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Erythroderma due to generalized pustular psoriasis in an infant: A case report of a rare condition

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Case Report

ABSTRACT

Erythroderma is a widespread redness of the skin accompanied by exfoliation that affects over 90% of the body surface. Its presence in infants is clinically important because it can be life-threatening, making it essential for paediatricians and dermatologists to be vigilant. Various skin diseases can cause erythroderma. Timely, accurate identification of the underlying cause results in improved treatment and prognosis. Generalized pustular psoriasis is one of many causes for erythroderma. Nevertheless, this condition is rare in infants, making it challenging to diagnose and treat. The current medical evidence for treatment is primarily derived from case reports rather than clinical trials. Hence, this case report aims to share the authors' experience in treating infants aged 6 months with erythroderma caused by generalized pustular psoriasis. In our case, the diagnosis was determined by completing the patient's medical history, performing a physical examination, and conducting a histopathological examination. The treatment involved the topical application of moisturizers containing ceramide as well as the systemic administration of steroids and vitamin D. Due to the restricted availability of systemic retinoids, the authors administered steroids in this particular case, resulting in an improved outcome.

INTRODUCTION

Erythroderma is characterized by diffuse erythema and scaling involving more than 90% of the body surface.[1] It is a relatively rare condition among children.[2] Its occurrence in infants is clinically significant due to its potential to be life-threatening, thus necessitating awareness among paediatricians and dermatologists.[2] Commonly thought to be a rare phenomenon, infantile erythroderma can result from a wide range of underlying conditions and may indicate both harmless skin issues and serious systemic diseases.[3] Due to an impaired skin barrier, an impacted infant faces risks of losing fluid and energy through their skin, experiencing hypothermia, suffering from hypernatremic dehydration, and being vulnerable to infections caused by viruses, bacteria, or fungi.[3] The causes of erythroderma in newborns and infants are challenging to identify and are frequently postponed because the clinical, biochemical, and histological signs are not very specific. Furthermore, the aetiology for erythroderma in this age group is quite distinctive.[2] Specific skin conditions known to trigger erythroderma in infants include infantile seborrheic dermatitis, atopic dermatitis, psoriasis, pityriasis rubra pilaris, generalized mastocytosis, erythrodermic ichthyosis, Netherton syndrome, and ectodermal dysplasia.[3,4] Timely identification of the underlying cause results in improved treatment outcomes

and prognosis. Currently, neonatal erythroderma is managed individually on a case-by-case basis.[4]

Generalized pustular psoriasis (GPP) is an uncommon and serious type of pustular psoriasis marked by extensive and recurring outbreaks of sterile pustules filled with neutrophils in the epidermis. This condition may also be associated with fever and systemic inflammation.[5-7] GPP in infants is extremely rare, accounting for only 0.6% of pustular psoriasis cases in both paediatric and adult populations.[6] Due to its rarity, both diagnosis and treatment are challenging for clinicians, with current management largely based on case reports and series rather than randomized clinical trials. Given the uncommon nature of erythroderma in infants and, even more so, of generalized pustular psoriasis as its cause, this case report aims to share the authors' clinical experience in diagnosing and treating such cases with improved outcomes.

CASE DESCRIPTION

Patient information: A 6-month-old female infant was brought by her parents with complaints of generalized skin peeling. Symptoms began at 2 weeks of age as an erythematous rash on the neck, followed by the development of pustules. The infant was treated by a paediatrician with unknown topical and oral medications, resulting in temporary improvement. 1.5 months before presentation at Dr. Sardjito Hospital, pustular lesions and fever reappeared and progressively worsened, spreading to the scalp, limbs, and trunk. The infant was previously treated with 1% hydrocortisone cream and oral betamethasone for two weeks without improvement. There was no history of allergies or similar conditions in her family. The infant's only known exposures included bar soap and baby shampoo.

Clinical findings: On admission, the patient presented with Cushingoid features and erythroderma. Vital signs were stable (HR: 125 bpm, RR: 32/min, Temp: 37°C, SpO2: 97%). Anthropometrics showed stunting and underweight (length: 60 cm, and weight: 6 kg). Dermatological examination revealed generalized erythematous patches with scaling and pustules, forming "lakes of pus" on the cheeks and feet (Figure 1).

Timeline: The patient's skin symptoms began at 2 weeks of age, with the appearance of a red rash on the neck, followed by pustules. She was taken to a paediatrician and treated with unspecified topical and oral medications, leading to temporary improvement. However, approximately 1.5 months before admission (around 4.5 months of age), the pustular rash on the neck recurred, and this time it was accompanied by fever. The parents sought treatment at a local hospital, where the infant was prescribed 1% hydrocortisone cream and oral betamethasone syrup for two weeks. Despite this treatment, her condition worsened, with skin peeling and erythematous lesions spreading to the scalp, limbs, and trunk. As her symptoms progressed, she was referred to Dr. Sardjito General Hospital at 6 months of age for further evaluation and management.

Diagnostic assessment: Differential diagnoses included erythroderma infantum secondary to generalized pustular psoriasis, atopic dermatitis, seborrheic dermatitis, Netherton syndrome, and congenital ichthyosiform erythroderma. Laboratory findings included normocytic normochromic anaemia (Hb 8.5 g/dL), normal WBC with neutrophilia (60.9%) and relative lymphopenia (33.5%), hypocalcaemia (2.17 mmol/L), vitamin D deficiency (14.6 ng/mL), elevated IgE (509 IU/mL), and hypoalbuminemia (2.65 g/dL). ANA was negative. KOH skin scraping and microscopic hair examination were unremarkable (Figure 2). Skin biopsy revealed parakeratosis, psoriasiform acanthosis with club-shaped rete ridges, suprapapillary thinning, Munro microabscesses, and superficial dermal infiltrates (Figure 3), supporting a diagnosis of generalized pustular psoriasis.

Therapeutic intervention: Initial management included supportive care: IV fluids (570 cc/day), IV ampicillin-sulbactam (300 mg q6h), oral paracetamol (990 mg q8h prn) (since during hospital admission our patient sometimes experienced fever with temperature ranged from 37 – 38.2^{0}C and withdrawn after 24 hours period of free fever), vitamin D (2000 IU/day), vitamin C (50 mg/day), zinc (20 mg/day), albumin transfusion, Cleopatra bath (fresh milk, olive oil, warm water), and ceramide-containing moisturizer twice daily. On day 3, worsening pustulosis led to the initiation of systemic corticosteroids (oral methylprednisolone 0.25 mg/kg/day, divided q8h),

resulting in significant clinical improvement within 3 days (Figure 4). The systemic corticosteroid was tapered off over 2 weeks of administration.

Follow-up and outcomes: By day 7, the patient revealed marked improvement and was discharged with ongoing Cleopatra baths, topical moisturizer, and tapering doses of oral steroids. Follow-up confirmed further clinical improvement without new pustular lesions or complications.

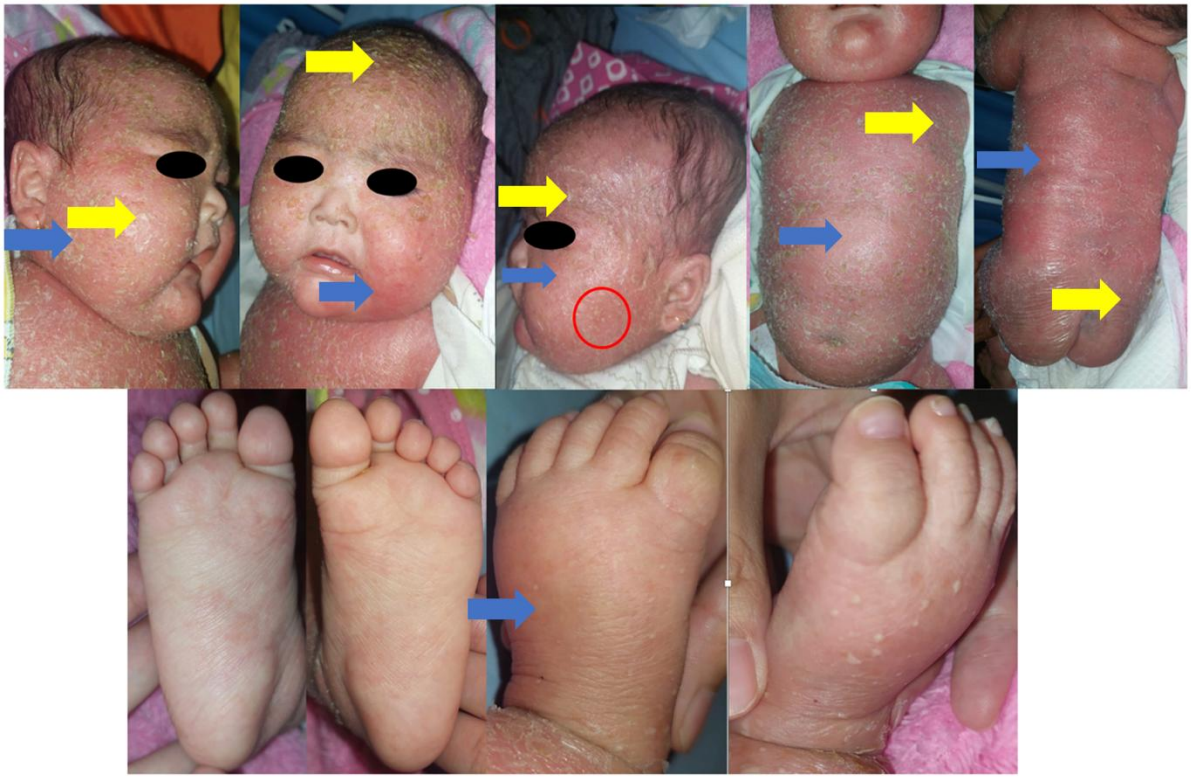

Figure 1. Clinical manifestation of the patient (Blue Arrow: erythematous patches, Yellow Arrow: scales, Red Circle: pustular formation)

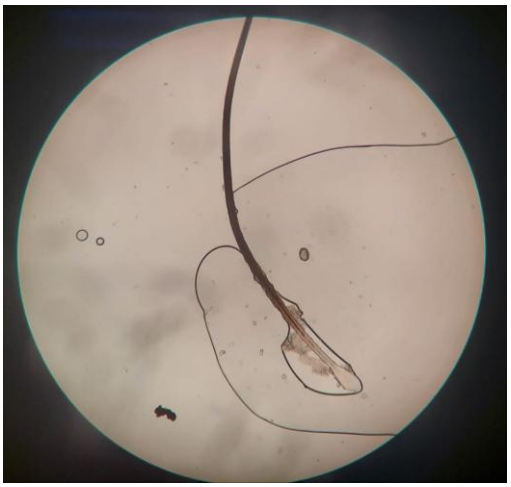

Figure 2. Microscopic examination of hair showed no sign of trichorrhexis invaginata

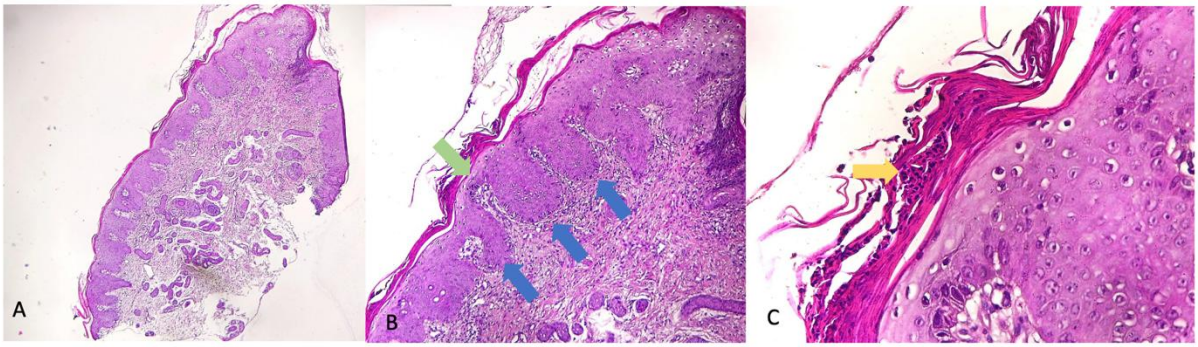

Figure 3. Histopathological examination (A. 40x magnification, B. 100x magnification, C. 400x magnification). Blue Arrow: psoriasiform acanthosis, Green Arrow: suprapapillary thinning, Yellow Arrow: Munro's microabscess.

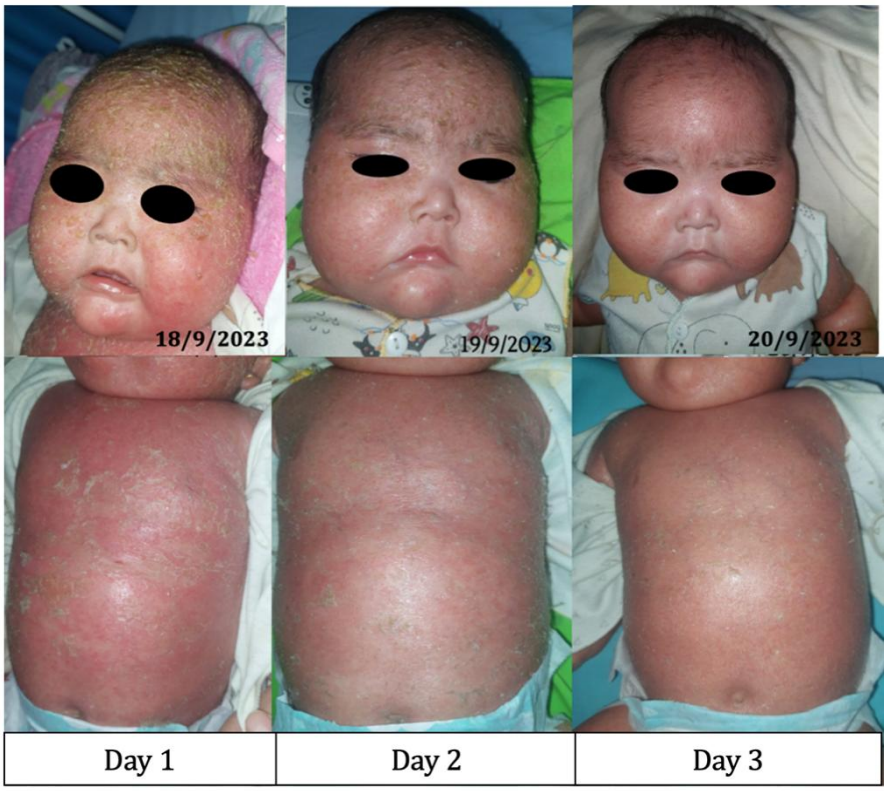

Figure 4. Clinical improvements (Day 1: erythematous patches and scales were prominent, Day 2: erythematous patches started to fade and scales started to decrease as compared to day 1, Day 3: further improvement in terms of scales as compared to day 1 and 2).

DISCUSSION

Erythroderma in infants is a rare but potentially life-threatening condition with a wide range of causes. There are six primary categories related to the causes: congenital ichthyoses (which includes Netherton syndrome (NS)), primary immunodeficiencies (PIDs), metabolic disorders, medication use, skin infections, and other causes.[3] Cuperus et al. reported that erythroderma in infants is most frequently due to ichthyosis, primary immunodeficiencies, and severe dermatitis.[4] Meanwhile, a review article by Swarnkar and Sarkar found that the leading causes were immunodeficiency, ichthyosis (simple and complex), Netherton syndrome, eczematous and papulosquamous dermatitis, and erythroderma of unknown aetiology.[2] Both studies highlight that psoriasis is a rare cause of erythroderma in infants.

In our case, differential diagnoses were based on the most common aetiologies described above. Among dermatitis-related causes, both atopic and seborrheic dermatitis were considered.

Atopic dermatitis can present in infancy, typically after the first month of life, but rarely progresses to erythroderma. Lesions are usually vesicular and exudative, often located on the cheeks, scalp, and extensor surfaces.[8] Infantile seborrheic dermatitis, which can overlap with atopic dermatitis, presents with yellowish scales and often involves the scalp and facial areas.[9] However, in our patient, no family history of atopy was noted, clinical features were more widespread, and histopathological findings did not show the expected spongiotic changes, making both atopic and seborrheic dermatitis unlikely. Another important consideration in infants with erythroderma is Netherton syndrome, a congenital disorder characterized by a triad of ichthyosiform erythroderma, *trichorrhexis invaginata* (bamboo hair), and atopic diathesis.[10,11] While the elevated IgE levels in our patient could suggest atopic predisposition, *trichorrhexis invaginata* was not observed, and histopathological features did not reveal parakeratosis, hyperkeratosis, or hypogranulosis, thereby ruling out Netherton syndrome.[10,11]

Histopathological examination revealed diffuse parakeratosis, regular acanthosis with club-shaped rete ridges (psoriasiform pattern), suprapapillary thinning, and neutrophilic exocytosis forming Munro microabscesses. The dermal papillae showed dilated capillaries with a mild perivascular infiltrate of lymphocytes, histiocytes, and neutrophils. No abnormalities were found in hair follicles or eccrine glands. These findings are consistent with a diagnosis of psoriasis.[12,13]

Managing erythroderma in infants presents unique challenges due to increased risks of fluid loss resulting from a high body surface area-to-volume ratio.[2] Supportive care should focus on fluid balance, electrolyte correction, monitoring vital signs, and preventing hyperpyrexia. In our case, fluid requirements were met with 570 cc/day, and paracetamol was administered at a dose of 15 mg/kg every 8 hours when needed (temperature exceeded 37.5^0C during hospital admission). There is currently no universally recognized treatment guideline for Generalized Pustular Psoriasis (GPP).[14] The treatment for acute GPP flare-ups relies on how severe the skin and overall health issues are. Generally, it involves a combination of topical treatments, systemic medications, and supportive care, which is often delivered in a specialized hospital setting.[14] Considering the possibly serious risks associated with GPP flares, the most suitable treatment options should provide a quick onset of action, a swift resolution of the disease, the capability to prevent future flares, and an acceptable safety profile.[3,15]

For generalized pustular psoriasis, treatment typically involves both topical and systemic therapies. Recommended topical options include emollients, corticosteroids, coal tar, dithranol, calcipotriol, and calcipotriene.[14] Our patient received a ceramide-based emollient twice daily and Cleopatra baths (milk and olive oil in lukewarm water), which have been shown to aid in desquamation and soothe inflamed skin.[16,17] Cleopatra bath is known to contain lactic acid, a natural alpha hydroxy acid (AHA), which is able to chemically peel the skin and aid in skin rejuvenation.[17] In our patient, the use of Cleopatra baths has proved to help soothe the skin (Figure 4).

Systemic retinoids are considered the first-line treatment for generalized pustular psoriasis in children. Srivatsa et al. demonstrated a successful case of infantile pustular psoriasis treated with oral acitretin at 0.56 mg/kg/day, tapered over 3–4 months, with remission at 10 months.[18] However, due to the unavailability of systemic retinoids in this setting, the patient initially received only topical therapy. Since we did a biopsy on the second day (not done on the first day since we needed to consult an anaesthesiologist to aid the sedation process), corticosteroid was withheld to avoid obscuring the histopathological findings. Then on a third day, with no clinical improvement, oral methylprednisolone was initiated at 0.25 mg/kg/day (equivalent to 0.5 mg every 8 hours), resulting in significant improvement. The steroid was tapered over two weeks, and the patient remained stable at follow-up.

Vitamin D supplementation was also provided due to a documented deficiency. Vitamin D has antiproliferative and immunomodulatory effects, including downregulation of pro-inflammatory cytokines such as IL-3, IFN-γ, IL-6, and IL-8, and inhibition of dendritic cell activation and T-cell maturation.[19,20] Additionally, vitamin C and zinc were administered to support immune function and reduce the risk of secondary infections. After all the interventions, the patient's condition was improved, as shown in Figure 4.

CONCLUSION

A case of erythroderma due to generalized pustular psoriasis in a 6-month-old female infant has been reported, showing a positive response to a combination of topical therapy using a ceramide-containing moisturizer, Cleopatra baths (milk and olive oil), and systemic corticosteroid therapy with methylprednisolone at a dose of 0.25 mg/kg/day, tapered weekly over a total treatment duration of two weeks. The case was managed through a multidisciplinary approach involving dermatologists and paediatricians, resulting in significant improvement of the skin lesions.

CONFLICT OF INTEREST

The authors declare that there is no conflict of interest regarding the publication of this case report. Informed consent was obtained from the patient's legal guardian (parents) for publication of this case.

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The authors thank the parents for their permission to publish this case as a case report.

DATA AVAILABILITY STATEMENT

The data supporting the findings of this case report are not publicly available due to privacy and ethical restrictions. However, the data can be made available from the corresponding author upon reasonable request.

SUPPLEMENTARY MATERIAL(S)

None.

AUTHORS CONTRIBUTIONS

MA, YWW, and RD contributed equally to the conception, clinical management, and writing of this case report. HTR was responsible for the histopathological examination and interpretation. MA was primarily responsible for drafting the manuscript and compiling patient data. RD supervised the clinical analysis, provided critical revisions, and approved the final version of the manuscript. All authors have read and agreed to the final manuscript.

DECLARATION OF USING AI IN THE WRITING PROCESS

The authors declare that no artificial intelligence (AI) or AI-assisted technologies were used in the writing or preparation of this manuscript. All content was generated and reviewed solely by the authors.

LIST OF ABBREVIATIONS

ANA: Antinuclear Antibody, HR: Heart Rate, RR: Respiratory Rate, SpO_2: Peripheral Oxygen Saturation, Hb: Haemoglobin, WBC: White Blood Cell, IgE: Immunoglobulin E, IV: Intravenous, q6h: Every 6 hours, q8h: Every 8 hours, IU: International Unit, mg/kg: Milligrams per kilogram, °C: Degrees Celsius.

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