

Ecthyma gangrenosum in a 3-month-old, previously healthy infant

A Case Report

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Abstract

Rationale: Ecthyma gangrenosum (EG) is an aggressive cutaneous disease caused by local or systemic infection with *Pseudomonas aeruginosa*. EG is characterized by cutaneous manifestations ranging from nodule and papule, to necrotic ulceration with surrounding erythema, especially with black eschar or central crust. EG presents with characteristic skin lesions which is important to establish diagnosis of sepsis caused by *P. aeruginosa*, a serious condition that can be treated efficiently if diagnosed early.

Patient concerns: A 3-month-old female infant was presented with characteristic skin lesions of EG and developed sepsis 3 days later.

Diagnoses: Ecthyma gangrenosum and sepsis caused by *Pseudomonas aeruginosa*.

Interventions: Meropenem was used in combination with ceftazidime at first and excision of necrotic skin lesions was performed later.

Outcomes: Cure.

Lessons: Early recognition of EG plays an important role in providing appropriate empiric antibiotic treatment at early stage of sepsis, and improves the prognosis. Surgical excision may be helpful if no improvement was achieved via antibiotic treatment.

Abbreviation: EG = ecthyma gangrenosum.

Keywords: ecthyma gangrenosum, previous healthy infant, *Pseudomonas aeruginosa*, pseudomonas sepsis

1. Introduction

Ecthyma gangrenosum is a skin lesion that results from either primary skin infection or hematogenous seeding by a bacterium. EG gangrenous ulcer is characterized by a black eschar or central crust and may be surrounded by a red halo.^[1–3] EG is mainly caused by infection with *Pseudomonas aeruginosa* (*P. aeruginosa*), which has a high mortality. EG is clinically relevant not only because of its potentially fatal prognosis, but also because it may signal the presence of a predisposing condition.^[4] EG has rarely been reported in previously healthy individuals. Here, we

report a case of severe *P. aeruginosa* sepsis in a 3-month-old, previously healthy infant.

2. Case presentation

A 3-month-old female infant was admitted to our Pediatric Intensive Care Unit (PICU), with a 3-day history of fever, cough, and widespread skin lesions, as well as diarrhea that has persisted for 1 day. Oral betamethasone was initiated for 3 days prior to admission, because of the skin lesions had become more and more necrotic, leading to a clinical suspicion of pyoderma gangrenosum. On admission, the patient appeared toxic, with high fever of approximately 40.3°C. Physical examination highlighted multiple skin lesions, 2 to 5 cm in diameter, which were in different stages of evolution and presented as round, ulcerated, necrotic papules, and nodules with central crust. The lesions were distributed all over the body (Fig. 1A and 1B). The laboratory results were as follows: hemoglobin, 9.7 g/dL; white blood cells, 9700/mm³; absolute neutrophil count, 1756/mm³ (18.1%); platelets, 264,000/mm³. Gram staining of showed Gram-negative bacilli. Computed tomography of the lungs indicated a large opacity in the basal-rear segments of both lungs, with pleural effusion. Empiric treatment with intravenous meropenem (15 mg/kg per day) was initiated after collecting blood, cerebrospinal fluid, and skin lesion (pustule) samples. The patient's general condition quickly improved, and fever decreased. On day 2 after admission, the diarrhea and respiratory symptoms resolved. The cultures of blood and skin lesion grew *P. aeruginosa*. A repeated blood test performed on day 3 after admission showed: hemoglobin, 9.75 g/dL; white blood cells, 35,800/mm³; absolute neutrophil count, 17,560/mm³ (68.3%); and normal red blood cells. Thus, the diagnosis of EG and *P. aeruginosa* sepsis was confirmed. According to the results of the

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Figure 1. *P. aeruginosa* ecthyma gangrenosum ulcer in the face, abdomen, and extremities (A, B).

drug susceptibility test, intravenous ceftazidime (50 mg/kg, twice a day) was added. Daily drainage of the lesions was started and topical antibiotic therapy with iodine and fusidic acid cream (20%) was instituted twice a day. Serology tests were negative for HIV, herpes simplex virus, cytomegalovirus, Epstein-Barr virus, and mumps/measles/rubella. Immunologic workup showed the following: CD3 cells, 70% (normal range, 54–80%); CD4 cells, 45.34% (normal range, 25–40%); CD8 cells, 12.51% (normal range, 18–30%); CD4/CD8 ratio, 3.62 (normal range, 0.98–1.94); CD16+CD56 cells (natural killer cells), 2.62% (normal range 8.1–25.6%); CD19A cells (B lymphocytes), 25.87% (normal range, 7–20%); IgG antibodies, 415 mg/dL (normal range, 750–1560 mg/dL); IgA antibodies, 9.5 mg/dL (normal range, 3–82 mg/dL); IgM antibodies, 57.3 mg/dL (normal range, 92–204 mg/dL). The treatment continued for a month, during which it was gradually tapered off without recurrence of sepsis; repeat blood counts were carried out every 3 days for 3 weeks, and consistently showed absolute counts of white blood cells $< 9700/\text{mm}^3$. However, there was no significant improvement of the lesions. Antibiotic treatment was extended for another 2 weeks, but no improvement was achieved with respect to the lesions. Finally, excision of necrotic plaque was performed. The pathology of the lesion showed acute inflammatory cell infiltration and vascular proliferation in the dermis (Fig. 2A). Abscesses due to abundant necrotic adipose and fibrous tissues were noted in the subcutis (Fig. 2B). Gram staining of the biopsy indicated a few Gram-negative bacilli in collagen fiber

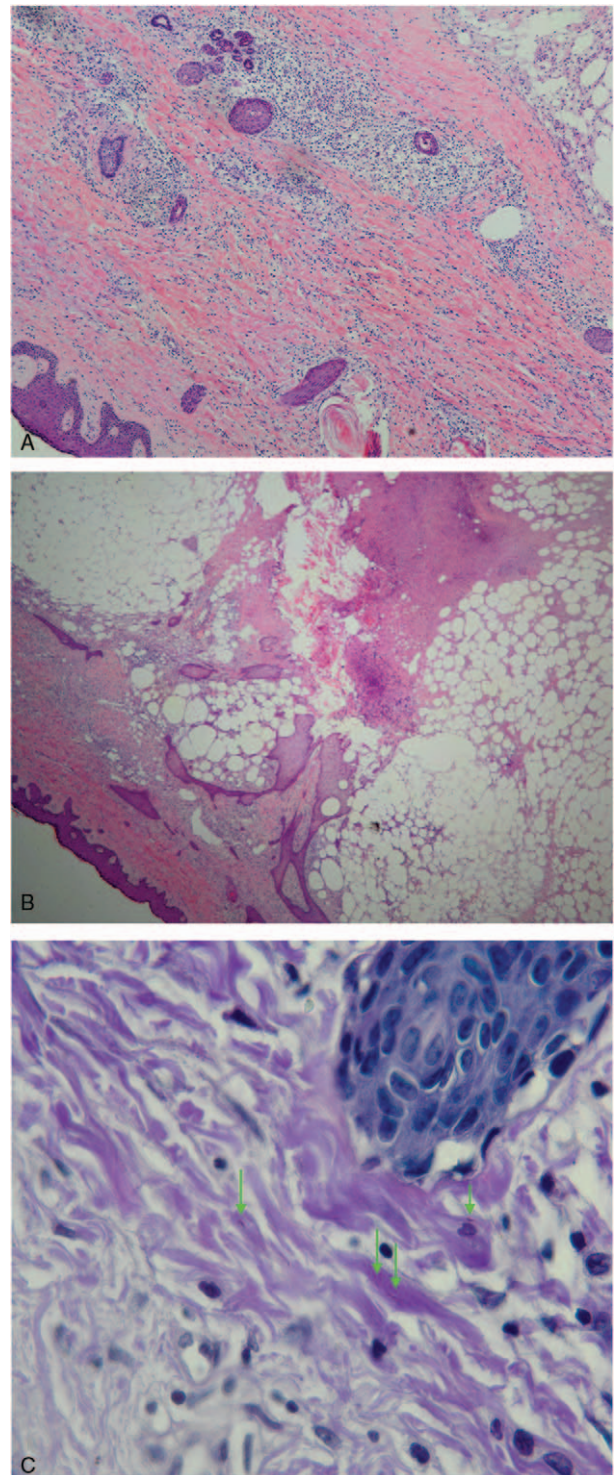


Figure 2. Microscopic examination revealing acute inflammatory cell infiltration and vascular proliferation in the dermis (HE; original magnification x40) (A). Abundant adipose and fibrous tissues, and formation of abscesses in the subcutis (HE; original magnification x40) (B). Gram staining of the biopsy indicated a few Gram-negative bacilli in collagen fiber bundles (Gram stain; original magnification x1000) (C). In the report, we just retrospectively analyzed the patient's diagnosis and treatment process. All the laboratory tests and all the medicine used were according to the patient's condition. There was not any clinic research of the report. This case report just informs clinical practice and enhances critical thinking. The ethical approval was not necessary. The patient's parent has already given informed consent; we can provide if requested. H&E = hematoxylin and eosin.

bundles (Fig. 2C). The patient was discharged 10 days after the excision. Small scars were left on the abdomen, extremities, and buttocks. The follow-up examination 2 year after the initial visit revealed no sign of recurrence or new lesions.

3. Discussion

Ecthyma gangrenosum is the cutaneous manifestation of pseudomonas infection in patients with sepsis that can develop all over the body.^[1–3] Patients with chronic diseases and immunodeficiency are typically at risk for developing EG. However, there have been rare case reports of EG happened in previously healthy children.^[4–7] It was suggested that these patients may have had risk factors for the development of EG or unrecognized underlying medical conditions.^[4] Our patient had been previously healthy, but her immunologic workup indicated that she was mildly immunocompromised. Most previous reports mentioned that the skin lesions usually occur in the gluteal and perineal regions (57%), or in the extremities (30%).^[6] It was peculiar for our patient without history of chronic diseases, immunodeficiency, or recent previous antibiotic therapy to suffer EG, with such an extremely wide distribution of the EG lesion.

Two pathogenic mechanisms of EG have been well described, namely the bacteremic and the nonbacteremic form.^[8] In the classic or bacteremic form, the skin lesions represent the hematogenous dissemination of the organism to the skin. Therefore, in such patients, blood cultures are positive. In the nonbacteremic form, the patients may tend to have a better prognosis than those who are septicemic^[9]; however, secondary bacteremia may occur if treatment is delayed. Our patient initially presented with fever, diarrhea, and respiratory symptoms, characteristic of the bacteremic form. Sepsis developed later, as proven by the growth of *P aeruginosa* in the skin lesion and blood cultures. Some authors have reported *P aeruginosa* sepsis in previously healthy children without underlying medical problems. Different clinical manifestations were observed in these children, the most relevant being skin lesions, fever, diarrhea, pneumonia, and shock. The mortality rate was approximately 55%.^[10] It is thus important to notice such manifestations early, so that appropriate antibiotic treatment can be instituted timely. This is especially true for patients such as ours, who had normal white blood cell count and was mildly neutropenic at the time of presentation, but later developed sepsis. Qualitative neutrophil defects represent significant risk factors for sepsis. EG lesions usually present before the results of the blood and lesion cultures and help to administer appropriate antimicrobial therapy without delay.^[11] In our patient, widespread EG lesions were characteristic of *P aeruginosa* septicemia. Antibiotic treatment is usually composed of cephalosporins, aminoglycosides, or penicillins, alone or in combination. For our patient, meropenem combined with ceftazidime resulted in a quick cure.^[10] Additionally, although ulcer and eschar were characteristic lesions in our child, bacterial cellulitis, group A beta hemolytic deep impetigo, leukocytoclastic vasculitis-like vasculi-

tis, and malignancy should also be included in the differential diagnosis.^[7,11]

EG treatment has 3 stages: initial empiric antibiotic therapy is administered as soon as infection is suspected; when the etiology is established, aggressive antibiotic or antifungal treatment is administered; finally, surgical excision is often necessary, because EG manifests as a necrotizing soft-tissue lesion.^[12] The treatment plan of our patient was made according to the 3 stages. Based on the prognosis and outcome of our patient, surgical excision may be helpful if no improvement was achieved with respect to the lesions in the process of conventional antibiotic treatment.

4. Conclusion

Early recognition of EG plays an important role in providing appropriate empiric antibiotic treatment early in the development of sepsis, and improves the outcome. Clinical awareness of *P aeruginosa* infection in previously healthy children should be increased. Surgical excision may be helpful if no improvement was achieved with respect to the lesions in the process of conventional antibiotic treatment, since extended course of the antibiotic treatment was not effective in our case.

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