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# Isolated Renal Zygomycosis Caused by *Syncephalastrum* Species in an Immunocompetent Host: A Case Report

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#### **ABSTRACT**

A 39-year-old male presented with left flank pain, intermittent high-grade fever, and passage of whitish flakes in the urine for the last 6 months. Urine culture was sterile. Fluffy whitish material, suggestive of fungal elements, was discovered during cystoscopy and ureteroscopy. On direct microscopy, this material showed fungal hyphae that were suggestive of Zygomycosis. The material was identified as *Syncephalastrum* species on Sabouraud dextrose agar (SDA) culture media. The patient was managed by endoscopic removal of the fungus and antifungal medication. He was doing well at the 1, 3, and 6-month follow-up evaluations. Renal Zygomycosis caused by *Syncephalastrum* species in an immunocompetent host is extremely rare and can be managed successfully by endoscopic removal and antifungal medication.

### INTRODUCTION

Zygomycosis is an opportunistic fungal infection that rarely occurs in healthy individuals. The exact route of pathogen entry is unknown, but it is undoubtedly exogenous. Possible sources of the infections include adhesive dressings, air conditioning filter units, food, and cutaneous trauma. The pathogenesis of renal mucormycosis is not clear, although hematogenous dissemination and a lower urinary tract infection have been suggested. Isolated organ involvement is extremely rare.

Only few cases of isolated renal involvement have been reported. The authors report an unusual case of renal Zygomycosis caused by *Syncephalastrum* spp in a patient with no predisposing factors. No previously published reports of this condition were found.

### **CASE REPORT**

A 39-year-old male presented with left flank pain for the last 6 months. Pain was intermittent and dull aching. There was a history of severe pain in the suprapubic area before passage of urine. The pain was relieved after urination. The patient had been passing whitish flakes in the urine. He also had intermittent high-grade fever with chills and rigor. There was no history suggestive of crystalluria or extramarital contact.

Physical examination was unremarkable. Urine analysis revealed 3-4 pus cells, and the urine culture was sterile. A peripheral smear for the malaria parasite was negative. Hemogram and blood biochemical reports were normal. Ultrasonography of the kidney, ureter, and bladder region showed only left proximal hydroureteronephrosis. Intravenous urography showed only a poorly visualized left

**KEYWORDS**: Renal Zygomycosis; *Syncephalastrum* species; Immunocompetent host

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#### **Abbreviations and Acronyms**

DTPA = diethylene-triamine-pentaacetic acid

RGP = retrograde pyelography



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kidney with hydronephrotic change. A diethylene-triaminepentaacetic acid (DTPA) renal scan showed that the left kidney was hydronephrotic with poor perfusion and an obstructive pattern with no significant diuretic washout. The differential function was 22.58% on the left side.

Retrograde pyelography (RGP) was planned. However, fluffy whitish material was discovered during cystoscopy, which was removed by cystoscopic bladder wash. Ureteroscopy revealed similar whitish flakes in the upper ureter that were also removed and sent for microscopy and culture. The RGP was abandoned.

The whitish material showed sparsely spaced fungal hyphae on direct microscopy, morphologically suggestive of Zygomycetes (Figure 1). The material was identified as *Syncephalastrum* species on Sabouraud dextrose agar (SDA) culture media (Figure 2). It showed broad, sparsely spaced hyphae with irregular side walls and obtuse branching. It also showed 2 fruiting-like bodies with zygospore formation.

Postoperatively, the patient was empirically put on intravenous fluconazole. The authors received the direct microscopy report suggestive of Zygomycetes 24 hours after surgery. The patient was then put on amphotericin-B. However, he had an acute, severe reaction to the drug so it was stopped prematurely. In concert with internal medicine colleagues, the patient was again put on intravenous fluconazole; the patient responded

to this treatment. Thereafter, he was put on oral fluconazole for 14 days.

Follow-up examinations were completed at 1, 3, and 6 months. The patient was asympatomatic and an intravenous urogram (IVU) showed improvement in renal function. At the 6-month evaluation, the DTPA renal scan showed that the left kidney was mildly hydronephrotic with moderate perfusion and no obstruction; differential function was 37.6%.

#### **DISCUSSION**

Zygomycosis is an uncommon fungal infection of the order *Mucorales*. It is most commonly caused by an organism belonging to the genus *Rhizopus*. Other genera include *Absidia*, *Mucor*, *Apophysomyces*, and *Saksenaea vasiformis* [1]. Most cases of Zygomycosis are known to occur in individuals with a predisposing factor such as acquired immune deficiency syndrome (AIDS), diabetes mellitus, or drug abuse; transplant recipients are also at risk [2-5]. It is not known whether Zygomycosis can present with minimal constitutional symptoms in a healthy individual. It is also not known whether a course of amphotericin-B after endoscopic removal of fungal bezoars would be sufficient treatment in a healthy patient.

Renal involvement in disseminated mucormycosis occurs in up to 19% of cases [6]. Isolated renal Zygomycosis is extremely

Figure 1. Direct Microscopy Showing Sparsely Spaced Fungal Hyphae, Suggestive of Zygomycosis (lactophenol cotton blue stain, 50x).

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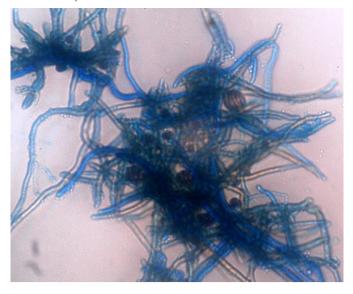
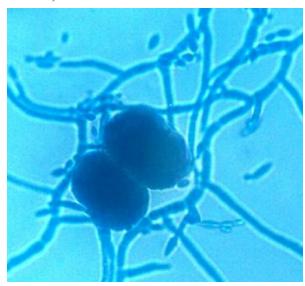


Figure 2. *Syncephalastrum* Species: Broad Hyphae With Irregular Side Walls, Obtuse Branching, and 2 Fruiting-Like Bodies With Zygospore Formation (high magnification). doi: 10.3834/uij.1944-5784.2010.08.03f2





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rare, particularly in healthy individuals [7,8]. Only a few cases caused by various species have been reported. This is first known case of renal Zygomycosis caused by *Syncephalastrum* species.

Syncephalastrum is a saprophytic organism that has been isolated from environmental sources. It has been found in soil in India, the Southern United States, Panama, and Israel. Whether or not this fungus actually causes disease in humans has been a topic of debate. A case report of a cutaneous Syncephalastrum of the finger is the only detailed case published in the literature [9]. Definitive diagnosis of the disease due to Syncephalastrum requires demonstration of Zygomycete fungal elements in a tissue section or other clinical specimen and diagnostic culture findings, as in the present case. This fungus probably has a low pathogenic potential in a competent host. The ability of Syncephalastrum to grow at or above 37°C may be important in its ability to cause human disease.

Successful therapy for invasive zygomycete infection usually involves a combined approach consisting of early diagnosis, correction of the underlying predisposing condition, aggressive surgical debridement, and early systemic amphotericin therapy. In contrast, *Syncephalastrum* spp (which has a low pathogenic potential in a healthy person) can be successfully treated by aggressive surgical debridement or endoscopic removal. Further studies are needed to know the first-line antifungal against *Syncephalastrum*. It is not known whether amphotericin-B or fluconazole is equally effective against this genus.

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