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A Rare Case of Ecthyma Gangrenosum Caused by *Proteus vulgaris* and *Candida albicans* in a Patient with Castleman Disease

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Ecthyma gangrenosum (EG) is an ulcerative pyoderma of the skin that extends into the dermis and subdermal tissue with necrotic changes, and is generally described as a round, deep, punched-out ulceration with a central black eschar surrounded by an erythematous halo [1]. Development of EG is most commonly associated with *Pseudomonas aeruginosa* and is often seen in immunocompromised patients. However, cases of EG caused by other bacterial, viral, and fungal pathogens have been reported in the literature, albeit rarely. Prompt surgical debridement with appropriate antibiotic therapy is crucial for lowering the mortality rate in EG patients [1]. We describe an interesting case of EG caused by *Proteus vulgaris* (*P. vulgaris*) and *Candida albicans* (*C. albicans*) in a patient with multicentric Castleman disease (MCD), which involves hyperactivation of the immune system and multiple organ system dysfunction. Our patient showed extensive lesions involving necrotic ulcerative changes on the lower abdomen and on the right upper arm. To the best of our knowledge, this was a rare case of EG associated with *P. vulgaris* and *C. albicans*.

A 57-year-old woman presented to the emergency department with multiple skin lesions involving her trunk and upper extremities. Six months previously, the patient had developed a perianal fistula and a colostomy was performed. The patient noticed an erythematous skin rash on her right arm that spread to her trunk and gradually progressed into necrotic bullae. Her past history indicated that she had been diagnosed with MCD three years previously, was taking immunosuppressive agents with oral steroids, and had no history of taking antibiotics. The patient had a mild temperature of 38.0°C, and the laboratory findings showed leukopenia with thrombocytopenia. A skin examination revealed a deep punched-out ulceration on the abdomen near the colostomy site and a large, palm-sized eschar formation with an erythematous halo on her right arm (Figs. 1, 2). Under suspicion of EG, the patient received cefepime to treat the most likely cause, *Pseudomonas aeruginosa* infection. Superficial wound swabs and tissue for Gram staining and culture were obtained before antibiotic

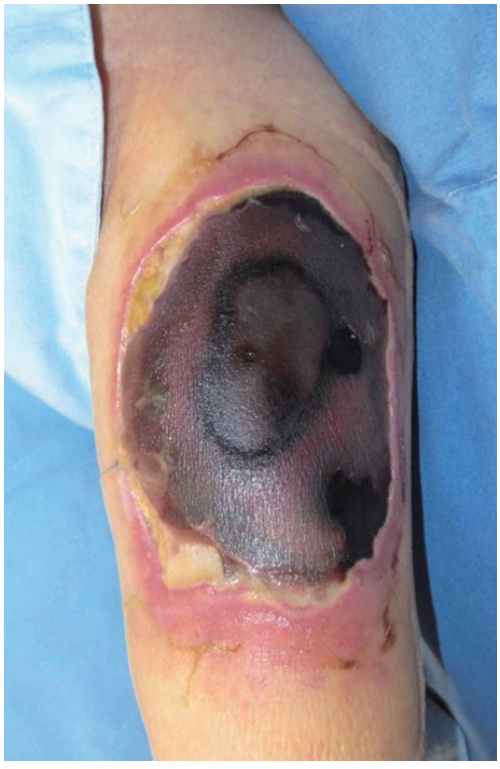

Fig. 1.
A large palm-sized eschar formation with erythematous halo changes on the patient's right arm.

Images

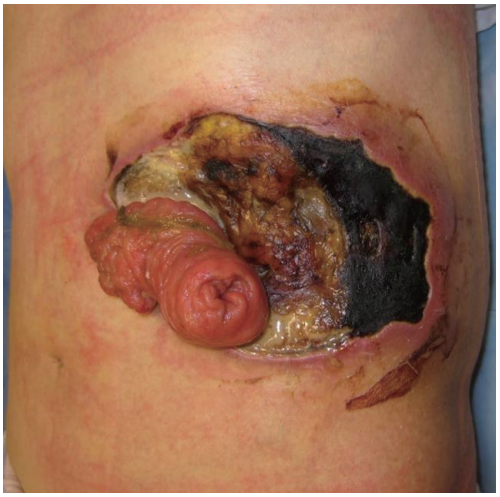

Fig. 2.
A deep punched-out ulceration lesion near the colostomy site with eschar formation.

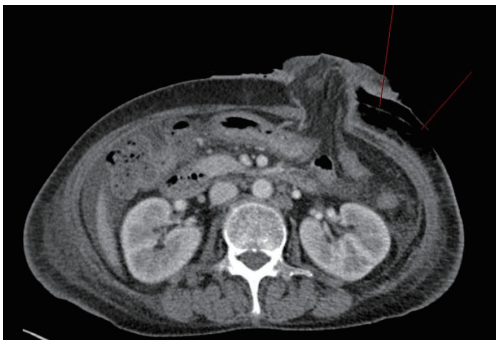

Fig. 3.
Abdominal computed tomography showed a subcutaneous emphysema without deep fascial enhancement.

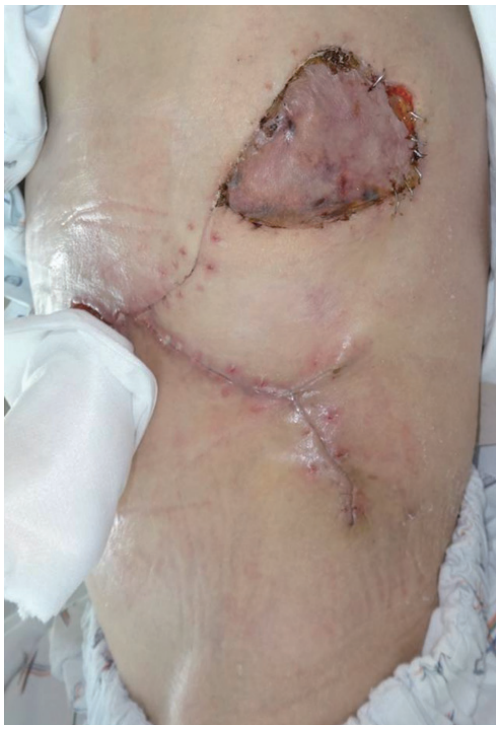

Fig. 4.
A completely healed lesion on the left flank of the patient, after surgical debridement and local flap coverage with a split-thickness skin graft were performed.

treatment, and Gram-negative bacilli were found and identified as *P. vulgaris*. Abdominal computed tomography showed subcutaneous emphysema without deep fascial enhancement (Fig. 3).

Surgical debridement and wound irrigation were performed to remove the necrotic tissue. Moreover, a follow-up culture was taken and a tissue biopsy was obtained from the necrotic lesion. The previously noted organism, *P. vulgaris*, was found along with *C. albicans*. Appropriate antibiotic and antifungal treatment was initiated, including ceftazidime and teicoplanin with fluconazole. After changing antibiotics and adding an antifungal agent, the lesions gradually improved. The erythematous changes around the eschar subsided and the central necrosis stopped progressing. A week after surgical debridement reconstruction was planned, local flap coverage with a split-thickness skin graft was performed to cover the defect. The patient completed three weeks of treatment with intravenous antibiotic and antifungal agents with no signs of fever, and the wound healed completely, with no complications (Figs. 4, 5).

EG is a relatively uncommon condition and the precise pathogenesis of EG is not well understood. However, it is considered to be the result of a perivascular invasion of numerous viable bacteria, leading to infection and necrosis of dermal and subdermal tissue [1]. Both bacteremic and nonbacteremic types of EG have been described. Nonbacteremic EG is considered to be the result of direct inoculation of the infected organism into the skin site. Nonbacteremic EG patients have a lower mortality rate of 16%, compared to 38%–96% for bacteremic EG patients [2]. Our patient's blood cultures were negative, which suggested the nonbacteremic type of EG. However, the presence of lesions in different anatomic locations suggests the possibility of a low-grade or transient bacteremia that was undetectable in blood cultures. Moreover, the initiation of empirical antibiotic treatment at the first sign of infection may have prevented septicemia in this patient. The organism most commonly associated with EG is *Pseudomonas aeruginosa*, but reports have demonstrated various other pathogenic etiologies for EG, including *Aeromonas hydrophila, Chromobacterium violaceum, Citrobacter freundii, Corynebacterium diphtheria, Escherichia coli, Klebsiella*

pneumoniae, *Morganella morganii*, *Neisseria gonorrhoeae*, *Serratia marcescens*, *Xanthomonas maltophilia*, and *Yersinia pestis* [3]. In a review study, Vaiman et al. [4] analyzed 167 cases of EG from 1975 to 2014. *Pseudomonas aeruginosa* was detected in 123 cases (73.65%), and other pathogenic etiologies were detected in 41 cases (26.35%). This suggests that a broader definition of EG is necessary, expanding the previous definition that identified the main pathogen as *Pseudomonas aeruginosa*. In order to confirm the pathogen involved in EG, a *Pseudomonas aeruginosa* antigen study can be performed.

Several other diseases involving cutaneous ulceration resemble EG, such as ecthyma, pyoderma gangrenosum, and necrotizing fasciitis, which may cause EG-like ulcerations. It is necessary to consider the differential diagnosis, because the presence other diseases involving cutaneous ulcerations that resemble EG may lead to the misdiagnosis of EG, and improper treatment may lead to a higher mortality rate. For example, pyoderma gangrenous is treated with systemic immunosuppressive therapy, such as oral or intravenous pulse corticosteroids, and cyclosporine, which has harmful side effects [5]. EG is typically found in immunocompromised individuals, most commonly in patients with a prior diagnosis of hematologic malignancies, and the prognosis depends on the patient's general health and immunological status [1]. The patient in our case was susceptible to infection due to underlying Castleman disease and had multiple large lesions caused by the microorganisms *P. vulgaris* and *C. albicans*, which are a rare finding. Due to prompt treatment with appropriate antibiotics and surgical debridement of all necrotizing tissues, septicemia was prevented and the patient was discharged from the hospital 31 days after admission.

In conclusion, our case emphasizes that although EG is considered pathognomic for *Pseudomonas aeruginosa*, other pathogens, including fungal or viral organisms, should be considered. Correspondingly, the choice of an initial antibiotic or antifungal agent may vary depending on the case. The high mortality rate of EG means that early diagnosis and proper treatment are very important for decreasing EG-related mortality, even when the diagnosis has not yet been confirmed.

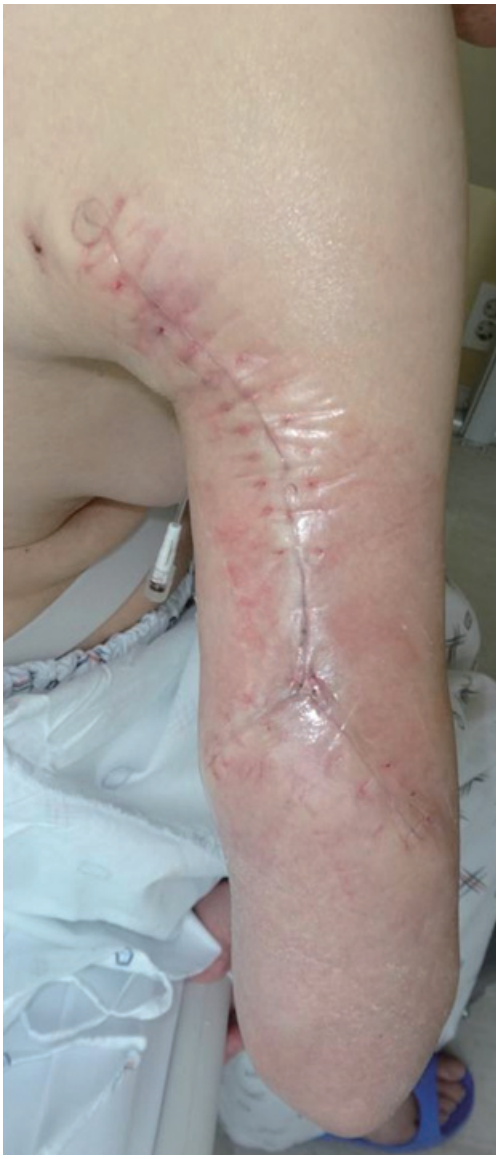

Fig. 5.
A completely healed lesion on the right arm of the patient, after surgical debridement and local flap coverage with a split-thickness skin graft were performed.

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