A case of perineal ecthyma gangrenosum

Zeynep Meltem Akkurt¹, Veysi Fidan¹, Derya Uçmak¹, Tuba Dal², Selvi Kelekçi³, Haydar Uçak¹

¹Department of Dermatology and Veneral Diseases, Dicle University School of Medicine, Diyarbakır, Turkey

²Department of Microbiology, Dicle University School of Medicine, Diyarbakır, Turkey

³Department of Pediatrics, Dicle University School of Medicine, Diyarbakır, Turkey

Abstract

Ecthyma gangrenosum is a skin lesion associated with *Pseudomonas aeruginosa*. A previously healthy one-year-old boy who had been diagnosed with varicella 10 days ago was admitted to our hospital with complaints of diarrhea, green ear discharge and new lesions in the diaper area. Intravenous meropenem and amikacin had been previously initiated. Physical examination revealed greenish, well bordered necrotic ulcers on both gluteal areas and the perianal area. *Pseudomonas aeruginosa* grew in wound culture. A colostomy was opened due to recalcitrant diarrhea and ulcers. The patient remitted in one month and was discharged. T lymphocyte subgroups and immunoglobulins were found to be normal on immunologic evaluation. We presented this case to draw attention to the fact that ecthyma gangrenosum due to *Pseudomonas aeruginosa* may occur in the diaper area and that sepsis due to *Pseudomonas* should be investigated. (Turk Pediatri Ars 2016; 51: 46-8)

Keywords: Ecthyma gangrenosum, pseudomonas, sepsis

Introduction

Pseudomonas aeruginosa is an aerob, gram negative bacterium and grows in humid environments. It leads to very severe infection in individuals who have immunological disorders and who have undergone invasive therapeutic procedures. In addition, it also causes to severe nosocomial infections (1). Ecthyma gangrenosum is a cutaneous finding related with *Pseudomonas aeruginosa*. Underlying sepsis should be investigated in patients who are thought to have ecthyma gangrenosum, because the mechanism of development includes septic vasculitis in cutaneous vessels.

Case

A previously healthy one-year old male patient had been diagnosed with varicella in another center and was referred to our hospital after intravenous meropenem (20 mg/kg/day) and amikacin (15 mg/kg/day) were initiated when diarrhea, green ear discharge and lesions different from varicella lesions in the diaper area occurred 10 days after the diagnosis of varicella. On physical examination, greenish, sharply-circumscribed necrotic ulcers were observed in both gluteal areas and perianal area (Figure 1). Dermatology Clinic was consulted. Ecthyma gangrenosum was considered and it was recommended that

wound culture be performed and wet dressing and silver sulphadiazine cream be applied two times a day. The laboratory values during this period were as follows: WBC: 8.26 K/uL Hb: 11.6 g/dL Hct: %33.3 platelets: 454 K/uL lymphocyte: 6.05 K/uL, neutrophil: 0.052 K/uL (2.00-6.90), urea: 5 mg/dL, creatinine: 0.3 mg/dL ALT: 10 U/L, AST: 24 U/L, CRP: 1.76 mg/dL, IgG: 1.620 mg/dL, IgM: 312 mg/dL, IgA: normal C3, C4: normal, IgE: normal. Cryptosporidium and rotavirus antigens in stool were found to be positive. Meropenem was discontinued and ceftazidim at a dose of 30 mg/kg/day was added. Metranidazole at a dose of 30 mg/kg/day was added because of diarrhea and the lesions in the gluteal area. After one week, vancomycin at a dose of 15 mg/kg/day was added considering nosocomial gram positive infection in the patient who had fever. *Pseudomonas aeruginosa* was grown in wound culture and it was learned that it was resistant to ceftazidim. Ceftazidim was discontinued and treatment was continued with amikacin, vancomycin and metranidazole. The patient was also consulted with Pediatric Surgery Clinic and a colostomy was opened. One month after hospitalization, the patient whose general status and lesions improved was discharged. Colostomy was closed after two months. In the immunological evaluation, T lymphocyte subgroups and immunoglobulins were found to be normal. Informed consent was obtained for this case presentation.

Address for Correspondence: Zeynep Meltem Akkurt, E-mail: meltem@doctor.com

Received: 21.05.2013 **Accepted:** 29.07.2013

©Copyright 2016 by Turkish Pediatric Association - Available online at www.turkpediatriarsivi.com

DOI: 10.5152/TurkPediatriArs.2016.1300

Figure 1. Sharply-circumscribed necrotic ulcers with greenish centers in the perineal area

Discussion

The agent could not be shown in blood cultures, because intravenous antibiotics were initiated in another center, but we considered pseudomonas sepsis related with transient immunosuppression following varicella infection and related ecthyma gangrenosum.

Frequent cutaneous findings related with pseudomonas include folliculitis, wound site infection and green nail syndrome. Ecthyma gangrenosum which is a bacterial vasculitis is a rare cutaneous finding manifested by bacterial invasion of the venous and arterial walls and subcutaneous tissue in pseudomonas sepsis. This finding is observed in 1-3% of the patients who have pseudomonas sepsis. It is especially observed in the genital region, legs, abdomen and extremities. Necrosis involves small veins more frequently compared to arteries. The lesion which starts as an erythematous and purpuric macule transforms into a hemorrhagic bulla and a gangrenous ulcer with a black center in 12-24 hours. Some authors have described atypical endurated nodules with irregular borders (1-4). Multiple lesions, delayed treatment and neutropenia have been identified as predictors of poor prognosis (3).

Although ecthyma gangrenosum has been reported most commonly in relation with *Pseudomonas aeruginosa*, it may also be related with other bacteriae including *Aeromonas hydrophila*, *Staphylococcus aureus*, *Serratia marcescens*, *Escherichia coli*, *Klebsiella pneumonia* and *Citrobacter freundii* and even fungi (*Aspergillus spp*, *Mucor spp*, *Candida spp*) (5, 6). On histopathological examination, marked vesiculation in the epidermis and diffuse dermal necrosis are observed similar to pyoderma gangrenosum. A picture of vasculitis which invades into the middle and outer vascular layers and which does not involve the intima is observed. Intravascular thrombus which leads to regional necrosis is built up by the rods, immune complexes and/or bacterial exotoxins/endotoxins in the

vascular wall. Thus, obstruction and regional infarction is observed in the vessel. This characteristics explain rapid progression of the lesions to necrosis. The bacteriae may be observed as bluish clusters (4, 7).

In healthy children, sepsis related with pseudomonas species occurs rarely. However, increasing case reports suggest that pseudomonas sepsis in infants is not recognized sufficiently.

Ecthyma gangrenosum is a helpful skin lesion in the diagnosis of *Pseudomonas aeruginosa* sepsis. Suspicion is strengthened if otitis media, fever, pneumonia, gastrointestinal involvement, neutropenia and coagulation defects are present additionally. These clinical features necessitate initiation of appropriate antibiotics which would rapidly affect *Pseudomonas aeruginosa* (1). It is known that ceftazidim (alone) or combination of piperacillin-tazobactam and aminoglycoside is effective. The lesions have resolved by leaving scar despite antibiotic treatment in some case reports (7).

Seventy three cases were reported in an article which summarized *Pseudomonas aeruginosa* sepsis reported in healthy children in the literature (1). Most of the patients were reported to be male and below the age of one year. Skin lesions, fever, diarrhea, pneumonia and shock were the most commonly observed findings. The mortality rate was found to be about 55%. In the study of Huang et al. (8), pseudomonas infections which occurred in two hospitals in a period of 16 years were reported. In this study, it was reported that 43 of 121 children were healthy previously and most of them were boys aged below one year. The most commonly observed findings included fever and diarrhea.

In most of the cases, the cause of tendency to infection is not known, but transient immunosuppression related with drugs or infection may be present. On the other hand, *Pseudomonas aeruginosa* is present in the intestines in approximately 5-30% of healthy individuals. It has been proposed that antibiotic treatment may increase the relative intensity of *Pseudomonas aeruginosa* and predispose these individuals to infection. In a case report, the authors proposed that previous viral infection might have led to otitis media and pneumonia related with *Pseudomonas aeruginosa* and later to bacteriemia by weakening the mucosal barrier in a child. Another view is that there may be underlying hypogammaglobulinemia, cyclic neutropenia or neutrophil dysfunction in healthy children who develop pseudomonas sepsis (1, 3, 7).

However, it has been reported that benign ecthyma gangrenosum related with transient bacteriemia or

infection limited to skin may occur without sepsis in some publications. The reason for this may be a change in the intestinal flora and irritation in the diaper area. It is thought that the mortality rate is lower and clinical prognosis is better in these patients (9, 10).

In conclusion, this case was reported to remind that *Pseudomonas aeruginosa* infections may also occur in healthy children in contrast to the general opinion and to emphasize that ecthyma gangrenosum is a finding which leads to diagnosis.

Informed Consent: Written informed consent was obtained from patients' parents who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - Z.M.A., T.D.; Design - V.F., H.U.; Supervision - T.D., H.U., S.K.; Materials - Z.M.A., V.F., D.U.; Data Collection and/or Processing - Z.M.A., V.F., D.U.; Analysis and/or Interpretation - Z.M.A., D.U.; Literature Review - Z.M.A., V.F.; Writing - Z.M.A., D.U., H.U.; Critical Review - T.D., S.K.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study has received no financial support.

References

1. Viola L, Langer A, Pulitano S, et al. Serious pseudomonas aeruginosa infection in healthy children: case report and review of literature. *Pediatr Int* 2006; 48: 330-3. [\[CrossRef\]](#)
2. Pacha O, Hebert AA. Ecthyma gangrenosum and neutropenia in a previously healthy child. *Pediatr Dermatol* 2013; 30: e283-4. [\[CrossRef\]](#)
3. Wu CT, Huang JL. Multiple ecthyma gangrenosum in a healthy infant with community-acquired *Pseudomonas aeruginosa* sepsis. *Pediatr Emerg Care* 2010; 26: 750-1. [\[CrossRef\]](#)
4. Goolamali SI, Fogo A, Killian L, et al. Ecthyma gangrenosum: an important feature of pseudomonal sepsis in a previously well child. *Clin Exp Dermatol* 2008; 34: e180-2. [\[CrossRef\]](#)
5. Pulido J, McMahon P, Treat JR, et al. Labial ecthyma gangrenosum in an immunocompromised infant with leukemia: heightening awareness for the urologist. *Urology* 2012; 80: 1366-8. [\[CrossRef\]](#)
6. Fang LC, Peng CC, Chi H, Lee KS, Chiu NC. *Pseudomonas aeruginosa* sepsis with ecthyma gangrenosum and pseudomembranous pharyngolaryngitis in a 5-month-old boy. *J Microbiol Immunol Infect* 2014; 47: 158-61. [\[CrossRef\]](#)
7. Koo SH, Lee JH, Shin H, Lee JI. Ecthyma gangrenosum in a previously healthy infant. *Arch Plast Surg* 2012; 39: 673-5. [\[CrossRef\]](#)
8. Huang YC, Lin TY, Wang CH. Community-acquired pseudomonas aeruginosa sepsis in previously healthy infants and children: analysis of forty-three episodes. *Pediatr Infect Dis J* 2002; 21: 1049-52. [\[CrossRef\]](#)
9. Huminer D, Siegman-Igra Y, Morduchowicz G, Pitlik SD. Ecthyma gangrenosum without bacteremia. Report of six cases and review of the literature. *Arch Intern Med* 1987; 147: 299-301. [\[CrossRef\]](#)
10. Taieb A, Boisseau AM, Sarlangue J, et al. Gangrenous ecthyma of the diaper area in infants. *Ann Pediatr (Paris)* 1992; 39: 443-6.