

Ecthyma Gangrenosum about a Case at the Centre Hospitalier Roi Baudouin de Guédiawaye in Senegal

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How to cite this paper: Diouf, J.B.N., Barry, H.M., Sarr, L.P. and Ba, N.D. (2022) Ecthyma Gangrenosum about a Case at the Centre Hospitalier Roi Baudouin de Guédiawaye in Senegal. *Open Journal of Pediatrics*, 12, 652-656.

<https://doi.org/10.4236/ojped.2022.124066>

Received: June 23, 2022

Accepted: August 29, 2022

Published: September 1, 2022

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Abstract

Background and Aim: Ecthyma gangrenosum (EG) is a rare necrotic skin infection caused by *Pseudomonas aeruginosa* occurring on healthy skin mainly, but not exclusively, in immunocompromised patients. We report the case of EG of unusual presentation in a 12-month-old infant. **Case Presentation:** This was a 12-month-old male infant who was referred from a peripheral structure for the treatment of facial swelling associated with fever. In his antenatal and intra nasal history, there was a notion of hospitalization for 15 days for early neonatal infection and a delay in psychomotor development. The admission examination showed the fair general condition, hyperthermia at 39°C, tachycardia at 136 beats/minute, and polypnea at 48 cycles/minute. The weight-for-height index with a z-score of 3DS was also found. The dermatological examination at the entrance found an extensive brownish inflammatory tense infiltrated plaque taking the mandibular, submental, retro-auricular occipital regions rapidly evolving towards necrosis, a significant swelling predominant on the upper right eyelid preventing the opening of the associated eyes to mouth ulceration. Biological abnormalities were predominantly neutrophilic leukocytosis, elevated CRP, and elevated muscle enzymes. Microbiological examination of skin samples had isolated *Pseudomonas aeruginosa*. He had received antibiotic therapy, and a necrosectomy under general anesthesia. The evolution was favorable and he was discharged from the hospital on D26 of hospitalization. **Conclusion:** This observation is rare due to its location, its early onset, and non-neutropenic background.

Keywords

Ecthyma Gangrenosum, *Pseudomonas aeruginosa*, Infant

1. Introduction

Ecthyma gangrenosum (EG) is a rare skin infection [1]. It may be a systemic infection occurring on healthy skin mainly, but not exclusively, in immunocompromised patients; thus, it may be a manifestation of an underlying clinical entity [1] [2] [3]. The pathogen of ecthyma gangrenosum is *Pseudomonas aeruginosa*, which is a gram-negative, aerobic coccobacillary bacterium [4]. EG initially presents as a round, painless erythematous macule that later becomes nodular bullous or pustular with an indurated base and margin [1] [4] [5]. EG is clinically important not only because it is an infection with a potentially serious outcome, but may also signal the presence of a predisposing condition. The most common risk factor for EG is neutropenia in the context of malignancy or immunosuppressive therapy. EG has rarely been reported in healthy individuals [5]. Treatment is based on parenteral antibiotic therapy adapted to the antibiogram (third-generation cephalosporin, fluoroquinolones). We report the case of a 12-month-old infant who presented with EG without known predisposing factors, and who developed neutropenia during admission.

2. Clinical Case

This was a 12-month-old male infant referred from a peripheral facility for management of facial swelling with fever. His ante- and perinatal history included a 15-day hospitalization for early neonatal infection. He was breastfed for six months before diversification and his vaccination status was up to date according to the Senegalese EPI. In addition, there was a delay in psychomotor development (no sitting or standing at his current age). The examination on admission showed a fair general condition, hyperthermia at 39°C, tachycardia at 136 beats/minute, and polypnoea at 48 cycles/minute. The weight-for-height index with a z-score of 3DS was also found. The dermatological examination on entry revealed a brownish inflammatory infiltrated placard extending to the mandibular, sub-chin, and retro-auricular occipital regions and rapidly evolving towards necrosis (Figure 1), a large swelling predominantly on the upper right eyelid preventing the opening of the eyes associated with oral ulceration. The rest of the physical examination was normal. Biological abnormalities were hyperleukocytosis (27,500/mm³) with a predominance of neutrophils (19,780/mm³ or 72.0%), an elevated CRP of 198.3 mg/L, and an increase in muscle enzymes (CPK: 295 U/L, LDH: 1136 U/L). Renal function, blood ionogram, HIV-PCR, serum protein electrophoresis, CD4 and CD8 lymphocyte counts were normal. The blood culture was negative and the microbiological examination of the skin samples showed *Pseudomonas aeruginosa* sensitive to fosfomycin, gentamicin, and tobramycin and intermediate to imipenem, aztreonam, and ciprofloxacin. He was treated with intravenous antibiotics (amoxicillin + clavulanic acid 80 mg/kg/day), local care, and nutritional management. He then underwent necrosectomy under general anaesthesia (Figure 2) and antibiotic therapy was secondarily adapted to the antibiogram (imipenem + gentamicin).



Figure 1. Tense brownish inflammatory infiltrated plaque extending to the mandibular, submental, retro-auricular and occipital regions rapidly progressing to necrosis.



Figure 2. Lesions after necrosectomy.

The evolution was favourable with good healing of the lesions and stable apyrexia. The patient was discharged from hospital on day 26, with per on antibiotics and care every two days.

3. Discussion

Pseudomonas aeruginosa is one of the bacteria frequently implicated in opportunistic infections and responsible for many hospital-acquired infections. The infection is represented in its most severe form by ecthyma gangrenosum [6]. Ecthyma gangrenosum (EG) is a severe skin infection caused by *Pseudomonas aeruginosa*, most often occurring in an immunocompromised environment [6]. In children, it occurs in the context of primary immunodeficiency, cancer (50%) and in particular acute leukaemia, acquired neutropenia and haemophagocytic syndrome [7]. In our patient, severe malnutrition was present. Its occurrence in immunocompetent patients is rare, although some case reports describe occur-

rences in healthy children [8]. A review of the literature showed that most of these patients, although previously healthy, had risk factors for the development of ecthyma gangrenosum or had unrecognised underlying medical conditions. Our age was 12 months, whereas Zommodi A *et al.* [8] found that of the 18 children, 16 were infants under 22 months of age. The patient reported in this study had lesions located on the face. This finding was also found in the study by César AML *et al.* [9] who also reported a facial location in 3 cases out of their series of 8 patients. Other locations described were anogenital and axillary. EG typically begins with erythematous macules followed by the formation of bluish haemorrhagic bullae that rupture and give rise to necrotic ulcerations surrounded by an erythematous halo. The patient described here had an unusual clinical presentation with the appearance of a rapidly necrotic lesion progressively extending to the entire sub-mandibular area, reflecting the great clinical polymorphism of this condition. The most common risk factor for EG is neutropenia in immunocompromised patients. Neutrophils are the main host defence against *Pseudomonas* infection and their qualitative and quantitative deficiency are important risk factors for the development of ecthyma. Neutrophil values below $500/\text{mm}^3$ present the greatest risk of severe infection [10]. We found the patient to have neutropenia of $1000/\text{mm}^3$ during hospitalisation. A review of the English literature showed that many apparently healthy patients with ecthyma gangrenosum were neutropenic at the time of presentation [8]. Although neutropenia is a risk factor for the development of ecthyma gangrenosum, it has been suggested that *Pseudomonas* infection can cause a transient neutropenic state by producing a toxin that inhibits neutrophil migration into infected areas and decreases the number of circulating neutrophils [8]. Of the five children described by Neta C *et al.* [10], four were neutropenic with an absolute neutrophil count $< 1500 \text{ mm}^3$ after presenting with ecthyma gangrenosum. This indicates that although the absolute neutrophil count at presentation may be near normal, it may collapse soon after to severe neutropenia and sometimes remain severe and even require treatment. This may be partly explained by the fact that the *Pseudomonas* infection itself reduces the number of circulating neutrophils [10]. Another possible explanation is that these patients were neutropenic before presentation and had a proliferative response to the acute infection resulting in a temporarily normal absolute neutrophil count. Our patient was undernourished with a z-score of 3DS. César A M L *et al.* [9] in their series of 8 cases found undernutrition as a risk factor.

4. Conclusion

This observation is rare because of its location, its early occurrence, and its non-neutropenic background.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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