DOI: 10.4274/ejbh.galenos.2024.2024-5-10



Rare Breast Emergency: A Case of Necrotizing Fasciitis of the Breast in a Lactating Patient

- 📵 Gökhan Giray Akgül¹, 📵 Sümeyra Güler¹, 📵 Simay Akyüz², 📵 Duygu Bayram³, 📵 İbrahim Burak Bahçecioğlu¹,
- 📵 Müjdat Turan², 📵 Hikmet Erhan Güven², 🔟 Mehmet Ali Gülçelik¹, 📵 Kerim Bora Yılmaz²

ABSTRACT

Necrotizing fasciitis is a rare but potentially lethal infection of the skin and soft tissue, commonly seen in the perianal and gluteal regions. Concomitant diabetes is a predisposing factor. Primary necrotizing fasciitis of the breast is rare in healthy women. In this article, we present a very rare case of breast necrotizing fasciitis in the context of the literature. We report the case of a 35-year-old female patient who had given birth two months prior to admission and developed necrotizing fasciitis of the breast during lactation. The patient presented to the emergency department with sepsis. Examination revealed widespread erythema, dark discoloration, edema, and necrotic areas indicative of wet gangrene and crepitation in the left breast. Necrotizing fasciitis is a rapid and aggressive disease that can be fatal, and delayed diagnosis may unfortunately result in death. Therefore, careful evaluation of all suspected cases, especially for patients with risk factors, is crucial for early diagnosis and timely treatment. This case highlights the importance of recognizing necrotizing fasciitis of the breast in lactating women to ensure prompt and appropriate management, potentially saving lives.

Keywords: Necrotising fasciitis; lactation; bioactive wound dressings; negative pressure wound therapy; split thickness skin graft

Cite this article as: Akgül GG, Güler S, Akyüz S, Bayram D, Bahçecioğlu İB, Turan M, Güven HE, Gülçelik MA, Yılmaz KB. Rare Breast Emergency: A Case of Necrotizing Fasciitis of the Breast in a Lactating Patient. Eur J Breast Health. 2024; 20(4): 309-312

Kev Points

- Primary necrotizing fasciitis of the breast is extremely rare in healthy women.
- The progressive infection developing due to trauma during lactation can lead to significant mortality and morbidity if diagnosed late. This underscores the need for clinicians to consider progressive infections in lactating patients in the differential diagnosis, even in the absence of common predisposing factors such as diabetes.
- The patient's breast was preserved through emergency surgical debridement, negative pressure wound therapy, and bioactive wound dressings.

Introduction

Necrotizing fasciitis (NF) is an aggressive, necrotic, and lifethreatening infection of the soft tissues. It is progressive by nature and is accompanied by arterial thrombosis, leading to gangrene of the skin and subcutaneous tissues, as well as manifestations of severe sepsis, multiple organ failure, and death (1). The progressive nature of the disease is characterized by an increase in pressure caused by the infection in the closed fascial plane and its ability to spread towards low-pressure areas along the fascial plane and affect surrounding tissues in other areas (2, 3). Disease progression and local regional damage are determined by compartment syndrome and ischemic necrosis at the capillary level due to increased pressure.

Treatment of these infections is primarily surgical, and debridement, abscess drainage, and pressure reduction are necessary to prevent disease progression. Septic shock and its associated complications are linked to mortality rates of almost 90% following treatment delays (4). Primary necrotizing fasciitis of the breast (PNFB) is extremely rare and NF is most commonly observed in the extremities, perineum, and abdominal wall. In recent years, PNFB cases have been presented in the literature more often, and this increase may be related to the rising incidence of diabetes mellitus, which is considered an important comorbidity of NF (5).

In this study, we report a case of NF in a lactating patient which was thought to have developed as a result of trauma due to breastfeeding.

> Received: 29.05.2024 Accepted: 15.06.2024 Available Online Date: 26.09.2024

Corresponding Author: Kerim Bora Yılmaz; kerimbora.yilmaz@sbu.edu.tr



¹Department of Surgical Oncology, Gülhane Training and Research Hospital, University of Health Sciences Turkey, Ankara, Turkey ²Department of General Surgery, Gülhane Training and Research Hospital, University of Health Sciences Turkey, Ankara, Turkey

³Department of Pathology, Gülhane Training and Research Hospital, University of Health Sciences Turkey, Ankara, Turkey

The patient underwent a breast-conserving procedures and skin graft reconstruction.

Case Presentation

A 35-year-old female patient presented to the emergency department two months after giving birth to her third child. She had a nipple fissure caused by breastfeeding trauma and was unable to breastfeed for three days. One week before admission, she experienced pain, swelling, and increased breast temperature. She had a history of irregular and short-term amoxicillin and clavulanic acid use.

In the left breast, there was widespread erythema, dark color changes, edema, and necrotic areas, consistent with wet gangrene, as well as crepitation (Figure 1).

Hospitalization was recommended for this patient with a pre-diagnosis of sepsis and elevated acute-phase reactants levels [C-reactive protein (CRP) 305 mg/dL; white blood cell (WBC) count 20.4×10^3 cells/ μ L; hemoglobin A1c 5.2%]. Urgent debridement was planned during hospitalization, but the patient refused treatment. On the night of the same day, the patient was re-admitted to the emergency department because of the progression of her complaints. Based on physical examination findings, the patient was admitted to the general surgery department. The Laboratory Risk Indicator for Necrotizing Fasciitis (LRINEC) score was 8 at the time of hospitalization (6). With the diagnosis of NF, necrotic areas up to a depth of approximately 3 cm from the subcutaneous tissue were debrided under emergency conditions, and negative pressure wound therapy (NPWT) was



Figure 1. A, B. There was widespread erythema, dark colour changes, oedema, and necrotic areas consistent with wet gangrene



Figure 2. A, B. Application of negative pressure wound therapy after debridement

applied using Genadyne Silver Foam® for infection and exudate control in the wound area. The patient underwent repeated debridement and breast-conserving surgery followed by NPWT. The patient was spared from mastectomy through effective surgical debridements with clear surgical margins and the application of NPWT (Figure 2). Empirical antibiotic therapy was initiated with piperacillin tazobactam (4×4.5 mg) and teicoplanin (1×400 mg). Streptococcus pyogenes (group A Streptococcus, teicoplanin-sensitive) was detected in the tissue culture. Cabergoline (Dostinex 0.5°) was administered in tablet form (0.5 mg) based on the recommendation of the endocrinology department to stop lactation, which impairs wound healing. Hyperbaric treatment, which was deemed appropriate, could not be administered because of patient refusal. The patient, whose infection findings regressed with repeated debridements and who developed healthy granulation tissue, was planned for reconstruction with a graft (Figure 3). During outpatient follow-up, the patient received intermittent NPWT and bioactive wound dressings were applied. A collagen/laminin-based dermal matrix containing resveratrol-loaded microparticles was used to fill the tissue defects and promote further granulation (Figure 3). During outpatient follow-up, granulation in the tissue defect reached the skin level, and reconstruction via grafting was performed (Figure 3). Figure 4 (A) and (B) show pathological specimens obtained from the patient, with one sample showing severe inflammation and a duct rich in neutrophils that had broken down the breast lobules. Written informed consent was obtained from the patient.



Figure 3. A. Increasing granulation with bioactive dressings; **B.** Reconstruction with a graft after the infection was brought under control; **C.** Epitelization

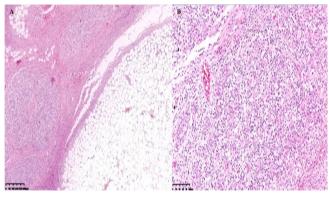


Figure 4. A. Severe inflammation that advanced into the adipose tissue disrupted the breast lobules (haematoxylin and eosin, magnification of $4\times$); **B.** Severe inflammation rich in neutrophils. Destruction of the duct is seen in the middle (haematoxylin and eosin, magnification of $10\times$)

Discussion and Conclusion

Although rare, NF is extremely aggressive. The course of the disease is characterized by different symptoms according to the area of disease involvement in the skin and subcutaneous and fascial tissues. Breast involvement is very rare, but can complicate the differential diagnosis of the disease. PNFB is often misdiagnosed and mistakenly confused with other breast diseases, such as cellulitis, mastitis, abscess, or inflammatory breast cancer (7, 8). The mortality rates reported in the literature are generally related to delayed diagnosis and treatment, as was the case for two patients in our previous series of five patients (2). However, the development of this disease remains unclear. Late diagnosis and inadequate treatment of primary breast infections may result in cellulitis, breast abscess, or the progression of an infectious disease to PNFB. If the infection in our lactating patient had been treated with an appropriate surgical method and antibiotic therapy, we might not have encountered a progressive infection. A history of fissures due to breastfeeding in our patient was associated with trauma. Post-traumatic NF of the breast tissue has been reported in the literature, but it has been described most frequently after surgical interventions (7, 9).

Advanced age, diabetes mellitus, chronic alcoholism, obesity, immunosuppression, vascular disease, malignancy, skin biopsies, and trauma are risk factors for the development of NF (2, 7, 8). Our patient was not diabetic, but was lactating, and she was referred to our hospital for a breast infection. Broad-spectrum antibiotics are preferred for the initial administration of empirical antibiotics. Definitive antibiotic therapy should be administered based on the microbial results obtained from tissue cultures after intraoperative debridement. It must be noted that the disease has a progressive nature and is a clinical entity that can be controlled surgically.

Our patient was diagnosed with type II monomicrobial infection secondary to group A beta-hemolytic Streptococcus (9). In such cases, treatment approaches include appropriate fluid and electrolyte administration under emergency and intensive care unit conditions, broad-spectrum antibiotic therapy until culture results are available, and timely application of aggressive surgical debridement. Very high mortality rates have been reported in cases of delayed treatment, especially in patients with comorbidities, which are related to the diagnosis time. The extent of surgery was determined based on the principle of not leaving any necrotic tissue. The circummammary ligament anchors the superficial fascia of the breast to the deep fascia of the chest at the perimeter. Cooper ligaments, which are specialized vertical cutaneous ligaments that anchor the skin, travel from the posterior lamina fascia through the breast gland to the anterior lamina. When planning treatment, these anatomical structures and fascial connections should be considered as they may influence the progression and spread of NF (10). To date, various operations have been performed in such cases, ranging from selective debridement to radical mastectomy, as reported in the literature. This wide range of treatments is due to differences in the spread of necrotic tissue and efforts to control the spread of infection. In the clinical stage at which treatment was initiated for the patient presented here, a response to infection was achieved with extensive surgical debridement, allowing the patient to be spared from radical mastectomy. NPWT is routinely used in these dressings, especially after surgical debridement of the infected tissues. NPWT products with instillation or products containing silver sponges are generally preferred after the first debridement. When the infection is brought under control, closure of the defect becomes a priority (11, 12). NPWT creates tension, which stimulates the production of granulation tissue and reduces wound size and bacterial load by contracting the wound (13). Upon increasing the microcirculatory blood supply with NPWT, inflammatory cells migrate to the wound region, resulting in the elimination of extravascular edema (14). Compared with traditional dressings, this approach also promotes and accelerates the formation of granulation tissue by removing bacteria, end products, exudates, and debris. Furthermore, it stimulates angiogenesis and secures wound coverage, thereby facilitating wound healing (15).

Detailed physical examination is required to diagnose NF in patients with basic skin changes. Laboratory tests and imaging studies may be necessary in cases with suspicious skin findings. Wong et al. (6) developed the LRINEC scoring system. Based on serum CRP, WBC count, hemoglobin, sodium, creatinine, and glucose values, the present case scored 8 points, putting the patient in the high-risk category at the time of diagnosis. Values of ≥8 increase the risk of NF development by 75% (16). Additionally, based on clinical findings, the case was classified as grade 3 (late stage) due to crepitation, darkening of the skin, and tissue necrosis reaching the gangrene level (17). Since the patient had an advanced clinical stage and a high LRINEC score, diagnostic imaging was not considered necessary, and it was not performed to avoid treatment delays and disease progression. The progressive nature of the disease, septic status, and related risks should be considered, and surgical consent for mastectomy and chest wall debridement should be obtained. Debridement of the surrounding tissues should also be performed as necessary when the disease spreads to the skin of the arm or abdomen (2, 18). Large tissue defects may occur after debridement, and interventions for vascular and neural structures may be required, especially in cases extending to the axillary region where vascular and neural structures are involved. If NPWT is applied in this region, barrier protectors for vascular and neural structures should be used (2).

Tissue-engineered biomaterials that play an active role in wound healing are called bioactive wound dressings. These materials, which contain natural extracellular matrix components and provide structural support for tissue repair owing to their biocompatible structures, contain polymers, such as collagen, hyaluronic acid, chitosan, and alginate.

In our patient, treatment with dipalmitoylphosphatidylcholine (DPPC)-based microparticles added to a 3- dimensional porous collagen laminin matrix was used to fill the tissue defect with granulation and prepare the wound bed for grafting after infection was controlled (19). The presence of the glycosaminoglycan derivative hyaluronic acid, collagen/hydrophilic properties, gelatine providing a 3-dimensional pore structure, laminin as a cell-binding protein, DPPC in the cell membrane, and resveratrol as an antioxidant in this wound dressing enabled the preparation of the wound bed after the infection was controlled and before grafting (20). Split-thickness skin grafting prevents the loss of protein by covering the granulated tissues and enables closure of the area in question to avoid infection and facilitate rapid epithelisation (21).

Although rarely reported in the literature, breast NF is an often-deadly disease that spreads rapidly and aggressively. Several confounding factors may have resulted in delayed diagnosis and mortality. For early diagnosis and timely treatment, it is essential that all suspected cases be evaluated carefully and thoroughly, regardless of the patient's

age. This is particularly important for patients with risk factors and comorbidities.

Informed Consent: Written informed consent was obtained from the patient.

Authorship Contributions

Surgical and Medical Practices: G.G.A., S.A., D.B., S.G., İ.B.B., M.T., H.E.G., M.A.G., K.B.Y.; Concept: S.A., İ.B.B., M.T.; Design: G.G.A., S.G., H.E.G.; Data Collection and/or Processing: D.B.; Analysis or Interpretation: S.G., M.A.G.; Literature Search: D.B., M.T.; Writing: İ.B.B., M.A.G., K.B.Y.

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

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

References

- Singh A, Ahmed K, Aydin A, Khan MS, Dasgupta P. Fournier's gangrene. A clinical review. Arch Ital Urol Androl. 2016; 88: 157-164. (PMID: 27711086) [Crossref]
- Yilmaz KB, Saydam M, Akinci M, Akkoca M, Arikok AT, Guler S, et al. Primary necrotizing fasciitis of the breast. Case series with 5 patients. J Infect Dev Ctries. 2022; 16: 902-908. (PMID: 35656964) [Crossref]
- Kim J, Yoo G, Lee T, Kim JH, Seo DM, Kim J. Classification Model for Diabetic Foot, Necrotizing Fasciitis, and Osteomyelitis. Biology (Basel). 2022; 11: 1310. (PMID: 36138789) [Crossref]
- Chernyadyev SA, Ufimtseva MA, Vishnevskaya IF, Bochkarev YM, Ushakov AA, Beresneva TA, et al. Fournier's Gangrene: Literature Review and Clinical Cases. Urol Int. 2018; 101: 91-97. (PMID: 29949811) [Crossref]
- Misiakos EP, Bagias G, Patapis P, Sotiropoulos D, Kanavidis P, Machairas A. Current concepts in the management of necrotizing fasciitis. Front Surg. 2014; 1: 36. (PMID: 25593960) [Crossref]
- Wong CH, Khin LW, Heng KS, Tan KC, Low CO. The LRINEC (Laboratory Risk Indicator for Necrotizing Fasciitis) score: a tool for distinguishing necrotizing fasciitis from other soft tissue infections. Crit Care Med. 2004; 32: 1535-1541. (PMID: 15241098) [Crossref]
- Cai Y, Cai Y, Shi W, Feng Q, Zhu L. Necrotizing Fasciitis of the Breast: A Review of the Literature. Surg Infect (Larchmt). 2021; 22: 363-373. (PMID: 33026953) [Crossref]
- Konik RD, Huang GS. Management of Primary Necrotizing Fasciitis of the Breast: A Systematic Review. Plast Surg (Oakv). 2020; 28: 215-221. (PMID: 33215036) [Crossref]
- Gupta A Dr, Gupta A, Ravi B Dr, Mundra M Dr, Sandhu H Dr, Agrawal S Dr, et al. Post-traumatic necrotising fasciitis of the breast: a case study with literature review. J Wound Care. 2019; 28: 775-778. (PMID: 31721667) [Crossref]

- Rehnke RD, Groening RM, Van Buskirk ER, Clarke JM. Anatomy of the Superficial Fascia System of the Breast: A Comprehensive Theory of Breast Fascial Anatomy. Plast Reconstr Surg. 2018; 142: 1135-1144. (PMID: 30511967) [Crossref]
- Khansa I, Schoenbrunner AR, Kraft CT, Janis JE. Silver in Wound Care-Friend or Foe?: A Comprehensive Review. Plast Reconstr Surg Glob Open. 2019; 7: e2390. (PMID: 31592393) [Crossref]
- 12. Bukovcan P, Koller J, Hajská M, Záhorec P. Clinical Experience With the Use of Negative Pressure Wound Therapy Combined With a Silver-impregnated Dressing in Mixed Wounds: A Retrospective Study of 50 Cases. Wounds. 2016; 28: 255-263. (PMID: 27560468) [Crossref]
- 13. Hu J, Goekjian S, Stone N, Nelson A, Cooper MJ. Negative Pressure Wound Therapy for a Giant Wound Secondary to Malignancy-induced Necrotizing Fasciitis: Case Report and Review of the Literature. Wounds. 2017; 29: E55-E60. (PMID: 28862979) [Crossref]
- Apelqvist J, Willy C, Fagerdahl AM, Fraccalvieri M, Malmsjö M, Piaggesi A, et al. EWMA Document: Negative Pressure Wound Therapy. J Wound Care. 2017; 26: S1-S154. (PMID: 28345371) [Crossref]
- Normandin S, Safran T, Winocour S, Chu CK, Vorstenbosch J, Murphy AM, et al. Negative Pressure Wound Therapy: Mechanism of Action and Clinical Applications. Semin Plast Surg. 2021; 35: 164-170. (PMID: 34526864) [Crossref]
- Bechar J, Sepehripour S, Hardwicke J, Filobbos G. Laboratory risk indicator for necrotising fasciitis (LRINEC) score for the assessment of early necrotising fasciitis: a systematic review of the literature. Ann R Coll Surg Engl. 2017; 99: 341-346. (PMID: 28462647) [Crossref]
- Chaomuang N, Khamnuan P, Chuayunan N, Duangjai A, Saokaew S, Phisalprapa P. Novel Clinical Risk Scoring Model for Predicting Amputation in Patients With Necrotizing Fasciitis: The ANF Risk Scoring System. Front Med (Lausanne). 2021; 8: 719830. (PMID: 34869417) [Crossref]
- Ucar EA, Durur-Subasi I, Yilmaz KB, Arikok AT, Hekimoglu B. Quantitative perfusion parameters of benign inflammatory breast pathologies: A descriptive study. Clin Imaging. 2020; 68: 249-256. (PMID: 32911313) [Crossref]
- Eroğlu İ, Gökçe EH, Tsapis N, Tanrıverdi ST, Gökçe G, Fattal E, et al. Evaluation of characteristics and in vitro antioxidant properties of RSV loaded hyaluronic acid-DPPC microparticles as a wound healing system. Colloids Surf B Biointerfaces. 2015; 126: 50-57. (PMID: 25543983) [Crossref]
- Gokce EH, Tuncay Tanrıverdi S, Eroglu I, Tsapis N, Gokce G, Tekmen I, et al. Wound healing effects of collagen-laminin dermal matrix impregnated with resveratrol loaded hyaluronic acid-DPPC microparticles in diabetic rats. Eur J Pharm Biopharm. 2017; 119: 17-27. (PMID: 28461085) [Crossref]
- Saydam M, Yılmaz KB, Bostancı MT, Turan M, Akıncı M, Yılmaz İ, et al. The use of autologous epidermal grafts for diabetic foot ulcer emergencies: A clinical study. Ulus Travma Acil Cerrahi Derg. 2022; 28: 262-267. (PMID: 35485568) [Crossref]