

Stenotrophomonas maltophilia as a rare cause of meningitis and ventriculoperitoneal shunt infection

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Abstract

Stenotrophomonas maltophilia is an extremely rare pathogen responsible for ventriculoperitoneal shunt infection and meningitis. This young female patient with history of multiple shunt revisions in the past, came to us with shunt dysfunction and exposure of the ventriculoperitoneal shunt tube in the neck. The abdominal end of the shunt tube was seen migrating into the bowel during shunt revision. The cerebrospinal fluid analysis showed evidence of *Stenotrophomonas maltophilia* growth. This is the first reported case of *Stenotrophomonas maltophilia* meningitis associated with ventriculoperitoneal shunt migration into the bowel.

INTRODUCTION

Stenotrophomonas maltophilia (*S. maltophilia*), is a Gram-negative bacterium that has rarely been reported to cause meningitis [1, 2] and ventriculoperitoneal (VP) shunt infections [1]. We report a case of VP shunt infection and meningitis caused by *S. maltophilia* in a 24-year-old female who presented with VP shunt dysfunction and migration of the distal end of the shunt tube into the bowel. So far only 35 cases of *S. maltophilia* meningitis have been reported in the literature [1, 3–9]. The intention of this article is to present this rare case and to briefly review the relevant literature.

CASE REPORT

A 24-year-old female was referred to our hospital from another hospital where she presented with fever, headache and exposure of the shunt tube at the neck through a skin erosion. Considering the possibility of meningitis, the shunt tube was externalized at its cranial end and connected to an external ventricular drainage (EVD). Cerebrospinal fluid (CSF) study was suggestive of pyogenic meningitis. The patient was started on injection ceftriaxone and vancomycin empirically as culture reports were pending.

The patient's past history was significant for congenital hydrocephalus for which she underwent VP shunt at the age of 6 months. Since then, she has had multiple shunt revisions at the ages of 8, 15 and 23 at different hospitals, details of which were unavailable.

On admission to our hospital, the patient had no neurological deficits. The shunt tube was seen exposed through an erosion in the neck. The cranial end of the shunt tube was connected to an EVD.

The empirical antibiotics were continued. We then performed an endoscopic third ventriculostomy (ETV). During ETV, the ventricular catheter of the VP shunt was visualized with the aid of a flexible endoscope. The catheter was seen surrounded by a tuft of choroid plexus and hence it was retained. The distal end of the shunt tube was stuck within the abdomen. Laparoscopic visualization of the abdomen by gastro surgeons showed that the abdominal end of the shunt tube had penetrated the jejunal wall with a well-formed track covering the tube from the anterior abdominal wall to the jejunum (Fig. 1). After removing the shunt tube, the bowel opening was surgically closed.

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Abbreviations: CNS, central nervous system; CSF, cerebrospinal fluid; CT, computed tomography; ETV, endoscopic third ventriculostomy; EVD, external ventricular drainage; LV, left ventricle; SC, stress cardiomyopathy; *S. maltophilia*, *Stenotrophomonas maltophilia*; TMP/SMX, trimethoprim/sulfamethoxazole; VP, ventriculoperitoneal.

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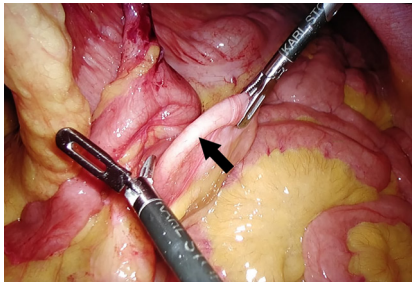


Fig. 1. Showing the abdominal end of the VP shunt tube (black arrow) entering the jejunum.

Her CSF culture sent from the first hospital grew *S. maltophilia*, sensitive to levofloxacin, minocycline and trimethoprim/sulfamethoxazole (TMP/SMX). Hence, the empirical antibiotics were stopped and she was started on TMP/SMX. About a week after the ETV, she developed severe headache. Computed tomography (CT) brain showed worsening of hydrocephalus, indicating failed ETV. After an emergency right frontal EVD, she was continued on TMP/SMX for 10 days.

After obtaining a sterile CSF culture, she was taken up for left VP shunt. After completion of the VP shunt, she developed bradycardia and cardiac arrest, while on the operating table. After 15 min of resuscitation, she was revived. Postoperative CT brain showed satisfactory placement of the shunt tube. The electrocardiogram done showed ST-T changes in a non-territorial distribution of coronary arteries and prolonged QTc. The echocardiogram showed hypercontractility in the basal aspects of the left ventricle and akinetic mid, distal and apical segments with overall reduced left ventricular (LV) ejection fraction. Cardiologists made a diagnosis of stress cardiomyopathy (SC). She was electively ventilated for 2 days and extubated without any neurological deficits. On subsequent echocardiograms her LV ejection fraction normalized.

Patient was discharged without any deficits. During follow-up patient developed an allergic reaction to TMP/SMX and hence the drug was changed to minocycline to complete an 8 week antibiotic therapy. She remained asymptomatic at 18 months follow-up.

DISCUSSION

It is ubiquitous in the environment and notorious for producing serious multidrug resistant opportunistic infections in humans [10]. The organism is found in aqueous habitats, plants, animals, food and water sources [10]. It is known to produce polymicrobial infections commonly in the respiratory tract in patients with cystic fibrosis as a cocolonizer with *Pseudomonas aeruginosa* [10]. Infection by this organism is currently a major health concern due to its presence in nonclinical environments, increased incidence of community acquired infections and its ability to spread in a clinical setting.

S. maltophilia very seldomly causes central nervous system (CNS) infections like meningitis [1, 2] and meningoencephalitis [6]. *S. maltophilia* meningitis is believed to be acquired secondarily to neurosurgical interventions [1], long hospital stays [11], prolonged intensive care unit exposures [11] and presence of intravascular or intracranial devices [1]. Our case had two of these risk factors as (i) she underwent multiple ventriculoperitoneal shunt surgeries in the past and (ii) had intracranial devices for prolonged period.

Khanum *et al.* reviewed 30 cases of *S. maltophilia* meningitis in 2020 [5]. In addition to this we could identify five more cases of *S. maltophilia* meningitis [4, 6, 8, 9]. The age of the 35 previously reported patients ranged from preterm to 73 years with a M:F ratio of 24:11. Among these patients, prior neurosurgical interventions were performed in 28 (80%) and placement of an intracranial implant was done in 22 (63%). There were a total of 24 implants in these 22 patients. These include ten (42%) EVD, nine (37%) VP shunt, three (13%) ommaya reservoir, one (4%) aqueduct stent and one (4%) aneurysm clip [1, 3–9]. Of the 35 cases reported six (17%) patients succumbed to death.

The ability of the organism to change trait together with virulence associated factors have made them difficult to treat pathogens [2]. The organism has the ability to affect both the immunocompromised as well as immunocompetent humans making it both an opportunistic and also a true pathogen [2]. It is known to produce respiratory tract infections, urinary tract infections, surgical site infections, ophthalmic infections, septic shock and medical implant infections in immunocompromised individuals [1]. Pathogenesis of *S. maltophilia* is by colonization, rather than infection on implants [12]. Presence of shunts and tubes for a prolonged period of time allows formation of a biofilm around them [2]. The ability of the organism to produce various beta-lactamase enzymes, biofilm formation and presence of natural multidrug resistance efflux systems contribute to its potential to develop multidrug resistance [2].

TMP/SMX is the most frequently used antibiotic followed by quinolones in *S. maltophilia* infections [1]. Chung *et al.* documented higher *in vitro* susceptibility of *S. maltophilia* to minocycline compared to TMP/SMX [13]. In severe cases intraventricular colistin has also been tried [6]. In literature there is no consensus regarding the exact duration of treatment. We gave an 8 weeks course of antibiotics with TPM/SMX and minocycline.

The most common neurological condition associated with SC is subarachnoid haemorrhage [14]. Other less common causes are stroke, seizures, encephalitis, meningitis and head trauma [14]. The exact cause of SC in our patient is not known.

The authors postulate that the possible risk factors of *S. maltophilia* meningitis in the present case are (i) faecal contamination of the distal end of the shunt tube that perforated the bowel (ii) contamination of the exposed shunt tube at the neck either from the skin or external environment and (iii)

performance of multiple shunt revisions in the past. However, we accept that none of these could be confirmed.

In conclusion, although the literature on *S. maltophilia* infection has increased in the past decade, the disease still remains a rare cause of CNS infection. We aim to discuss the methods employed by our team in combating this rare infection and to instil the importance of high clinical suspicion towards the *S. maltophilia* in a patient who had undergone multiple neurosurgical procedures. Early recognition and appropriate management can reduce morbidity and mortality.

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Author contributions

All of the authors have equally contributed in the conceptualization, preparation and verification of the manuscript.

Conflicts of interest

The authors declare that there are no conflicts of interest.

Ethical statement

The study followed the ethical standards of the institution and patient confidentiality was kept. The consent was obtained.

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