



Rare Central Nervous System Infection in an Infant with Immunodeficiency

Ketan Kumar¹ · Richa Singh¹ · Mukta Mantan¹

Received: 24 August 2023 / Accepted: 27 September 2023 / Published online: 19 October 2023
© The Author(s), under exclusive licence to Dr. K C Chaudhuri Foundation 2023

To the Editor: A 45-d-old boy was referred to us with a persistent central nervous system (CNS) infection. Born to second degree consanguineous parents, the child had received treatment for early onset sepsis and pneumonia, followed by poor weight gain at home. On day 24, he was re-admitted with apnea and cyanosis. Cerebrospinal fluid (CSF) examination suggested meningitis. Blood and CSF cultures grew *Candida pelliculosa* and *Burkholderia cepacia* respectively. There was ventriculomegaly with CSF ooze and diffuse meningeal enhancement on neuroimaging. At presentation, the child had no CNS deficits.

He was started on intravenous levofloxacin, co-trimoxazole and amphotericin B. HIV serology of the mother was non-reactive. Workup was sent for chronic granulomatous disease (CGD).

After 7 d of therapy, there was further elevation in CSF protein. Culture grew *Pseudomonas aeruginosa*. Even after changing antibiotics, the child developed raised intracranial tension. MRI brain showed multiple brain abscesses in bilateral frontotemporal and left parietal lobes. The child succumbed to septic shock on day 65 of life.

Before the child's demise, we received the results of dihydrorhodamine (DHR) test, suggestive of CGD. The family, however, declined further genetic testing. They were advised to go for a formal prenatal genetic consultation in any future pregnancy.

CGD and cystic fibrosis (CF) are known to increase susceptibility to *Burkholderia*. However, CNS involvement

is uncommon. Among the three largest series of CGD patients, a single case of *B. cepacia* meningitis was reported from the United States [1–3]. The organism was reportedly isolated from the CSF in three neonates at a tertiary NICU in our country [4]. It is unclear whether they were worked up further for any underlying condition.

This case report highlights that *B. cepacia* is a potential CNS pathogen and its isolation warrants aggressive treatment along with early testing for CGD.

Declarations

Conflict of Interest None.

References

1. Winkelstein JA, Marino MC, Johnston RB Jr, et al. Chronic granulomatous disease. Report on a national registry of 368 patients. *Med (Baltim)*. 2000;79:155–69.
2. van den Berg JM, van Koppen E, Ahlin A, et al. Chronic granulomatous disease: the european experience. *PLoS ONE*. 2009;4:e5234.
3. Rawat A, Vignesh P, Sudhakar M, et al. Clinical, immunological, and molecular profile of chronic granulomatous disease: a multi-centric study of 236 patients from India. *Front Immunol*. 2021;12:625320.
4. Patra S, Bhat YR, Lewis LE, et al. *Burkholderia cepacia* sepsis among neonates. *Indian J Pediatr*. 2014;81:1233–6.

Publisher's Note Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.

✉ Ketan Kumar
ketankrpatwari@gmail.com

¹ Department of Pediatrics, Maulana Azad Medical College and Associated Hospitals, 2 – Bahadur Shah Zafar Marg, New Delhi 110002, India