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Case Report

A rare case of blood stream infection due to *Candida ciferii* in an immuno-compromised patient

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ABSTRACT

Invasive infections related to yeast are increasingly observed in immune-compromised patients in hospitals. Fungal infections have increased morbidity and mortality and prolonged hospital stay which can lead to rise in medical care costs. Non-albicans Candida species have been increasingly found as causative agents in human infections with important therapeutic implications. We present a case of a 37-year-old, female patient, known case of B cell Acute Lymphoblastic Leukaemia admitted in a tertiary care hospital in central India for supportive care and chemotherapy. Patient was responding well to chemotherapy. On post induction day 20, she complained of high-grade fever with abdominal pain. Two sets of blood culture were sent to Microbiology Diagnostic Laboratory for diagnosis. She was started on Injection Magnex Forte (Cefoperazone-Sulbactum) empirically. The Gram stain of positive blood culture showed Gram positive budding yeast like cells in all four bottles. The organism was identified as *C. ciferrii* on Vitek 2 with 95% identification. Antibiotic susceptibility testing showed sensitive to Amphotericin B MIC ≤0.25 and voriconazole MIC ≤0.12. It was resistant to fluconazole MIC ≥64 µg/ml.

Keywords: Acute lymphoblastic Leukaemia, *Candida cifferi*, Febrile neutropenia

INTRODUCTION

Invasive infections related to yeast are increasingly observed in immune-compromised patients in hospitals.[1,2] Fungal infections have increased morbidity and mortality and prolonged hospital stay which can lead to rise in medical care costs.[3]

Non-albicans Candida species have been increasingly found as causative agents in human infections with important therapeutic implications.[4] The unusual yeast species *Candida ciferrii*, was first described in 1965, and it was named in honor of memory of Prof. Dr. R. CIFERRI.[5] It is the anamorph of *Stephanoascus ciferrii* and has been described as a pathogen in superficial mycoses and infrequently as a systemic disease.[1] *C. ciferrii* is an unusual species of Candida, which has been rarely reported as a cause of human infection mostly in patients with immune suppression.[1-3,5,6]

Authors report a case of a successfully treated patient with invasive disease due to this fungal pathogen**.**

CASE REPORT

A 37-year-old, female patient, known case of B cell Acute Lymphoblastic Leukaemia was admitted in a tertiary care hospital in central India for supportive care and chemotherapy. Her Complete Blood Count showed WBC count of 85360/cu.mm.

She was started on Induction Chemotherapy with following drugs- Vincristine, Prednisolone, Daunomycin, L-asparaginase and intrathecal methotrexate. Following induction chemotherapy, her WBC counts stared reducing upto 2230/cu.mm. Her USG abdomen and pelvis were performed and showed metastasis in kidneys and portal hypertension.

Patient was responding well to chemotherapy. On post induction day 20, she complained of high-grade fever with abdominal pain.

Two sets of blood culture were sent to Microbiology Diagnostic Laboratory for diagnosis. She was started on Injection Magnex Forte (Cefoperazone-Sulbactum) empirically.

All four bottles were positive the next day on the automated Bact T Alert system for blood cultures by Biomerieux. The Gram stain of positive blood culture showed Gram positive budding yeast like cells in all four bottles.

They were plated on SDA (Sabouraud's Dextrose agar), Blood agar and Mac-Conkey agar at 37^0 Celsius in the incubator. Another SDA was kept at 25^0 Celsius at room temperature. Colonies on next day were whitish, creamy, pasty in SDA at both temperatures and also in Blood agar and Mac-Conkey agar. Gram stain of colonies showed Gram positive budding yeast like cells. Germ tube was negative after 2 hours.

Repeat blood culture was advised and again all four bottles signaled positive. On both occasions, the organism was identified as *C. ciferrii* on Vitek 2 with 95% identification. Antibiotic susceptibility testing showed sensitive to Amphotericin B MIC ≤0.25 and voriconazole MIC ≤0.12. It was resistant to fluconazole MIC ≥64 µg/ml.

Following the report, she was started on Amphotericin B. She responded to the drug and became afebrile henceforth. Repeat blood cultures were consistently negative. Recovery thereafter was uneventful.

RESULTS

C. ciferrii is an unusual species of Candida, which has been rarely reported as a cause of human infection mostly in patients with immunosuppression.[1-3,5,6]. Most of the reported *C. ciferrii* cases include malignant otitis externa and onychomycosis.[7,8]

Saha et al, reported a fluconazole sensitive strain isolated in a diabetic chronic obstructive pulmonary disease patient presenting with pneumonia. She also had a wonderful recovery with fluconazole.[6]

Agin et al, reported a fatal candidemia case caused by *C. ciferrii* in an 8-year-old child in which isolated candida species were resistant to amphotericin-B (MIC >1 µg/ml), fluconazole, (MIC ≥64 µg/ml), caspofungin (MIC ≥32 µg/ml), and anidulafungin (MIC ≥32 µg/ml) but sensitive to voriconazole (MIC ≤0.12 µg/ml.[9]

Upadhyay S et al, studied a series of six cases of *C. ciferrii* infection in a tertiary care centre of north india. They found among the six cases, two cases were of candidemia, three from lower respiratory samples and one from drainage fluid. Among the six cases only two isolates was resistant to fluconazole. Rest were sensitive to other antifungals. These two cases were managed with caspofungin and all cases had an uneventful recovery.[10]

Hiram Villanueva-Lozano reported an unusual case of *C. ciferrii* fungemia in an immune-compromised patient with Crohn's and Mycobacterium bovis disease. In this case, fluconazole MIC = 32 µg/ml and the patient did not respond clinically. Finally, it was necessary to use posaconazole, a broadspectrum drug, to treat this patient.[11]

De Gentile L et al, report six cases of toe-nail onyxis due to an unusual yeast species, *Candida ciferrii*.[12]

Capoor et al, reported *C. ciferrii* in AML patient. The isolate was resistant to amphotericin B, but susceptible to fluconazole and itraconazole despite being on fluconazole prophylaxis.[13] In this study, it was resistant to fluconazole.

Gunsilius et al, reported a fluconazole-resistant invasive systemic mycoses due to *C. ciferrii* in AML patient who suffered a relapse after autologous peripheral blood progenitor cell transplantation.[14] Only very few case reports are available for the blood stream infections due to this pathogen in immune-compromised hosts.

In this case, as the patient was immune-compromised due to ALL and developed febrile neutropaenia after day 20 of induction chemotherapy. She was not responding to empirically started antibiotic therapy. So, it becomes important to consider unusual opportunistic pathogens, especially in such haematological malignancies.

CONCLUSION

It is important to consider unusual pathogens as the probable cause of infection, especially in immune-compromised, previously treated, or patients with a prolonged hospital stay. These rare pathogens in immune-compromised hosts cannot be neglected and suggest that in vitro susceptibility testing of isolated fungi should be performed for the selection of appropriate antifungal drugs.

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