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Editor: **Ali Akhaddar,** M.D., IFAANS Avicenne Military Hospital, Marrakech, Morocco

Case Report

Brain abscess caused by *Trueperella bernardiae* in a child

James Pan, Allen L. Ho[1], Arjun V. Pendharkar[1], Eric S. Sussman[1], May Casazza[1], Samuel H. Cheshier[1], Gerald A. Grant[1]

Department of Neurological Surgery, University of Washington School of Medicine, Seattle WA, [1]Department of Neurosurgery, Stanford University School of Medicine, Stanford, California, USA

E-mail: James Pan - jamespan@uw.edu; Allen L. Ho - aho5@stanford.edu; Arjun V. Pendharkar - apendhar@stanford.edu; Eric S. Sussman - esussman@stanford.edu; May Casazza - MCasazza@stanfordchildrens.org; Samuel H. Cheshier - cheshier@stanford.edu; *Gerald A. Grant - ggrant2@stanford.edu
*Corresponding author

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Abstract

Background: Recurrent intracranial abscesses secondary to refractory otitis media present a challenge which demands multidisciplinary collaboration.

Case Description: We present the first known case of pediatric brain abscess caused by a polymicrobial infection of *Trueperella bernardiae*, *Actinomyces europaeus*, and mixed anaerobic species resulting from acute-on-chronic suppurative left otitis media. This patient required two separate stereotactic abscess drainages and a complex course of antibiotics for successful management.

Conclusion: Surgery is essential in the management of cerebral abscess both in agent identification and therapeutic drainage. Management of abscesses secondary to unusual and polymicrobial organisms often requires consultation from other medical and surgical specialties.

Key Words: Brain abscess, neurology, neurosurgery, pediatrics, polymicrobial

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INTRODUCTION

Intracranial cerebral abscesses are life threatening conditions that require immediate medical and neurosurgical attention. If left untreated, permanent neurological damage and result from damage to the brain parenchyma, or resulting inflammation from ventricular dissemination can prove to be fatal. Identification of the etiology and organisms associated with the abscess is crucial to selecting an appropriate antibiotic regimen. Surgery plays an important role in initial biopsy for identification of organisms, and resection can be considered for residual disease. We review an unusual case of pediatric brain abscess caused by Trueperella bernardiae and review both medical and surgical management strategies.

CASE REPORT

The patient is a 5-year-old male with a history of acute-on-chronic suppurative left otitis media status post bilateral percutaneous tympanostomy tube placement

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and multiple courses of antibiotics. He initially presented to his local emergency department with left-sided otalgia, lethargy, emesis, and decreased oral intake. As per the family, he did not have any focal neurological deficits or headaches at that time. He was diagnosed with suppurative acuteotitis externa and prescribed ciprofloxacin ear drops.

Five days later, he returned to his local emergency department with a tactile fever and seizures. Upon arrival, his temperature was 38.4°C and he was found to have tonic-clonic movements. A febrile seizure was suspected, successfully aborted with acetaminophen and lorazepam, and he was loaded on levetiracetam. A computed tomography (CT) scan demonstrated a low-density temporal lesion with left to right midline shift. There was increased density of the external auditory meatus, middle ear, and inner ear. Laboratory studies were notable for a white blood count (WBC) of 33.9 (82.3% neutrophils, 11% bands). He was given one dose of ceftriaxone and piperacillin/tazobactam, intubated, started on a midazolam infusion, and was transferred to our institution for further evaluation.

He was stabilized in our pediatric intensive care unit (PICU) with a non-focal neurologic examination. Vancomycin, cefepime, and metronidazole were started for empiric coverage. On hospital day 2, a magnetic resonance imaging (MRI) demonstrated left tympanomastoiditis with a 3.0 × 1.7 × 2.5 cm left temporal lobe brain abscess. Stereotactic burr hole aspiration yielded 3 mL of foul-smelling yellow-gray fluid, which was sent for cultures [Figure 1a]. The left external auditory canal was found to be filled with copious purulent otorrhea, the tympanic membrane was bulging with purulent middle ear effusion, and the middle ear space filled with granulation tissue. A left tympanoplasty tube was also placed.

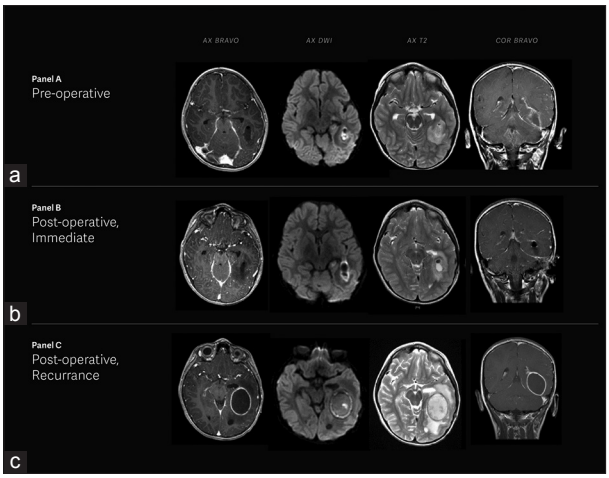

Figure 1: (a) Magnetic resonance imaging of the brain illustrating left temporal brain abscess, pachymeningitis, leptomeningitis, ependymitis, and left tympanomastoiditis. (b) Post-surgical changes showing interval drainage of left temporal lobe abscess with decreased amounts of perihilar material in the resection cavity. (c) Interval increase in size of left temporal lobe abscess with small satellite abscesses seen extending to the ependymal surface of the left lateral ventricle

The patient tolerated the procedure well and was extubated without any complication. Post-operative MRI demonstrated interval drainage of the left temporal lobe abscess [Figure 1b]. He recovered well neurologically with only a mild receptive and expressive aphasia. He continued to receive a combination of vancomycin, cefepime, and metronidazole. On hospital day 7, cultures from aspirated pus grew *Trueperella bernardiae*, *Actinomyces europaeus*, and mixed anaerobic Gram-positive cocci. Cultures from the left ear were positive for *Trueperella bernardiae*, *Corynebacterium amycolatum*, and *Corynebacterium aurimucosum*. These species were identified using matrix-assisted laser desorption ionization. Antibiotics were switched to intravenous meropenem 40 mg/kg every 8 h for a total of 6 weeks. The remainder of his hospital course was unremarkable, with no further febrile episodes or seizures. He was discharged on hospital day 13 at his baseline neurological status with a PICC line for continued meropenem administration.

After returning home, the patient was doing well until he presented to his ED the next day with persistent emesis and left ear pain with intermittent purulent drainage. An MRI obtained at this time demonstrated an interval increase in size of the left temporal lobe abscess with small satellite abscesses extending to the ependymal surface of the left lateral ventricle [Figure 1c]. His antibiotics were re-broadened to vancomycin, cefepime, and metronidazole. Out of concern for recurrence of the abscess, he was taken for stereotactic aspiration and mastoidectomy. Intraoperatively, 26 mL of thick purulent liquid was aspirated from the abscess.

Cultures of the repeat aspiration remained sterile. Our infectious disease colleagues recommended continuation of triple therapy of vancomycin, cefepime, and metronidazole for 6 weeks and he was discharged on hospital day 14 at his neurologic baseline. Susceptibility testing later revealed that this particular strain of *Actinomyces* was sensitive to the penicillin class; therefore, after he completed a 6-week course of triple therapy, and he was transitioned to oral amoxicillin monotherapy for 6 additional months.

DISCUSSION

The development of recurrent intracranial abscesses secondary to refractory otitis media presents a clinical challenge in terms of neurosurgical and antimicrobial treatment. Bacterial cerebral abscess formation can result from direct or hematogenous spread. The direct spread of bacterial agents from a contiguous site is responsible for

20%–60% of cases.[3] The most common parameningeal foci for contiguous spread include subacute and chronic otitis media and mastoiditis, frontal or ethmoid sinusitis, or dental infections. Abscess formation as a result of otolaryngologic complications has been decreasing, especially in developed countries with adequate access to antibiotics. Mortality from brain abscess due to complications from otolaryngologic sources has been estimated to be 3.8%–18.6%.[6,12]

Brain abscesses due to spread from parameningeal foci are often complex. A systematic review and meta-analysis identified *Streptococcus* as the most common agent from the middle ears, mastoids, and sinuses leading to brain abscess formation.[1] A 16S rRNA-based metagenomic analysis of cerebral abscesses from 51 patients also identified *Staphylococcus* and mixed flora responsible for abscess formation.[7] *Trueperella bernardiae* is a catalase-negative and Gram-positive coccobacillus which acts as an obligate parasite of the human pharynx and skin, and has been rarely reported in the literature as a pathogenic microorganism. Originally isolated from blood cultures, bone, wounds, abscesses, and skin infections, this bacterium was provisionally assigned to the CDC fermentative Coryneform group 2 and later to *Actinocytes bernardiae* sp. nov., based on 16S rRNA sequencing. In 1997, *Actinocytes Bernardiae* was re-assigned to the genus *Arcanobacterium* based on re-analysis of 16S rRNA gene sequences and comparative analysis. This species was again re-assigned to the *Trueperella* genus after another reassessment of pylogenetic positioning and chemotaxonomic characteristics.[11] The only known case of *Trueperella* associated with a brain abscess was reported in a 68-year-old woman with a history of diabetes insipidus, rheumatoid arthritis, and long-standing chronic suppurative otitis media.[8] *Trueperella bernardiae* has also been reported in a wound infection after laparoscopy[10] and in a prosthetic joint infection after total knee replacement.[4] To the best of our knowledge, this case is the first pediatric brain abscess with culture confirmed *Trueperella bernardiae* reported in the literature.

Antibiotic therapy should be started immediately when there is a clinical suspicion for brain abscess. A retrospective study examining factors associated with mortality from brain abscesses identified that a delay in initiating antibiotics resulted in an increased odds ratio of 1.5 per day of delay for in-hospital death.[5] Before culture identification of the infectious agent, empiric antibiotic therapy should be based on the presumptive source of infection. For otologic, oral, and sinus sources, metronidazole provides excellent abscess penetration and has bactericidal activity against anaerobic species. A third- or fourth-generation cephalosporin is recommended for coverage of aerobic Gram-positive and Gram-negative organisms. At our institution, we have elected to use cefepime due to *Pseudomonas* coverage and its association with otologic sources. Vancomycin should also be considered when there is high suspicion for *Staphylococcus aureus* and especially for drug-resistant subtypes.

Surgery is essential for identification of the causative organism since the abscess can be polymicrobrial and to reduce the size of the abscess when there is mass effect. Needle aspiration and surgical resection are both methods which can aid in diagnosis, and treatment even if performed after antibiotic treatment has been initiated. If the brain abscess is suspected to be secondary to hematogenous spread, antibiotics can initially be selected based on blood cultures. If neuroimaging does not show a central cavity in the abscess, stereotactic biopsy of the area of presumed cerebritis and administering empirical antibiotics with follow-up imaging should be considered.[2]

Stereotactic needle aspiration is generally preferred over surgical resection due to a lower complication rate and adverse neurological sequelae. With modern neurosurgical techniques, almost any abscess that measures at least 1 cm in diameter, regardless of location, is amenable to stereotactic aspiration.[2] Needle aspiration can be achieved relatively easily through a twist drill hole, at any stage of the abscess, even in severe cases. This technique can also be used to access technically challenging locations such as the brainstem and periventricular regions.[9] However, one study comparing 23 patients undergoing image-guided burr hole aspiration versus 22 patients receiving surgical resection of brain abscesses reported that needle aspiration is associated with a need for longer antibiotic therapy and prolonged neurological recovery time.

Surgical resection is a much more invasive approach and is now infrequently performed as first-line therapy. However, excision can be considered in cases where the abscess is superficial and not located in or near eloquent structures, and particularly when there is suspicion of fungal or tuberculous infection or of branching bacteria (e.g., *Actinomyces* or *Nocardia* spp.).[2] Surgical excision can also be considered after initial needle aspiration if there is no clinical improvement within 1 week, decline in neurological function, signs of increased intracranial pressure, or interval increase in the size of the abscess. If an abscess is located adjacent to the ventricular system, but has not yet ruptured into the ventricle, needle aspiration would be favored over surgical excision due to the risk of rupture and high morbidity and mortality associated with disseminated ventriculitis.

This rare case of a cerebral abscess due to *Trueperella bernardiae* clearly benefited from multidisciplinary collaboration among infectious disease specialists, otolaryngologists, and neurosurgeons to achieve the best

possible outcome in this child with a life-threatening condition.

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Conflicts of interest
There are no conflicts of interest.

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