

Successful Use of Negative-pressure Wound Therapy and Dermal Substitute in the Treatment of Gluteal Ecthyma Gangrenosum in a 2-year-old Girl

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Summary: Ecthyma Gangrenosum is a manifestation of *Pseudomonas Aeruginosa* infection, usually occurring in immunocompromised patients, which can be associated with *Pseudomonas Aeruginosa* bacteremia with potentially lethal outcome. The clinical appearance is of an inflammatory cutaneous lesion with a central necrotic spot; the lesion then rapidly progresses to a gangrenous ulcer with a gray-black eschar extending in the deep soft tissues. Treatment of Ecthyma Gangrenosum includes both aggressive systemic antibiotic therapy and surgical procedures. A 2-year-old girl affected by B-cell precursor acute lymphoblastic leukemia was admitted to our hospital for suspected sepsis; the diagnosis was later confirmed by blood cultures positive for *Pseudomonas Aeruginosa*. In the days following the diagnosis, the patient developed a necrotic lesion of the right gluteal area consistent with Ecthyma Gangrenosum. Aggressive surgical debridement was then performed, followed by negative-pressure wound therapy and reconstruction with dermal substitute and autologous skin graft, which were successful. Ecthyma Gangrenosum is a potentially lethal condition affecting especially immunocompromised patients; aggressive medical treatment with combination antibiotic therapy is warranted and multiple surgical procedures, including extensive surgical debridement and diverting colostomy, are needed. Various reconstructive techniques have been reported in the literature, although no gold-standard can be established to date. Since Ecthyma Gangrenosum lesions are characterized by the presence of both high inflammatory activity due *Pseudomonas* infection and extensive tissue loss, the association of negative-pressure therapy and dermal substitutes implant seem to have a rationale in the surgical treatment of Ecthyma Gangrenosum and should therefore be considered. (*Plast Reconstr Surg Glob Open* 2018;6:e1953; doi: 10.1097/GOX.0000000000001953; Published online 3 October 2018.)

Ecthyma Gangrenosum is a manifestation of *Pseudomonas Aeruginosa* infection, usually occurring in immunocompromised patients, such as children affected by hematologic malignancies.¹

The condition initially presents as an inflammatory cutaneous lesion with a central necrotic spot; the lesion then rapidly progresses to a gangrenous ulcer with a gray-black eschar extending in the deep soft tissues.^{2,3} The perineum and the

axillary region are the most common sites of infection, even though virtually any site can be affected.⁴ Ecthyma Gangrenosum can be associated with *Pseudomonas Aeruginosa* bacteremia, in which case the condition can be fatal.⁵

Treatment of Ecthyma Gangrenosum includes both aggressive systemic antibiotic therapy and surgical procedures.^{1,2} Surgical treatment includes debridement and skin graft²; colostomy is often necessary in deep perineal lesions.⁴

We present a case of successful treatment of gluteal Ecthyma Gangrenosum with the use of negative-pressure therapy, dermal substitutes, and autologous skin graft, with the aim to provide some supportive experience to the currently limited pediatric literature about this topic.

CASE REPORT

A 2-year-old girl affected by B-cell precursor acute lymphoblastic leukemia was admitted to our hospital in

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Received for publication April 28, 2018; accepted July 31, 2018.

Presented at the 5th International Society for Pediatric Wound Care Annual Meeting, July 18, 2017, London, United Kingdom.

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DOI: 10.1097/GOX.0000000000001953

Disclosure: The authors have no financial interest to declare in relation to the content of this article. The Article Processing Charge was paid for by the authors.

the Hematology-Oncology Department for a febrile episode during chemotherapy-induced pancytopenia. On admission, she was in poor general conditions, febrile (TC 39°C) and tachycardic (HR 160 bpm). On examination, a violet nodule on the right buttock and 2 perianal nodules were noted. In the suspicion of sepsis, broad-spectrum intravenous antibiotics were commenced. Blood cultures were reported positive for *Pseudomonas Aeruginosa* 48 hours after the admission; the patient was then diagnosed with sepsis caused by *Pseudomonas Aeruginosa* and treated with intravenous Teicoplanin and Meropenem.

In the following 7 days, general conditions worsened with persistent fever and tachycardia the development of hypoproteinemia-induced edema. The gluteal nodule progressed to an extensive necrotic area involving the whole right buttock, starting from the perianal region (Fig. 1). Local tissue swabs were positive for *Pseudomonas Aeruginosa*. On magnetic resonance, necrotic tissue reached the fascia of the gluteal muscle, which was intact; rectum and anal sphincter were not involved. The patient was then diagnosed with Ecthyma Gangrenosum and referred to Burn and Plastic surgical team.

On day 11 from admission, the patient underwent surgical debridement of the necrotic tissue (Fig. 2); negative-pressure therapy with VAC-System[®] was then started and continued for 33 days. A first reevaluation under general anesthesia, with a second surgical debridement, was

performed 72 hours after the first intervention. Careful wound inspections and VAC foam changing were performed under general anesthesia every 72 hours for the first week, then every 5 days. The authors took advantage of the narrow healthy tissue between the perianal area and the lesion to apply the sealing transparent dressing. One of the surgeons of the team inspected the dressing daily or even more frequently, if required, and the external dressing was changed with the patient awake whenever there was any loss of negative pressure, as a sign of external contamination, or evidence of contaminating material.

General conditions improved within 48 hours after second surgical debridement. The patient became afebrile and inflammatory markers progressively decreased. Sepsis was controlled and chemotherapy according to BFM AIEOP ALL 2009 protocol was resumed 18 days after first surgical debridement.

Given the extensive loss of subcutaneous tissue, a dermal substitute (Integra[®]) was implanted after negative-pressure therapy was finished, 33 days after surgical debridement, covered with silicone sheet and conventional dressing (Fig. 3). An autologous split skin graft was then performed 22 days after the implantation of the dermal substitute; the donor area was the posterior aspect of the right thigh.

Skin graft healing was achieved, and the patient was discharged 14 days after skin grafting procedure (Fig. 4). The patient has fully recovered from the event with no functional deficit and is currently still on standard protocol treatment for acute leukemia

DISCUSSION

Ecthyma Gangrenosum is a potentially lethal condition affecting especially immunocompromised patients.¹

Due to the high of mortality of the condition, reported as high as 54% in perineal lesions associated with bacteremia,⁵ aggressive medical treatment with combination antibiotic therapy is warranted,^{6,7} and multiple surgical



Fig. 1. Clinical appearance of the lesion.



Fig. 2. Wound bed after surgical debridement.



Fig. 3. Implantation of Integra[®].



Fig. 4. Result of autologous graft 14 months postoperative.

procedures, including extensive surgical debridement and diverting colostomy, are needed.^{2,4,8}

Various reconstructive strategies have been adopted, including split skin graft,³ myocutaneous flap⁹ and even posterior anorectoplasty for perineal lesions with rectal involvement.² Case series, however, are too limited to establish a gold-standard treatment.

Use of negative-pressure wound therapy followed by autologous skin graft has been reported¹⁰; negative-pressure wound therapy removes the excess wound fluid, diminishes absorbance of toxic substances, promotes separation of nonvital tissue and reduces bacterial load of the treated area,¹¹ and is therefore often used in the treatment of necrotizing soft-tissue infections.¹²

In our case, since the rectum and the anal sphincter were not involved, colostomy was not performed, to prevent spreading of *Pseudomonas* infection from the bowel to the abdominal wall.^{10,13} Due to the severity of *Pseudomonas Aeruginosa* infection and the extensive loss of subcutaneous tissue, we decided to use negative-pressure wound therapy to prepare the wound bed and then we implanted a dermal substitute followed by autologous split skin graft.

Dermal substitutes are bio-matrices that fulfill the functions of the cutaneous dermal layer: control of pain and scarring.¹⁴ They act as matrices or scaffolds and promote new tissue growth and enhance wound healing¹⁵ and are routinely used in the reconstruction of full thickness

burns.¹⁶ There are currently only few cases of reconstruction with dermal substitutes of postinfectious lesions reported in the literature.^{17,18}

To our knowledge, this is the first reported case of combined use of negative-pressure wound therapy, dermal substitutes, and autologous split skin graft in the treatment of a necrotizing soft-tissue infection. Since Ecthyma Gangrenosum lesions are characterized by the presence of both high inflammatory activity due *Pseudomonas* infection and extensive tissue loss, the association of negative-pressure therapy, dermal substitutes, and autologous split skin graft seems to have a rationale in the surgical treatment of Ecthyma Gangrenosum and should therefore be considered.

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