



Case report

Primary necrotizing fasciitis of the breast in a young patient, a case report

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ABSTRACT

Background: Necrotizing fasciitis is a life-threatening and potentially fatal condition which commonly affects extremities, abdominal wall and perineum. However, it can affect any body part. Breasts are very rarely affected, with most of the cases are following surgical intervention or trauma. Predisposing conditions include diabetes, chronic alcoholism, advanced age, vascular disease, and immunosuppression and many cases are preceded by an injury or invasive procedure. Here we present necrotizing fasciitis of the breast in a 28-year-old female patient with no identified risk factor.

Clinical presentation: A 28-year-old woman on her 4th month of lactation presented to the emergency department with a 4-day history of right breast pain and swelling. Associated with this, she had darkening of the skin of the right breast, high-grade fever, and foul-smelling discharge from the wound. On examination, she was febrile, tachycardic and hypotensive. Breast examination showed signs suggestive of NF. Subsequently, the patient was managed with broad spectrum antibiotics, supportive care, surgical debridement followed by partial mastectomy and finally the patient was discharged improved.

Conclusion: Although it is a rare phenomenon, necrotizing soft-tissue infections of the breast can progress rapidly and carry a high mortality rate. Therefore, early diagnosis and immediate surgical intervention are of paramount importance. Here, we aim to show the importance of early diagnosis and the need for appropriate therapy to avoid complications and death.

1. Introduction

Necrotizing fasciitis is a rare but life-threatening and potentially fatal condition which mainly involves the subcutaneous tissue and the fascia [1]. Although it can occur in any part of the body, extremities, abdominal wall and perineum are the most commonly affected areas [2]. Predisposing conditions include diabetes, chronic alcoholism, advanced age, vascular disease, and immunosuppression and many cases are preceded by an injury or invasive procedure (the breast journal) [1]. It rarely affects the breast, and it can happen following trauma, surgical intervention, or although it is extremely rare, it can happen in individuals without the mentioned risk factors just like our patient. Breast necrotizing fasciitis could differ from other body regions in etiology pattern and clinical presentation so that it can be easily misdiagnosed [2]. Because of the aggressive nature and high mortality of this disease, immediate surgical intervention, coupled with antibiotic therapy and physiologic support, is necessary to prevent complications and death. Unlike most reported cases, here we present necrotizing fasciitis of the breast in a healthy young mother with no identified risk

factors.

‘This case report has been reported in line with the SCARE CriteriaAgha RA, Franchi T, Sohrabi C, Mathew G, for the SCARE Group. The SCARE 2020 Guideline: Updating Consensus Surgical CAse REport (SCARE) Guidelines, International Journal of Surgery 2020;84:226–230 [3].’

2. Clinical presentation

A 28-year-old Ethiopian married housewife woman on her 4th month of lactation presented to the emergency department with a 4-day history of right breast pain and swelling. Associated with this, she had darkening of the skin of the right breast, high-grade fever, and foul-smelling discharge from the wound. The patient had given birth vaginally to a single child 4-month back with no peripartum complication. She has no history of trauma to the breast, previous nipple discharge, or any surgical intervention, and she has no family history of breast disease. She has no history of diabetes, hypertension, HIV infection, smoking or any other chronic illness. She was given oral ibuprofen 400 mg at the nearby

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health center and had taken it for two days prior to her presentation. Otherwise she has no history of drug intake. She is a mother of 3 children, 7 years, 3 years and 4 months old.

On examination, she had fever with a temperature of 38.7 °C, blood pressure 90/50, tachycardia with a pulse rate 120/min, and tachypnea with a respiratory rate 32/min. on breast examination, the whole right breast was swollen and the skin over the right breast, especially the central circum-areolar area, was grossly necrotic with ruptured and unruptured bullae (Figs. 1 and 2). There was no fluctuation, but the breast was extremely tender. No palpable masses in the right breast and no clinically significant palpable lymph nodes in the axilla. The left breast was normal.

On admission, laboratory evaluation revealed leukopenia (2200) with left shift (78 % neutrophil) and anemia (Hg of 10.7 g/dl). Organ function showed hypo-albuminemia of 2.1 g/dl and electrolyte studies showed hypokalemia of 3.22 mmol/l, but other laboratory parameters were normal. CRP determination was not done because it is not available in the hospital. Ultrasound of the breasts showed diffusely swollen and enlarged breast with stranded echogenic fatty component and hypo-echoic edematous glandular component, no abscess pocket, and normal contralateral breast.

With the initial assessment of necrotizing infection of the breast, the patient was kept NPO, intravenous ceftriaxone 1 g Iv BID, Metronidazole 500 mg Iv TID, and crystalline penicillin 4 * 10⁶ on QID were started, and surgical debridement was planned. Later, the patient was operated by a senior general surgeon and senior general surgery residents under general anesthesia and supine position at Gondar University Specialized Teaching Hospital. Intraoperatively, there was 12 * 13 cm circumferential grossly necrotic skin of the right breast, including the nipple-areola complex. Dark foul-smelling fluid came out upon opening the skin and there was pus mixed with milk (Fig. 3). There was necrosis of breast tissue extending to the pectoralis fascia, but the pectoralis muscle



Fig. 2. Gross picture of the breast.



Fig. 1. Gross appearance of the breast on physical examination.



Fig. 3. Intraoperative picture showing pus mixed with milk.

looked normal.

What was done was necrotic tissue debrided along with excision of the necrotic skin, the wound thoroughly washed with hydrogen peroxide and warm normal saline, and a sample taken for culture and

sensitivity, and histopathology analysis. Finally, the wound was left open for wound care and was covered with saline-soaked gauze. The second debridement was performed within 48 h of the initial operation. During the second operation, there was some necrotic tissue on the upper quadrants of the breast. Therefore, a partial mastectomy was performed. The histopathology of the specimen showed intense neutrophilic inflammation at areas forming micro-abscess, lobular destruction, and granulation tissue formation (Figs. 4 and 5). No features of neoplasm were seen. Microbiological culture results showed no microorganisms.

The patient's initial course was rough. However, she made a remarkable recovery after that, and the wound improved significantly and no unanticipated events happened (Fig. 6), and she was discharged after 10 days of hospital stay. On subsequent follow up the wound became well granulated (Fig. 7) and split thickness skin graft done. The patient had had a good follow-up and she reported that she was happy and thankful with the care she was provided.

3. Discussion

Necrotizing fasciitis was first described by Hippocrates and later defined by Wilson in 1952. It is characterized by necrosis of subcutaneous tissue and fascia with sparing of muscles and subsequent necrosis of the fascial planes and surrounding tissue [4]. NSTI has an incidence of approximately 1000 cases per year in the United States, or 0.04 cases per 1000 person years. The incidence of NSTI increased between 1980 and 2000, although the exact reason for this remains speculative. Possible explanations include increased microbial virulence and resistance because of excessive use of antibiotics, better disease reporting, or both. Regardless, although it remains rare, NSTI is a highly lethal condition requiring early aggressive intervention for salvage [5].

Microbial invasion of the subcutaneous tissues typically occurs through surgical site infection, blunt or penetrating trauma, but may be



Fig. 4. Pathology picture showing lobular destruction.

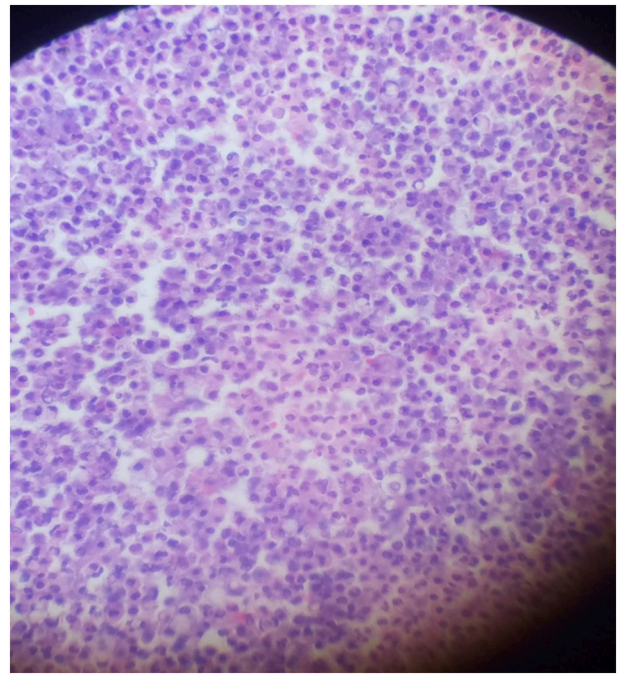


Fig. 5. Showing intense neutrophilic infiltration and granuloma formation.



Fig. 6. 5th post-operative day.

idiopathic like our patient. Bacteria rapidly track through the fascial tissue planes and produce *endo-* and *exotoxins* [6]. The key pathological characteristic in NF is thrombosis of cutaneous perforating vessels. Skin findings are much smaller than the actual extent of the disease because many dermal capillary beds must go through thrombosis before skin changes suggestive of necrosis can occur [7]. Based on the etiologic agents' necrotizing fasciitis can be classified into Four main categories, but the first two types are the most common: Type 1 necrotizing fasciitis is most commonly seen and tends to be poly-microbial infection including anaerobes, Type 2 which is caused by group A beta-hemolytic streptococci with or without staphylococci, Type 3 is caused by vibrio species and Type 4 which is caused by fulgal infection mainly candida, and it is usually associated with trauma [8]. NF can affect any part of the body, especially the trunk, lower and upper limbs, the perineum, and external genitalia (Fournier's gangrene) [4,6]. The risk factors of NF include surgery, trauma, obesity, hypertension, diabetes, asthma, and smoking. Postpartum period or breastfeeding can be



Fig. 7. 21st post-operative day.

a risk factor [7]. Wound dehiscence is another possible risk factor that allows bacteria to tract through the superficial layers, producing toxins that caused local tissue ischemia and necrosis, as well as systemic illness [7]. Reports of NF associated with NSAID use have been published in the literatures and several hypotheses were suggested about its relationship including suppression of symptoms resulting in delay in diagnosis, or enhanced bacterial virulence. However, current data do not support the casual role of NSAIDs in the development of NF [9].

The clinical presentation is often nonspecific, and a high clinical suspicion is therefore needed for timely diagnosis. Patients may present with systemic complaints of fever and malaise and only vague localizing symptoms. If treatment is delayed, patients invariably progress to sepsis, developing tachycardia, hypotension, and acute renal failure. The overlying skin may appear normal, or the local examination findings may be subtle and mistaken for those of cellulitis. The infection is not walled off as in an abscess and therefore lacks a clear boundary, making palpation challenging. This lack of a walled-off boundary accounts for both the rapid spread of disease and the difficulty in clinical diagnosis. Crepitus due to superficial fascial emphysema is only rarely palpable. However, the overlying skin is classically warm and indurated, so-called “wooden skin,” with mottled, purple patches and swelling and erythema of the skin, followed by the appearance of bullae filled with serous fluid. Extreme pain followed by anesthesia suggests the diagnosis [10,11]. As the condition progresses there will be extensive inflammation and necrosis along fascial planes, with systemic toxicity manifested as shock and organ failure [10].

Diagnosis is based on clinical examination showing signs of inflammation with skin color change and pain out of proportion with the local findings along with systemic toxicity and subcutaneous crepitation. However, due to paucity of the skin findings early in the course of the disease, diagnosis can be extremely challenging and a high index of suspicion is required. At times, the definitive diagnosis can only be made intraoperatively demonstrated by the lack of resistance of tissues adherent to the fascia to blunt dissection [1]. In case of NF of the breast, cutaneous findings may not be apparent because of the thicker tissue between the deep fascia and skin. By the time cutaneous signs are noticed, the damage is extensive and it may necessitate a mastectomy [12]. In the above case scenario, imaging studies such as CT scans or

MRI can be helpful [4]. Delay in diagnosis may result in sepsis, septic shock, and death [13]. NF can cause derangement of various biochemical and hematologic parameters, based on these parameters LRINEC score was developed which includes: blood glucose, total leukocyte count, hemoglobin, serum sodium, C-reactive protein and serum creatinine levels. A score of greater than 8 gives greater than 75 % of probability of a diagnosis of NF. Based on the above the criteria, our patient had a score of 4 (random blood sugar of 200 mg/dl and anemia 10.7 mg/dl and the others parameters were normal except C-reactive protein which is not determined since it is not done in our hospital) [8]. However, if there is a high suspicious based history and physical examination, the patient should go straight to operative debridement regardless of the LRINEC score.

The sonographic findings of necrotizing fasciitis include irregularity of the fascia, abnormal fluid collections along the fascial planes, and diffuse thickening of the fascia. These findings were not observed in patients without necrosis. Presence of gas in the fluid collection indicates further worsening of the situation. MRI is also an effective tool in diagnosing necrotizing fasciitis due to its superior soft tissue resolution. It has a high sensitivity but a low specificity (89–100 % versus 46–86 %). It has been described to demonstrate the extent of deep tissue spread much better than any other imaging modality. The MRI findings include thickening of subcutaneous tissues, fluid collection, and contrast enhancement of superficial tissue. In our case ultrasound showed a diffusely swollen and enlarged breast with hyper-echoic (echogenic) component as compared with the contralateral normal breast [14,15].

The most important thing in the management of NF is immediate surgery with aggressive debridement of the non-viable tissue, initial broad-spectrum antibiotics later guided by culture and sensitivity results, and repeated debridement until no more necrotic tissue is seen just like our patient. The above measures must continue until the progress of the disease stops along with other supportive measures such as nutritional rehabilitation. Once the clearance of devitalized tissue has been achieved, reconstructive measures aimed at skin closure can begin and generally it should be delayed until the patient completely recovers [10]. There have been few previously reported studies on the optimal method NF. In our experience since we do not have hyperbaric oxygen therapy and negative pressure wound therapy we use conventional wound treatment method. However although the daily cost of NPWT is higher than conventional wound treatments because healing is rapid and the length of hospitalization is reduced, overall healthcare costs may be reduced [8,16].

Majority of cases with primary breast NF are managed with a total mastectomy to gain source control and split-thickness skin grafts were the most common reconstructive technique performed. Other options included delayed primary closure, full thickness skin grafts, local tissue rearrangement, and pedicle flap reconstruction. Reconstruction should be patient dependent, but any of the techniques in the reconstructive ladder may be used [12]. NF of the breast is a very rare condition and only a few case reports have been published in the literature. As far as our knowledge is concerned no case reports have been published in Ethiopia so far.

4. Conclusion

Due to the rarity of necrotizing fasciitis of the breast, a high index of suspicion is required to diagnose it. NF of the breast can occur in individuals without risk factors. Aggressive surgical debridement coupled with initial broad spectrum antibiotics and supportive cares are the main stay of treatment. Early diagnosis and appropriate management significantly reduces morbidity and mortality in patients with necrotizing fasciitis. We present a patient with NF of the breast managed with a partial mastectomy and split thickness skin graft.

Abbreviations

| | |
|------|-----------------------------------|
| CT | computer tomography |
| NF | necrotizing fasciitis |
| NSTI | necrotizing soft tissue infection |
| IV | intravenous |

Ethical clearance

The case report has been submitted to the School of Medicine at University of Gondar for Ethical Board Review and approved as ethically sound report.

Consent

Written informed consent was taken from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review for the editor-in-chief of this journal.

Availability of data and materials

The authors of this manuscript are willing to provide any additional information regarding the case report.

Ethical approval

The case report has been submitted to the School of Medicine at University of Gondar for Ethical Board Review and approved as ethically sound report.

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Author contribution

All authors contributed to the conception, writing and editing of the case report. All authors are agreed to be accountable for all aspects of the manuscript.

Guarantor

Dr. Samrawit Andargie Kassa.

Research registration number

This is not the first in man case report.

Declaration of competing interest

No potential conflict of interest relevant to this article was reported.

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