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Case Report

Postoperative cerebrospinal fluid infection by *Ralstonia mannitolilytica*: Two case reports and a literature review

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ABSTRACT

Background: *Ralstonia* species are Gram-negative bacilli that are commonly found in moist environments, such as water and soil. They are opportunistic human pathogens, particularly found among immunocompromised patients, and are an infrequent cause of infection. The difficulty in correctly identifying and differentiating between *Ralstonia* species members using routine biochemical methods as well as their resistance to many classes of antibiotics poses a specific diagnostic and therapeutic challenge.

Case Description: We report two cases from our neurosurgical unit complicated by postoperative cerebrospinal fluid infection caused by *Ralstonia Mannitolilytica* that posed a therapeutic challenge.

Conclusion: Our hypothesis is contaminated irrigation fluids might be a significant cause of post-operative meningitis and prolonged hospital stay.

Keywords: Cerebrospinal fluid (CSF) infection, Meningitis, *Ralstonia mannitolilytica*

INTRODUCTION

Ralstonia mannitolilytica is an emerging opportunistic pathogen.[5] It is a Gram-negative organism that leads to a wide spectrum of infections in hospital settings.[1] *R. mannitolilytica* has been described as a cause of a wide spectrum of hospital-acquired infections due to their ability to survive in liquid media, including saline and chlorhexidine, and on medical devices.[1]

We report two cases from our neurosurgical unit that was complicated by postoperative cerebrospinal fluid (CSF) infection caused by *R. mannitolilytica*.

CASE PRESENTATION

Case 1

A 9-year-old boy, who is a known case of epilepsy, was admitted electively to our neurosurgery unit as a case of suprasellar cystic lesion, for surgical resection. Our working diagnosis was craniopharyngioma [Figure 1].

The patient underwent endoscopic cyst aspiration, biopsy, septostomy, and external ventricular drain (EVD) insertion. The surgery went well with no immediate postoperative complications. The patient was vitally stable, and he was transferred to pediatric intensive care unit for monitoring.

Day 1 postoperatively, the patient developed one spike of fever that responded to paracetamol. The patient was awake, conscious, and well-looking. His examination and basic laboratory investigations were within normal limits. The patient was on prophylactic cefazolin, as per our surgical prophylactic antibiotic protocol. Next day, the fever became more persistent. Septic workup was done, the patient was started on vancomycin and ceftazidime. The antibiotic regimen was modified to include gentamicin as the patient continued to have high grade fever. His inflammatory markers were within normal range. White blood cell count (WBC) was 12×10^9/l. Erythrocytes sedimentation rate (ESR) was 2 mm/h. C-reactive protein (CRP) was 1 mg/L. CSF examination taken from the EVD revealed WBC 392 cells/µL (74% polymorph), glucose 3.2 mmol/L, and protein 0.3 g/L. CSF culture resulted on day 5 postoperatively showing light growth of Gram-negative rods, namely, *R. mannitolilytica*. Other cultures were negative. The antibiotic regimen was down-graded to ceftazidime to complete 14 days. The EVD was removed and the tip was sent for culture. The same organism was isolated for EVD tip. Yet, the sensitivity report showed resistant development toward ceftazidime. Intravenous Ciprofloxacin was added to the regimen in light of the new sensitivity report. Antibiotics were stopped after clinical, laboratory, and radiological evidence of resolution.

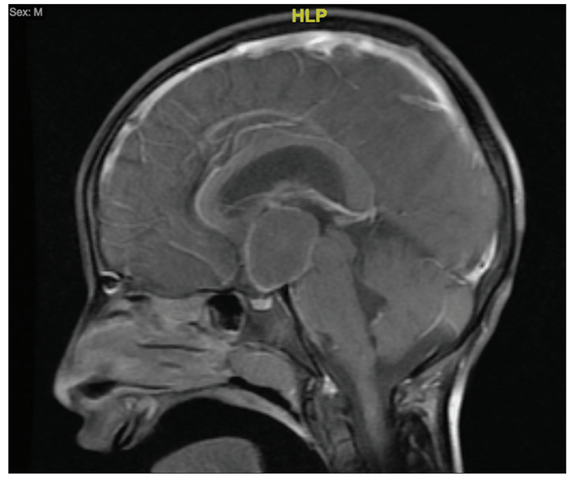

Figure 1: Magnetic resonance imaging of the brain, T1-weighted gadolinium-enhanced. Sagittal view, showing hypothalamic/suprasellar cyst, measuring 2.2 × 2.3 × 3.2 cm in its maximum transverse, anteroposterior, and craniocaudal dimensions, respectively, with peripheral thin enhancement of the cyst and solid enhancement of the right inferolateral. HLP=head left posterior.

Case 2

A 19-year-old female is known case of systemic lupus erythematosus and idiopathic intracranial hypertension (IIH). She underwent right ventriculoperitoneal (VP) Shunt insertion after she failed to respond to medical management. She was on prophylactic Cefazolin 25 mg/kg upon induction, and the same dose every 8 h 1 day postoperatively. The patient was discharged home with no immediate postoperative complications.

Two-week postoperatively, she presented to our emergency department with history of the right lower quadrant abdominal pain and high-grade fever for 3 days. On examination, she was febrile and tachycardic. Her Glasgow coma scale was 15/15. She had right lower quadrant tenderness. Her neurological examination was within normal limits, and no meningeal signs were observed. Septic screening was sent including CSF sample from shunt reservoir. Computed tomography (CT) of brain showed VP shunt in place and no collection. CT of the abdomen showed subcutaneous collection at VP shunt entry site measuring 3.7 × 6.0 cm with intrapelvic collection in the left supravesicular space measuring 4.1 × 5.3 cm [Figure 2].

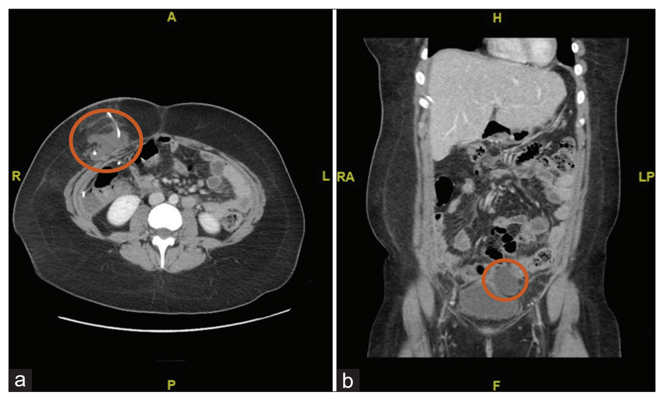

Figure 2: (a and b) Abdominal computed tomography image on (a) axial view Red circle: subcutaneous collection at shunt entry site (b) coronal view showing small subcutaneous collection at shunt entry site in the abdominal cavity measuring 3.7 × 6.0 cm, associated with small intrapelvic collection in the left supravesicular space causing minimal mass effect to the left lateral urinary bladder wall measuring 4.1 × 5.3 cm. RA= Right anterior / LP = left posterior / H=head/ F=feet, red circle: intrapelvic collection in the left supravesicular space causing minimal mass effect.

The patient was started empirically on Cefuroxime and Metronidazole. Yet the patient continued to have high-grade fever, so antibiotic was upgraded to Vancomycin and Ceftazidime. She underwent aspiration of abdominal collection and Jackson-Pratt drain was inserted, and fluids were sent for culture and sensitivity. Furthermore, VP shunt was removed, and both abdominal and cerebral tip were sent for culture. CSF culture resulted with heavy growth of

R. mannitolilytica. Aspirated abdominal fluid culture showed no growth after 7 days. Pipracillin/tazobactam was started and other antibiotics were stopped. Next day, the patient continued to have high-grade fever. We started the patient on Ciprofloxacin and metronidazole. The patient started to improve clinically and repeated CT abdomen showed resolution of the abdominal collection. The patient was discharged later on oral antibiotics to complete 14 days. The patient declined further surgical treatment for IIH.

DISCUSSION

Bacteremia and postoperative CSF infections are associated with a high mortality and morbidity outcomes but data about systematic approach to the diagnosis, treatment, and management of *R. mannitolilytica* bloodstream and postoperative CSF infections are lacking.[2] A few cases with *R. mannitolilytica* related hospital outbreaks have been reported in the literature.[4,6] However, the first report which revealed bacteremia and bacteriuria in 25 patients was due to contaminated parenteral fluids.[10] Although *Ralstonia* species Central nervous system (CNS) infection was reported in the literature, *R. mannitolilytica* postoperative meningitis was reported once in the literature.[10] We believe that the number of cases may be underestimated due to the difficulty of identification of *R. mannitolilytica*, and misidentification as *Ralstonia pickettii*, *Burkholderia cepacia* complex, and *Pseudomonas fluorescens* pointing toward the presence of a contamination.

R. mannitolilytica has been described as a cause of recurrent ventricular-atrial shunt-associated meningitis.[10] The ability of these bacteria to cause localized and systemic infections is likely due to their ability to produce biofilm, which, in turn, is key to their survival in the environment, their evasion of the host's immune response and their frequent antibiotic resistance.[1] *R. mannitolilytica* typically colonized in environmental habitats such as water, soil, and plants and it is classified as a Gram-negative, oxidase-positive, and non-fermentative rods.[2] The characteristic acidification of mannitol is the reason behind referring it to *R. mannitolilytica* and thus separating it from other described *Ralstonia* species.[5] Nowadays, *R. mannitolilytica* is considered as an emerging opportunistic pathogen that leads to nosocomial infection secondary to contamination of medical equipment, devices, water, or parenteral solutions.[5] Typical demographic of *R. mannitolilytica* infection is mainly immunocompromised patients.[1] *Ralstonia* bacteria have been associated with infections in hospital settings due to their ability to survive in liquid media, this is not surprising as even in cases of well documented hospital outbreaks the environmental origin was not identified.[1] The organism could not be eradicated using the manufacturer's recommended disinfection protocol. Although generally of low virulence, *Ralstonia* species can be a cause of potentially serious healthcare-associated infections.[6]

In spite of new antimicrobials and aseptic surgical techniques, postoperative meningitis (POM) results in considerable morbidity and mortality. Any neurosurgical intervention has a risk of developing POM with an incidence of 0.34–3.1%.[8] The spectrum of pathogens associated with postoperative meningitis is ever expanding. Limited data about antimicrobial susceptibility are reported.[1] *R. mannitolilytica* is known to be intrinsically resistant to colistin. Variable *R. mannitolilytica* susceptibility to ceftazidime, cefepime, and carbapenem and amino-glycosides resistance have been reported.[1-3,6,10] *R. mannitolilytica* seems to easily gain antimicrobial resistance presenting a susceptibility profile not far to a multi-drug resistant agent. It has been postulated that broad spectrum antimicrobial treatment should contribute to increase *R. mannitolilytica* strains resistance.[7,9] However, where reported, all *R. mannitolilytica* involved in bacteremia cases showed susceptibility to sulfamethoxazole-trimethoprim and most of the strains appeared susceptible to fluoroquinolones, cefotaxime, and piperacillin-tazobactam.[2]

CONCLUSION

We hypothesize that the infection was acquired following contamination by contaminated irrigation fluids during both procedures. *Ralstonia* species are not widely recognized as a major pathogen, despite their ability to induce serious multi-drug resistance infection. Clinician must consider them in differential diagnosis of postoperative CSF infections and sepsis to help in their early isolation and treatment.

Declaration of patient consent

Institutional Review Board (IRB) permission obtained for the study.

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Conflicts of interest

There are no conflicts of interest.

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