

**A Rare Case of *Burkholderia cepacia* Complex Bloodstream Infection in Rasmussen Encephalitis and Super Refractory Status Epilepticus****Krishna Sarkar<sup>a</sup>, Kirti Nirmal<sup>a\*</sup>, Hage Yaja<sup>a</sup>, Deeksha Chaudhary<sup>a</sup>, Shukla Das<sup>a</sup>**<sup>a</sup>Department of Microbiology, University College of Medical Sciences and Guru Tag Bahadur Hospital Delhi 110095, India.**Abstract**

**Background:** *Burkholderia cepacia* complex (BCC) is known for its resistance to antibiotics and sporadic occurrence in various infections, including bloodstream infections. Its association with neurological disorders, such as Rasmussen Encephalitis (RE), remains poorly understood.

**Case presentation:** We describe a 4-year-old girl with RE and Super Refractory Status Epilepticus, a condition characterized by frequent seizures that do not respond to conventional treatments. Despite initial treatment, her condition worsened, and blood cultures revealed the presence of *Burkholderia* species. Adjusting the antibiotic therapy to target this specific pathogen led to significant improvement in her clinical status.

**Conclusion:** Our case highlights the rare occurrence of BCC bloodstream infection in a child with complex neurological issues. Although the precise link between BCC and neurological disorders is unclear, our findings contribute to the growing body of evidence that BCC can cause severe infections beyond the respiratory system. This case underscores the importance of accurate diagnosis and tailored treatment in managing such challenging infections effectively.

**Keywords:** *Burkholderia cepacia* Complex: Bloodstream Infection: Neurological disorders: Rasmussen Encephalitis: Super Refractory Status Epilepticus.

**DOI:** 10.21608/SVUIJM.2024.296626.1893

**\*Correspondence:** [doctorkirtinirmal@gmail.com](mailto:doctorkirtinirmal@gmail.com)

**Received:** 1 June, 2024.

**Revised:** 7 June, 2024.

**Accepted:** 18 June, 2024

**Published:** 24 June, 2024

**Cite this article as:** Krishna Sarkar, Kirti Nirmal, Hage Yaja, Deeksha Chaudhary, Shukla Das. (2024). A Rare Case of *Burkholderia cepacia* Complex Bloodstream Infection in Rasmussen Encephalitis and Super Refractory Status Epilepticus. *SVU-International Journal of Medical Sciences*. Vol.7, Issue 2, pp: 197-201.

Copyright: © Sarkar et al (2024) Immediate open access to its content on the principle that making research freely available to the public supports a greater global exchange of knowledge. Users have the right to Read, download, copy, distribute, print or share link to the full texts under a [Creative Commons BY-NC-SA 4.0 International License](https://creativecommons.org/licenses/by-nc-sa/4.0/)

## Introduction

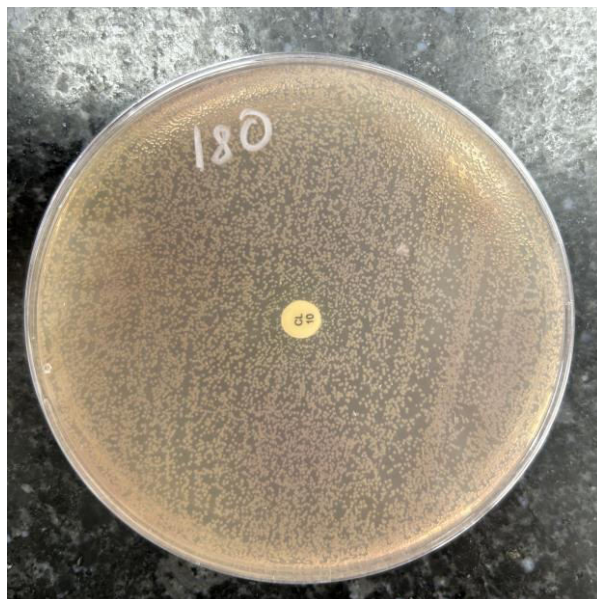
*Burkholderia cepacia* complex (BCC) encompasses a group of gram-negative bacteria known for their wide-ranging environmental distribution and inherent resistance to antibiotics (Tavares et al., 2020). While typically associated with respiratory tract infections in individuals with cystic fibrosis or chronic lung diseases, BCC infections have also been reported in various clinical settings, including bloodstream infections (Lord et al., 2020). However, the occurrence of BCC in conjunction with encephalitis, particularly Rasmussen Encephalitis (RE), represents a rare and intriguing clinical phenomenon. RE is a rare, chronic inflammatory brain disorder characterized by unilateral hemispheric inflammation, leading to progressive neurological deficits and intractable seizures (Varadkar et al., 2014). The relationship between BCC infection and encephalitis, including RE, remains poorly understood, with only a limited number of reported cases in the literature suggesting a potential association between BCC and neurological disorders. Therefore, elucidating the potential association between BCC infection and encephalitis, including the pathogenesis and clinical implications, warrants further investigation to enhance our understanding of this rare but significant clinical entity.

## Case Report

A 4-year-old female presented with a history of recurrent seizures despite receiving multiple antiepileptic medications. On admission, she was in a state of super refractory status epilepticus, experiencing frequent left-sided focal seizures refractory to conventional treatments. Additionally, she exhibited symptoms consistent with Rasmussen encephalitis, including progressive hemiparesis and cognitive decline. The patient also presented with elevated blood pressure readings, shock, and intermittent fever spikes. Examination revealed generalized edema and limb swelling, suggestive of systemic involvement. Laboratory investigations revealed hypokalemia and a positive urine culture for budding yeast cells, indicative of a urinary tract infection. CSF culture showed no growth. The patient was empirically started on meropenem and vancomycin; however, the patient's clinical condition continued to deteriorate. A set of blood cultures was obtained, both of which flagged positive for microbial growth. Subculture on nutrient agar revealed violet-coloured colonies (Fig. 1), revealing gram-negative bacilli on gram staining, resistant to colistin (Fig.2) and aminoglycosides, and sensitive to cotrimoxazole.



**Fig. 1.** Colony morphology of *Burkholderia cepacia* complex on nutrient agar showing violet-coloured colonies



**Fig.2. *Burkholderia cepacia* complex showing inherent resistance to colistin**

(Table.1) presents the findings of the conventional microbiological investigation, which point toward *Burkholderia* species. Identification was further confirmed by Vitek 2 Compact.

Based on sensitivity reports, meropenem and vancomycin were discontinued, and cotrimoxazole was initiated. Later on, ceftazidime was added, leading to improvements in the patient's sepsis.

**Table 1. Conventional biochemical tests for *Burkholderia cepacia***

Biochemical tests	Result
Catalase	Positive
Oxidase	Positive
Oxidation-Fermentation (OF) Test	Oxidative
Triple Sugar Iron (TSI) Agar Test	K/K ( gas, no H <sub>2</sub> S)
Methyl Red (MR) Test	Negative
Voges-Proskauer (VP) Test	Negative
Citrate Utilization Test	Positive
Urease Test	Negative
Nitrate Reduction Test	Negative
Phenylalanine Deaminase (PPA) Test	Negative
Indole	Negative
H <sub>2</sub> S	Negative
Motility	Motile
Colistin Susceptibility Test	Resistant
Gentamicin	Resistant
Cotrimoxazole	Sensitive

### Discussion

This case report illustrates a *Burkholderia cepacia* complex bloodstream infection in a paediatric patient with Rasmussen encephalitis and super refractory status

epilepticus. Our findings contribute to the limited body of literature documenting BCC infections outside the context of cystic fibrosis, particularly in patients with significant neurological involvement.

The relationship between BCC infection and neurological disorders, particularly RE, is not well-documented. The exact mechanisms by which BCC could influence neurological outcomes remain speculative. However, systemic infections and the resulting inflammatory response might exacerbate neurological symptoms (Boyd et al., 2022). Previous case reports have suggested that BCC can cause central nervous system infections, including meningitis and brain abscesses, albeit rarely (Dorsett et al., 2016; Peralta et al., 2018). Peralta et al (2018) reported a case where *Burkholderia multivorans* was isolated from a meningitis patient following two episodes of central line-associated bloodstream infections. Similarly, Crispim et al (2010) documented a case of *Burkholderia cepacia* meningitis in an infant. The potential for direct CNS involvement or secondary inflammatory effects warrants further investigation. *Burkholderia cepacia* is generally a colonizing organism rather than an infectious one, but it can be pathogenic in immunocompromised individuals if found in body fluids that are typically sterile. When identified in blood culture, it may signify an infection, a pseudo-infection, or contamination from intravenous fluids (Doit et al., 2004).

BCC is increasingly recognized as a nosocomial pathogen, particularly in immunocompromised individuals and patients with prolonged hospital stays (Kirmani et al., 2019). This organism is known for its ability to persist in moist environments, including hospital equipment and intravenous fluids, leading to hospital-acquired infections (Doit et al., 2004). Comparative analysis with recent literature reveals that BCC bloodstream infections in non-cystic fibrosis patients, particularly those with neurological conditions, are exceptionally rare. Recent studies have emphasized the role of *Burkholderia cepacia* in causing bacteremia, especially in intensive care settings and among patients with invasive

devices such as central venous catheters (Meena et al., 2019; Rastogi et al., 2019). In our case, the presence of BCC in the bloodstream points towards its potential to cause significant morbidity in hospitalized patients with complex medical histories, reinforcing the need for stringent infection control measures to prevent HAIs .

### Conclusion

Our case adds to the emerging evidence that BCC can cause severe infections beyond the respiratory system, affecting diverse patient populations, including those with neurological disorders. The transition from empiric broad-spectrum antibiotics to targeted therapy resulted in clinical improvement, emphasizing the importance of accurate microbiological diagnosis and tailored treatment plans.

### References

- Boyd RJ, Avramopoulos D, Jantzie LL, McCallion AS. (2022). Neuroinflammation represents a common theme amongst genetic and environmental risk factors for Alzheimer and Parkinson diseases. *Journal of Neuroinflammation*, 19(1):223.
- Bovolenta E, Zanon MP, Mondino S, Manfrin V, Rassu M, Diquigiovanni A, et al. (2023). Management of an Epidemic Outbreak of *Burkholderia Cepacia* in A Hospital in The North of Italy. *Journal of Food and Nutrition*, 2(1):2836-276.
- Crispim JN, Dâmaso C, Marques JG, Miguéns J, Valente P. (2010). *Burkholderiacepacia* meningitis: A case report. *Journal of Pediatric Infectious Diseases*, 5(4):393-6.
- Doit C, Loukil C, Simon AM, Ferroni A, Fontan JE, Bonacorsi S, et al. (2004). Outbreak of *Burkholderia cepacia* bacteremia in a pediatric hospital due to contamination of lipid emulsion stoppers. *Journal of clinical microbiology*, 42(5): 2227-30.

- **Dorsett M, Liang SY (2016).** Diagnosis and treatment of central nervous system infections in the emergency department. *Emergency Medicine Clinics*, 34(4):917-42.
- **Kirmani S, Khan F, Kaushal N, Shahid M, Taneja N, Ahmed A (2019).** Burkholderia cepacia complex bacteremia in pediatric intensive care unit in a tertiary care hospital in North India. *CHRISMED Journal of Health and Research*, 6(4):232-6.
- **Lord R, Jones AM, Horsley A (2020).** Antibiotic treatment for Burkholderia cepacia complex in people with cystic fibrosis experiencing a pulmonary exacerbation. *Cochrane Database of Systematic Reviews*, 2020(4): CD009529.
- **Meena S, Bir R, Sood S, Das BK, Kapil A (2019).** Emergence of Burkholderia cepacia in ICU Setting. Indian journal of critical care medicine: peer-reviewed, official publication of *Indian Society of Critical Care Medicine*, 23(9):423-426.
- **Peralta DP, Chang AY, Ariza-Hutchinson A, Ho CA. (2018).** Burkholderia multivorans: A rare yet emerging cause of bacterial meningitis. *IDCases*, 11:61-3.
- **Rastogi N, Khurana S, Veeraraghavan B, Inbanathan FY, Sekar SK, Gupta D, et al. (2019).** Epidemiological investigation and successful management of a Burkholderia cepacia outbreak in a neurotrauma intensive care unit. *International Journal of Infectious Diseases*, 79:4-11.
- **Tavares M, Kozak M, Balola A, Sá-Correia I. (2020).** Burkholderia cepacia complex bacteria: a feared contamination risk in water-based pharmaceutical products. *Clinical microbiology reviews*, 33(3):10-128.
- **Varadkar S, Bien CG, Kruse CA, Jensen FE, Bauer J, Pardo CA, et al. (2014).** Rasmussen's encephalitis: clinical features, pathobiology, and treatment advances. *The Lancet Neurology*, 13(2):195-205.