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Case Report

Brain abscess after meningioma removal caused by *Citrobacter freundii* infection in an adult

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ABSTRACT

Background: *Citrobacter* species are an unusual cause of cerebral abscess in infant. In particular, *Citrobacter freundii* can invade and replicate in human brain microvascular endothelial cells with a selective neurovirulence, producing ventriculitis and brain abscess mainly in the infant. A delayed brain abscess caused by *C. freundii* species in adult patients and after surgery is an occurrence that has not yet been reported in the literature.

Case Description: The authors reported a case of a 60-year-old patient that presented a delayed postoperative brain abscess following resection of a left parietal convexity meningioma. A resurgery was performed, with bone flap removal, debridement, and culture of the purulent content of the previous surgical cavity. The microbiological examination showed the isolation of *C. freundii*. Postoperatively, the patient improved, with progressive headache reduction and right upper limb weakness improvement. She was continued on medical therapy for 4 weeks, until inflammatory index and white blood cells count gained normal range, then, she was admitted to a neurorehabilitation center.

Conclusion: A delayed brain abscess caused by *C. freundii* in adult patients and after surgery is an occurrence that has not yet been reported in the literature, with a consequent complex management, due to the lack of clear guidelines.

Keywords: Abscess, Brain, Citrobacter, Meningioma

INTRODUCTION

Citrobacter freundii is a facultative anaerobic Gram-negative bacterium, part of the Enterobacteriaceae, physiologically found as commensal in the intestinal human tract. Unlike other organisms causing meningitis, *C. freundii* can invade and replicate in human brain microvascular endothelial cells with a selective neurovirulence, producing ventriculitis and brain abscess mainly in the infant, with a high mortality rate.[1,6] About 1.3% of neonatal meningitis cases are caused by *Citrobacter* infection, with a mortality rate approximately of 30%. Surviving children, in most cases, develop a wide range of neurological deficits.[4] Brain abscesses related to

this bacterium are rarely found in adulthood. Management of these infections may include surgical drainage with targeted antibiotic therapy for a minimum of 21 days.[6] Herein, the authors report the case of an adult patient with a delayed postoperative intracerebral *C. freundii* abscess with a relevant literature review.

CASE DESCRIPTION

A 60-year-old female patient was admitted to our unit after a right upper limb weakness and severe headache. She underwent brain computed tomography (CT) scan and a subsequent magnetic resonance imaging (MRI) that documented a left extra-axial parietal lesion with intense contrast-enhancement after gadolinium administration [Figures 1a-c]. Then, she underwent a left parietal craniotomy and a microsurgical removal of the lesion. Histological examination documented a meningothelial meningioma (WHO Grade I). She was discharged after 1 week in good clinical status and 1-month postoperative MRI documented a complete removal of the lesion [Figures 2a-c]. After 1 month, the patient was admitted to the emergency department of another institution because she presented headache, confusion, speech disorder, and a severe right upper limb hyposthenia. An urgent brain MRI showed a widespread and irregular contrast-enhancement around surgical resection boundaries with leptomeningeal and dural thickening as well as a cavity hyperintense on T1-WI, highly suggestive for a brain abscess [Figures 3a-c]. A resurgery was performed, with bone flap removal, debridement, and culture of the purulent content of the previous surgical cavity. The microbiological evaluation of the specimens showed the isolation of *C. freundii*. The patient started immediately a target antibiotic therapy with 3 times daily intravenous dose of meropenem 1 g and twice-daily intravenous dose of linezolid 600 mg.

Postoperatively, the patient improved, with progressive alleviation of headache and improvement of right upper limb weakness. She was continued on medical therapy for 4 weeks, until inflammatory index and white blood cells count gained normal range; then, she was admitted to a neurorehabilitation center. Three-month postoperative MRI documented a complete abscess removal and markedly reduced enhancement at surgical site [Figures 4a-c].

DISCUSSION

Citrobacter is facultative anaerobic Gram-negative bacilli, from the family of Enterobacteriaceae. The most common bacillus is represented by *Citrobacter koseri*, followed by *C. freundii*, that represents 4.6% of children meningitis. It is responsible for infections of multiple sites, especially in patients with immunodeficiencies, even the brain, causing meningitis or intracerebral abscesses. Vertical (mother to child with the onset of meningitis) and horizontal (nosocomial late occurrence of brain abscess) transmissions are the most common way of infections described. The mortality related to *Citrobacter* abscess is high with a rate up to 30%.[4] Nevertheless, it is highly recommended to perform

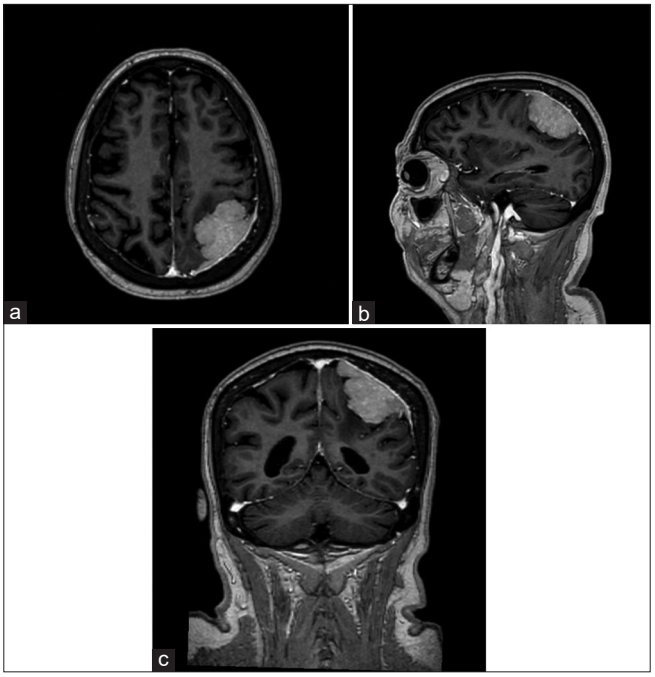

Figure 1: Brain magnetic resonance imaging T1-weighted axial (a), sagittal (b), and coronal (c) images with gadolinium documented a left postrolandic convexity meningothelial meningioma.

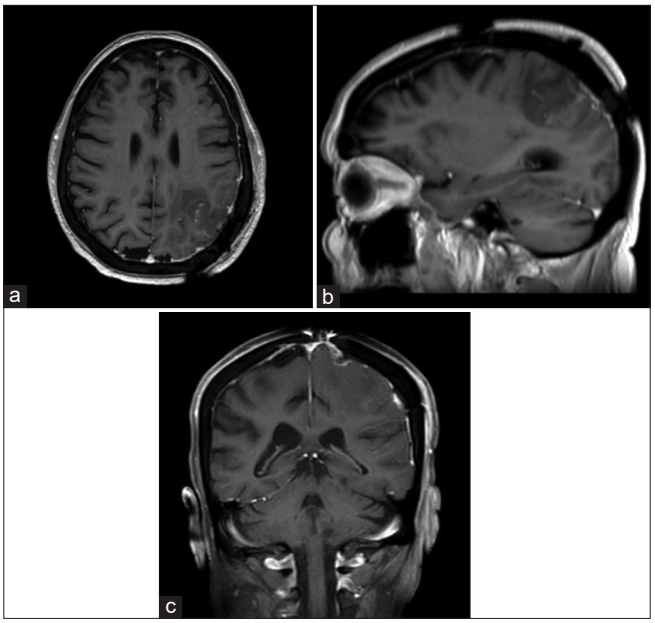

Figure 2: Early postoperative brain magnetic resonance imaging T1-weighted axial (a), sagittal (b), and coronal (c) images documented a complete meningioma removal.

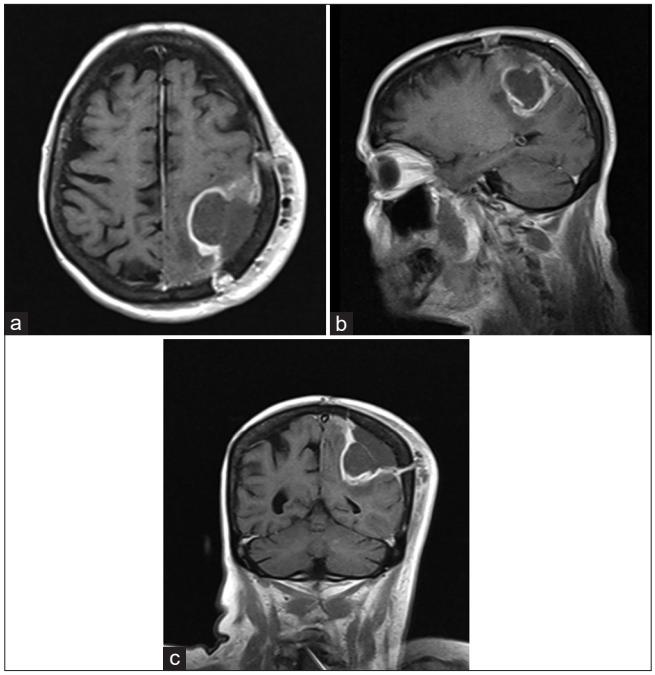

Figure 3: Brain magnetic resonance imaging T1-weighted axial (a), sagittal (b), and coronal (c) images with gadolinium documented an irregular contrast enhancement around surgical resection boundaries with leptomeningeal and dural thickening as well a uniform hyperintense surgical site, highly suggestive for brain abscess.

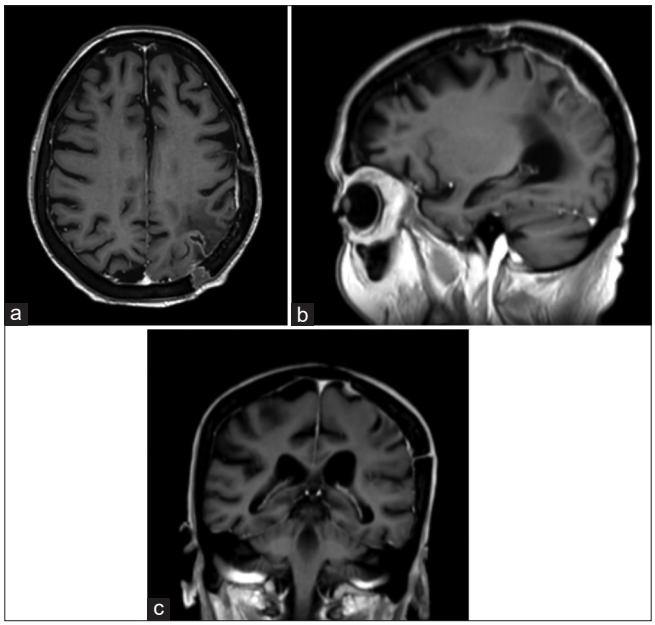

Figure 4: Postoperative brain magnetic resonance imaging T1-weighted axial (a), sagittal (b), and coronal (c) images with gadolinium documented a complete abscess removal and markedly reduced enhancement at surgical site.

brain imaging (MRI or CT scan) in all the infants with *Citrobacter* meningitis.[2-4]

Brain contrast-enhanced ultrasound is useful as a bedside or intraoperative tool, especially in clinically unstable patients.

As in the present case, the most common symptoms are fever, headache, and focal neurological deficits.

Drainage of the abscess and antibiotic therapy for a minimum of 21 days is mandatory to eradicate the infection. Indeed, the most used and effective antibiotics are cephalosporins, imipenem, chloramphenicol, trimethoprim-sulfamethoxazole, and gentamicin, defining as best combination of meropenem and ciprofloxacin due to an optimal penetration of blood–brain barrier and into neutrophils.[6]

In cases of sepsis, drugs with susceptibility can be chosen, adjusting the dose, treatment interval, and infusion time for nonsusceptible drugs, considering the toxicity in neonatal age. Another important point is to rationalize the use of antibiotics to reduce multidrug resistance.[6]

The goals of abscess surgical drainage include removal of the mass effect and related midline brain shift, and reduction of the bacterial load, thus improving efficacy of a systemic antibiotic therapy.[2,3]

The pathogenesis of abscess is probably related to a primary vasculitis of white matter with a consequent bacterial invasion. Indeed, according to Badger *et al*., the mechanism of infection and spreading of this bacterium is strictly related to the invasion of human brain microvascular endothelial cells through microfilaments, *de novo* proteins, microtubules, and endosome acidification, with a subsequent intracellular replication. An outer membrane protein in *Citrobacter* may also led to neurovirulence, producing ventriculitis and brain abscess. Hence, these findings could probably explain the high mortality related to this infection.[1]

As previously explained, *C. freundii* is frequently associated to severe meningitis and brain abscesses, due to its invasive ability, mainly in the infant.[1,3,5] Accordingly, the occurrence of a brain abscess in an adult patient can be considered infrequent and rarely mentioned. In rare cases, during the infancy, *C. freundii* meningitis may cause aqueduct stenosis and subsequent obstructive hydrocephalus.[4] A literature review identified just one case of *C. freundii* abscess that mimics a breast metastasis in an adult female patient.[2] Conversely, their management should follow the same recommendation reported for infant patients: brain imaging, immediate surgery, and target antibiotic therapy. Therefore, a prompt diagnosis should be mandatory to obtain the best outcome, considering that this *Citrobacter* spp. in rare cases can cause central nervous system infections even in adults.

CONCLUSION

The ability to invade and replicate in human brain microvascular endothelial cells makes *C. Freundii* an insidious and dangerous bacterium, rarely found in adulthood. A delayed brain abscess caused by *C. freundii* in adult patients and after surgery is an occurrence that has not yet been reported in the literature, with a consequent confused and difficult management, due to the lack of clear guidelines. Early diagnosis and effective antibiotic therapy are essential, but many cases also require surgical intervention.

Declaration of patient consent

Patient's consent not required as patient's identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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