

Fournier Gangrene Caused by *Candida albicans* in an Infant After Cardiac Surgery

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Abstract Fournier gangrene is a rare, rapidly progressive, life-threatening condition. We report a 23-day-old boy with pulmonary atresia and ventricular septal defect treated surgically, who developed Fournier gangrene. Emergency surgery was performed with tissue sampling for microbiological examination. *Candida albicans* was confirmed; caspofungin followed by fluconazole was administered with excellent results.

Keywords Fournier gangrene · *Candida albicans* infection · Pediatric cardiac surgery · Congenital heart defects · Echinocandins · Fluconazole

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Introduction

Fournier gangrene (FG) is a rare, rapidly progressive, life-threatening condition with necrotizing fasciitis of the perineum first mentioned by Baurienne in 1764, however, described and named after Jean Alfred Fournier and his report from the year 1883 [1, 2]. The disease mostly affects patients between 50 and 60 years of age and is uncommon in children. The typical flora involves polymicrobial bacteria, most commonly *Escherichia coli*, *Proteus*, *Staphylococcus*, *Enterococcus* and anaerobes; however, single cases of fungal etiology in adults were reported [2–5]. The risk factors of FG developing include diabetes, immunosuppression, chronic kidney disease, local trauma, urethral stricture and genitourinary infections [3]. It is essential to diagnose FG as early as possible, because the infection can rapidly spread from local necrosis to adjacent organs, leading to sepsis and death; mortality rates of 7.5–88% are reported [5–7]. We report a case of a male infant, who developed fungal FG after cardiac surgery.

Case Report

A 23-day-old male new-born weighting 3.1 kg with pulmonary atresia and ventricular septal defect was admitted to the Department of Pediatric Cardiac Surgery for treatment. The child was born hypotrophic (2.7 kg) at 38 weeks of gestation, and his mother

Fig. 1 Fournier gangrene due to *Candida albicans* in an infant ► after cardio-surgical treatment erythema and edema of scrotum, groins with extension toward left axilla after scrotum surgical examination and debridement (a), status post-second surgical intervention after next 24 h (b) and significant local improvement after 14 days of antifungal therapy (c)

suffered from myasthenia gravis. Routinely performed microbiological screening at admission revealed that the child's throat was colonized with methicillin-resistant *Staphylococcus aureus* (MRSA) and rectum with *Candida albicans*. Before the admission for surgery, there was difficulty with urinary catheterization probably due to urethral. However, after few attempts before the operation, a smaller sized catheter was successfully placed. Perioperative antibiotic prophylaxis consisted of vancomycin and gentamycin. After diagnostic catheterization and detailed pulmonary vessels, anatomy description unifocalization with aorto-pulmonary shunts was performed bilaterally (Gore-tex®, 3.5 mm in diameter). Significant blood steal syndrome occurred postoperatively, resulting in insufficient organ perfusion and requiring prolonged intensive care unit stay (ICU). Other complications included: prolonged mechanical ventilation, digestive problems requiring total parenteral nutrition, cholestasis and diaper rash. Reoperation due to left-sided shunt problems was necessary on the 7th postoperative day. After 2 weeks, on the 15th day after the first surgery clinical examination revealed painful erythema and edema of the scrotum; however, the child was not febrile. Scrotal examination showed mucopurulent tissue infiltration, and local debridement was performed; samples for microbiological examination were taken. Complete blood count examination revealed 11.16 G/l of white blood count, whereas C-reactive protein (CRP) and procalcitonin (PCT) were 68 mg/l and 0.55 ng/ml, respectively. *Candida* score was estimated as three points (colonization, total parenteral nutrition and surgery). Antibacterial (meropenem, vancomycin) and antifungal (fluconazole) empirical therapy was initiated. Within the next 24 h erythema and edema were spreading over both groins and toward the left axilla (Fig. 1). FG was diagnosed clinically, and the next emergency surgery was performed relieving edema in order to prevent microthrombosis of small



subcutaneous vessels and progressive necrotizing fasciitis. Tissue sampling for further microbiological examination was also performed. *C. albicans* was confirmed in all microbiological cultures from tissue samples, and blood samples, however, were negative for *Candida*. Because of instable clinical status, the child was transferred to ICU. Caspofungin was started (loading dose of 70 mg/m², then 50 mg/m²) and continued for 14 days followed by fluconazole with excellent results (Fig. 1). Follow-up period at 6 months remains uneventful.

Discussion

FG is a rare condition with the incidence of 1.6 cases per 100,000 males adult per year [10]. The disease mostly affects patients between 50 and 60 years of age, and in children FG is not common. However, FG is a fulminant and life-threatening disease, because although proper treatment mortality in these groups of patients remains high, mortality rates of 7.5–88% are reported [2, 5–8]. A literature review found only <60 pediatric cases of FG worldwide, with 66% of those in infants younger than 3 months [9].

Commonly, the etiology of FG is polymicrobial and the infection is most often caused by three or more microorganisms: *E. coli*, *Proteus*, *Staphylococcus*, *Enterococcus* and anaerobes [3–5]. However, single-case reports describing FG caused by fungi are reported. *C. albicans*, *Candida glabrata* and *Rhizopus microsporus* etiology are described in adults [5, 10, 11]. Our case report is unique because to the best of our knowledge there is no report of solitary *C. albicans* FG in children.

It is essential to diagnose FG as early as possible, because the infection can rapidly spread from local necrosis to adhering organs, causing sepsis and death. The diagnosis of FG is based on clinical symptoms such as intense pain, pruritus and tenderness in the genitalia, usually associated with edema of the overlying skin. It is worth emphasizing the role of round the clock access to microbiological laboratory with wide spectrum of diagnostics examination. The guidelines for FG treatment recommend hemodynamic stabilization, aggressive wide surgical debridement and empirical broad-spectrum antibiotics therapy. Also hyperbaric oxygen therapy is recognized as an additional form of FG treatment [2]. However, worldwide increase in fungal

infections incidence results in suggestion of some authors to add the empirical antifungal treatment, especially in patients at risk for fungal infections. This concerns mainly patients with primary and secondary immunodeficiencies. Fluconazole therapy could be a reasonable therapeutic option; however, due to widespread use of azoles a large number of non-*albicans Candida* species isolates in recent years suggest that echinocandins may be a good choice for empiric therapy, especially in hemodynamic unstable patients [3, 5, 12]. Also combined therapy of different antifungal therapeutics groups may be successful if monotherapy fails; echinocandin and liposomal amphotericin are suggested in this setting [13].

In our case, secondary immunodeficiency in hypotrophic infant was probably caused by significant blood steal syndrome that occurred postoperatively, resulting in vital organs hypoperfusion. Another risk factor for FG was preoperative traumatic urinary catheterization. However, the first symptoms of FG showed relatively late, on 15th postoperative day. Our patient was colonized with methicillin-resistant *S. aureus* (MRSA) as well as with *C. albicans* preoperatively; nevertheless, twice as surprisingly the fungi were an etiological factor for FG. One could explain this phenomenon that the perioperative antibiotic prophylaxis appeared effective against MRSA, while no prophylaxis against fungi was used. In our case, rapid diagnosis based on clinical signs, broad-spectrum empirical antibiotics and aggressive surgical approach with debridement and tissue sampling for microbiological identification of an etiological agent which guided our pharmacotherapy led to final therapeutic success.

In conclusion, the successful treatment of FG depends on early diagnosis and aggressive etiological management. Therapeutic strategy includes hemodynamic stabilization, broad-spectrum antibiotics and wide surgical debridement with maximum excision of all necrotic tissues. General increase in fungal infections and the incidence of fungal colonization worldwide may suggest that empiric antifungal treatment should also be considered in FG.

Compliance with Ethical Standards

Conflict of interest Dr. Jaworski reports and has become personal fee for lecture from MSD Polska, Dr. Irga-Jaworska reports and has become personal fee for lecture from MSD

Polska, Dr. Haponiuk has nothing to disclose, Dr. Naumiuk has nothing to disclose, and Dr. Chojnicki has nothing to disclose.

Informed Consent Informed consent from the patient's parents for the publication of this case report is provided. Informed consent for the publication of all patient's pictures from the patient's parents is provided.

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References

1. Fournier JA. Jean-Alfred Fournier 1832–1914. Gangrène foudroyante de la verge (overwhelming gangrene). *Sem Med* 1883. *Dis Colon Rectum*. 1988;31:984–8.
2. Sroczynski M, Sebastian M, Rudnicki J, Sebastian A, Agrawal AK. A complex approach to the treatment of Fournier's gangrene. *Adv Clin Exp Med*. 2013;22:131–5.
3. Perkins TA, Bieniek JM, Sumfest JM. Solitary *Candida albicans* infection causing Fournier gangrene and review of fungal etiologies. *Rev Urol*. 2014;16:95–8.
4. Shyam DC, Rapsang AG. Fournier's gangrene. *Surgeon*. 2013;11:222–32.
5. Loulergue P, Mahe V, Bougnoux ME, Poiree S, Hot A, Lortholary O. Fournier's gangrene due to *Candida glabrata*. *Med Mycol*. 2008;46:171–3.
6. Laor E, Palmer LS, Tolia BM, Reid RE, Winter HI. Outcome prediction in patients with Fournier's gangrene. *J Urol*. 1995;154:89–92.
7. Bjurlin MA, O'Grady T, Kim DY, et al. Causative pathogens, antibiotic sensitivity, resistance patterns, and severity in a contemporary series of Fournier's gangrene. *Urology*. 2013;81:752–8.
8. Rouzrokh M, Tavassoli A, Mirshemirani A. Fournier's gangrene in children: report on 7 cases and review of literature. *Iran J Pediatr*. 2014;24:660–1.
9. Pais VM. Fournier gangrene. <http://emedicine.medscape.com/article/2028899-overview#a7>. Accessed 1 Apr 2016.
10. Jensen P, Zachariae C, Larsen FG. Necrotizing soft tissue infection of the glans penis due to atypical *Candida* species complicated with Fournier's gangrene. *Acta Derm Venerol*. 2010;90:431–2.
11. Johnin K, Nakatoh M, Kadowaki T, et al. Fournier's gangrene caused by *Candida* species as the primary organism. *Urology*. 2000;56:153.
12. Jaworski R, Irga N, Haponiuk I, et al. Candidemia in children after complex congenital heart defects surgery treated with caspofungin—our own experience and a review of literature. *Med Sci Monit*. 2011;17:PH35–9.
13. Jaworski R, Haponiuk I, Irga-Jaworska N, et al. Fungal infections in children in the early postoperative period after cardiac surgery for congenital heart disease: a single-centre experience. *Interact Cardiovasc Thorac Surg*. 2016;23:431–7.