Eradication of *Cokeromyces recurvatus* in a pregnant patient: A case report

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**A B S T R A C T**

A 31-year-old woman, gravida 3 para 1, at 10 weeks of gestation presented to a vulvar specialty clinic with a 6-month history of vulvar pruritus that had not responded to the treatment prescribed by her referring gynecologist. Examination was consistent with vaginitis and vulvar lichen simplex chronicus. Fungal cultures ultimately revealed *Candida albicans* and *Cokeromyces recurvatus*. The organisms were eradicated with oral fluconazole and intravaginal terazol. Few cases of isolation of *C. recurvatus* in humans have been reported. Both immunosuppressed and immuno-competent hosts have been identified, with the most common sites of isolation being the genitourinary and gastrointestinal tracts. Asymptomatic colonization and superficial disease have been noted; no invasive cases of *C. recurvatus* have been described. Even fewer cases of *C. recurvatus* infection during pregnancy have been reported, and symptomatic vulvovaginal disease has been described only once prior to this case report.

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1. Introduction

The first case of eradication of culture-proven infection with *Cokeromyces recurvatus* in a symptomatic pregnant patient is reported here. It is only the second case of symptomatic vaginitis with *C. recurvatus* in a pregnant patient in the literature.

2. Case

A 31-year-old woman, gravida 3 para 1, at 10 weeks of gestation, was referred to a university vulvar specialty clinic for evaluation of chronic vulvar pruritus. She reported vulvar itching of six months' duration. The patient had been treated by the referring physician with topical and oral antifungals, topical steroids, and emollients without improvement. She stated that fluconazole had initially helped with her symptoms, but the pruritus quickly returned. She had undergone biopsy four months earlier, which showed benign squamous epithelial hyperplasia with chronic inflammation; silver stain showed surface hyperplasia and yeast fungal elements consistent with candida organisms. At the time of her intake visit, she was using emollient skin protectant only. Her medical and family history were non-contributory.

Her examination showed normal female architecture with erythema and lichenification of the perineal body. The vestibule was diffusely erythematous, and thick white vaginal discharge was noted upon speculum placement. A wet mount demonstrated many pseudohyphae and a few budding growths of yeast. No white blood cells or parabasal cells were noted. Lactobacilli were present. A vaginal specimen was obtained for yeast culture with sensitivities. The patient was given a prescription for fluconazole, 2 doses, 72 h apart, and then weekly doses for three additional weeks. She was also given a prescription for ketoconazole shampoo to cleanse the perineum daily until symptoms improved. She was leaving to travel abroad for six weeks the afternoon of her visit, so follow-up was arranged for after her return. Six days after collection, the vaginal fungal culture was positive for *Candida albicans*, which would also be susceptible to fluconazole.

At her subsequent visit, now at 16 weeks of gestation, the patient reported a partial improvement in symptoms. She had completed the prescribed regimen as described above. She also continued to use the ketoconazole shampoo biweekly as prophylaxis. Although she reported improvement in symptoms, her examination remained abnormal. The labia minora and vestibule were erythematous and the labia minora were edematous. The thick white vaginal discharge was unchanged. Wet mount demonstrated no pseudohyphae, but copious white blood cells were present. Large “mariner’s wheel” yeast buds were noted. No parabasal cells were seen and lactobacilli were present. A vaginal swab was again collected for fungal culture. While awaiting the culture result, the patient was empirically treated with a 7-day course of intravaginal teraconzaole. Ten days later, the vaginal fungal culture returned positive for *Cokeromyces recurvatus*.

Due to pregnancy complications, the patient was lost to follow-up until after delivery. Her return visit was 5 months later. She reported complete resolution of symptoms. Her examination was normal, without erythema, edema, lichenification, or vaginal discharge. Her wet
monia [4,5]. Even fewer cases of ranging from spontaneous resolution without treatment to a fetal pneumaticorganism has been isolated from the pulmonary system with outcomes on pap smear. Very few cases of asymptomatic colonization have been reported, all of which have occurred in North America [1]. Immunosuppressed and immuno-competent hosts have been identified; however, invasive disease has been described only in immunosuppressed patients [4–6], with one attributable death [5]. The most common sites of isolation are the genitourinary and gastrointestinal tracts [1–3], with the most frequent scenario presenting as asymptomatic colonization found on pap smear. Very few cases of asymptomatic colonization have been reported, and none has been treated. Less often, the appearance to Paracoccidioides brasilensis. The yeast are thick walled, with the largest yeast cells surrounded by a crown of smaller yeast buds [1], often compared to a mariner’s wheel (Fig. 1).

Few cases of isolation of C. recurvatus from humans have been reported, all of which have occurred in North America [1]. Immunosuppressed and immuno-competent hosts have been identified; however, invasive disease has been described only in immunosuppressed patients [4–5], with one attributable death [5]. The most common sites of isolation are the genitourinary and gastrointestinal tracts [1–3], with the most frequent scenario presenting as asymptomatic colonization found on pap smear. Very few cases of asymptomatic colonization have been reported, and none has been treated. Less often, the organism has been isolated from the pulmonary system with outcomes ranging from spontaneous resolution without treatment to a fetal pneumonia [4,5]. Even fewer cases of C. recurvatus in pregnancy have been reported, and the case presented is only the second report of asymptomatic vulvovaginal disease during pregnancy [7]. In the first case the C. recurvatus was isolated together with Clamidia trachomatis, and so the etiology of that patient’s symptoms remains unclear.

This case highlights the importance of wet-mount microscopy (WMM) in identifying nontraditional causes of vaginitis. As many women’s health providers move away from WMM in favor of PCR testing for the diagnosis of vaginitis [8], the diagnosis of nontraditional causes of vaginitis may be delayed. Had the unusual mariner’s wheel morphology not been seen, a repeat culture may not have been collected.

Limitations of the case report include the loss of follow-up from 16 weeks of gestation until after delivery. Fluconazole is not the first-line choice in treating candidial vaginitis in pregnancy; however, the patient was exposed to fluconazole prior to establishing care at the vulvar clinic and accepted the small risk of pregnancy loss due to the severity of her vaginitis symptoms. Another limitation is lack of microscopy photos from this case—in response to this, the author has established a system for collecting WMM clinical images.

In conclusion, Cokeromyces recurvatus is an uncommon cause of vaginitis, but should be considered, especially when mariner’s wheel yeast cells are noted on wet-mount microscopy.

Contributors

M. Luann Racher is the sole author of this case report.

Conflict of interest

The author has no conflict of interest regarding the publication of this case report.

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Patient consent

Obtained.

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This case report was peer reviewed.

References

[1] Julie A. Ribes, Carolyn L. Vanover-Sams, Doris J. Baker, Zygomycetes in human disease, Clin. Microbiol. Rev. 13 (2) (2000) 236–301.
[2] Cherie Paquette, et al., Cokeromyces recurvatus in a cervical papanicolaou test: A case report of a rare fungus with a brief review of the literature. Diagn. Cytopathol. 44 (5) (2016) 419–421.
[3] Shelley I. Odronic, et al., Two cases of Cokeromyces recurvatus in liquid-based Papanicolaou tests and a review of the literature, Arch. Pathol. Lab. Med. 136 (12) (2012) 1593–1596.
[4] Maksim Agaronov, et al., Cokeromyces recurvatus Identified in Lung Biopsy: Case Report of a Non-pathogenic Fungus, Highlighting Its Potential Histologic Mimics, Ann. Clin. Lab. Sci. 45 (2) (2015) 229–214.
[5] Lori J. Ryan, et al., Fatal Cokeromyces recurvatus pneumonia: report of a case highlighting the potential for histopathologic misdiagnosis as Coccidioides, Int. J. Surg. Pathol. 19 (1) (2011) 373–376.
[6] T.W. Tsai, et al., Cokeromyces recurvatus infection in a bone marrow transplant recipient, Bone Marrow Transplant. 19 (3) (1997) 301.
[7] Deanna A. McGough, Annette W. Fothergill, Michael G. Rinaldi, Cokeromyces recurvatus poitras, a distinctive zygomycete and potential pathogen: criteria for identification, Clin. Microbiol. Newsl. 12 (15) (1990) 113–117.
[8] NK. Lowe, JL. Neal, NA. Ryan-Wenger, Accuracy of the clinical diagnosis of vaginitis compared with a DNA probe laboratory standard, Obstet Gynecol. 113 (1) (2009) 89–95, https://doi.org/10.1097/AOG.0b013e3181909f63.