

## Case Report

## Asian Pacific Journal of Tropical Medicine

journal homepage: www.apjtm.org



doi: 10.4103/1995–7645.284646

Impact Factor: 1.77

*Trichosporon asahii* ankle cavity effusion infection in a patient with severe aplastic anemia

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## ABSTRACT

**Rationale:** *Trichosporon*, an anamorphic fungus, proliferates under high humidity, causing serious opportunistic infections collectively called trichosporonosis. Among the *Trichosporon* species causing trichosporonosis are *Trichosporon (T.) asahii*, *T. asteroides*, *T. cutaneum* etc.

**Patient concerns:** A 38-year-old Chinese male with severe aplastic anemia was admitted due to multiple joints pain, poor appetite, and right ankle swelling. One year earlier he had undergone allogeneic hematopoietic stem cell transplantation.

**Diagnosis:** *T. asahii* infection and severe aplastic anemia.

**Interventions:** Combined treatment of amphotericin B liposomes (55 mg/24 h) and voriconazole (200 mg/12 h) for 8 days.

**Outcomes:** The symptoms of the patient's ankle were relieved and effusion cultures showed no *T. asahii*.

**Lessons:** To the best of our knowledge, *T. asahii* ankle cavity effusion infections are rare. *Trichosporon* infections may be attributed to risk factors such as improper long-term use of antimicrobials for an underlying disease (e.g., anemia, hypoalbuminemia). Attention should be paid to prevent and control *Trichosporon* infections in order to avoid comorbidities.

**KEYWORDS:** *Trichosporon asahii*, Ankle cavity effusion, Aplastic anemia

## 1. Introduction

*Trichosporon* species are widespread and have been isolated from a wide range of substances, including soil, cheese, cabbage, parrot droppings, human hair and nails, and scarab beetles[1]. *Trichosporon* infections have been the subject of clinical research in association with cancer, organ transplants, hypoinmunity, burns, peritoneal dialysis, and prolonged use of antibiotics[2]. Poor prognosis of the infection was well illustrated by the report of a fatal outbreak of

trichosporonosis in neonates under antifungal therapy[3]. However, *Trichosporon (T.) asahii* infection in the presence of aplastic anemia has rarely been reported[4]. We therefore report a case of a 38-year-old man who presented with severe aplastic anemia and *T. asahii* ankle cavity effusion infection at our tertiary care institution. The study was approved by the Institutional Review Board of Yijishan Hospital of Wannan Medical College (Protocol No.: WF2019016).

## 2. Case report

A 38-year-old man from Anhui Province presented with right ankle swelling for 1 year after having undergone allogeneic hematopoietic stem cell transplantation were admitted to our hospital. In January 2018, he presented with a 10-day history of dizziness and fatigue. Bone marrow evaluation verified the diagnosis of severe aplastic anemia. Afterwards he underwent sibling-donated allogeneic hematopoietic stem cell transplantation without complication. Subsequently, he was diagnosed with Epstein-Barr virus and cytomegalovirus infections for which he was treated with rituximab,  $\gamma$ -globulin, and ganciclovir. Five months later, he presented with chills and fever accompanied by mid-upper abdominal pain. Clinical laboratory results showed procalcitonin: 0.16 ng/mL, C-reactive protein: 34.53 mg/L, white blood cell count:

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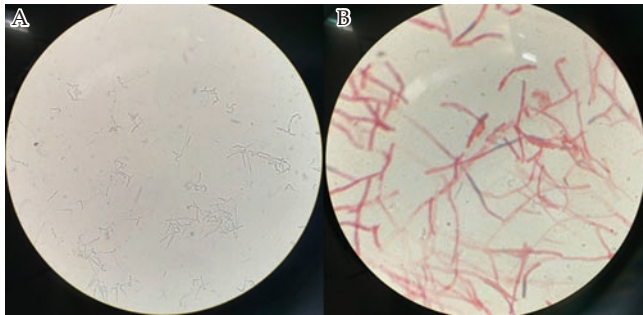
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**How to cite this article:** Zhang P, Yang ZH, Li J, Li XN. *Trichosporon asahii* ankle cavity effusion infection in a patient with severe aplastic anemia. Asian Pac J Trop Med 2020; 13(6): 278-280.

**Article history:** Received 15 May 2019      Revision 17 October 2019  
Accepted 10 December 2019      Available online 3 June 2020

$3.0 \times 10^9/L$ , neutrophils: 85.1%, and platelets:  $29 \times 10^9/L$ . He was treated with aztreonam combined with amikacin. Eight months later, he presented with poor appetite and gastrointestinal complaints. As graft-versus-host disease was possible, he was given tacrolimus and methylprednisolone. In January 2019, after long-term use of antibiotics and immunosuppressive agents, he complained of pain in multiple joints, especially the right ankle and knee. MRI indicated right ankle swelling with exudation of surrounding soft tissue. Considering a possible ankle cavity infection, we extracted effusion for microorganism culture and identification.



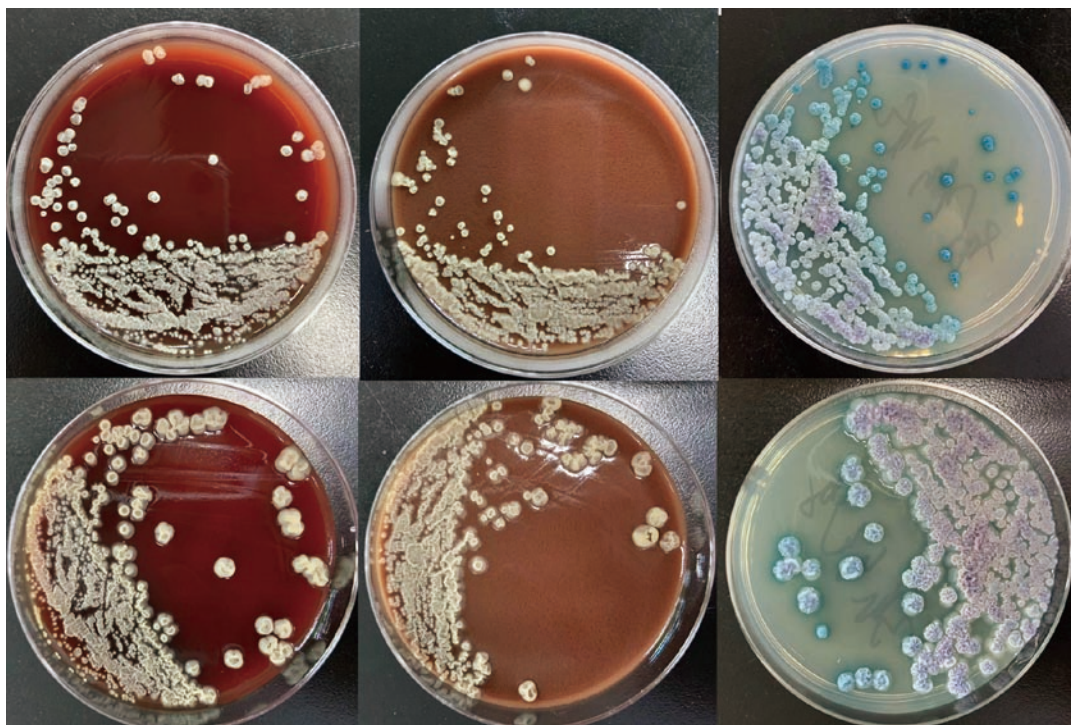
**Figure 1.** Routine microscopy (A) and Gram stain (B) of *Trichosporon asahii* isolated from ankle cavity effusion from a 38-year-old male with severe aplastic anemia and multiple joint show, respectively, yeast cells without budding but with septate hyaline hyphae with arthroconidia. Magnification:  $10 \times 40$  (A) and  $10 \times 100$  (B).

The effusion sample underwent routine microscopy, gram stain, and subculture. Routine microscopy revealed yeast cells without budding, and Gram stain revealed septate hyaline hyphae with

arthroconidia (Figure 1). On Sabouraud's dextrose agar at  $37^\circ\text{C}$ , the fungal colonies appeared curdy, white, and cottony (Figure 2). VITEK 2 compact automated system and VITEK MS (bioMérieux SA, Marcy l'Etoile, France) identified *T. asahii* (Supplementary Figure 1). Antifungal susceptibility was tested using ATBTM FUNGUS 3 (bioMérieux, La Baume-les Grottes, France) for 5-fluorocytosine, amphotericin B, fluconazole, itraconazole, and voriconazole. Minimum inhibitory concentration for 5-fluorocytosine, amphotericin B, fluconazole, itraconazole, and voriconazole were 4.0, 0.5, 2.0, 0.25, and 0.125 mg/L, respectively. Prompt treatment with amphotericin B liposomes (55 mg/24 h *i.v.*) and voriconazole (200 mg/12 h *p.o.*) for 8 days decreased the patient's ankle symptoms, with negative effusion culture results for *T. asahii*.

### 3. Discussion

According to the National China Hospital Invasive Fungal Surveillance Net program, 133 *Trichosporon* isolates were identified by sequencing the IGS1 region. The results showed that the most common species was *T. asahii* (108, 81.2%), followed by *T. dermatis* (7, 5.3%), *T. asteroides* (5, 3.8%), *T. inkin* (5, 3.8%), and *T. dohaense* (3, 2.3%)[5]. Although invasive trichosporonosis has been studied for many years, there are no standard guidelines for clinical interpretation of *Trichosporon* infection. Commonly, infections occur in patients who have hematologic malignancies with neutropenia and other risk factors such as a tumor, use of an immunosuppressor, antibiotics, uremia, chronic respiratory failure, and diabetes-related foot problems. *Trichosporon* is the second next to *Candida* species as



**Figure 2.** *Trichosporon asahii* isolated from ankle cavity effusion from a 38-year-old male Chinese with severe aplastic anemia and multiple joint pain growing on blood agar (left), chocolate (middle), and Sabouraud's dextrose agar (right) at  $37^\circ\text{C}$  incubation for 24 h (top row) and 48 h (bottom row).

the the most frequent cause of fungemia in patients with hematologic malignancies.

The invasive trichosporonosis diagnosis is of two types: proven and probable. A proven diagnosis requires (1) *trichosporon* organism growth in blood cultures, cerebrospinal fluid cultures, or biopsy specimen cultures; and (2) related symptoms. Probable diagnosis requires the patient to have a predisposing host factor and clinical evidence of fungal disease. In our case, the patient's ankle was swollen, and his MRI indicated right ankle swelling and exudation of the surrounding soft tissue. Subsequent cavity effusion cultured on Sabouraud's dextrose agar displayed *T. asahii* via the VITEK 2 compact automated system and VITEK MS. The culture affirming *T. asahii* and the associated symptoms met the criteria for a proven diagnosis.

Upon treatment, most *Trichosporon* cases occur as an accidental infection despite standard prophylactic antifungal regimens. Mortality rates are reportedly >80%[6]. In a small-case study, only 23% of patients with hematologic malignancies and invasive trichosporonosis treated with amphotericin B therapy survived. Flucytosine did not offer additional benefit. The echinocandins have little effect on *Trichosporon* species and are not recommended for treating these infections. Despite some contradictory evidence about *Trichosporon* treatment, amphotericin B is commonly considered the initial management. To date, however, the treatment and management of invasive trichosporonosis has begun to shift from amphotericin to azoles. Suzuki *et al.* reported that, among hematologic malignancy patients infected with *Trichosporon*, those treated with azoles (*versus* those who were not) exhibited significantly better survival[7]. Clinical practice pattern data supported the use of azoles as well. Azoles such as voriconazole, posaconazole, and fluconazole may be effective[8]. Amphotericin B combined with azoles may also be effective[9]. Our patient's antifungal susceptibility test showed five antifungal drugs with sensitivity. Therefore, we prescribed amphotericin B liposomes combined with voriconazole. Eight days later, after repeated isolation and effusion fluid cultures, no *T. asahii* were evident, and the patient's ankle symptoms were alleviated. Thus, we believe that management and therapy of trichosporonosis infection must include an azole[10].

#### 4. Conclusion

This case of *T. asahii* infection in ankle cavity of the male patient with severe aplastic anemia presented questions about this rare fungus and its ability to invade the immunosuppressed host. Dedicated efforts by clinicians and microbiologists to target fungal infections are suggested, as well as further studies to manage and control *Trichosporon* infections.

#### Conflict of interest statement

The authors declare that there is no conflict of interest.

#### Authors' contributions

PZ performed clinical data collection, data analysis and drafted the article. ZHY and JL were responsible for photo taking and identification of fungus. PZ and XNL contributed to the final version of the manuscript.

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