insertion as inpatients (Rojas et al, 2009; Sood et al, 2013).

*S. maltophilia* is resistant to several antibiotics used empirically for nosocomial infections. Inducible beta-lactamase activity, efflux pumps and extracellular glycocalyx formation are cited resistance mechanisms (Nicodemo and Paez, 2007). Co-trimoxazole is the empirical and definitive choice for clinically suspected and culture-proven infections respectively, exhibiting more than 90% susceptibility (Falagas et al, 2008).

In both the current cases, the British Society for Antimicrobial Chemotherapy method for antimicrobial susceptibility testing was used with a reported minimal inhibitory concentration of 4 mg/litre (Wheat, 2001). Antibiotic combinations with ticarcillin/clavulanate or ciprofloxacin may combat resistance with clinical success rates ranging from 67% to 100% (Vartivarian et al, 1994; Carroll et al, 1998; Zelenitsky et al, 2005). While the 21-day course was completed to ensure full treatment, external ventricular drain sampling in both cases demonstrated that 18 days of antimicrobial therapy achieved suppression of CSF bacterial growth. This may be considered in future as longer bacterial viability may be of concern.

Overall mortality of *S. maltophilia* meningitis is 17% (5/30), reflecting the microorganism's pathogenicity. Both the current cases had a successful outcome with the timely commencement and completion of a 21-day course of co-trimoxazole following microbiology consultation. BJHM

Figure 1. Gram stain of CSF from Case 1, showing evenly stained Gram-negative straight, or slightly curved, rods of *Stenotrophomonas maltophilia*.

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### CASE REPORT 1

A 10-week-old boy who was born at term and who had previously undergone placement of a ventriculoperitoneal shunt for post-infective hydrocephalus (raised white cell count on lumbar puncture but no organisms detected on Gram stain or culture) re-presented 6 weeks after the procedure with irritability, drowsiness and pyrexia of 39°C. Peripheral blood tests showed a mildly raised white cell count at $17x10^6$/litre and mildly raised C-reactive protein level at 21 mg/litre.

A shunt tap suggested CSF infection with a raised white cell count at $643x10^6$/litre (differential count 89% neutrophils, 11% lymphocytes), raised protein count at 0.55 g/litre, and low glucose level at 1.7 mmol/litre, although no organisms were detected on Gram staining. His shunt was removed and replaced with an external ventricular drain, and he was initially started on empirical intravenous meropenem and vancomycin antibiotic therapy.

Peripheral blood cultures, CSF and the shunt tip all grew Gram-negative bacilli (*Figure 1*) and so on advice from the microbiology consultant, intrathecal gentamicin was added. The final cultures grew *Stenotrophomonas maltophilia* sensitive to co-trimoxazole. He was started on intravenous co-trimoxazole at 14 mg trimethoprim/70 mg sulfamethoxazole (the baby weighed 5.5 kg) and the meropenem and gentamicin were discontinued.

As per the authors' institution's practice, immediately following the full course (21 days) of co-trimoxazole treatment together with normal CSF analysis (CSF white cell count $2x10^6$/litre) and negative cultures, a new ventriculoperitoneal shunt was inserted. He made a full recovery, with no relapse of infection and no neurological or neuro-developmental deficit at 6-month follow up.

## CASE REPORT 2

An ex-premature boy (born at 32 weeks' gestation) underwent insertion of a ventriculoperitoneal shunt for post-haemorrhagic hydrocephalus secondary to intraventricular haemorrhage. He re-presented 2 months later at a corrected age of 12 weeks with poor feeding, lethargy and pyrexia of 38.6°C.

Peripheral blood tests showed mildly raised white cell count at $15x10^6$/litre and mildly raised C-reactive protein level at 12 mg/litre. His shunt was tapped and CSF analysis revealed an elevated white cell count at $16x10^6$/litre (differential count 82% neutrophils, 18% lymphocytes), raised protein count at 0.49 g/litre, slightly reduced glucose at 2.0 mmol/litre and Gram-negative bacilli were seen on Gram staining. His shunt was removed, an external ventricular drain inserted, and he was commenced on intravenous vancomycin and meropenem antibiotic therapy.

The cultures from the CSF confirmed *Stenotrophomonas maltophilia* infection sensitive to co-trimoxazole and so co-trimoxazole at a dose of 12.5 mg trimethoprim/65 mg sulfamethoxazole (the baby weighed 5.0 kg) was instigated for a total of 3 weeks, and vancomycin and meropenem were discontinued. Immediately after completion of antibiotics with a repeat normal CSF analysis, a new ventriculoperitoneal shunt was inserted. He also made a full recovery with no relapse of infection and no neurological deficit at 6-month follow up.

## LEARNING POINTS

- *Stenotrophomonas maltophilia* meningitis is extremely rare with only 30 previously reported cases in the literature, and is rarer still in children.
- Despite its rarity, *S. maltophilia* meningitis can complicate neurosurgery in infants as well as adults.
- The severe clinical course and high mortality rate mandate consideration of *S. maltophilia* in the differential diagnosis of meningitis in patients with predisposing factors and who do not respond to empirical treatment.
- When *S. maltophilia* is cultured, antibiotic treatment should initially consist of co-trimoxazole. Treatment must be adjusted according to antimicrobial sensitivities to ensure resolution of infection since resistance to co-trimoxazole is an increasing problem.


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