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Case report

Abiotrophia defectiva meningitis following ventriculoperitoneal shunt repair: Case report and literature review

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ABSTRACT

Introduction: Abiotrophia defectiva is an uncommon and potentially severe cause of bacterial meningitis that can be seen in the setting of neurosurgical procedures. We describe here a case of *A. defectiva* meningitis in a patient with VP shunt revision.

Case report: The patient is a 59-year-old female with VP shunt placed several years prior in the setting of normal pressure hydrocephalus. She underwent successful shunt repair following worsening gait abnormalities. On post-operative day 8 she was brought to the emergency room with fevers, photophobia and altered mentation. CSF cultures returned positive for *A. defectiva* and she was treated with intravenous vancomycin. Due to the presence of retained hardware she was discharged on chronic suppressive therapy with oral amoxicillin.

Conclusion: This case illustrates *A. defectiva* as a cause of bacterial meningitis following neurosurgical intervention. It is important to consider this fastidious organism and tailor antimicrobial therapy appropriately to prevent significant morbidity and mortality. As was the case with our patient, suppressive therapy may be warranted in setting of retained hardware.

Introduction

The development of bacterial meningitis as a complication of neurosurgical procedures or intracranial trauma is associated with significant morbidity and mortality. *N. meningitis* and *S. pneumoniae* represent the most common organisms involved in the general adult population, though patients with penetrating trauma or following neurosurgery tend to have increased isolation of anaerobic gram-negative bacilli, *S. aureus* and *S. epidermidis* [1]. *Abiotrophia defectiva* is a fastidious gram-positive bacterium previously classified as nutritionally variant streptococci (NVS) which has only rarely been identified as the causative agent in a central nervous system (CNS) infection [2]. We describe here a case of bacterial meningitis due to *A. defectiva* in a patient that underwent revision of a ventriculoperitoneal (VP) shunt. We provide a discussion of the rare organism as well as a review of all cases described to date.

Case description

Our patient is a 59-year-old female with mental disability and seizure disorder with a history of right VP shunt placed several years prior for confirmed normal pressure hydrocephalus. She initially presented to the emergency room (ER) for shunt interrogation and revision following worsening gait abnormalities and mental status. She received IV cefazolin peri-operatively and was placed on oral cephalexin to continue for 7 days post-operatively per the Neurosurgery team. She tolerated the procedure well, however on post-operative day 3, a cerebrospinal fluid (CSF) leak was noted from her right clavicular incision. Shunt series was obtained, and she was taken to the operative room for wound exploration and tie-off of the right parietal VP shunt catheter at the clavicle. Gait and mentation improved, and she was discharged with outpatient follow up. She was brought back to the ER the next morning (post-operative day 8) following development of fever, chills, acute confusion as well as photophobia and neck pain.

She was noted to be febrile and tachycardic on initial evaluation with leukocytosis and elevated inflammatory markers on blood work. CT head was obtained which showed stable ventricular system but increased soft tissue edema at the area of parietal suture along the catheter distal tract. She was started on intravenous vancomycin, ampicillin, and ceftazidime in the ER. Blood cultures remained negative. Her VP shunt was drained with clear CSF return noted. CSF studies revealed 53 nucleated cells with 50% neutrophils, glucose of 60 and

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Table 1
Susceptibilities of *Abiotrophia defectiva* isolated from patient CSF culture. S (susceptible); I (intermediate); R (resistant). Please see supplemental material for CLSI breakpoints and interpretative criteria.

Agent	MIC	Susceptibility
Cefepime	≤ 0.500	S
Ceftriaxone	≤ 0.120	S
Chloramphenicol	4	S
Meropenem	≤ 0.250	S
Penicillin	0.25	I
Vancomycin	≤ 0.500	S

protein of 52.1.

Given concern for infectious etiology, the patient underwent right frontal VP shunt removal. However, a portion of the parietal catheter could not be retrieved as it was adhered, and hardware could not be completely removed without significant damage to the tissue. CSF culture returned positive for A*biotrophia defectiva*. Patient had developed a macular rash for which Dermatology was consulted with suspicion that this was cephalosporin-induced and so ceftazidime and ampicillin were discontinued once CSF culture speciation finalized. In setting of her allergic reaction concern and pending sensitivities, vancomycin monotherapy was continued inpatient. Results of sensitivities (Table 1) were unfortunately delayed and when finally available, the patient had completed 19 days of vancomycin and could be transitioned to oral therapy. She was transitioned to amoxicillin 1000 mg twice daily on which she was discharged indefinitely.

Patient was evaluated in clinic several weeks later with improved gait and mentation. She had been tolerating the suppressive regimen without adverse effects with no evidence of infection recurrence.

Discussion

There have been several descriptions of *Abitropha defectiva* causing infective endocarditis [3] however very few cases involving the organism as a cause of meningitis have been reported to date. *A. defectiva* is a gram-positive aerobe that was previously classified as nutritionally variant streptococcus along with *Granulicatella adiacens, Granulicatella elegans* and *Granulicatella balaenopterae* [4]. These organisms can be found in the normal oral flora, as well as gastrointestinal and genital flora. Infections have also been reported in relation to septic arthritis and bacteremia [5]. The first case describing this aerobic bacterium as a potential causative organism of CNS infection was noted by Schelgel et al. in a patient that underwent CT guided myelography [6]. The source of infection was presumed to be from the oropharyngeal flora of the physician doing to procedure at a time when surgical masks were not obligatory.

Upon review of recent literature there have only been five reported cases of CNS *A defectiva* infections which are summarized in Table 2. Most cases appear to be nosocomial and introduced at the time of a neurosurgical intervention. It is likely that our patient was infected at the time of her VP shunt revision or following the wound exploration for the CSF leak. To our knowledge there have been no other reported cases of successful continued management of *A defectiva* meningitis with suppressive therapy in the setting of retained hardware.

There are no clear recommendations on the ideal duration of treatment and agent of choice for NVS infections. Empiric treatment for both gram positive and negative organisms as well as anaerobes is usually recommended pending culture speciation and sensitivities. A review by Albertri et al. evaluated the various antimicrobial sensitivities of NVS and found all to be susceptible to vancomycin and none with aminoglycoside resistance. Resistance to penicillin was noted particularly in *A. defectiva* compared to *G. adiacens* however resistance to ceftriaxone was more pronounced in *G. adiacens*. Several isolates showed resistance to levofloxacin, erythromycin, and clindamycin [10].

Though *Abiotrophia* remains an unusual cause for neurological

Table 2
List of *Abiotrophia defectiva* meningitis cases to date and infection characteristics. WBC (white blood cell count); PMN (polymorphonuclear cells); N/A (data not available) F (female); M (male). Immunocompromised – defined as conditions that weaken the body's immune response to disease processes. This includes both congenital immunodeficiency (such as X-linked agammaglobulinemia) and acquired immunodeficiency (such as HIV, hematopoietic malignancy or use of immune suppressing drugs).

Case	Age	Sex	Surgical procedure	Perioperative antibiotics received	Immunocompromised status	Bacteremia present	Type of infection	Time from procedure	CSF evaluation	Penicillin sensitive	Treatment used	Clinical outcome
Schegel et al.[6]	49	F	CT-guided myelography	No	No	Unknown	Meningitis	4	WBC: 320/mm3 (90% PMNs), glucose: 3 mM, protein: 55 mg/dL	Yes	Vancomycin/fosfomycin/cefixime/rifampin (10 days)	Stable
Cercero et al.[2]	53	F	MCA aneurysm clipping and VP shunt placement	Yes (cefazolin)	No	Unknown	Meningitis and brain abscess	11	WBC: 630/mm3 (78% PMNs), glucose: 53 mg/dL, protein: 190 mg/dL	Yes	Penicillin/gentamicin (28 days)	Stable
Zerone et al.[7]	66	M	None	On ofloxacin for bacteremia	Yes	Yes (Salmonella typhimurium)	Brain abscess		N/A	Yes	Amoxicillin/rifampin (10 days) followed by amoxicillin/clindamycin (35 days)	Stable
Levin et al.[8]	58	F	None	Unknown	No	Unknown	Brain abscess		N/A	N/A	Ceftriaxone/vancomycin/metronidazole (died before treatment was complete)	Death
Tena et al.[9]	64	M	Spinal anesthesia	Yes (cefazolin)	No	No	Meningitis	4	WBC: 345/mm3 (70% PMNs), glucose: 15 mg/dL, protein: 234 mg/dL	Yes	Ampicillin/vancomycin/ceftriaxone (10 days)	Stable
Current report	59	F	VP shunt revision	Yes (cefazolin)	No	No	Meningitis	8	WBC: 53 cells/mm3 (50% PMNs), glucose: 60 mg/dL, protein: 52 mg/dL	Intermediate	Vancomycin (14 days) followed by Amoxicillin (indefinitely)	Stable

infections, there have been an increasing number of cases of infective endocarditis in both immunocompromised and immunocompetent hosts [11,12] and so this fastidious aerobe should not be overlooked, particularly in the setting of negative cultures.

Conclusion

Abiotropha defectiva is a rare and severe cause of meningitis that is difficult to isolate given its fastidious nature. It is important to consider this bacterium in CNS infections especially following neurosurgical instrumentation. Our case illustrates such a situation and reminds us of the importance of appropriate antimicrobial coverage particularly in the setting of retained hardware and need for chronic suppressive therapy.

Author statement

Thomas Erwes - writing.
Casey Godshall - writing.

Conflict of interest

No conflicts of interest to disclose.

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Consent

Consent unable to be obtained. No identifying characteristics used in case description.

Ethical approval

Not applicable.

Appendix A. Supporting information

Supplementary data associated with this article can be found in the online version at doi:10.1016/j.idcr.2023.e01708.

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