Received: 30 August 2023 | Revised: 9 October 2023 | Accepted: 13 October 2023

DOI: 10.1002/ccr3.8111

CASE REPORT

Clinical Case Reports Open Access WILEY

Management strategy in actinomycosis brain abscess

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Key Clinical Message

We reported herein a case of isolated cerebral actinomycosis in a 54-year-old immunocompetent man. Brain MRI showed a left frontal intra-axial lesion and perilesional edema. We performed an open biopsy of the left frontal enhancing lesion. Intraoperative findings showed a yellowish, malleable, and capsulated lesion that was well defined with surrounding normal tissue within pus inside and lacked any necrotic content. MR spectroscopy showed a high level of choline, lactate, and lipid peaks with a choline/N-Acetylaspartic acid ratio of 1.8. The diagnosis was confirmed histologically, and the patient was treated successfully for 3 months after surgical aspiration. Surgical management allowed to confirm the diagnosis with a shorten antibiotics, a rapid resolution of symptoms, and a complete recovery.

KEYWORDS

actinomycosis, brain abscess, magnetic resonance imaging, surgery

1 | INTRODUCTION

Cerebral or brain actinomycosis is a rare bacterial infection that affects the brain and surrounding tissues. It is caused by the *Actinomyces* species which are anaerobic Gram-positive bacteria. They are commonly found in the cervicofacial and digestive tract.[1] This bacterium can enter to human body through a wound or injury, or they can spread from an existing infection. Cerebral actinomycosis is most commonly reported in patients with immunosuppression, such as patients living with human immunodeficiency virus (HIV) or patients with underlying malignancies, or those who have had a recent surgery or dental procedure.[2] Cerebral actinomycosis can lead to severe complications if not treated promptly. In this current paper, we reported a case of cerebral actinomycosis in immunocompetent patient with no risk factors diagnosed on histological examination after elective biopsy.

2 | CASE REPORT

A 54-year-old man with no past medical history was admitted to our department for a first episode of generalized tonic-colonic seizures. There are no ocular symptoms neither gait disorders; neurologic examination showed a Glasgow Coma Scale score of 15/15 (Eye opening = 4, Verbal response = 5, Motor response = 6) with normal physical examination. On admission, he is afebrile, with a blood pressure of 140/80 mm Hg, a regular heartbeat at 80/min and normal oxygen saturation. The patient displayed only neurologic presentation, with no concomitant respiratory or gastro-intestinal clinical features or symptoms.

Routine laboratory findings such as complete blood count, kidney and liver function tests were normal. C-reactive protein value was elevated at 45 mg/L. Peripheral blood cultures were negative. Further investigations including serology for HIV, hepatitis B and C hepatitis and toxoplasmosis were negatives. QuantiFERON-TB Gold test was also negative. An initial brain computed tomography (CT) scan showed the presence of an intra-axial left frontal tissue lesion of 22.7×18.5 mm with cocardial enhancement surrounded by perilesional edema without mass effect (Figure 1). A follow-up brain MRI examination performed 1 week later showed a left frontal intra-axial lesion and perilesional edema. The lesion showed ring or nodular enhancement, and there was no diffusion restriction (Figure 2). Magnetic resonance spectroscopy (MRS) showed an increased choline level and elevated lactate and lipid peaks with a choline/N-Acetylaspartic acid (NAA) ratio of 1.8. A lumbar puncture (LP) was not performed. We conducted an elective biopsy for the lesion in the left front lobe. The intraoperative diagnosis revealed a yellowish, malleable, and capsulated lesion that was well defined with surrounding normal tissue within pus inside and lacked any necrotic content. Gross examination showed an abscessed cystic specimen measuring 3×3×2 cm with purulent contents and surrounded by a thick fibrous capsule. Upon histopathological analysis, microscopy revealed a brain parenchyma remodeled by foci of suppurative necrotizing processes. Filamentous organisms appearing as radiating rosettes and staining positively with periodic acid-Schiff (PAS) are observed in the central areas of the abscess, thus corresponding to sulfur granules of actinomycoses (Figure 3). These findings were consistent with actinomycosis brain abscess. No granulomas or tumor proliferation were seen. Unfortunately, direct bacteriological and mycological examinations were not performed. The patient started antimicrobial drugs (amoxicillin clavulanate 6 g by intravenous (IV) infusion/d) for 2 weeks associated with levetiracetam (500 mg twice/day) orally. The outcome was favorable, with a progressive improvement of his neurologic status. The patient was discharged on the same oral treatment was maintained for a total of 3 months with a complete recovery, with no evidence of relapse afterwards; the patient received oral levetiracetam, and IV infusion of amoxicillin was changed to oral administration. Thus, the patient was regularly seen at our outpatient clinics. Follow-up brain MRI at 3 months did not show recurrence of cerebral lesions. He was free of symptoms with a normal neurological examination.

3 | DISCUSSION

Cerebral actinomycosis is a rare but serious condition that is caused by the bacteria *Actinomyces. A. israelii* is the most common causative strain isolated from human specimens,[3,4] which is commonly found in the oral cavity.[1] Incubation for at least 10 days in strictly anaerobic culture conditions is required for the isolation of *Actinomyces* spp.[5] The biggest limitation is that bacteriological culture was not obtained to confirm *Actinomyces* spp. This would have allowed antimicrobial susceptibility testing to guide treatment. Bacteriological culture should be attempted even if *Actinomyces* is fastidious to grow. This is essential for definitive diagnosis and tailoring antimicrobial therapy.

The oro-cervical variety is approximately 50% of all reported instances.[6] It is typically caused by dental intervention or trauma to the mouth. Anamnestic investigations in our patient concluded to the absence of recent dental procedure. Other localizations include thoracic (15%–20%) and abdomino-pelvic (20%) actinomycosis, whereas the central nervous system (CNS) is uncommon and the

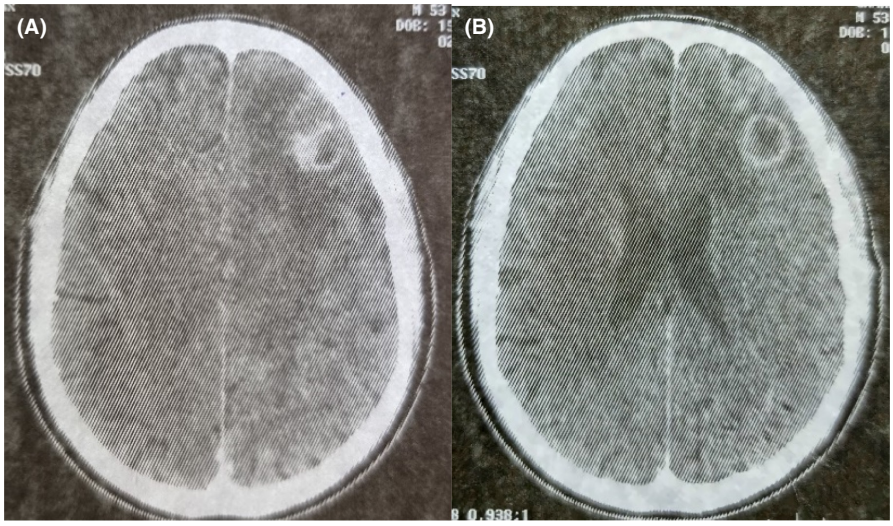

FIGURE 1 Cerebral computed tomography (axial section) (pretreatment) showed an intra-axial localized lesion in the left frontal tissue with cocardial enhancement surrounded by perilesional edema without mass effect.

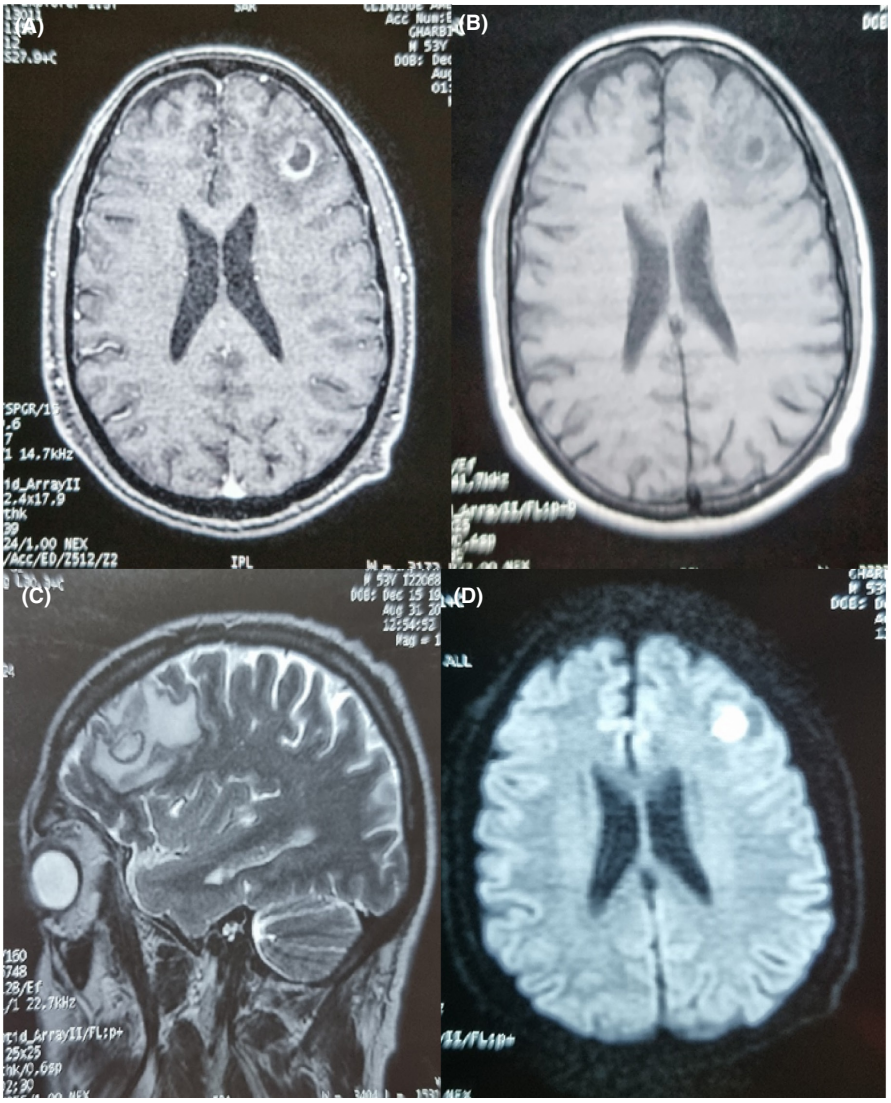

FIGURE 2 Cerebral magnetic resonance imaging (MRI) concluded that lesions are hypointense on T1-weighted images and hyperintense on T2-weighted images with mild perilesional edema (A–B–C). The lesions show ring or nodular enhancement, and there is no diffusion restriction (A–D).

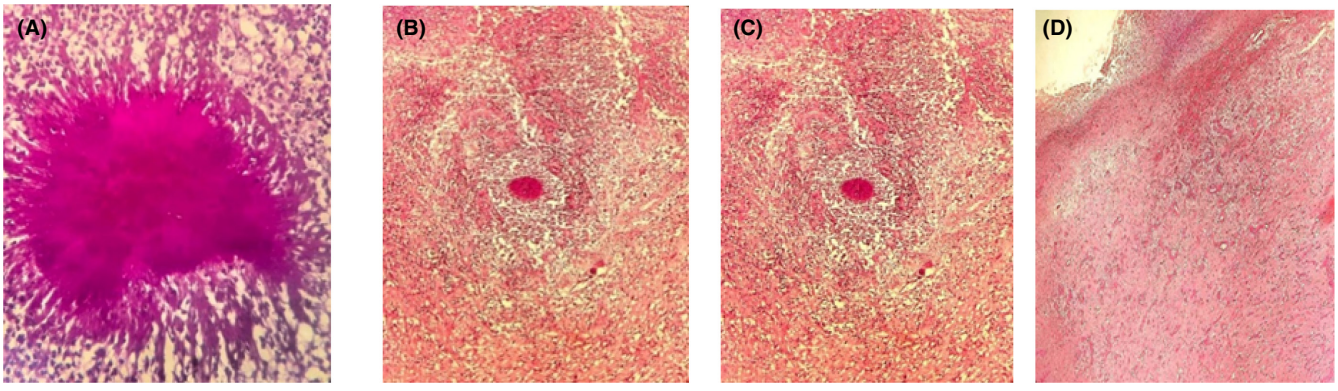

FIGURE 3 Acute inflammation of brain parenchyma (A), low power view of fibrillary organism surrounded by a mixed inflammatory infiltrate (HE stain) (B), (inset) high power view (C), and periodic acid-Schiff stain (D).

infection is typically caused by hematogenous spread or direct extension from the oro-cervical region.

CNS actinomycosis can present brain abscess, meningitis/meningoencephalitis, actinomycoma, subdural empyema, or spinal and cranial epidural abscess.[7–9] *Actinomycosis* can cause disseminated forms, by hematogenous spread and systemic involvement. In our case, CNS localization was primitive and no other site of infection was found. One important aspect of this condition is the diagnosis difficulty. The symptoms of cerebral actinomycosis can be similar to

those of other neurological disorders, and imaging studies may not always provide a suggestive or definitive diagnosis. Medical literature regarding or studying radiological findings of actinomycotic brain abscesses is scarce.[12,13] Imaging descriptions in case reports frequently lack specificity about defining radiological characteristics.[11,14] Actinomycotic brain abscesses have been described as having a hypointense core and a rim that is hyperintense on T1 non-contrast imaging[12,13] and hypointense on T2 images. They have also been described as being irregular, thick and nodular,[13] or thin[12,14] peripherally enhancing lesions.[13] Although restricted core diffusion is frequently observed,[12,13] it is not the norm. The dissemination may not be constrained by the perimeter.[12,13] In our patient, MRS showed a high level of choline, lactate, and lipid peaks, which could be observed in swollen demyelinating lesions and high-grade tumors. Additionally, the histopathological diagnosis of actinomycosis is difficult because tissue specimens have typically few sulfur grains, and because cultures are negative in approximately 70% of the cases.[10,11]

The standard course of treatment for actinomycetal infections consists of 2–6 weeks of intravenous penicillin followed by 6–12 months of oral penicillin or amoxicillin. Erythromycin, tetracycline, doxycycline, minocycline, and clindamycin are alternatives for patients who are allergic to penicillin. All of the antibiotics described above, however, have varying degrees of CNS permeability.[15] In case of brain abscesses caused by *Actinomyces*, surgery leads to identify the pathogen and their susceptibility, lowers the bacterial load and environmental stress, avoids hernias, raises the oxygen tension in sick tissue, and facilitates antibiotic penetration.[15] The aspiration of brain abscess using the burr hole technique is said to enhance the penetration of antibiotics into the abscess cavity and may shorten the course of antibiotic therapy, according to a prior case report of *A. israelii* brain abscess.[16] For our patient, the surgical team decided to perform a craniotomy and excision and we decided to treat for a shorter period of 3 months after surgery. The rationale for the three-month antibiotic duration is based on the fact that the associated surgery was considered successful, with a reduction of bacterial inoculum following the procedure, as is the case with brain abscesses caused by bacterial pathogens. Although no consensus regarding treatment has been established, associated surgical management allowed this patient to be successfully treated with a shorter course of antibiotics.

4 | CONCLUSION

Cerebral actinomycosis can occur in immunocompetent patients despite the absence of an obvious portal of entry. This rare pathology raises a real diagnostic problem regarding the absence of specific imaging findings. Surgery management can help physicians by confirming the diagnosis, draining the bacterial inoculum, and thereby shortening the duration of antimicrobial drugs.

AUTHOR CONTRIBUTIONS

Mohamed Dehmani Yedeas: Conceptualization; data curation; investigation; resources; writing – original draft. **Mohamed Amine Rachdi:** Methodology; resources; software; writing – original draft. **Souheil Zayet:** Writing – review and editing. **Rahma Yaiche:** Resources; software; supervision; validation; visualization. **Ridha Chkili:** Supervision; validation; visualization; writing – review and editing.

FUNDING INFORMATION

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

Data are available on request due to privacy restrictions. The data presented in this case study are available on request from the corresponding author.

ETHICS STATEMENT

Research Ethics Committee of the Military Hospital of Tunis has approved the work and confirmed that it does not transgress ethics.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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How to cite this article: Yedeas MD, Rachdi MA, Zayet S, Yaiche R, Chkili R. Management strategy in actinomycosis brain abscess. *Clin Case Rep*. 2023;11:e8111. doi:10.1002/ccr3.8111