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#### CASE REPORT

# *Stenotrophomonas maltophilia*-associated odontogenic cerebral abscess in an immunocompetent patient: A case report

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### **Key Clinical Message**

*Stenotrophomonas maltophilia* can cause rare odontogenic brain abscesses in immunocompetent patients, highlighting the importance of considering uncommon pathogens in central nervous system infections. With only three reported cases of cerebral abscesses and one pituitary abscess caused by this microorganism, tailored diagnostic methods and individualized treatment regimens are crucial for accurate management.

### Abstract

Brain abscesses present diagnostic and therapeutic challenges, with *Stenotrophomonas maltophilia* infections being exceptionally rare in the central nervous system. We present a case of odontogenic brain abscesses caused by *S. maltophilia* in an immunocompetent patient, highlighting the rarity and complexity of such infections. A 66-year-old male presented with spatial-temporal disorientation and left-sided weakness. Radiological investigations revealed an expansive lesion in the right posterior frontal region. A craniotomy and drainage were performed, identifying *S. maltophilia* in the purulent material. The patient responded well to tailored antibiotic therapy. *S. maltophilia* related central nervous system infections are infrequent, emphasizing the need for a heightened clinical suspicion in atypical cases. This case contributes to the literature, emphasizing the importance of a multidisciplinary approach for successful diagnosis and management.

#### K E Y W O R D S

brain abscess, immunocompetent, multidisciplinary approach, odontogenic infection, *Stenotrophomonas maltophilia* 

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# **1** | INTRODUCTION

Brain abscesses are infections affecting the central nervous system (CNS), presenting significant challenges in both diagnosis and treatment.<sup>1,2</sup> Obtaining samples from the abscess and conducting microbiological analyses are crucial for guiding appropriate treatment. However, bacterial diagnosis is achieved in only approximately 80% of cases. Typically, streptococci, enterobacteriaceae, Bacteroides spp., and staphylococci are identified as causative bacteria. Stenotrophomonas maltophilia, a gram-negative bacterium, is an infrequent cause of acute bacterial meningitis.<sup>3</sup> In fact, literature reports only three cases of cerebral abscesses and one pituitary abscess caused by this microorganism, indicating an exceptionally low incidence of S. maltophilia infections in these anatomical locations (Table 1).<sup>4–6</sup> Infections involving S. maltophilia are associated with notable morbidity and mortality, and the optimal management of such cases remains unclear. In this context, we present a case of an immunocompetent patient who developed odontogenic brain abscesses with S. maltophilia. This case is accompanied by a comprehensive review of relevant literature, shedding light on the rarity and complexity of such infections. The findings underscore the need for a better understanding of S. maltophilia-related CNS infections to improve diagnostic accuracy and treatment strategies in these challenging cases.

## 2 CASE HISTORY/EXAMINATION

A 66-year-old immunocompetent male patient presented with clinical onset characterized by spatial-temporal

# 3 | METHODS (DIFFERENTIAL DIAGNOSIS, INVESTIGATIONS, AND TREATMENT)

sive formation in the right posterior frontal region.

After the computed tomography (CT) scan, a brain magnetic resonance imaging (MRI) with gadolinium was conducted, confirming the presence of an expansive lesion in the right posterior frontal region. The lesion exhibited fluid/overfluid content, marked proton diffusion restriction, and marginal contrast enhancement. T2/FLAIR images indicated significant hyperintensity in the perilesional white matter, suggesting vasogenic edema and mass effect on adjacent structures (Figure 1). Considering the patient's medical history, an odontogenic source for the cerebral abscess was hypothesized, potentially linked to a previous tooth decay.

Based on the diagnostic findings, the patient underwent a right fronto-parietal craniotomy and drainage of the intracerebral lesion, which was found to contain purulent material. The collected material was sent to the laboratory for culture.

Prior to obtaining culture results, empirical antibiotic therapy with ceftriaxone, vancomycin, and metronidazole was initiated.

Selective culture was performed using the following media:

**TABLE 1** Clinical characteristics, treatments, and outcomes of patients with *Stenotrophomonas maltophilia*-associated brain abscesses and related pathologies reported in case studies.

Authors, year	No. patients	Gender	Age	Site/type of pathology	Clinical presentation	Treatment type
Issaoui et al., 2012 <sup>4</sup>	1	Male	11 days	Meningitis, subdural empyema, multile intraparenchymal abscesses	Hyperbilirubinemia, respiratory distress, seizures, neurological deterioration, increased cranial circumference	Pharmacological
Rémi et al., 2019 <sup>5</sup>	1	Male	74 years	Brain abscesses	Right-sided hemiparesis, clonic seizures, dysphagia, aspiration pneumonia, worsening of post-stroke hemiparesis	Pharmacological, device removal
Yang et al., 2021 <sup>6</sup>	1	Female	30 years	Pituitary abscess (previous surgery for pituitary spindle cell oncocytoma, intrasellar cystic lesion with suprasellar extension)	Nasal stuffiness, progressive headache, reduced visual acuity	Surgical treatment (endoscopic endonasal transshenoidal approach—EETA)

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- 1. CNA (Columbia CNA Agar with 5% sheep blood): Selective medium for isolating gram-positive bacteria.
- 2. MSA (mannitol salt agar): Used to isolate staphylococci.
- 3. MacConkey agar: Selective medium for gram-negative bacteria.
- 4. Sabouraud dextrose agar: Used for the isolation of fungi and yeasts.
- 5. Schaedler blood agar: For gram-negative and obligate anaerobic microorganisms.

The samples were incubated overnight at 37°C for aerobic growth, and at 37°C in special containers for anaerobic conditions (anaerobes require longer growth times, up to 72 h).

After 24 h, growth was assessed for common microorganisms. If no growth was present, the sample was reincubated for an additional 24 h (total of 48 h) before final reporting of no growth. In the present case (cerebral abscess sample), growth was observed on MacConkey agar, specific for gram-negative bacteria, after 24 h; the colonies appeared morphologically homogeneous. Growth was also observed on Sabouraud agar, specific for fungi.

Microorganism identification was performed using MALDI-TOF methodology based on mass spectrometry for rapid partial results communication to the department, followed by antibiogram execution.

Culture results revealed the presence of *S. maltophilia*, a gram-negative bacterium, and *Candida tropicalis*, a yeast (Figure 2).

Antibiogram was conducted using a specific panel for gram-negative bacteria through the broth microdilution method. Results indicated nearly total resistance of *S*.

*maltophilia* to all tested antibiotics, with an intermediate response to Trimethoprim/Sulfametoxazole.

The patient was transferred to neurointensive care and later to the neurosurgery department. Postoperative recovery was marked by gradual neurological improvement. The patient demonstrated regression of the left hemisomatic motor deficit and maintained a seated position.

Following culture results, antibiotic therapy was adjusted to linezolid and levofloxacin.

## 4 | CONCLUSION AND RESULTS (OUTCOME AND FOLLOW-UP)

The patient was discharged on the twentieth postoperative day with a prescription for oral antibiotic therapy, including linezolid 600 mg (twice daily) and levofloxacin 500 mg (twice daily) for up to 6 weeks from the start of antibiotic therapy. A 1-month follow-up brain MRI postoperatively showed significant volumetric reduction in the right frontal cortico-subcortical region of the surgical cavity residue, with reduced enhancement of margins after gadolinium administration (Figure 3).

### 5 | DISCUSSION

*S. maltophilia* is a gram-negative bacillus that thrives in aerobic, non-fermentative conditions and is commonly found in diverse aquatic and humid environments, including drinking water supplies. This bacterium's ability

Treatment administered	Treatment response	Follow-up	Outcome
Ceftriaxone (initial), thiamphenicol (subsequently)	Stabilization after antibiotic change, improvement in respiratory and neurological status	Discharge after 54 days of hospitalization, with residual hypotonia and ventricular dilation	Discharge with neurological improvement but residual hypotonia
Initial: piperacillin/tazobactam, Adapted to: meropenem, vancomycin, Further adapted to: intravenous moxifloxacin and trimethoprim/sulfmethoxazole	Abscess size reduction documented, seizures controlled	64 days of antibiotic therapy, follow-up MRIs	Seizure-free, weaned off ventilator, transferred to rehabilitation hospital, able to stand and walk with walker, thalamic pain lessened
EETA, removal of abscess, irrigation with antibiotics and saline, dural graft, spraying fibrin sealant, 6-week antibiotic	Complete resolution of clinical symptoms	No further headaches or other symptoms occurred	Complete recovery



**FIGURE 1** Preoperative axial T1-weighted MRI with gadolinium revealing a hypointense lesion in the right posterior frontal region with peripheral contrast enhancement (A). Axial and sagittal T2-weighted images confirm the presence of an expansive lesion in the right posterior frontal region. The lesion exhibits significant hyperintensity in the perilesional white matter on T2 images, suggesting vasogenic edema and mass effect on adjacent structures (B).



**FIGURE 2** In the culture plate image, colonies representing *Stenotrophomonas maltophilia* were observed on MacConkey Agar, specific for gram-negative bacteria, after 24 h; the colonies appeared morphologically homogeneous.

to form biofilms facilitates easy adherence to surfaces and medical devices in hospital settings, contributing to increased transmission and antibiotic resistance.<sup>7</sup> In recent years, there has been a growing incidence of *S. maltophilia* infections, particularly on foreign materials such as dialysis equipment and cardiac pacemakers.<sup>7</sup> The production of biofilms plays a crucial role in the organism's adherence to foreign materials, posing concerns with the increasing use of neurostimulation devices in neurological and psychiatric treatments.<sup>5,8</sup>

When diagnosing or suspecting an abscess, *Stenotrophomonas* should be considered due to its biofilm capabilities and problematic resistance spectrum. Traditional antibiotics like meropenem and vancomycin/linezolid may prove ineffective, necessitating alternative treatments such as moxifloxacin, trimethoprim/ sulfamethoxazole, ceftazidime, and tigecycline.<sup>7</sup> *S. maltophilia* is now recognized as a significant nosocomial pathogen, particularly affecting individuals with immunosuppression, prolonged hospital stays, indwelling devices, and extended antibiotic use. It causes various infections, including pneumonia, bacteremia, urinary tract infections, endocarditis, meningitis, and more.<sup>9,10</sup>

## 6 | CHALLENGES IN DIAGNOSIS

Diagnosing *S. maltophilia* meningitis presents several challenges due to its relatively low prevalence and non-specific clinical manifestations. Clinical symptoms such as fever, headache, altered mental status, and focal neurological deficits are common but can mimic other CNS infections or non-infectious etiologies.<sup>11</sup> Neuroimaging studies, including CT or MRI, may reveal abscess formation, meningeal enhancement, or hydrocephalus, but these findings are not specific to *S. maltophilia* infections.<sup>12</sup> Moreover, conventional microbiological techniques, including culture-based methods, may fail to isolate the organism, leading to delayed or missed diagnoses.<sup>11</sup> Enhanced awareness among healthcare providers and utilization of advanced diagnostic methods

**FIGURE 3** One-month postoperative brain MRI images revealed a significant reduction in the volume of the lesion within the right frontal cortico-subcortical region, indicating complete resolution. Additionally, there was nearly complete disappearance of enhancement along the margins after gadolinium administration.



TABLE 2	Overview of central	l nervous system involvemen	t of Stenotrophomonas	maltophilia Infection.
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Variable	Description
Pathogenesis	<i>S. maltophilia</i> thrives in diverse environments and forms biofilms, facilitating adherence to surfaces and medical devices, increasing transmission and antibiotic resistance
Epidemiology	Central nervous system (CNS) infections caused by <i>S. maltophilia</i> are infrequent, with only a few reported cases of brain abscesses and pituitary abscesses attributed to this pathogen
Clinical presentation	Clinical manifestations of <i>S. maltophilia</i> CNS infections are nonspecific and may include fever, headache, altered mental status, and focal neurological deficits
Diagnostic challenges	Diagnosing <i>S. maltophilia</i> meningitis is challenging due to its low prevalence, nonspecific symptoms, and difficulties in isolating the organism using conventional microbiological techniques
Diagnostic methods	Advanced diagnostic methods such as PCR assays, MALDI-TOF MS, and NGS can facilitate timely and accurate identification of <i>S. maltophilia</i> infections in the CNS
Treatment strategies	Empirical antibiotic therapy often involves combination regimens, guided by local susceptibility patterns and the severity of the infection. Surgical intervention may be necessary for source control
Antimicrobial resistance	<i>S. maltophilia</i> exhibits intrinsic resistance to many antibiotics, including traditional agents like meropenem and vancomycin/linezolid, necessitating alternative treatment options such as moxifloxacin, trimethoprim/ sulfamethoxazole, ceftazidime, or tigecycline
Prognosis	Despite aggressive management, mortality rates for <i>S. maltophilia</i> CNS infections remain substantial, highlighting the need for further research into novel antimicrobial agents and treatment strategies

such as polymerase chain reaction (PCR) assays, matrixassisted laser desorption/ionization time-of-flight mass spectrometry (MALDI-TOF MS), and next-generation sequencing (NGS) can facilitate timely and accurate identification of *S. maltophilia* meningitis, improving patient outcomes.<sup>11–13</sup>

# 7 | TREATMENT MODALITIES AND INDICATIONS

Treatment of *S. maltophilia* meningitis poses significant challenges due to the organism's intrinsic resistance to many antibiotics.<sup>12</sup> Empirical therapy often involves

combination regimens to broaden the antimicrobial coverage, but the choice of agents should be guided by local susceptibility patterns and the severity of the infection.<sup>11</sup> While trimethoprim/sulfamethoxazole (TMP-SMX) remains a cornerstone of treatment, its efficacy may be limited in some cases, necessitating alternative antimicrobial agents such as moxifloxacin, ceftazidime, or tigecycline.<sup>11,12</sup> The duration of therapy is typically prolonged, ranging from 14 to 21 days, and may require surgical intervention for source control, such as drainage of abscesses or removal of infected devices.<sup>12,13</sup> Despite aggressive management, mortality rates remain substantial, underscoring the need for further research into novel antimicrobial agents and treatment strategies tailored to S. maltophilia meningitis.<sup>11</sup> To provide a comprehensive understanding of the CNS involvement of S. maltophilia infection, we present a concise overview summarizing the key variables associated with these infections (Table 2).

## 8 | CONCLUSION

In conclusion, our case report highlights the rare occurrence of odontogenic brain abscesses caused by *S. maltophilia* in an immunocompetent patient. The diagnosis and management of such infections pose significant challenges, necessitating a comprehensive approach that includes clinical, radiological, and microbiological assessments.

The case presentation underscores the importance of considering unusual pathogens, such as *S. maltophilia*, in the context of brain abscesses, especially when traditional causative agents are not identified. The patient's clinical course, successful surgical intervention, and tailored antibiotic therapy demonstrate the importance of a multidisciplinary approach in achieving positive outcomes.

This report contributes to the existing literature by providing insights into the diagnostic and therapeutic complexities associated with *S. maltophilia*-related CNS infections. Further research is warranted to better understand the epidemiology, pathogenesis, and optimal management strategies for such rare cases.

### AUTHOR CONTRIBUTIONS

Gianluca Scalia: Conceptualization; data curation; methodology; supervision; validation; visualization; writing – original draft; writing – review and editing. Giancarlo Ponzo: Conceptualization; data curation. Massimiliano Giuffrida: Data curation; formal analysis. Domenico Patanè: Conceptualization; data curation; investigation. Marcello Filippo Riso: Conceptualization; data curation; formal analysis; investigation. Adriana Garozzo: Supervision; validation; visualization. Bipin Chaurasia: Supervision; validation. **Giuseppe Emmanuele Umana:** Supervision; validation; visualization. **Giovanni Federico Nicoletti:** Supervision; visualization.

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The authors declare no conflicts of interest.

### DATA AVAILABILITY STATEMENT

Data sharing not applicable—no new data generated, or the article describes entirely theoretical research.

### ETHICS STATEMENT

This case report was compiled after obtaining informed consent from the patient for the disclosure of clinical history and management with the intention of publication. All attached imaging and clinical materials were deidentified to ensure patient anonymity.

### CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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