References

1. Heffner, R.R., Armbrustmacher, V.W. & Earl, K.M. Focal myositis. *Cancer* 1977, **40**: 301–306.
2. Hansen, L.B., Baekgaard, N. & Reske, N.E. Focal myositis of the rectus abdominal muscles. *Acta Chir Scand* 1985, **151**: 77–80.
3. Isaacson, G., Chan, K.H. & Heffner, R.J. Focal myositis. A new cause for the paediatric neck mass. *Arch Otolaryngol Head Neck Surg* 1991, **117**: 103–105.
4. Shapiro, M.J., Applebaum, H. & Besser, A.S. Cervical focal myositis in a child. *J Pediatr Surg* 1986, **21**: 375–376.
5. Azuma, T., Komori, A. & Nagayama, M. Focal myositis of the tongue. *J Oral Maxillofac Surg* 1987, **45**: 953–955.
6. Takita, M.A., Kawamoto, T.O., Nogami, H., Takada, M., Onishi, S. & Sakuda, M. Focal myositis of the tongue: report of a case. *J Osaka Univ Dent Sch* 1985, **25**: 161–169.
7. Ellis, G.L. & Brannon, R.B. Focal myositis of the perioral musculature. *Oral Surg* 1979, **48**: 337–341.
8. Maguire, J.J., Milford, L.W. & Pitcock, J.A. Focal myositis in the hand. *J Hand Surg Am* 1988, **13**: 140–142.
9. Chiba, S., Miyano, N., Matsumoto, H., Chiba, T. & Wakabayashi, J. [A case of isolated focal myositis successfully treated by arterial injection of prednisolone—its findings by CT scan and MRI]. *Rinsho Shinkeigaku* 1990, **30**: 1261–1265.
10. Agrawal, B.L. & Giessen, P.C. Eosinophilic myositis. An unusual cause of pseudotumour and eosinophilia. *JAMA* 1981, **246**: 70–71.
11. Li, J.T., Garrity, J.A., Kephart, G.M. & Gleich, G.J. Refractory periorbital oedema in a 29-year-old man [clinical conference]. *Ann Allergy* 1992, **69**: 101–105.

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Candida lusitaniae causing fatal meningitis

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Summary: Fatal meningitis due to *Candida lusitaniae* in a 35 year old previously healthy man is described. *C. lusitaniae* is an opportunistic fungal pathogen reported infrequently in the English literature. This is the third case report of meningitis and the first fatal infection in an adult from Central India due to *C. lusitaniae* known to the authors.

Introduction

Candida lusitaniae van Uden et do Carmo-Sousa, originally isolated from the gastrointestinal tracts of warm-blooded animals,¹ has recently been found to colonize rarely the gastrointestinal, respiratory and urinary tracts and skin of hospitalized patients.² This organism shares morphological, biochemical and other characteristics with other species of the genus *Candida*, such as *C. tropicalis* and *C. parapsilosis*.³ More recently, *C. lusitaniae* has been recovered from a variety of human clinical specimens such as respiratory and genitourinary secretions, stool, blood, pleural fluid, kidneys, lungs, bone^{2,4,5} and cerebrospinal fluid.^{6,7}

The reports of a premature infant⁷ and a 2 year old white male⁶ with *C. lusitaniae* sepsis and meningitis are the only examples of meningeal infection caused by this organism, of which we are aware. Blinkhorn *et al*.⁸ in their review did not include any additional patient with this form of *C. lusitaniae* disease. The present report describes a third case of meningitis and a first fatal case caused by this emerging pathogen in a non-white patient from central India.

Case report

A 35 year old man was admitted as an emergency to the Jawaharlal Nehru Hospital and Research Centre, Bhilainagar, Madhya Pradesh, India, with a 8 day history of fever and unconsciousness of one

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day's duration and three episodes of generalized convulsions. The patient had no history of medical illness prior to hospitalization. He had received intravenous (i.v.) ciprofloxacin and gentamicin for 3 days from a private medical practitioner. On examination, he was found to be unconscious, febrile (38.3°C) and mildly dehydrated. The spleen had enlarged 2 cm below the costal margin and liver 2 cm below the right costal margin. He had no significant lymphadenopathy. Signs of meningeal irritation were present and plantar reflexes were bilateral extensor. There were no cranial nerve palsy or motor deficits. His haemoglobin was 12.3 g/dl, white cell count $11.9 \times 10^9/l$ (neutrophils 76%), and platelets $204 \times 10^9/l$. The hepatic and renal function tests were normal. A sickling test was negative and urinalysis, skull and chest X-rays were normal. Lumbar puncture yielded haemorrhagic cerebrospinal fluid (CSF) with white cells of $500 \times 10^9/l$ (neutrophils 70% and mononuclears 30%) and red cells of $25 \times 10^9/l$. The concentration of CSF glucose was 1.96 mmol/l (serum glucose 5.32 mmol/l) and that of protein was 105 mg/dl. No bacteria were seen on a Gram-stained smear of CSF sediment. His blood, CSF and urine cultures grew no bacteria. He received i.v. fluids, i.v. chloramphenicol, gentamicin and crystalline penicillin. Two days later the CSF grew a yeast subsequently identified as *C. lusitaniae*, which was susceptible to amphotericin B and flucytosine tested at the Mycotic Diseases Branch, Centers for Disease Control, Atlanta, USA.

The colonies of the organism were cream-coloured, soft, smooth and glistening. After 3–4 days of growth in glucose–yeast-extract–peptone water at 25°C, cells of *C. lusitaniae* were subglucose to ovoid with a size range of $1.5\text{--}5\text{ }\mu\text{m} \times 2.5\text{--}10\text{ }\mu\text{m}$. The pseudomycelium on cornmeal agar was composed of branched chains of slender, often curved pseudohyphae that bore short chains and clusters of blastoconidia.⁹ The separation of *C. lusitaniae* from similar yeast such as *C. parapsilosis*, *C. tropicalis*, *C. freyschusii* and *C. obtusa* is necessary.⁸ Tests used for differentiation included fermentation of cellobiose, glucose, galactose and trehalose, lack of growth over cyclohexamide-containing media and negativity for urease production.^{7,8} The yeast was appropriately identified with the API 20C system (Analytab Products, Plainview, NY, USA).⁸

Over the next day, the neurological condition remained the same with no improvement. He remained febrile and within 34 hours of hospitalization, the patient expired due to sudden cardiorespiratory arrest. Postmortem examination was refused but a postmortem sample of brain was scanty and revealed no evidence of tissue invasion in histological sections stained with haematoxylin and eosin and Gomari's Methanamine Silver.

Discussion

Pappagianis *et al.*⁴ and Holzschu *et al.*¹⁰ published the first documented case of opportunistic infection caused by *C. lusitaniae* in a patient with leukaemia and also demonstrated the capacity of this strain to develop resistance to amphotericin B. Merz and Sandford¹¹ reported a polyene-resistant variant of *C. tropicalis* that was later reidentified as *C. lusitaniae*.

During the last 13 years after the publication of the first documented case of opportunistic infection caused by *C. lusitaniae*,⁴ 18 cases have been reported in the English literature and over 100 clinical isolations of *C. lusitaniae* have been described.^{8,12} Of the 18 cases reported until now, 13 patients were adults,⁸ two were neonates,^{3,7} and one was a teenager.¹¹ Most patients with *C. lusitaniae* fungaemia were immunocompromised, usually owing to an underlying malignancy^{2–4,6,12–15} and transplantation.¹³ Not surprisingly, the use of broad-spectrum antibiotics (14 of 18 patients), the presence of an intravascular catheter (nine of 18), the use of cytotoxic or corticosteroid therapy (nine of 18), and the occurrence of granulocytopenia (five of 18) were noted frequently, while the presence of diabetes mellitus was noted infrequently (one of 18).¹³ Total parenteral nutrition, a known risk factor for the development of fungaemia caused by other candida species¹⁶ could not be assessed as a predisposing factor because of a lack of documentation in previous reports. However, Terreni *et al.*¹⁷ reported *C. lusitaniae* septicemia in a 34 year old male undergoing home i.v. hyperalimentation and suggested hyperalimentation as one of the predisposing factors which might have contributed to their patient's acquisition of *C. lusitaniae* fungaemia. In addition, there has been at least one reported instance of transient septicemia in which the yeast was isolated only once and no treatment was instituted.¹⁸

The first successful therapy of *C. lusitaniae* fungaemia was reported in 1985.¹⁹ Before 1989, with the exception of a case of transient fungaemia not requiring therapy,¹⁸ all reported infections with *C. lusitaniae* required antifungal therapy.^{3,6,8,13,17,19,20} and resistance to amphotericin B played a role in the fatal¹⁷ outcome of the disease.^{2,4,11,14,20}

Our patient received broad-spectrum antibiotics (ciprofloxacin and gentamicin) for 3 days prior to hospitalization. We believe that broad-spectrum antibiotic usage was the predisposing factor in our patient who was otherwise healthy causing immunocompromisation contributing to the development of fatal *C. lusitaniae* meningitis.

To date, out of more than 100 clinical isolations of *C. lusitaniae* reported in the English literature,^{8,12} three-quarters of the isolates have been recovered from the blood, urine or respiratory tract.⁸ Positive

extravascular cultures in patients with *C. lusitaniae* fungaemia reported in the English literature were obtained mostly from the respiratory tract and pharynx, stool, urine, soft tissue, skin, vagina or intravascular catheter.⁸ Only two isolates of *C. lusitaniae* have been recovered from CSF.^{6,7} Our case report describes the recovery of the third isolate of *C. lusitaniae* from CSF.

Until now, human infections due to *C. lusitaniae* have been reported from North America and Europe. Our report indicates that *C. lusitaniae* is not necessarily restricted geographically to the developed world. It is important that the clinical laboratory recognizes this emerging pathogen when it is isolated from clinical material, especially from individuals already debilitated by malignant disease, sepsis, cytotoxic and steroid therapy, usage of broad-spectrum antibiotics and diabetes mellitus. Thus it appears that *C. lusitaniae* is similar to

other yeast species in its ability to colonize individuals and cause opportunistic infections in compromised hosts.^{2,8} This species does not seem to differ from other medically important yeasts in the development of resistance to amphotericin B *in vivo*.² In conclusion, *C. lusitaniae* should be considered as an opportunistic pathogen in both immunocompromised adults and children when isolated in the appropriate clinical setting, with no prejudice to geographical localization and restriction.

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References

1. Van Uden, N. & Buckley, H. Genus berkhout. In: Lodder, J. (ed.) *The Yeasts - A Taxonomic Study*, 2nd ed. Elsevier/North-Holland Biomedical Press, Amsterdam, 1970, pp. 994-996.
2. Merz, W.G. *Candida lusitaniae*: frequency of recovery, colonization, infection and amphotericin B resistance. *J Clin Microbiol* 1984, **20**: 1194-1195.
3. Christenson, J.C., Guruswamy, A., Mukwaya, G. & Rettig, P.J. *Candida lusitaniae*: an emerging human pathogen. *Pediatr Infect Dis J* 1987, **6**: 755-757.
4. Pappagianis, D., Collins, M.S., Hector, R. & Remington, J. Development of resistance to amphotericin B in *Candida lusitaniae* infecting a human. *Antimicrob Agents Chemother* 1979, **16**: 123-126.
5. Schiltzer, R.L. & Ahearne, D.G. Characterization of atypical *Candida tropicalis* and other uncommon clinical yeast isolates. *J Clin Microbiol* 1982, **15**: 511-516.
6. Leggiadro, R.J. & Collins, T. Postneurosurgical *Candida lusitaniae* meningitis. *Pediatr Infect Dis J* 1988, **7**: 366-367.
7. Sanchez, P.J. & Cooper, B.H. *Candida lusitaniae* sepsis and meningitis in a neonate. *Pediatr Infect Dis J* 1987, **6**: 758-759.
8. Blinkhorn, R.J., Adelstein, D. & Spagnuolo, P.J. Emergence of a new opportunistic pathogen, *Candida lusitaniae*. *J Clin Microbiol* 1989, **27**: 236-240.
9. Libertin, C.R., Wilson, W.R. & Roberts, G.D. *Candida lusitaniae* - an opportunistic pathogen. *Diagn Microbiol Infect Dis* 1985, **3**: 69-71.
10. Holzschu, D.L., Presley, H.L., Miranda, M. & Phaff, H.J. Identification of *Candida lusitaniae* as an opportunistic yeast in humans. *J Clin Microbiol* 1979, **10**: 202-205.
11. Merz, W.G. & Sandford, G.R. Isolation and characterization of a polyene-resistant variant of *Candida tropicalis*. *J Clin Microbiol* 1979, **9**: 677-680.
12. Goodman, J.L., Winston, D.J., Greenfield, R.A. *et al.* A controlled trial of fluconazole to prevent fungal infections in patients undergoing bone marrow transplantation. *N Engl J Med* 1992, **326**: 845-851.
13. Baker, J.G., Nadler, H.L., Forgacs, P. & Kurtz, S.R. *Candida lusitaniae*: a new opportunistic pathogen of the urinary tract. *Diagn Microbiol Infect Dis* 1984, **2**: 145-149.
14. Hadfield, T.L., Smith, M.B., Winn, R.E., Rinaldi, M.G. & Guerra, C. Mycoses caused by *Candida lusitaniae*. *Rev Infect Dis* 1987, **9**: 1006-1012.
15. Winn, R.E., Hadfield, T.L., Smith, M.B. *et al.* *Candida lusitaniae* mycoses. *Abstracts of the 86th Annual Meeting of the American Society for Microbiology*, Washington, D.C., March 23-28, 1986, p. 403.
16. Curry, C.R. & Quil, P.G. Fungal septicemia in patients receiving parenteral hyperalimentation. *N Engl J Med* 1971, **285**: 1221-1225.
17. Terreni, A.A., Strohecker, J.S. & Dowda, H. *Candida lusitaniae* septicemia in a patient on extended home intravenous hyperalimentation. *J Med Vet Mycol* 1986, **25**: 63-65.
18. Bradsher, R.W. Transient fungaemia due to *Candida lusitaniae*. *South Med J* 1985, **78**: 626-627.
19. Pensler, M.I., Krawczyk, P. & Lebar, W.D. *Candida lusitaniae* septicemia. *Clin Microbiol Newslett* 1985, **7**: 12.
20. Guinet, B., Chanas, J., Goullier, A., Bonnefoy, G. & Thomas, P.A. Fatal septicemia due to amphotericin B resistant *Candida lusitaniae*. *J Clin Microbiol* 1983, **18**: 443-444.