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Case Report / Приказ случаја

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Necrotizing soft tissue infection in pregnancy

Некротизирајућа инфекција меких ткива код трудница

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SUMMARY

Introduction Necrotizing soft tissue infection (NSTI) is a life-threatening condition, characterized by widely spread necrosis of skin, subcutaneous fat, fascia and muscles. Treatment involves surgical debridement and broad- spectrum antimicrobial therapy. Mortality is still high due to diagnostic delays. NSTI is rare in general population, there are even less literature data of this condition in pregnancy. As far all, timely diagnosis and therapy is crucial for outcome of these patients. Clinicians should have in mind NSTI in patients with perianal infections, especially in cases where immunosuppressive role of pregnancy is present.

Case outline We present a case of 21 year old pregnant woman with NSTI spreading from perianal region. Patient was admitted to hospital in 31 week of otherwise healthy twin pregnancy one day after incision of perianal abscess. At the admission she was examined by gynecologist, vitals were stable, laboratory results showed presence of infection. She was referred for another surgical procedure and broad spectrum antibiotics were prescribed. Next morning patient complained on intense abdominal pain. Clinical exam revealed only discrete redness of the skin which was tender on palpation, crepitating**.** She was immediately referred to surgery. Intraoperative findings revealed massive soft tissue infection spreading up to the chest wall. Wide skin incisions and debridement were performed. Patient developed septic shock and after initial resuscitation gynecologist confirmed intrauterine death of twins and indicated labor induction. Over the next few days' general condition improved. On several occasions wounds were aggressively debrided under general anesthesia which left patient with large abdominal wall defect. Twenty three days after initial operation the defect was reconstructed with partial thickness skin grafts, results were satisfactory.

Conclusion Diagnosis and outcome of NSTI are challenging for many reasons. Course of the disease is rapid and hidden. Chances of survival depend on early recognition and prompt treatment.

Keywords: necrotozing soft tissue infection; necrotizing fasciitis; pregnancy

САЖЕТАК

Увод Некротизирајућа инфекција меких ткива (НИМТ) је по живот опасно стање које карактерише опсежна некроза коже, поткожног масног ткива, фасције и мишића. Лечење је хируршко уз антибиотике широког спектра. Морталитет је висок услед касног постављања дијагнозе. НИМТ је ректа у општој популацији и још ређа код трудинца. На време постављена дијагноза и започињање терапије су кључни за исход ових болесника. На НИМТ се мора посумњати код перианалних инфекција, нарочито у трудноћи, као имуносупресивном стању.

Приказ случаја Приказан је случај 21-годишње труднице са НИМТ која полази од перианалне регије. Примљена је у болницу у 31. недељи некомпликоване близаначке трудноће, дан после инцизије перианалног абсцеса. На пријему је стабилних виталних параметара, прегледана и од гинеколога. Лабораторијски налази указују на присуство инфекције. Урађена је још једну хируршка интервенција и укључени антибиотици широког спецтра. Следећег јутра болесница се жали на јак бол у трбуху. Клиничким налаз показује дискретно црвенило коже која је јако осетљива уз присуство крепитација. Индикована је хитна операција и интраоперативно је нађена масивна инфекција меких ткива која се пружа до зида грудног коша. Изведене су широке инцизије уз дебридман. Болесница развија септични шок и након иницијалне ресуцитације гинеколог ехосонографски је утврдио интраутерину смрт оба плода и индовао индукцију порођаја. Током наредних дана опште стање болеснице се поправило. У неколико наврата рађен је агресивни дебридман у условима опше анестезије што је довело до великог дефекта предњег трбушног зида. Двадесет три дана после иницијалне операције дефект је реконструисан коришћењем кожног графта са задовољавајућим резултатом.

Закључак Дијагноза и преживљавање НИМТ зависе од времена постављања дијагнозе и почетка третмана јер је ток болести брз и скривен.

Кључне речи: некротизирајућа инфекција; некротизирајући фасциитис; трудноћа

INTRODUCTION

Necrotizing soft tissue infection (NSTI) is a life-threatening condition, characterized by widely spread necrosis of skin, subcutaneous fat tissue, fascia and muscle [1]. In literature, it is also often called a necrotizing fasciitis (NF), or Fournier's gangrene, which is only one of the forms of necrotizing infection of soft tissues. Its diagnosis and outcome are challenging for many reasons. The course of the disease is rapid and hidden, and its rarity further complicates diagnosis and onset of treatment. Even in cases with optimal treatment morbidity and mortality can be as high as 35%. [2].

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Of around 28 million patients in the NIS database (Nationwide Inpatient Sample of the Healthcare Cost and Utilization Project) in US only 0.04% were identified as having a NSTI [3]. Chances of survival depend on early recognition and prompt treatment. Because of the importance of early diagnosis primary care physicians need to maintain high index of suspicion for these infections and should be aware of possible presenting features [4]. There are less data in literature on NSTI in pregnancy. As for all, timely diagnosis and therapy is crucial for outcome. Clinicians should have in mind NSTI in patients with perianal infections, especially in cases where immunosuppressive role of pregnancy is present.

CASE REPORT

A 21-year-old woman was admitted to hospital in 31 week of otherwise healthy spontaneously conceived twin pregnancy, which was regularly checked, according to patient. Past medical and family history was unremarkable. She was referred from regional hospital one day following incision of perianal abscess at the right side. Complains started seven days prior to admission, described as discomfort and edema around anus. She denied any recent trauma. Antibiotics were prescribed, but since there was no improvement, after 4 days she was hospitalized in regional hospital for perianal abscess. Incision was performed under local anesthesia.

At admission obstetrics ultrasonography examination confirmed living intrauterine fetuses of 31weeks gestational age. Patients' vital signs were stable, body temperature was normal, laboratory results showed following abnormalities: WBC 8.5, RBC 2.88, Hgb 75 g/l, HCT 26.9%, CRP 294.2 mg/l, total protein 43.7 g/l, albumin 17.4 g/l. Clinical examination revealed: incision on the right side of the anus with secretion of small amount of pus, with signs of cellulitis on the left side spreading up toward left vulva. Due to unsatisfactory clinical finding, the same day she was referred to another surgical procedure. After incision, antibiotics were prescribed (Meropenem 1g/8h and Ampicillin 1g/8h), as well as paracetamol and fluids in consultation with gynecologist. During the morning round, patient complained on intense abdominal pain. Body temperature was still normal. Clinical examination revealed only discrete redness of the skin which was tender on palpation and crepitating (Figure 1). She was immediately referred to surgery, since NSTI was suspected. Intraoperative findings revealed massive soft tissue infection spreading from left

Figure 1. Clinical presentation prior to surgery.

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perineal region up to the chest wall predominantly on the left side (Figure 2). Wide skin incisions and excisions followed by necrectomy and debridement to the anterior abdominal wall were performed. Septic shock developed immediately requiring mechanical ventilation in the postoperative course.

Figure 2. Intraoperative finding.

Patient was hypotensive (80/40 mmHg), heart rate 128/min, and arterial blood gas confirmed metabolic acidosis. Laboratory results showed Hemoglobin 62 g/l, HCT 18 %, total protein 35.1g/l, albumine 12.5 g/l, CRP 206.7 mg/l, while body temperature rose to 38.6 °C. After initial resuscitation, one day after admission, gynecologist confirmed with ehosonography intrauterine death of twins and indicated labor induction. Over the next few days patient was treated in ICU with broad spectrum antibiotics, and general condition improved. Wound dressing was performed at least twice a day. Wound cultures identified Acinetobacter sp. and Enterococcus faecalis, antibiotics were switched accordingly. On seventh postoperative day mechanical ventilation was no longer needed. On several occasions wounds were aggressively debrided under general anesthesia which left patient with large abdominal wall defect. After stabilization she was referred to Clinic for Plastic and Reconstructive surgery for further treatment. Twenty three days after initial operation skin defect was reconstructed with partial thickness skin grafts (Figure 3). Eleven days post transplantation results were satisfactory. She was discharged from hospital 44 days after admission (Figure 4).

Figure 3. Abdominal wall reconstruction with partial thickness graft .

Figure 4. Abdominal wall at hospital discharge.

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DISCUSION

According to largest published retrospective population-based cohort study from Texas USA, in ten year period (2001-2010) there were 4,060,201 pregnancy associated hospitalizations-of which 148 were due to necrotizing infection. Only minority of women (17.6%) were reported to have chronic comorbid conditions, diabetes mellitus was the most common (50%). Drug and tobacco abuse were rare, while obesity was reported in 22.3% [5]. Publish data on NSTI in general population show that 52.7- 82% have at least one risk factor like diabetes mellitus or immunodeficiency of various degrees [6-8]. In systematic review of Angolues et al.[9], diabetes mellitus was predominant risk factor in 31%, smoking in 27%, alcoholism in 17%, cirrhosis in 8%, HIV in 6%, various stages of malignancy in 3%, corticosteroid therapy and chronic kidney insufficiency in 3% NSTI cases. In this case patient past medical history was unremarkable, suggesting that pregnancy might be as risk factor for necrotizing infection. We found only one similar case report on NSTI in pregnancy published in English language, presenting 15-year-old primigravid in 29th week of pregnancy [10], since the majority of necrotizing infections related to pregnancy appears during the postpartum period (82.4%) [5]. In the presented case report by Nikolau et al, diabetes was diagnosed incidentally at the time of hospital admission. In addition to diabetes mellitus, pregnancy was suggested as risk factor for necrotizing infection due to suppression of immune system during the second and third trimester and in postpartum period [10]. This argument should be carefully considered since pregnancy is not a state of generalized immunosuppression, but instead, immune response is modulated in both, systemic and, more effectively, local manner, which is focused at the maternal–fetus interface [11].

Course of NSTI varies, is often deceitful and 35% of patients are inicialy misdiagnosed. The beginning ailment may suggest many other conditions, e.g., cellulitis, erysipelas, phlebitis, etc. [12]. A cardinal early symptom is disproportionately strong pain in comparison to clinical finding at examination. In a publication by Goh et al. [13], in nine studies swelling was the most common presenting symptom (80.8%), followed by pain (79%) and erythema (70.7 %). Inicial finding in this case was not suggestive for NSTI, but rather for perianal abscess of cryptoglandular etiology, with one destinction, unuasually intensive pain. In the previosly mentioned case report diagnosis was established on third day of hospitalisation after unsuccesful treatment with incision and antibiotics, and after MRI confirmation [10]. According to data from literature, imaging techniques could be useful. Ultrasound or plain X ray cannot reliably detect NSTI. Fascial thickening on T2-weighted magnetic resonance imaging (MRI) has a sensitivity of 90% to 100%, but a specificity of only 50% to 85% for NSTIs. Computerized tomography should only be considered as a diagnostic aid and when it can be obtained very quickly, having in mind that it may miss 1 in 5 cases of deep NSTI. Most reliable seems to be macroscopic finding such as pasty gray necrotic tissue, thin purulent fluid with a gray-brown “dishwater” appearance, a lack of resistance to digital pressure against fascial planes (the finger test), a generalized lack of bleeding, visibly thrombosed vessels, and/or muscle that does not

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contract to electrocautery stimulation [2]. Although diagnosis of NF is clinical, it is often delayed, because the infection begins and progresses in the deep layers of subcutaneous tissues, giving initially a false impression of a typical cellulitis [14]. Meanwhile, infection spreads fast with speed of 2-3 cm per hour in anorectal region [6, 15], as seen in this case. In less than 24 hours infection spread more than 50 cm, from perianal region to the anterior chest wall.

The treatment of NSTI considers wide incisions and excisions of the affected region, operative debridement, tissue decompression and use of broad- spectrum antibiotics.

The historical data report that the exclusive use of antibiotics leads to 100% mortality, indicating the necessity of surgical intervention, which substantially decreased mortality [7]. Probably, more important is the time of intervention. Multiple studies confirmed that mortality is increased when surgical treatment is delayed as well in cases in which repeated excisions are needed [16]. According to Gallup et al, any patient with inordinate pain and unilateral edema in the pelvis, especially in the puerperium, should be suspected of necrotizing infection. The triad of pelvic pain, edema, and any sign of septicemia carries an extremely grave prognosis and mandates immediate surgical intervention [17].

Unfortunately, rapidly progressive infection, treatment delay and development of septic shock led to intrauterine death. In other reported case by Nikolaou et al., necrotizing infection caused preterm delivery of viable male fetus weighing 1470 grams by normal labor despite tocolytic therapy. Baby died due to septicemia after 48 hours [10]. Described complications of sepsis during pregnancy are: increased rates of premature births, fetal infection, hypoxia and acidosis, higher fetal mortality and increased probability for cesarean section. In the obstetric context, the assessment of fetal vitality has particular relevance, as the balance between fetal oxygen supply and consumption might be severely altered. No study has yet analyzed the best approach for fetal vitality assessment under this circumstance [18]. The best approach to ensure fetal vitality is to stabilize the mother's condition. The base treatment, which also applies to pregnant women with sepsis, is provided by the therapeutic guidelines based on the Surviving Sepsis Campaign [19]. The aim of initial hemodynamic resuscitation is to restore tissue perfusion to an adequate level and to ensure that cell metabolism and oxygen supply return to normal levels to avoid acidosis and consequent multiorgan dysfunction. In pregnancy, one further aim of initial hemodynamic resuscitation is to improve fetal vitality [18,19].

Empirically selected antibiotics must be initiated immediately. A wide variety of pathogens has been reported to be responsible for NSTI. The recent clinical classification distinguishes four types: Type I (70-80%, polymicrobial/synergistic) as in this case, type II (20% of cases; usually monomicrobial), type III (gram-negative monomicrobial, including marine-related organisms) and type IV (fungal) [16].

After massive debridement and repeated surgery like in case presented here, patient is left with defect. When primary closure is not possible soft tissue reconstruction can be considered, after stabilization of patient. Usually it's been performed using skin grafts and myocutanesous flaps, as in

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burn reconstruction. In cases with excessively large amounts of soft tissue involvement (>25% Body Surface Area), autograft reconstruction may be restricted by limited donor-site availability [20].

CONCLUSION

Necrotizing soft tissue infection is a rapidly progressive, life-threatening condition that requires early aggressive treatment. Clinical findings on presentation are crucial for diagnosis. It should be suspected in pregnancy and postpartum period in cases with unusually intensive pain, local edema and systemic signs of infection. Postponing treatment leads to septic shock with high mortality. The treatment is based on „source control" principle with aggressive surgical debridement, broad spectrum antibiotics and resuscitation. In cases such as described above, the assessment of fetal vitality is relevant, demanding joint efforts of surgeon, obstetrician and intensive care specialists.

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