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Case Report

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Conservative Surgical Management of Uterine Incisional Necrosis and Dehiscence After Primary Cesarean Delivery Due to *Proteus mirabilis* Infection: A Case Report and a Review of Literature

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Abstract

We hereby describe the conservative surgical management of a case of infected uterine incisional necrosis and dehiscence after primary cesarean delivery, and report our brief review on risk factors, physiopathology and the management of this postpartum complication. We encountered a 25-year-old woman presenting to our emergency department (ED) with severe suprapubic pain and high grade fever. She had an urgent cesarean delivery performed 10 days prior to presentation due to fetal distress. At the ED, CT scan of pelvis was ordered and showed an intraperitoneal collection anterior to the uterus at the level of the uterine cesarean scar. Exploratory laparotomy showed a uterine rupture at the previous incision site. We performed resection of necrotic edges, peritoneal lavage, approximation of uterine edges with separate interrupted sutures, placement of a suction drain in the *cul-de-sac* and a passive drain inside the uterine cavity through the cervix and vagina. Postpartum uterine scar rupture secondary to infection and necrosis is a rare but serious complication of cesarean delivery. Conservative management by drainage and resection of necrotic edges in addition to intravenous antibiotics may be considered as an option before resorting to hysterectomy in selected young patients. A low threshold to diagnose this complication is warranted.

Keywords: Cesarean delivery; Complication; Endomyometritis; Scar necrosis; Bladder flap hematoma

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Introduction

Cesarean delivery is one of the oldest procedures in the surgical field [1], with rates reaching lately up to 40% of all deliveries in the USA [2]. This route of delivery has been shown to be linked with multiple short- as well as long-term complications [2]. One of these complications is puerperal cesarean scar necrosis and dehiscence. This is rare and difficult to diagnose and the patient usually presents with a picture of endomyometritis which will prove difficult to treat. Hereby we report a case of uterine incision necrosis and rupture, occurring 10 days following primary intrapartum cesarean delivery. The case was managed conservatively with drainage and debridement with the aim of preserving fertility.

We also reviewed all available data pertaining to risk factors, physiopathology and management of this rare condition. We conducted a literature search on MEDLINE database between 1998 and 2017 to identify articles reporting similar cases. All articles published in English were included. The search terms included: "Uterine scar" OR "Uterine incision" AND "necrosis" AND "infection". Twenty-six articles were found. The lists of references of these articles were also reviewed. In total, we retrieved 17 articles reporting 23 cases similar to the present case [3-19] and one review article about the infected uterine incisional necrosis and dehiscence written by Rivlin et al in 2004 [20].

Case Report

A 25-year-old G5P5A0, previously healthy woman presented to the emergency department for severe suprapubic pain associated with high grade fever and yellowish malodorous vaginal discharge of 2 days duration. She reported that she underwent an intrapartum primary cesarean delivery in a different hospital, after receiving prophylactic dose of antibiotic (amoxicillin-clavulanic acid 1.2 g intravenous), 10 days prior to presentation. The procedure was performed in active labor at full term with cervical dilation of 9 cm, because of severe fetal bradycardia. Her past obstetrical history was only significant for four uneventful full-term normal vaginal

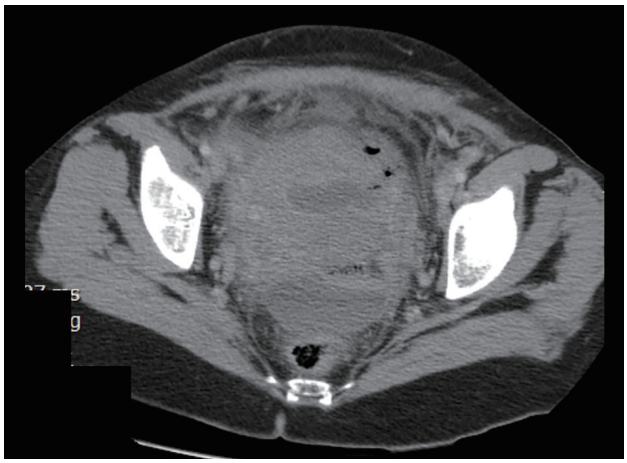

Figure 1. CT scan of the abdomen and pelvis with intravenous and oral contrast showing an intramyometrial gas formation, a collection of 5 × 3 × 3 cm anterior to the uterine scar and a second collection of 5 × 4 × 3 cm in the Douglas pouch.

deliveries. On admission, she was febrile (oral temperature: 39 °C), tachycardic (HR: 105 beats/min) and normotensive (BP: 110/60 mm Hg). The physical examination disclosed a guarded abdomen, severe suprapubic tenderness, and a foul smelling yellowish discharge from the cervix. Laboratory results showed a white blood count of 15,000 cells/mm^3 with 80% neutrophils, a C-reactive protein level of 340 mg/L (normal < 5 mg/L), in addition to normal electrolytes, creatinine, and urine analysis.

Broad-spectrum intravenous antibiotics were started (meropenem 1 g every 8 h) for endometritis treatment; however, the symptoms did not improve after 48 h. As a result, a CT scan of the abdomen and pelvis with intravenous and oral contrast was performed which showed a collection of 5 × 3 × 3 cm anterior to the uterine scar, and a second collection of 5 × 4 × 3 cm in the Douglas pouch (Fig. 1).

Due to the deteriorating condition of the patient and the imaging findings, an exploratory laparotomy was performed. Three hundred and fifty milliliters of pus were removed, after which a ruptured uterine scar with necrotic edges was revealed (Fig. 2a). Given the young age of the patient and her desire to preserve fertility, we decided not to perform a hysterectomy. Instead, we resected the necrotic edges (Fig. 2b), placed an intrauterine passive drain through the vagina, closed the large defect with three separate simple sutures (braided poly-filament number 2.0) because of tissue friability, and placed a suction drain in the Douglas pouch through the abdominal wall. We informed the patient about the risks of this conservative management and the possible resort to hysterectomy in case of failure.

The pus culture grew multi-resistant *Proteus mirabilis* sensitive to meropenem. The patient started to improve on the second post-operative day. The intrauterine drain was removed via vaginal route on the fifth post-operative day and the intra-abdominal drain on day 6. She was discharged at day 8 post-operatively. The patient was doing well at clinical follow-up 4 months after the procedure.

At 1-year follow-up, magnetic resonance imaging (MRI) of the pelvis showed an intact uterine serosa, a normal endometrial thickness and only a small indentation visible at the level of the uterine scar (Fig. 3).

Discussion

The rate of cesarean delivery rose dramatically from 4.5% in 1970 to 32.8% of total deliveries in 2010 in the United States [2]. It is even higher in some developing countries [21]. Several factors were identified to cause this high rate, many of which are avoidable [2]. This route of delivery is associated with multiple short- as well as long-term serious complications [22]. Puerperal infection is one of the most common morbidities. It is estimated to occur three times higher in low-risk patients undergoing planned cesarean delivery compared to those undergoing planned vaginal delivery (0.6% to 0.21%, respectively) [22]. The American College of Obstetricians and Gynecologists recommends a single-dose broad-spectrum antimicrobial prophylaxis (usually first-generation cephalosporin) for all patients within 60 min before the start of cesarean delivery [23]. Despite this practice, puerperal infection can

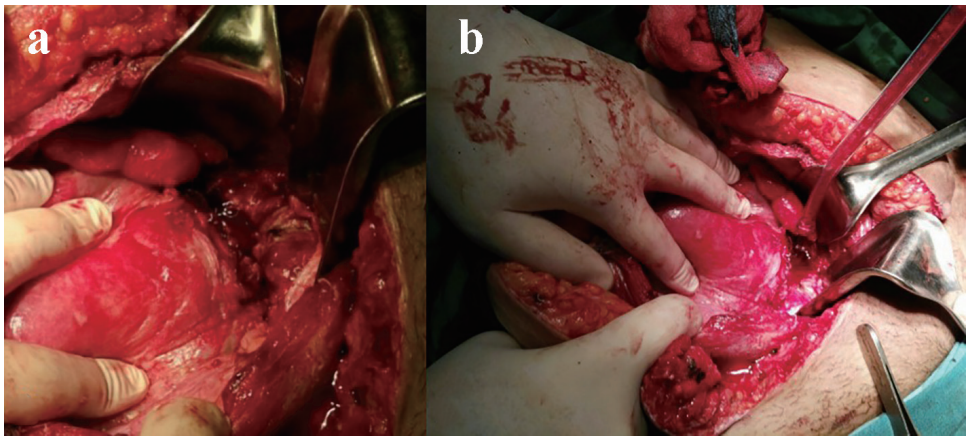

Figure 2. Exploratory laparotomy. (a) Ruptured uterine scar and infected necrotic edges. (b) Debridement of the uterine scar edges.

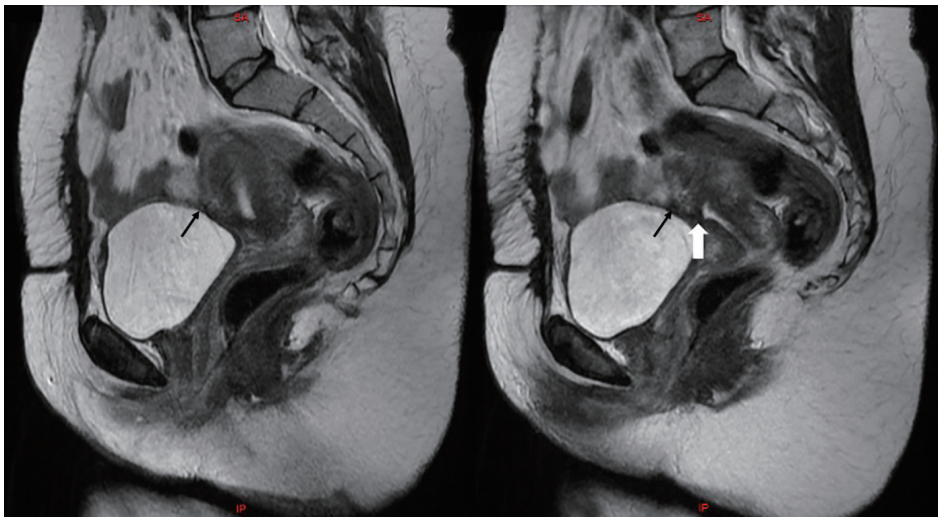

Figure 3. Two consecutive sagittal views on T2-weighted MRI of the pelvis showing an intact uterine serosa (black arrows), a normal endometrial thickness and a small indentation at the level of the uterine scar (white arrow).

still occur [24].

Definition and incidence

Infected uterine incisional necrosis and dehiscence is an extremely rare but a potentially lethal complication of cesarean delivery. It was defined by Rivlin et al [20] as the surgical evidence of uterine incision necrosis with or without separation of the edges of the uterine incision, subsequent to an acute infection. Due to the rarity of these cases, the exact incidence cannot be estimated.

Pathophysiology

The necrosis and the separation of uterine incision may be caused by the low perfusion to the edges due to overzealous suturing. Uterine scar weakness and dehiscence highly occur in patients with locked suturing of the myometrium compared to those with unlocked closure [25-27]. Rivlin et al [20] attribute the condition to the presence of suture material as a foreign body which constitutes a nidus for bacterial growth and subsequent cellulitis. A second proposed mechanism is that of Faro [28] who considers hematoma collection at the site of uterine incision -bladder flap hematoma - as risk factor of contamination by bacteria either directly inoculated at the time of cesarean delivery or climbing from the genito-urinary tract [29-31]. This may lead to abscess formation, myonecrosis and uterine rupture. In a study of 50 women having persistent postpartum fever, MRI showed a bladder flap hematoma in 32 patients (64%), parametrial edema in three (6%), and a pelvic hematoma in two (4%) [32]. Furthermore, the development of a severe or a sub-optimally treated endomyometritis may lead to necrosis and subsequent rupture of the uterine scar followed by formation of pelvic abscess collection. In fact, no mechanism *per se* can explain the exact sequence of events. Hence, it is a multifactorial condition.

Patients' characteristics and risk factors

The mean age of patients in the 23 reported cases we reviewed was 27.7 ± 6.6 years. Most patients were primipara. Several risk factors may be involved including those predisposing to postpartum endomyometritis or postoperative hematoma formation [33-39]. In most cases, cesarean delivery was done in an emergency settings following the onset of labor and after the rupture of membranes (35% versus 22%), similar to the present case.

Clinical presentation and timing

The clinical presentation of an infected uterine incisional necrosis and dehiscence may vary widely from abdominal pain and fever, to wound or vaginal discharge, heavy vaginal bleeding and sometimes to an overt peritonitis and shock if left untreated. After studying the characteristics of our patient and the 23 reviewed cases, we found that the most consistent symptoms reported were abdominal pain and/or fever not responding to intravenous broad-spectrum antibiotics for more than 48 h (16 of 23 patients; 69.6%). As previously described, a pelvic abscess following cesarean delivery should be suspected in case of persistence of fever despite the use of intravenous antibiotics for more than 72 h [40, 41]. The onset of symptoms ranged from 2 to 15 days after the cesarean delivery in 16 cases but it reached 6 - 10 weeks in seven cases (Table 1) [3-19].

Pathogens

Postpartum endomyometritis is usually a polymicrobial infection [42]. Several germs may be involved which enhance bac-

Table 1. The Presentation and the Outcome of 23 Patients Having Infected Uterine Incisional Necrosis and Dehiscence Found in the English Literature

Case	Presentation	Onset of symptoms after delivery	Hysterectomy	Culture
1 [3]	Wound infection; fever	15 days	Yes	NA
2 [4]	Fever	2 days	No	*Escherichia coli*
3 [5]	Wound infection; fever	7 days	No	*Staphylococcus aureus*
4 [6]	Vaginal discharge	15 days	Yes	NA
5 [7]	Abdominal pain; fever	9 days	Yes	*Escherichia coli*
6 [8]	Heavy vaginal bleeding	10 weeks	Yes	*Escherichia coli*
7 [9]	Abdominal wall dehiscence	3 days	No	*Streptococcus anginosus*
8 [10]	Abdominal pain; fever	15 days	No	NA
9 [11]	Abdominal pain; fever	8 weeks	No	*Staphylococcus aureus*
10 [12]	Heavy vaginal bleeding	6 weeks	Yes	NA
11 [13]	Heavy vaginal bleeding	11 days	No	*Corynebacterium sp, Prebotella bivia*
12 [14]	Wound infection; vomiting	6 days	No	*Staphylococcus, Enterococci*
13 [14]	Abdominal pain; fever; vomiting	10 weeks	No	*Escherichia coli*
14 [14]	Wound infection; fever; nausea	12 days	Yes	*Staphylococcus aureus*
15 [15]	Abdominal pain; fever	3 days	Yes	NA
16 [15]	Abdominal pain; fever	3 days	Yes	NA
17 [15]	Abdominal pain; fever	3 days	Yes	NA
18 [15]	Abdominal wall dehiscence	5 days	Yes	NA
19 [15]	Heavy vaginal bleeding	6 weeks	Yes	NA
20 [16]	Abdominal pain; vomiting	8 weeks	No	NA
21 [17]	Abdominal pain	5 days	Yes	*Pseudomonas aeruginosa, Staphylococcus, Citrobacter roseri*
22 [18]	Heavy vaginal bleeding; fever	14 days	Yes	NA
23 [19]	Heavy vaginal bleeding	6 weeks	Yes	NA
The present case	Abdominal pain; fever	10 days	No	*Proteus mirabilis*

NA: not available.

terial synergy. Aerobic and anaerobic bacteria were identified including: *Streptococci, Staphylococci, Enterococcus, Escherichia coli, Klebsiella, Proteus mirabilis, Gardnerella, Peptostreptococcus, Peptococcus, Clostridium, Bacteroids* and others [42]. However, the cultures of eight cases out of 23 grew a single germ: *Staphylococcus* (cases 3, 9 and 14), *Streptococcus* (case 7), and *Escherichia coli* (cases 2, 5, 6 and 13). Three cases had polymicrobial infection (case 11: *Corynebacterium sp* and *Prebotella bovi*; case 12: *Staphylococcus* and *Enterococcus*; case 21: *Staphylococcus, Pseudomonas and Citrobacte*r) as shown in Table 1. There are no previous papers reporting *Proteus mirabilis* as a single agent responsible of this complication.

Imaging studies

Computed tomography (CT) scan and MRI are both helpful in diagnosing pelvic masses and fluid collection [43]. CT scan findings poorly correlate with surgical findings concerning uterine incision dehiscence: an apparent discontinuity of the myometrium at the level of the incision may represent edema, and can be seen after an uncomplicated cesarean delivery [44, 45]. Another point worth mentioning is that the presence of a collection in proximity of the cesarean incision site, as seen with ultrasound or CT scan, does not mean the presence of dehiscence, and it might only reflect a hematoma at the bladder flap. In these cases, only the clinical course can determine the severity of the situation. However, compared to CT scan, MRI is more sensitive and specific in diagnosing dehiscence because it clearly delineates the uterine serosa layer [45, 46]. Nonetheless, the definitive diagnosis of infected uterine incisional necrosis and dehiscence is made during surgical exploration and it should not be based solely on imaging studies.

Treatment

The infected uterine incisional necrosis and dehiscence is a se-

rious complication of cesarean delivery, delayed treatment of which may result in septic shock and death. Since there are no treatment guidelines based on a good level of evidence, the surgical treatment should be tailored to patients on individual basis (e.g. clinical presentation, surgical findings and patient desire to preserve fertility). In most cases, total or subtotal hysterectomy and surgical debridement with conservation of the unaffected adnexa remain the gold standard approach according to Cunningham et al [47]. Among the 23 cases reviewed, 14 underwent hysterectomy because of severe pelvic adhesions, severe peritonitis, extensive involvement of pelvic organs and heavy vaginal bleeding. In selected cases, similar to the present case, it may be possible to preserve the uterus if the patient is stable and wishes to preserve her fertility, and if the uterus and intra-abdominal organs are minimally involved by the infection. A possible surgical approach consists of abscess drainage, necrotic edges debridement, placement of an intrauterine drain through the vagina and closure of the defect.

Conclusion

In conclusion, clinicians should have a low threshold to diagnose this complication as early as possible in patients who fail to respond rapidly to broad-spectrum antibiotics. Imaging studies help clinicians to exclude several serious cesarean complications keeping in mind that the presence of a collection anterior to the uterine incision can harbor uterine scar dehiscence. Hence, the clinical presentation and the high suspicion should dictate treatment strategies. Conservative management in properly selected patients is a valid choice for cases keen to preserve their fertility instead of resorting directly to hysterectomy. However, preserving the uterus does not necessarily mean preserving fertility especially with the risk of intrauterine adhesion formation preventing conception, or cesarean scar pregnancy and uterine rupture in subsequent pregnancy.

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