Management of congenital cutaneous candidiasis in a healthy term baby: A case report

SAGE Open Medical Case Reports JCMS Case Reports Volume 7: 1–3 © The Author(s) 2019 Article reuse guidelines: sagepub.com/journals-permissions DOI: 10.1177/2050313X19876707 journals.sagepub.com/home/sco



Sophia Colantonio¹, Erin Hedin², Heidi Oi-Yee Li³ and Geneviève Gavigan^{1,4}

Abstract

The management of congenital cutaneous candidiasis in a healthy term neonate of normal birth weight is unclear. Often, healthy term neonates undergo extensive evaluation followed by systemic treatment, which may not be clinically warranted. Here, we present a case of a healthy term neonate with congenital cutaneous candidiasis, whose work-up was minimally invasive and was successfully treated with one oral dose of antifungals and topical antifungals, as well as a review of the literature.

Keywords

Congenital candidiasis, neonate, treatment, management

Introduction

There is debate among clinicians regarding the management of a healthy term neonate with congenital cutaneous candidiasis (CCC). Investigations and treatment options range from a full septic work-up with parenteral therapy to skin scrapings and swabs with no therapy or topical antifungals. In this article, we describe less invasive management of a healthy term neonate with CCC.

Case report

A well 3-day-old male presented to the children's emergency department with a pustular eruption on his palms, soles, face and trunk. He was born at 40 + 0 weeks via spontaneous vaginal delivery. His birth weight was 3.3 kg. Immediately after birth, pustules were noted on his face and chest. He was monitored for a 24-h period, during which he was afebrile and neurologically appropriate. He was diagnosed with neonatal acne and discharged home post-partum without any treatment.

On examination, the patient was well and alert. He did not show any signs of increased work of breathing. The patient had numerous 1-3 mm erythematous macules and pustules on his palms (Figure 1(a)), soles (Figure 1(b)), arms, legs, chest, abdomen and face (Figure 1(c)). There was no evidence of oral thrush or cutaneous candidiasis of the genitals or perianal area.

At the bedside, two skin scrapings of the pustules were collected and fixed in ethanol. Routine hemotoxylin and eosin (H&E) staining was performed on one slide and Grocott staining on the other. The H&E stain showed keratinocytes and abundant neutrophils. The Grocott stain showed both yeast and pseudohyphal forms. The family was informed of the diagnosis of congenital candidiasis within 1 h of performing the skin scrapings. In addition, bacterial, viral and fungal cultures were taken from the pustules at the time of our initial visit. The results were available 2 days later and confirmed the sole growth of *Candida albicans*. Since a diagnosis was already made using the skin scrapings, a skin biopsy was not performed. Retrospective analysis of placental pathology was attempted, but due to the hospital's policy of keeping placentas from normal births for only 2 weeks, this was not possible.

Corresponding Author:

Geneviève Gavigan, Division of Rheumatology and Dermatology, Department of Pediatrics, Children's Hospital of Eastern Ontario, 401 Smyth Road, Ottawa, ON KIH 8LI, Canada. Email: genevieve.gavigan@gmail.com

Creative Commons Non Commercial CC BY-NC: This article is distributed under the terms of the Creative Commons Attribution-NonCommercial 4.0 License (http://www.creativecommons.org/licenses/by-nc/4.0/) which permits non-commercial use, reproduction and distribution of the work without further permission provided the original work is attributed as specified on the SAGE and Open Access pages (https://us.sagepub.com/en-us/nam/open-access-at-sage).

Division of Dermatology, Department of Medicine, University of Ottawa, Ottawa, ON, Canada

²Department of Pediatrics, University of Ottawa, Ottawa, ON, Canada
³Faculty of Medicine, University of Ottawa, Ottawa, ON, Canada
⁴Division of Rheumatology and Dermatology, Department of Pediatrics, Children's Hospital of Eastern Ontario, Ottawa, ON, Canada



Figure 1. Erythematous macules and pustules on (a) palms, (b) soles and (c) face and chest.



Figure 2. Treatment algorithm for congenital candidiasis.

Given that the patient was a well full term neonate who had been afebrile for all 3 days of life, his lack of systemic symptoms and his normal physical examination, routine blood work, blood cultures, urine cultures and lumbar puncture were deferred.

The initial treatment plan was to treat topically with nystatin 100,000 units/g cream four times per day until all the lesions had resolved. Due to a shortage of topical nystatin cream at the pharmacy, the patient was given one dose of oral fluconazole 3 mg/kg/day po in the emergency department until the nystatin cream could be obtained the following day. He was then treated with topical nystatin cream 4 times daily for 1 week. The rash resolved completely with this treatment regimen.

Discussion

Clinical presentation

CCC is an invasive fungal infection of the dermis and epidermis¹ caused by an ascending exposure to *Candida* spp. in utero.² Most CCC cases present with lesions at birth or within the first few days of life, but onset can be as late as 6 days of life.² The evolution of CCC first begins as an extensive monomorphous maculopapular eruption that later evolves to pustules which resolve with desquamation.^{1–4} Lesions are polymorphic with different stages often being observed at the same time.^{2,5} Additional clinical features may include diffuse erythematous macules and papules, bullae, onychia, paronychia, funisitis and acute chorioamnionitis with yellow-white plaques on the placenta and umbilical cord.^{2,4,6} Although skin involvement can be diffuse and involve any cutaneous portion of the body, commonly affected regions include the face, trunk, back, extensor surfaces of extremities, and intertriginous areas.² The diaper area and oropharynx are typically not implicated in CCC and involvement of these regions is more characteristic of neonatal and chronic mucocutaneous candidiasis.⁶ Pustules are typically present on palms and soles, differentiating CCC from miliaria and erythema toxicum neonatorum.^{4,5} CCC patients may also present with signs of systemic infections such as respiratory distress and leukocytosis accompanied by a burn-like dermatitis⁶ and erosive lesions. These cases have a much higher incidence among unwell or preterm neonates compared to healthy term neonates.^{2,4}

Management

Treatment recommendations for CCC depend on the presentation of the neonate (Figure 2). For a well term neonate, there are differing opinions regarding the treatment of choice. However, it is clearer that an unwell term baby or a preterm, low birth weight baby requires a full septic work-up and parenteral therapy.^{2,7}

Although this case of CCC was treated with a single oral dose of both antifungal and topical therapy, the use of topical therapy alone may be sufficient, as in vaginal candidiasis. It is difficult to determine, from this case study, if the successful treatment outcome was due to the combination of oral and topical treatments or if a single dose of oral fluconazole was sufficient; thus, further cases treated with topical therapy alone are necessary. Based on this case, we hypothesize that topicals alone may be sufficient treatment; however, we have not had a subsequent case yet to test this monotherapy treatment for CCC.

A retrospective cohort study, performed by Kaufman et al. at two major academic neonatal intensive care units (NICU) in the United States, examined infants admitted for CCC from 2004 to 2015. They found 21 cases of CCC out of all total of 19,303 admissions. All cases were due to C. albicans. Most of the cases were early preterm babies (average 26 weeks + 3 days gestation) with extremely low birth weight (average 950 g). All but one patient received intravenous systemic antifungal therapy and they advocated for this to be the mainstay treatment for all preterm or unwell neonates with CCC. On one occasion, they used and recommended oral therapy in a stable term neonate.¹ In this study, there was a significant selection bias for sicker patients who required NICU admission. Their cohort was heavily skewed to early preterm extremely low birth weight or unwell babies, for which intravenous therapy is the ideal treatment option. Only two patients were full term and normal birth weight; one of whom received intravenous and the other, oral therapy. Therefore, their conclusions cannot be generalized to healthy full term neonates who do not require NICU admission.

In a 2000 review of all the English literature that reported birth weight and outcome of neonates with CCC, Darmstadt et al. published management guidelines along with their own case report. This yielded 63 infants, 27 of which were larger than 2500 g. In infants above 2500 g, three had culture proven evidence of extra-cutaneous involvement (urine, cerebrospinal fluid (CSF)) and one was treated with intravenous therapy. The remainder had either oral, topical, a combination, or no therapy. All but one infant survived. They concluded that in full term well infants, CCC is nearly always self-limited and extensive evaluation is not warranted. Although some authors recommend topical or oral therapy in this population,^{7,8} Darmstadt et al. suggest no treatment, as they found no proven benefit. The exception is in a neonate presenting with burn-like dermatitis, which in their review was highly associated with invasive disease and mortality.

Recently, new case reports have emerged supporting less intensive therapies; including successful oral therapy in a neonate presenting with CCC and tachypnea⁶ and significant improvement of one neonate with CCC and severe respiratory distress prior to initiation of any treatment.⁹

In conclusion, clinicians should have a high index of suspicion for the development of invasive disease in CCC; however, in healthy term neonates, extensive evaluation and treatment may not always be warranted.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

Informed consent

The patient's mother provided written permission for publication of this case report and associated images.

References

- Kaufman DA, Coggins SA, Zanelli SA, et al. Congenital cutaneous candidiasis: prompt systemic treatment is associated with improved outcomes in neonates. *Clin Infect Dis* 2017; 64(10): 1387–1395.
- Darmstadt GL, Dinulos JG and Miller Z. Congenital cutaneous candidiasis: clinical presentation, pathogenesis, and management guidelines. *Pediatrics* 2000; 105(2): 438–444.
- Praag MCG, Van Van Rooij RWG, Folkers E, et al. Fetal and neonatal investigations and reports diagnosis and treatment of pustular disorders in the neonate. *Pediatr Dermatol* 1997; 14(2): 131–143.
- 4. Oza V, Asch S and Mathes EF. Three-day-old boy with palmar pustules. *JAMA Pediatr* 2016; 170(2): 171–172.
- Santos LA, Beceiro J, Hernandez R, et al. Congenital cutaneous candidiasis: report of four cases and review of the literature. *Eur J Pediatr* 1991; 150(5): 336–338.
- Tieu KD. Congenital cutaneous candidiasis in two full-term infants. *Pediatr Dermatol* 2012; 29(4): 507–510.
- Rowen J. Mucocutaneous candidiasis. *Semin Perinatol* 2003; 27: 406–413.
- Smolinski KN, Shah SS, Honig PJ, et al. Neonatal cutaneous fungal infections. *Curr Opin Pediatr* 2005; 17: 486–493.
- Aldana-Valenzuela C, Morales-Marquec M, Castellanos-Martínez J, et al. Congenital candidiasis: a rare and unpredictable disease. *J Perinatol* 2005; 25: 680–682.