Case Report

A case report of osteoarticular mucormycosis in post-COVID-19 patient

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ABSTRACT

Mucormycosis is a rare invasive fungal disease often seen in immunocompromised individuals. The mucormycosis cases increased in COVID-19 patients from March to June 2021. Here, we report the case of a 61-year-old male hypertensive patient with COVID-19 who was treated with antibiotics and steroids for recovery. After treatment, he developed a secondary infection of osteoarticular mucormycosis which is uncommon and rare. We report this case here in detail.

Key words: COVID-19, Osteoarticular mucormycosis, Treatment

The outbreak of coronavirus disease as a pandemic has spread rapidly on a global scale. With more than 1 year into this pandemic, the definitive treatment of COVID-19 continues to be controversial. However, steroids have shown some survival benefits. On the other side, the widespread use of steroids (glucocorticoids) has led to secondary bacterial or fungal infections [1]. The bacterial and fungal infection may be associated with pre-existing morbidity (diabetes mellitus and lung disease) or may develop as a hospital-acquired infection such as ventilator-associated pneumonia [2]. Recently, several cases of mucormycosis in people with COVID-19 have been increasingly reported worldwide, particularly from India. Mucormycosis or zygomycosis, also called phycomycosis, initially described in 1885 by Paltauf, is an uncommon and aggressive fungal infection that usually affects patients with alteration in their immune system (immunocompromised) [3]. The major risk factors for mucormycosis included uncontrolled diabetes mellitus in ketoacidosis, other forms of metabolic acidosis, use of corticosteroids, organ transplantation, trauma, malignant hematologic disorders, and deferoxamine therapy in patients receiving hemodialysis [4,5]. This angioinvasive fungal disease can present with varied clinical manifestations, including rhino-orbital-cerebral, pulmonary, cutaneous, gastrointestinal, disseminated, and miscellaneous forms [6] in which rhinocerebral presentation was the most common form of mucormycosis [7]. The mortality rate depends on the underlying disease of the infection site [8]. Globally, the prevalence of mucormycosis varied from 0.005 to 1.7 per million populations, while its prevalence is nearly 80 times higher (0.14/1000) in India as compared to developed countries, as shown in a study conducted in the year 2019–2020 [3,9].

We report a case of osteoarticular mucormycosis with soft-tissue involvement which is an uncommon manifestation seen in a patient with COVID-19.

CASE REPORT

A 61-year-old male hypertensive patient presented to the tertiary care hospital with the complaint of fever along with weakness and dyspnea for the past 7 days and 2 days, respectively, before admission.

On admission, his temperature was 100.8° F, pulse rate was 79/min, blood pressure was 164/90 mmHg, respiratory rate was 20/min, and SpO_2 was 88% on ambient air.

Then, his blood samples were collected for routine pathological testing and a nasopharyngeal swab was sent for COVID reverse transcription-polymerase chain reaction, which later on came positive for COVID-19. He was monitored in intensive care unit (ICU) and along with supportive therapy, intravenous dexamethasone was given for 7 days as a part of COVID-19 management. After 7 days stay in the ICU, he was shifted to the general ward. On discharge, oral corticosteroids were continued for 3 weeks in tapering dosage. Dexamethasone was administered by another hospital before presenting to us.

After 2 weeks from discharge, the patient had a complaint of back pain along with pain over the right lower limb. Magnetic

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resonance imaging (MRI) of the lumbosacral spine with whole spine screening was suggested on consulting with a local physician. Results showed no other abnormalities except age-related degenerative changes. In view of persistent symptoms, MRI screening of both hip and sacroiliac joints was repeated after 2 weeks which shows marrow edema along the periarticular sacral and iliac aspect of the right S.I joint suggestive of acute sacroiliitis, edema with small loculated fluid collection (abscess formation) in the right piriformis muscle, marrow edema involving right acetabulum, moderate synovial effusion in the right hip joint, and soft-tissue edema in the right gluteal and adductors muscles. Sonography of the right hip joint showed thickening of anterior recess with fluid collection suggestive of infective synovitis. There was also fluid collection underneath the gluteus muscle on the right side measuring approximately 3 mm × 19 mm × 12 mm.

USG-guided pus aspiration was done and sent for the microbiological examination which includes KOH, Gram stain, fungal culture, Gene-Xpert, and acid-fast bacillus Mycobacteria Growth Indicator Tube culture for *Mycobacterium tuberculosis*. All microbiological tests came negative and no causative agents were detected by these tests. Empirical AKT (HRZE) was initiated by a local doctor. The patient was not improving and consulted various doctors in which various antibiotics (cefoperazone+sulbactum and amikacin) were prescribed. Yet after that, symptomatically patient deteriorates.

The patient was then referred to an infectious disease specialist and it was almost 2 months from the initiation of complaints. On presentation, the patient had complaints of pain and tenderness over the right lower limb including the right hip joint and surrounding areas. He was in agony due to pain. There was the presence of tachycardia (pulse rate – 116/min) and elevated systolic blood pressure (BP – 140/80 mmHg), while the rest of the systemic examination was unremarkable. On local examination, there were tenderness and restriction of movement on the right hip joint.

On investigation, blood reports showed leukocytosis (TC – 10,930/mm^3) with relative eosinophilia (9.1% [1–6%]), with raised erythrocyte sedimentation rate of 88 mm/hr (range 2–20) and raised C-reactive protein – 32 (<6). His current MRI of the spine shows heterogeneous alteration in the sacrum and right pelvic bone. There was also periosseous collection, cortical, and marrow erosions with inflammatory soft-tissue edema (Fig. 1). These findings were suggestive of progression as compared to previous radiological reports.

The patient's Cat-I antitubercular medications were stopped and were prescribed analgesics and anti-allergic drugs for symptomatic management. After 5 days of off antibiotics, USG-guided aspiration of pus was done for microbiological testing which includes KOH, fungus culture, Gram-stain, AFB culture, Bactec Aerobic, Bactec Myco-F cultures, and Gene-Xpert. From which, Gram stain, AFB culture, and Gene-Xpert were negative, whereas, his Bactec fluid culture was positive. Gram-stain from positive Bactec Myco-F shows branched septate hyphae. Fungus culture of fluid by direct platting on Sabouraud Dextrose Agar (SDA) also shows grayish-white fluffy hyphal growth suggestive of mucormycosis (Fig. 2a). Fungus culture plated directly on solid media previously during the first aspiration of SDA, Rose Bengal Chloramphenicol Agar, and Potato Dextrose Agar also grew delayed mucor (5 days) from the same sample. Lactophenol cotton blue mount from growth revealed broad aseptate, branched, ribbon-like hyphae suggestive of *Mucor* species (Fig. 2b). *Mucor* in both specimens confirmed microbiological diagnosis. He was advised for IV amphotericin B along with wide surgical debridement. Debridement was done and the patient was put on IV amphotericin B. As per the infectious disease consultant, the patient was doing well. However, due to cost constraints, he went to a local hospital for further treatment and did not turn up for further follow-up.

DISCUSSION

Mucor species and *Rhizopus* species are saprophytic fungus/mold, therefore, when mucor/rhizopus is isolated in a clinical specimen, clinical correlation is a must. Growth of the same fungus from a repeat specimen is confirmatory of the presence of that specific fungus.

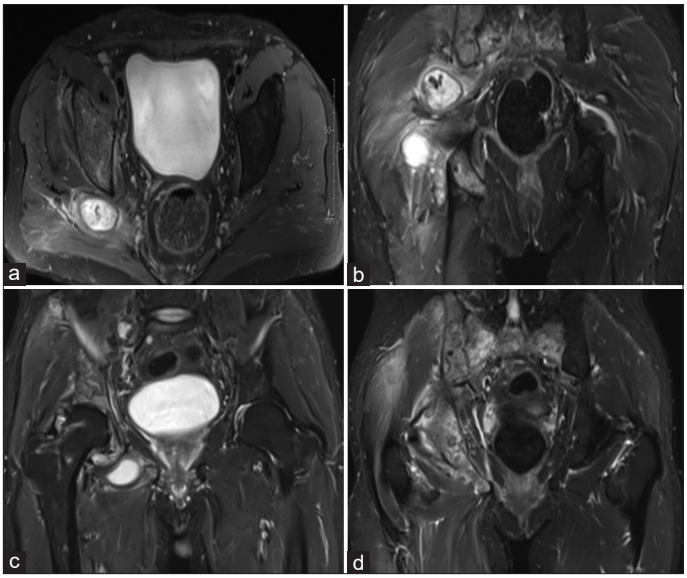

Figure 1: (a-d) Magnetic resonance imaging images of hips and joints showed heterogeneous alteration in sacrum and the right pelvic bone. There was also periosseous collection, cortical, and marrow erosions with inflammatory soft-tissue edema

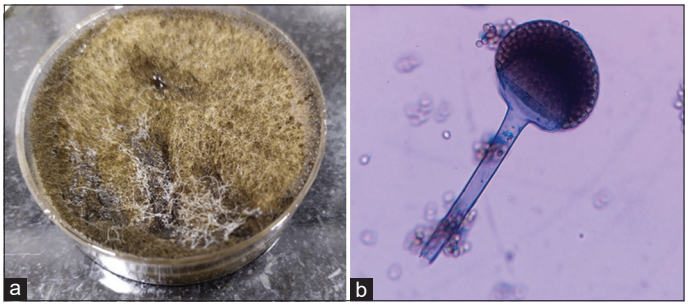

Figure 2: (a) Growth of mucor on Sabouraud Dextrose Agar plate. (b) Microscopic examination showed broad aseptate, branched, ribbon-like hyphae – *Mucor*

Mucormycosis is a rare, invasive fungal disease often seen in immunocompromised individuals; in patients with diabetic ketoacidosis, and in patients with concomitant use of steroids [9]. There were increasing case reports of mucormycosis among patients who were undergoing treatment of COVID-19. The COVID-19 infection is caused by the novel severe acute respiratory syndrome coronavirus 2 [2]. COVID-19-infected patients who were treated with the widespread use of steroids and recovered, developed secondary bacterial or fungal infection [10]. The most common cause for the rise of mucormycosis in COVID-19 patients are uncontrolled diabetes, the excessive use of corticosteroids, and long-term stays in the ICU.

The important characteristic of mucormycosis infections is that it can infect a wide variety of bones and joints with no real preferred site of infection. The site depends on the mechanism of infection, affecting the long bones after trauma or surgery, or a wide variety of bones after hematogenous dissemination [11]. The infection apparently could present in any clinical form, but disseminated mucormycosis was common and the mortality rate was high.

As in the present case, COVID-19 recovered patient was affected with mucormycosis. The site of infection was the right hip joint and the mechanism of infection can be disseminated. The involvement of the hip is an extremely unusual location for the development of mucormycosis. The reason behind mucormycosis was the use of corticosteroids during COVID-19 treatment. Initially, the patient has less pus collection in the right hip joint, which increased in volume suggesting active infection. In most of the cases, the time interval between COVID-19 and the initial mucormycosis was 10–15 days. This can be correlated with the present case where the patient had symptoms after 2 weeks of COVID-19. The patient did not have a history of previous surgery. The learning lessons here were: (i) All sites of the body are prone to mucormycosis. (ii) When a load of pus or debris is higher at the site of infection, there is a better recovery of microorganisms when loaded in automated blood culture bottles. (iii) Reporting such unusual cases after active communication with the clinician is the key to collect and disseminate such data among colleagues.

For the treatment of osteoarticular mucormycosis, antifungal therapy along with amphotericin B and surgical intervention is required in most cases. In the European Society of Clinical Microbiology and Infectious Diseases and Emergency and Critical Care Medicine joint guidelines, liposomal amphotericin B is recommended as the principal first-line agent for the treatment of mucormycosis [6,12]. COVID-19 followed by mucormycosis carries a very high mortality rate and timely detection, antifungal therapy, and aggressive surgical debridement remain key factors in the management [13].

CONCLUSION

We all need to stay current, read, and share data. Microbiology diagnostic used wisely can save lives. Trust the laboratory that you work with. Share interesting data regularly.

REFERENCES

1. Hughes S, Troise O, Donaldson H, Mughal N, Moore LS. Bacterial and fungal co-infection among hospitalized patients with COVID-19: A retrospective cohort study in a UK secondary-care setting. Clin Microbiol Infect 2020;26:1395-9.
2. Mehta S, Pandey A. Rhino-orbital mucormycosis associated with COVID-19. Cureus 2020;12:e10726.
3. Singh AK, Singh R, Joshi SR, Misra A. Mucormycosis in COVID-19: A systematic review of cases reported worldwide and in India. Diabetes Metab Syndr 2021;15:102146.
4. Spellberg B, Edwards J Jr., Ibrahim A. Novel Perspectives on Mucormycosis: Pathophysiology, Presentation, and Management; Microbiol Rev 2005;18:556-69.
5. Ibrahim AS, Spellberg B, Walsh TJ, Kontoyiannis DP. Pathogenesis of mucormycosis. Clin Infect Dis 2012;54 Suppl 1:S16-22.
6. Bhatt M, Soneja M, Fazal F, Vyas S, Kumar P, Jorwal P, *et al*. Two cases of osteoarticular mucor menace: A diagnostic and management conundrum. Drug Discov Ther 2018;12:374-8.
7. Gamaletsou MN, Sipsas NV, Roilides E, Walsh TJ. Rhino-orbital-cerebral mucormycosis. Curr Infect Dis Rep 2012;14:423-34.
8. Hong HL, Lee YM, Kim T, Lee JY, Chung YS, Kim MN, *et al*. Risk factors for mortality in patients with invasive mucormycosis. Infect Chemother 2013;45:292-8.
9. Skiada A, Pavleas I, Drogari-Apiranthitou M. Epidemiology and diagnosis of mucormycosis: An update. J Fungi (Basel) 2020;6:E265.
10. Singh Y, Ganesh V, Kumar S, Patel N, Aggarwala R, Soni KD, *et al*. Coronavirus disease-associated mucormycosis from a tertiary care hospital in India: A case series. Cureus 2021;3:e16152.
11. Taj-Aldeen SJ, Gamaletsou MN, Rammaert B, Sipsas NV, Zeller V, Roilides E, *et al*. Bone and joint infections caused by mucormycetes: A challenging osteoarticular mycosis of the twenty-first century. Med Mycol 2017;55:691-704.
12. Cornely OA, Arikan-Akdagli S, Dannaoui E, Groll AH, Lagrou K, Chakrabarti A, *et al*. ESCMID and ECMM joint clinical guidelines for the diagnosis and management of mucormycosis 2013. Clin Microbiol Infect 2014;20 Suppl 3:5-26.
13. Chauhan K, Soni D, Sarkar D, Karuna T, Sharma B, Singh S, *et al*. Mucormycosis after COVID-19 in a patient with diabetes. Lancet 2021;398:e10.

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