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CASE REPORT

Congenital cutaneous candidiasis in a term newborn

Candidíase cutânea congénita num recém-nascido de termo

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Abstract

Congenital cutaneous candidiasis (CCC) is a rare condition in neonates, mainly in term neonates, that develops in the 1st week of life. Its broad clinical spectrum makes it challenging to differentiate it from other exanthemas in the newborn. The involvement of palms and soles and the presence of pustules are important clinical clues for the differential diagnosis, with cultural examination confirming the diagnosis by identification of *Candida* spp. Treatment of clinically stable term neonates without evidence of invasive disease is currently controversial. We report a case of CCC in a term newborn with no evidence of invasive disease that evolved into a clinical cure after systemic and topical antifungal treatment.

Keywords: Congenital cutaneous candidiasis. Exanthema. Term newborn. Pediatric dermatology. Neonatal dermatology.

Resumo

A candidíase cutânea congenita é uma condição rara em recém-nascidos, principalmente em recém-nascidos de termo, que se desenvolve na primeira semana de vida e pode estar presente nas primeiras horas. Apresenta um amplo espectro de sintomas e pode ser difícil diferenciar de outros exantemas no recém-nascido. O envolvimento das palmas das mãos e plantas dos pés é uma característica importante no diagnóstico diferencial, e o exame cultural confirma o diagnóstico. O tratamento de recém-nascidos de termo clinicamente estáveis sem evidência de doença disseminada é controverso, e relatamos um caso de candidíase cutânea congênita extensa com resolução completa após tratamento sistêmico.

Palavras-chave: Candidíase cutânea congénita. Exantema. Recém-nascido termo. Dermatologia pediátrica. Dermatologia neonatal.

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Introduction

Congenital cutaneous candidiasis (CCC) is a rare condition in neonates. It is the result of a *Candida* spp. infection acquired in utero and is thought to occur *via* an ascending pathway from candida vulvovaginitis[1]. One of the most accepted risk factors is the presence of an intrauterine foreign body (intrauterine device or cervical cerclage)[2]. CCC can occur in infants regardless of the type of delivery, duration of rupture of membranes, or maternal diagnosis of candida vaginosis[3]. It is more common in preterm neonates under 27 weeks of gestational age, as well as in those with extremely low birth weight[4]. Immature, compromised mucocutaneous barriers and poor systemic host defences are thought to be predisposing factors in preterms. Extensive CCC in term neonates is rare, with only a few published reports in the literature.

Clinical case

A full-term (38 weeks) female infant was born by vaginal delivery with an appropriate birth weight for the gestational age (3044 gm). She was born to a healthy mother with a positive history of vaginal candidiasis before pregnancy. Serologies and prenatal ultrasounds were normal. There was no maternal history of herpes simplex infection, intrauterine device, cervical cerclage or amniocentesis. The mother tested positive for group B *Streptococcus* and received adequate antibiotic prophylaxis before delivery.

The Apgar score was 9/10/10 at 1, 5 and 10 minutes at birth. She had a regular clinical examination at birth.

At 24 hours of life, a few papules were noticed on the head and trunk, which rapidly and progressively worsened over the next 48 hours. On the third day, she developed a generalised papulopustular eruption involving the face, neck, trunk and extremities, including palms and soles, as well as diffuse bright erythema with erosions in the perineal area, multiple white plaques on the oral mucosa (Figs. 1 to 3), and purulent ocular exudate. There was no fever or other systemic symptoms. Based on clinical findings and skin examination, the diagnosis of CCC was strongly considered. Laboratory tests showed no evidence of systemic infection and a complete blood count revealed 18100/µL white blood cells (9500/uL neutrophils) and negative serum C-reactive protein concentration (0.03 mg/dL). Skin, ocular and blood cultures were collected. After multidisciplinary discussion, taking into consideration the extensive cutaneous and

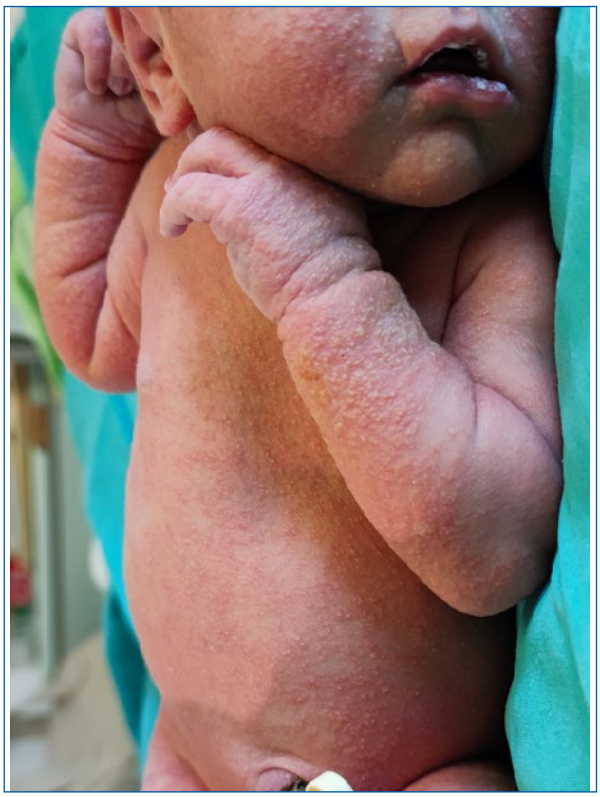

Figure 1. Diffuse papulopustular eruption involving the entire integument.

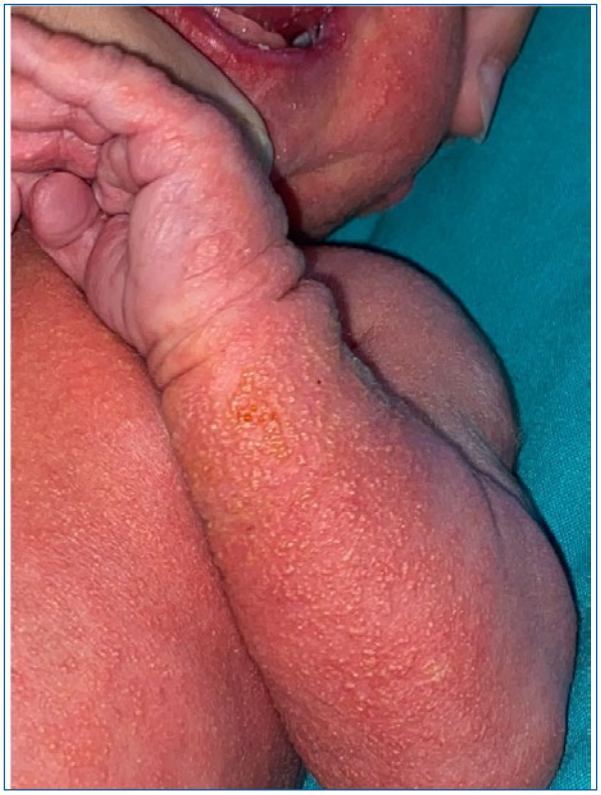

Figure 2. Presence of papules and pustules on the palms.

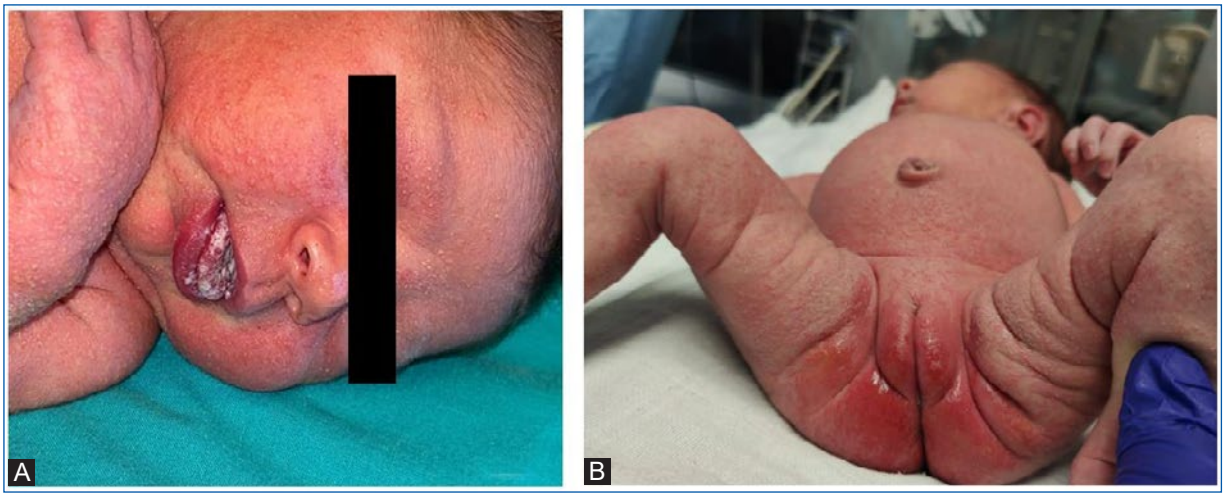

Figure 3. **A:** oral mucosal and **B:** perineal area.

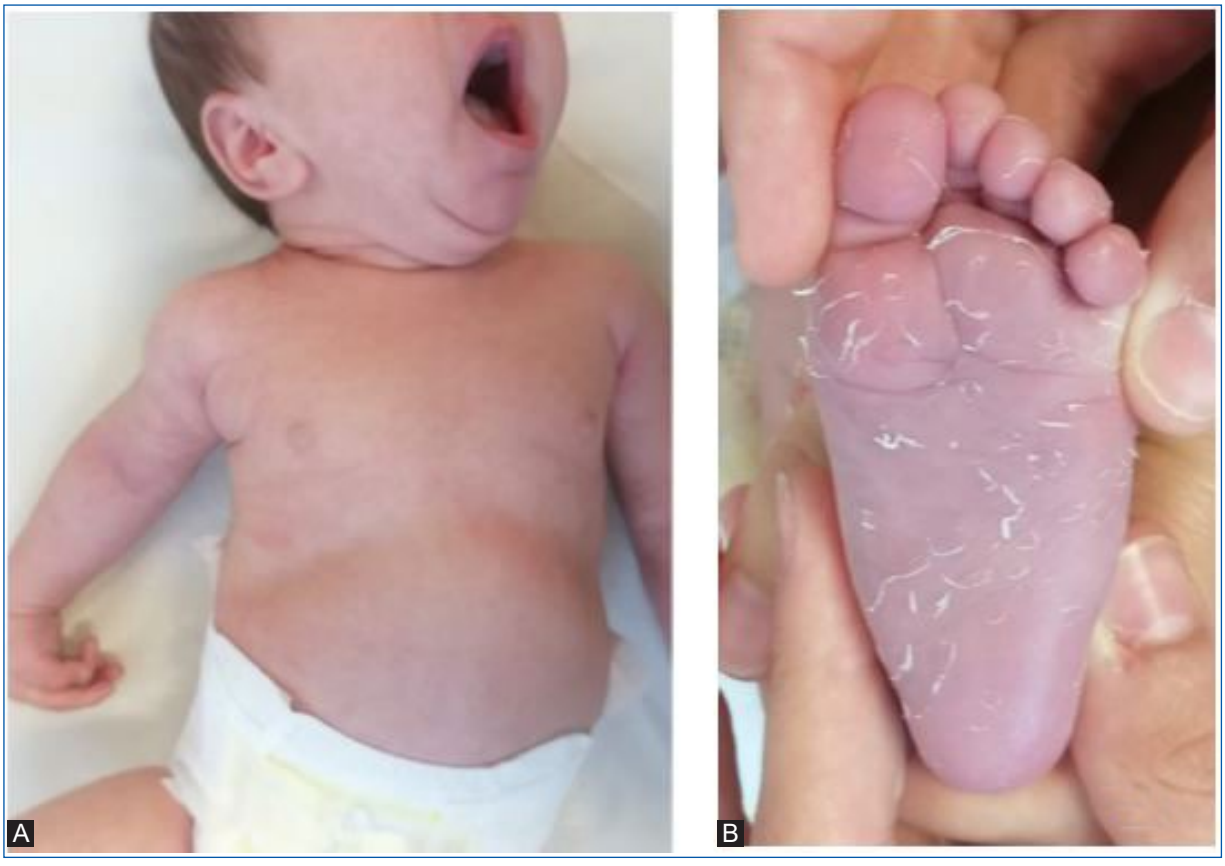

Figure 4. **A** and **B:** resolution with desquamation after therapy.

mucous membranes involvement, in the absence of systemic signs of infection, empiric treatment with intravenous fluconazole (12 mg/kg/day), oral nystatin and clotrimazole cream was started. Cerebrospinal fluid was not collected due to the lack of lesion-free areas at the puncture sites. Considering CCC as the most likely diagnosis and the absence of clinical or laboratory evidence of systemic infection, no antibacterial nor antiviral therapy was added, pending clinical evolution and culture results. After a few days, the

culture of pustules and ocular exudate identified *Candida albicans* confirming the diagnosis. Blood cultures were negative. The mother's vaginal evaluation did not show any signs of active vulvovaginal candidiasis and vaginal swab cultures were negative.

Oral candidiasis resolved on the third day of treatment, and skin lesions progressively resolved with desquamation after fourteen days of intravenous fluconazole and topical clotrimazole (Fig. 4).

There was no evidence of immunodeficiency in the initial investigation, and the patient is currently under clinical follow-up.

Discussion

We present a case of extensive mucocutaneous candidiasis appearing in the first 48 hours of life. The involvement of the palms and soles, as well as the involvement of the oral mucosa and perineal region, were essential for suspecting the diagnosis. There was no evidence of invasive disease, and there was a good evolution under topical and intravenous antifungal agents.

Congenital cutaneous candidiasis (CCC) develops in the 1st week of life and may be present in the first few hours[5]. It presents a broad spectrum of symptoms, from rash to severe invasive disease. It is clinically characterised by a generalised exanthema, including erythematous macules and papules, vesicles and pustules that later undergo desquamation[1,5]. The entire body can be affected, often involving the palms and soles, which is an essential feature for the differential diagnosis[6]. The most affected areas are usually the back, extensor surfaces and skin folds.

It is necessary to consider other differential diagnoses of exanthemas in newborns. Neonatal candidiasis differs from CCC by appearing after the first week of life and usually affects only the perianal region and oropharynx[1]. Other differential diagnoses of CCC are extensive-erythema toxicum neonatorum, transient neonatal pustular melanosis, several infectious causes (group B *Streptococcus*, *L. monocytogenes*, herpes simplex virus, varicella zoster-virus, among others), miliaria or even Langerhans cell histiocytosis[5]. Some features help in the differential diagnosis—the presence of plaques in the umbilical cord (funisitis) should raise the suspicion of CCC but also of *L. monocytogenes* infection; lesions on the palms and soles are characteristic of *Candida* infection. Due to the difficulty in the differential diagnosis, it is imperative to confirm the diagnosis by cultural examination with isolation of *Candida* spp.

Usually, it is self-limiting but can progress to invasive diseases such as candidemia, pneumonia, arthritis and endocarditis[4]. The probability of invasive disease and the mortality rate is inversely proportional to birth weight and gestational age.

Treatment of clinically stable term neonates without evidence of disseminated disease can be controversial. Nonetheless, studies also indicate that topical and shorter treatment cycles (< 14 days) are risk factors for the dissemination of *Candida* into the bloodstream[3]. In our case, due to the extensive mucocutaneous involvement with a higher risk of invasion, it was decided to initiate systemic therapy. Systemic treatment with fluconazole, amphotericin B deoxycholate and liposomal amphotericin B has been demonstrated to be effective[3]. In our hospital, amphotericin B deoxycholate is not available. Due to the low rate of fluconazole resistance in our unit and since liposomal amphotericin B is not the first choice in case of urinary tract infection[7] (which could not be excluded due to the great mucocutaneous involvement contraindicating aseptic collection), it was decided to use fluconazole as first choice with excellent clinical outcome.

Funding

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Conflicts of interest

None.

Ethical disclosures

Protection of human and animal subjects. The authors declare that no experiments were performed on humans or animals for this study.

Confidentiality of data. The authors declare that they have followed the protocols of their work centre on the publication of patient data.

Right to privacy and informed consent. The authors have obtained the written informed consent of the patients or subjects mentioned in the article. The corresponding author is in possession of this document.

References

1. Florens A, Maggiolino G, Kamat M, Joseph T, Piette W, Pyati S. A full-term infant with rash at birth. Pediatr Ann. 2010;39:551-4.
2. Roqué H, Abdelhak Y, Young BK. Intra amniotic candidiasis. Case report and meta-analysis of 54 cases J Perinat Med. 1999;27:253-62.
3. Kaufman DA, Coggins SA, Zanelli SA, Weitkamp JH. Congenital cutaneous candidiasis: prompt systemic treatment is associated with improved outcomes in neonates. Clin Infect Dis. 2017;64:1387-95.
4. Aruna C, Seetharam K. Congenital candidiasis. Indian Dermatol Online J. 2014;5:44-7.
5. Darmstadt GL, Dinulos JG, Miller Z. Congenital cutaneous candidiasis: clinical presentation, pathogenesis, and management guidelines. Pediatrics. 2000;105:438-44.
6. Rowen JL. Mucocutaneous candidiasis. Semin Perinatol. 2003;27:406-13.
7. Unni JC. Review of amphotericin B for invasive fungal infections in neonates and children. Pediatr Infect Dis. 2020;2:114-7.