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PAGES: 6-12

ORIGINAL PDF URL: <https://dergipark.org.tr/tr/download/article-file/519799>



Çocuklarda Ecthyma Gangrenosum'un Cerrahi Tedavi Yöntemleri: 3 Olgu Sunumu ve Pediatrik Hastalar için Literatürün Gözden Geçirilmesi

Surgical Treatment Modalities of Ecthyma Gangrenosum in Children: Report of 3 cases and Review of the Literature for Pediatric Patients

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ÖZ

Ecthyma Gangrenosum (EG) genellikle çeşitli semptom ve bulgularla vücudun çeşitli bölgelerini etkileyen ani başlangıçlı bir hastalıktır. Sorun Pseudomonas aeruginosa gibi enfeksiyonların bir sonucu gibi görünse de, bazı öngörülemeyen nedenler buna dahil olabilir. Hastalığın hızlı başlangıcı nedeniyle, uygulayıcılar genellikle EG için hazırlıksızdır. Yanıkların neden olduğu ancak EG'nin neden olduğu lezyonları tedavi etmek için de kalifiye olduk. EG tedavisi için de anlaşılmasa da, tesisimiz deneyimli üçüncül bakım çocuk hastanelerinden biriydi. Bu nedenle, bu 3 hastada problemi başarabildik. Burada EG'nin tedavi alanlarımızı paylaştık. Klinik ilerlemede tedavi hayal kırıklıklarının görüldüğü durumlarda bu deneyimlerin alternatif yönetim protokollerini kavramaya yardımcı olacağına inanıyoruz.

Anahtar Kelimeler: Cerrahi, Tedavi, Ecthyma Gangrenosum, Çocuklar

ABSTRACT

Ecthyma Gangrenosum (EG) is usually a sudden-onset disease affecting multiple regions of body with various symptoms and findings. Although the problem is seemed to be consequence of infections like Pseudomonas aeruginosa, some unpredictable causes may involve in it. Due to the rapid onset of the disease, practitioners are often unprepared for the EG. We are also qualified for treating lesions caused by burns but not EG. Although we were also unrehearsed for EG treatment, our facility was one of the experienced tertiary care children's hospitals. Therefore, we were able to accomplish the problem in these 3 patients. Herein, we shared our treatment plots of EG. We believe these experiences will be helpful to comprehend alternative management protocols when treatment disappointments were observed during clinical progress.

Keywords: Surgery, Treatment, Ecthyma Gangrenosum, Children

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Başvuru Tarihi/Received: 30-08-2017

Kabul Tarihi/Accepted: 25-04-2018



INTRODUCTION

Ecthyma Gangrenosum (EG) is a skin reaction of sepsis. Veins, arteries and subcutaneous tissue are inflamed with microorganisms. EG affected especially immune compromised patients, and it is a rare disease for disease categories (1-5). Endogenous mechanisms in infections like septicemia might be the cause (3). Somehow, the implantation of microorganisms in the skin is the key fact of the disease. Still, pathogen specification may be difficult in EG because of this type of penetration (3).

Injection sites, areas of minor trauma and invasive procedures like bone marrow biopsies are also claimed for EG (3). For miscellaneous situations, congenital heart aneurysm was related to be a possible EG initiation (2). There can be various beginning points for EG.

Blood cultures are important to find definite microorganism for EG. *Pseudomonas aeruginosa* is the most seen agent pathogen (1,2,5). However, some other organisms like *E.coli*, *Citrobacteria freundii*, *K.pneumonia*, *Morganella morganii* and fungal pathogens can be reasons (3,5).

Lesions usually resemble burns (1). Fever and diarrhea mostly associated with the EG (1). When the lesions are abundant, the remission time of the disease will be longer (4).

EG may need serious surgical treatments because necrotic tissue and debris may be mortal. In literature, aggressive debridement was reported (3). In our evaluation, we showed that different surgical procedures, including escharotomy, fasciotomy, debridement, amputation, flap and graft managements, had to be performed. Our main purpose is to emphasize that EG is a disease forming a clinical situation like in burns, and to ensure authors for the treatment EG within the concept of burn treatment.

The literature of last 5 year is documented for EG patients in Table-1.

Case 1

A 3-year-old girl referred to our hospital. She had been healthy before the disease. Her right hand was indurated, and demarcated lesion was detected (Figure 1).

Figure 1.



Laboratory studies revealed transaminase elevations (Aspartate transaminase: 4800 U/L, and alanine transaminase: 6300 U/L). Lactate Dehydrogenase level was 62000 U/L (N: 180-430). White blood count is high (21000 / μ L), and all culture evaluations resulted normal. Necrosis progressed in edematous lesion in a few days. At first, escharotomy and fasciotomy was performed (Figure 2).

Figure 2.



Broad-spectrum antibiotic, which was also affective for *P.aeruginosa* infections, was started, and wound care was performed with antibiotic dressings. In addition, negative pressure wound therapy was performed for 2 weeks. During this time, the newly formed necrotizing tissue was also resected. After these procedures, medicated paraffin dressing was performed for 3 weeks. When edema solved and all necrotic tissue was dissected, artificial skin dressing (Pelnac[®]) was performed (Figure 3). Finally, skin grafting was performed (Figure 4 and 5). Grafting resulted successfully, and patient was discharged with physiotherapy protocol.

Figure 3.



Figure 4



Figure 5.



Case -2

A 16-year-old boy was admitted to our hospital with no previous complaint. He had multiple necrotic skin lesions located in various regions. Most of them were spotted on left tibia and left ankle (Figure 6).

Figure 6.



He had lesion on back, on left and right arm, and also on right leg (Figure 7). His laboratory examinations revealed elevation of transaminase levels (Aspartate transaminase: 340, Alanine transaminase: 280 U/L) normal

tests, white blood count (34400 / μ L). Stool culture resulted as Vancomycin resistant enterococci, and other culture evaluations were normal. Initially, ulcerated lesions were treated with silver sulfadiazine wound dressing, and indurated lesions limited with necrosis were managed by medicated paraffin dressings. After demarcation lines of necrosis were confirmed, aggressive escharotomy was performed including the left ankle and pretibial regions (Figure 8).

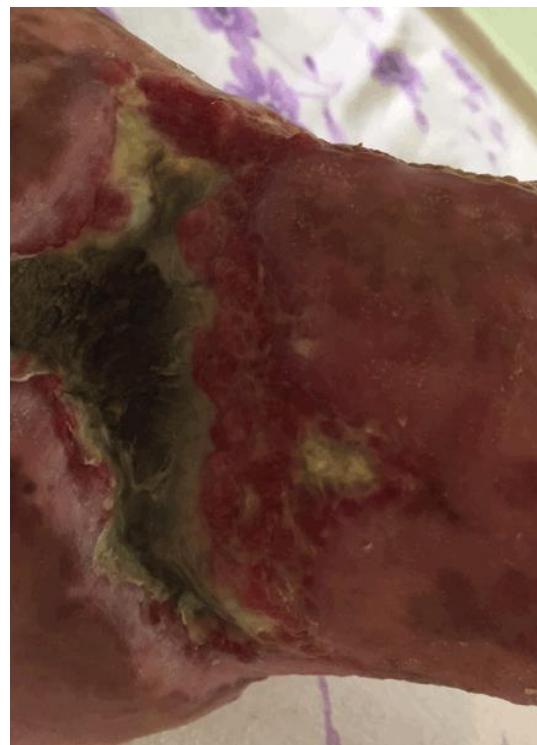
Figure 7.



Figure 8.



Figure 9.



As the area of the necrotic lesion was too large, flap procedures failed. Consequently, wound therapy was performed by wound dressings consisting in silver sulfadiazine for 3 weeks. During this period, the major lesion shrank to half diameter. Briefly, negative pressure wound therapy was added for granulation (Figure 9). Finally, flap replacement and skin grafting was performed. Treatment procedures of the patient are continuing when the report is written.

Table-1: Pediatric cases, including ours, of ecthyma gangrenosum.

Case	Year	Age/Gender	Site	Microorganism	Progress	Ref
1	2010	6-month/?	Bilat.upper and lower extr.	P.aeruginosa	Remission with scar	6 (Bakalli et al)
2	2011	7-month/F	Head,trunk,limb	P.aeruginosa	Remission with scar	4 (Yan et al)
3	2012	5-month/M	Right lower extr.	P.aeruginosa	Remission	7 (Fang et al)
4	2012	8-month/F	Neck,perineum	S.aureus	Remission	8 (Pechter et al)
5	2012	11-month/F	Labium	P.aeruginosa	Remission	9 (Pulido et al)
6	2013	7-month/F	R.pretibial	P.aeruginosa	Exitus	2 (Pach et al)
7	2013	12-hour/M	Left lower extr.	P.aeruginosa	Remission with scar	10 (Pathak et al)
	2014	23-day/M	Perineum	K.oxytoca	Remission	11(Prindaville et al)
4	2015	9-month/F	Face upper extr.	P. aeruginosa	Remission with scar	12(Gargouria et al)
5	2015	5-month/M	Abdomen	P.aeruginosa	Exitus	12(Gargouria et al)
6	2015	12-month/M	Right lower extr.	P.aeruginosa	Exitus	12(Gargouria et al)
12	2015	3-year/F	Right upper hand	?	Remission with scar	our case 1
13	2015	16-year/M	Multiple locations	Vancomycin resistant enterococci	Remission with scars	our case 2
14	2015	21-day/F	Hands and feet	K.pneumonia	Remission with amputation and scar	our case 3

Case-3

A 21-day Down syndrome girl applied to our hospital because of vomiting. On the following days, vomiting continued. Duodenal atresia was diagnosed. Duodenoduodenostomy was performed. Sepsis was added after awhile. White blood count was high (33000/μL), and other laboratory evaluations were normal. Klebsiella pneumonia was found in urine culture. Other cultures were negative. EG was diagnosed at palmar and tarsal region, and antibiotic absorbed sponges were started for treatment. Indurations and demarcation were seen in physical examinations. Left foot had the worst appearance, and unfortunately, foot had to be amputated at the end (Figure-10). Right foot was better except fingers. After dissecting necrotic regions, negative pressure wound therapy was performed for 2 weeks. Stump grafting was performed (Figure 11 and 12), and patient was taken into follow-up program.

Figure 10.



Figure 11.



Figure 12.



DISCUSSION

EG is a serious clinical situation that may have mortal consequences. When the disease is diagnosed, partial loss of skin will be the least outcome. Although treatment is possible, handling algorithms are not clear in EG. Main reason of indistinct treatment is the insufficient numbers of patients. If we evaluated the literature properly, we would find out that treatments had been performed according to the emerging situation, and various surgeries had been used but all these therapies had been performed haphazardly (3). This was the reason for us to evaluate the spectrum of treatments in literature. On the other hand, surgical procedures have not been explored sufficiently.

At the beginning, assessment of the “necrotic skin diameter” is important (3). In most centers, conservative treatments are chosen first before surgical procedures. A few studies supported surgery but numbers of the cases are few, and a detailed analysis of surgery on this subject has not been formulated yet (3). In usual, skin grafting is the choice for treatment (3). For necrotic lesions, early debridement is suggested (3).

We suggest performing escharotomy or debridement of necrotic skin at first since necrosis occurred quickly. If needed, fasciotomy might be added. In the second stage, handling with wound-dressing

procedures is appeared to be sufficient to clean up microbial colonization and to stimulate granulation reactions. According to us, at this part of the treatment, negative pressure wound therapy may be another instrument for same purposes mentioned above. If healing reactions of the tissue is slower than predicted, negative pressure therapy will be a pleasing option. At the final stage of the wound therapies, skin grafting or flap procedures must be done.

Treatment may not be finished with grafting. Amputation, unfortunately, may be needed against all efforts. Mercifully, we had no mortality. Nevertheless, all patients had long hospitalization days in intensive care units, which meant that the disease was high risk of mortality. Hence, therapy tools at all kind must be used for EG patients. This may be a problem for a hospital that does not have any preparation in this regard. Furthermore, caregivers may be unqualified or inexperienced.

One of our advantages is having a capacity of tertiary burn care unit. We are famous on this issue in our country. Therefore, we are qualified for surgical managements in EG patients. We believe that experience like this may be the reason of effective treatment. In another point of view, having less experience on burn treatment may be a disadvantage for other hospitals. Related to experience shortness, other caregivers in different hospitals may be unwilling to treat EG patients.

Although there are a lot of EG articles in literature, surgical procedures of these patients have been evaluated rarely. We are aware of only one article directly evaluating surgery (3). Most of the other literature mentioned about surgery as a part of the EG treatment but no further details were given. This is the major limitation of the discussion section. However, our experience on burns

treatment is supportive in EG treatment since clinical findings are almost same as burns.

CONCLUSION

EG treatments may be tailored in various types. Nevertheless it will be wiser to choose surgery firstly, and then medical treatments may be added. If healing periods prolong, medical devices like negative pressure therapies are the options to reinforce the treatment.

Surgery usually consists in excision of necrotic tissue and grafting. Although, healing is almost certain, scar formation is inevitable. With a medical treatment before surgery, morbidity and even mortality may be expected to be higher. In EG patients, we have a clear conscious for treatment procedures, and we announce our experience to help authors seeking a treatment for these kinds of patients.

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