

Case Report

Cryptococcus neoformans infection in a post-operative mastoid cavity of an immunocompetent individual: a rare case report

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Received: 15 May 2020

Accepted: 12 June 2020

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ABSTRACT

This is a case report of isolated cryptococcosis in the operated mastoid cavity of an immunocompetent individual. A 24 year old immunocompetent male who underwent modified radical mastoidectomy presented to the OPD with complaints of otalgia and recurrent otorrhoea, not responding to regular medical line of treatment and aural toileting. Microscopy and culture of the ear discharge revealed *Cryptococcus neoformans*. The patient was treated with oral and topical fluconazole and recovered completely. Although rare, cryptococcal mastoiditis should be considered even in an immunocompetent patient, as any delay in diagnosis and treatment can lead to fatal complications.

Keywords: Cryptococcosis, Cryptococcal mastoiditis, Fluconazole, Immunocompetent host, Fungal mastoiditis

INTRODUCTION

Open cavity mastoidectomy or canal-wall down procedure is one of the main techniques in cholesteatoma surgery. The perceived advantage is that it has a lower rate of recurrent and residual cholesteatoma compared to intact canal wall mastoidectomy.¹ However, the disadvantage is that there may be recurrent accumulation of cerumen and debris in an open mastoid cavity that requires regular cleaning.² Another problem of an open mastoid cavity is discharge and infections.³ Usually, this discharge is tissue fluid that leaks from the open mastoid cavity, which acts as a rich medium for bacterial and fungal growth. Unlike other areas of the head and neck, the mastoid, especially the sclerotic mastoid is not well vascularised. The combination of weeping tissue fluid and mediocre blood supply makes controlling infections of the mastoid challenging. Of all the factors associated with infections in the mastoid cavity, moisture plays the most important role along with debris.^{4,5} This case report describes a rare isolated infection of *Cryptococcus*

neoformans in the mastoid cavity of an immunocompetent individual who was operated 12 years back.

Cryptococcosis, formerly known as torulosis, European blastomycosis, or Busse-Buschke disease, is caused by *Cryptococcus neoformans*. *Cryptococcus* is a spherical-to-oval, encapsulated, yeast-like fungus that is widespread in spoiled milk, soil and bird droppings, especially pigeon excreta.

Fungal mastoiditis is usually seen in an immunocompromised host, though few cases have been reported in immunocompetent hosts as well.^{6,7} *Cryptococcus neoformans* as a cause of fungal mastoiditis is extremely rare and all cases reported till now have been in immunocompromised hosts; however to our knowledge, this is the first reported case in an immunocompetent individual. Recurrent otorrhoea and other signs and symptoms suggestive of mastoiditis, which are not responding to regular medical line of

treatment, should raise a high clinical suspicion in the minds of the surgeon and should include cryptococcal mastoiditis as a differential diagnosis in such cases.

CASE REPORT

A 24 year old male who had undergone modified radical mastoidectomy 12 years back at our institute reported to the OPD with complaints of left otalgia and otorrhoea since 1 month. Patient was asymptomatic prior to these complaints; however, he did not have any follow-up with us in all these years after the surgery.

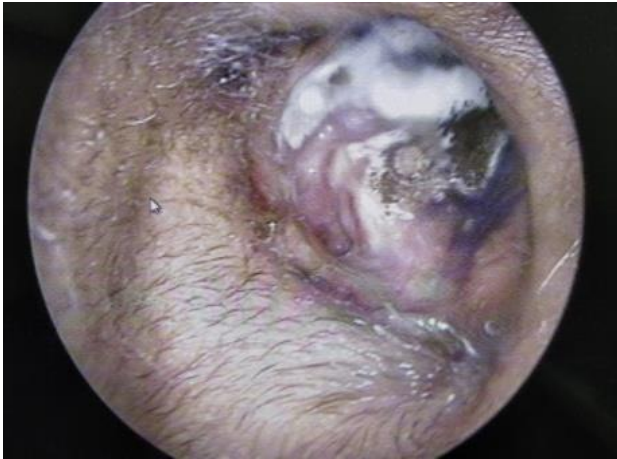


Figure 1: Otoendoscopy of the left ear mastoid cavity showing fungal mastoiditis.

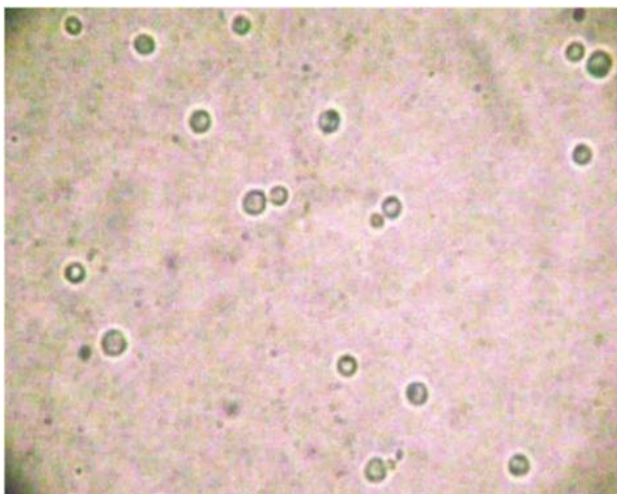


Figure 2: *Cryptococcus neoformans* on India ink staining.

Aural toileting was done and regular oral and topical antibiotic and topical antifungal (clotrimazole) along with oral analgesics were prescribed for the otalgia and otorrhoea. This went on for another 2 visits within a span of 2 weeks, however the patient had no relief in his symptoms; hence a swab was taken for staining and culture. Initial reports of KOH mount and negative India ink staining were suggestive of “budding forms of

cryptococcus”. Culture confirmed the growth of *Cryptococcus neoformans* (Figure 1-3).



Figure 3: *Cryptococcus neoformans* colonies on sabourauds dextrose agar.

He was further evaluated for any causes of immunocompromised states. Routine blood investigations included a normal complete blood haemogram and normal blood sugar levels. Neither he nor his family had any history of blood dyscrasias; there was no history of steroid use, cancer, chemotherapy or radiotherapy. He was serologically non-reactive for HIV.

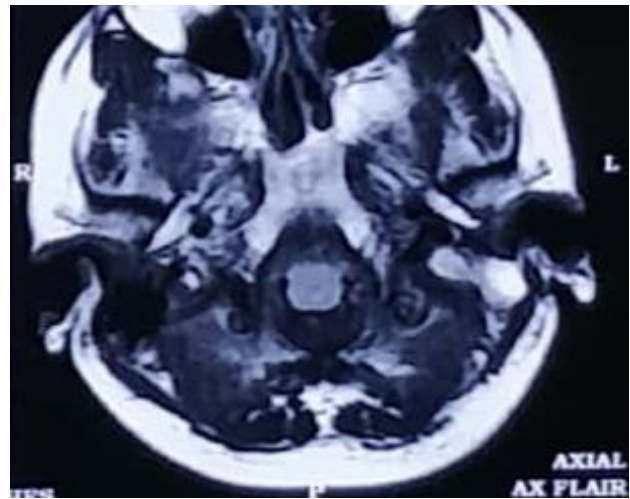


Figure 4: MRI brain with temporal bone showing mild soft tissue thickening along the walls of left mastoid cavity.

Cavity was viewed microscopically and endoscopically. It was found to be well epithelialised without any evidence of granulation tissue or gross bony erosions. High resolution computed tomography of the temporal bone showed no evidence of bony erosion, similarly Magnetic resonance imaging of the brain and temporal bones revealed mild soft tissue thickening along the walls

of the cavity with intact inner ear apparatus. No intracranial extension or lesions were noted in the brain parenchyma. Neurological examination revealed no abnormality. Radiograph of the chest was normal suggesting no pulmonary involvement (Figure 4).

Patient was started on oral fluconazole 200 mg once daily and topical fluconazole ear drops for two weeks with aural toileting every week. As patient showed remarkable response and complete relief of symptoms within two weeks of starting oral fluconazole therapy, it was decided to continue the same for two more weeks. There was complete remission of symptoms and swab culture done four weeks after starting the initial therapy did not show any growth of organism. Patient responded well to the therapy without any adverse effects. He was asked to follow up every month subsequently and advised to strictly keep the ear dry with due water precautions to be taken. To rule out window period of HIV infection, he was tested again three months after completion of therapy and was reported as non-reactive again. He has been symptom free for the past 6 months, without any evidence of fungal infection.

DISCUSSION

Cryptococcus neoformans is an opportunistic yeast that has predilection for the pulmonary and the central nervous system.⁸ It mainly affects the immunocompromised patients, especially those having HIV. Skeletal cryptococcosis is a rare entity that can either spread secondary to disseminated cryptococcosis or as a result of direct inoculation through fungal spores.⁹ This case appears to be a result of direct inoculation of fungal spores into the mastoid cavity. The factors that might have contributed to the manifestation of the fungus within the cavity could be an optimum body temperature of 37° C, no regular cleaning of the cavity which led to accumulation of debris, moisture and poor patient hygiene.

The usual problems of the post operative mastoid cavity like inadequately lowered facial ridge, unsaucerized cavity, narrow meatus and deep mastoid tip were not encountered in this case. Cases of osteomyelitis of the skull have been reported due to cryptococcosis leading to cryptococcal meningitis,⁹ but in this case no bony erosion or intracranial extension was noted and the same was confirmed radiographically.

Amphotericin B and flucytosine are the recommended treatment options along with surgical debridement in immunocompromised and severe cases.^{9,10} However considering the immunocompetent status of the patient and as the infection was limited to the mastoid cavity, we started the patient on oral fluconazole 200 mg single daily dose initially for two weeks, continuing the same for two more weeks after completion of the initial fourteen days of the therapy, along with topical fluconazole and weekly aural toileting.¹⁰ Other newer

antifungals highly active against cryptococcus which could have been tried are posaconazole and voriconazole, but clinical experience with these agents is limited.¹⁰

But as our experience with fluconazole was so emphatic and satisfying, the clinician should consider this option as the first line of treatment in a case of isolated fungal mastoiditis instead of experimenting with other higher and more expensive antifungals like amphotericin B straight away.

CONCLUSION

Although cryptococcus can affect any organ or part of the body, the mastoid cavity is an unusual site of infection. A strong clinical suspicion should arise in the minds of the surgeon when dealing with operated mastoid cavities which are chronically discharging since direct inoculation of cryptococcus in the cavity might lead to manifestation of the disease and not necessarily spread via the hematogenous route or because of disseminated cryptococcosis. Immune status and severity governs the treatment of such extra pulmonary cases without central nervous system involvement. Our patient having an intact immune status responded well to oral and topical fluconazole therapy and we recommend the same as the first line of management in such cases.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

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Cite this article as: Gosavi RS, Gupta AD, Gosavi SS, Phatak JU. *Cryptococcus neoformans* infection in a post-operative mastoid cavity of an immunocompetent individual: a rare case report. *Int J Otorhinolaryngol Head Neck Surg* 2020;6:1393-6.