CASE REPORT

Giant uterine fibroid complicated by aseptic necrobiosis and cutaneous fistulation: A case report and brief review of literature

Fibrome utérin géant compliqué d'une nécrobiose aseptique et d'une fistule cutanée: Un rapport de cas et une brève revue de la littérature

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Abstract

Background: Uterine fibroids are the most common tumors of the female reproductive system. Although often asymptomatic, they can lead to complications such as compression of adjacent organs, torsion, or aseptic necrosis. Cutaneous fistulation, though rare, is a complication presented in this case.

Case presentation: We reported the case of a 41-year-old female with no significant medical history who presented to the emergency department with a 4-month history of pelvic pain and purulent discharge from the supraumbilical region. Physical examination revealed an enlarged abdomen and a 3-cm supra-umbilical parietal mass with fistulation to the skin, discharging pus. Abdominal computed tomography imaging demonstrated a 30-cm uterine fibroid complicated by aseptic necrobiosis and cutaneous fistulation. The patient was started on antibiotic therapy, followed by myomectomy and resection of the fistulous tract by laparotomy.

Conclusion: Cutaneous fistulation of a uterine fibroid is a rare complication that requires prompt detection and an accurate diagnosis for effective management.

Key words: Aseptic Necrosis, Case report, Leiomyoma, Myomectomy

Résumé

Introduction: Le fibrome utérin est la tumeur la plus fréquente de l'appareil reproducteur féminin. Il peut être asymptomatique mais peut essentiellement se compliquer d'une compression des organes voisins, d'une torsion ou d'une nécrose aseptique. La fistule cutanée est une complication rare décrite dans notre cas.

Présentation du cas: Nous avons rapporté le cas d'une femme de 41 ans, sans antécédents pathologiques, qui s'est présentée aux urgences pour des douleurs pelviennes évoluant depuis 4 mois et un écoulement de pus dans la région sus-ombilicale. L'examen clinique a révélé une augmentation du volume abdominal et une tuméfaction pariétale sus-ombilicale de 3 cm fistulisée à la peau et accompagnée d'un écoulement de pus. Un fibrome utérin de 30 cm compliqué d'une nécrobiose aseptique et d'une fistule cutanée ont été observés au scanner abdominal. La patiente a été mise sous antibiotiques puis une myomectomie et une résection du trajet fistuleux ont été réalisées par laparotomie.

Conclusion: La fistule cutanée d'un fibrome utérin est une complication rare qui doit être détectée et diagnostiquée pour un traitement efficace.

Mot clés: Leimyome, Myomectomie, Nécrose aseptique, Rapport de cas

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Introduction

Uterine fibroids are the most common tumors of the female reproductive tract, affecting 25 to 40% of females of reproductive age. [1]. While often asymptomatic, fibroids can present with symptoms such as pelvic pain, menometrorrhagia, or abdominal distention [2].
The main complications include the mass effect that leads to compression of adjacent organs, torsion, anemia, aseptic necrobiosis, and potential impacts on fertility [3]. Histopathological examination provides a definitive diagnosis, confirming a benign tumor originating from the smooth muscle of the uterus [4].
This case aimed to discuss a giant uterine fibroid complicated by aseptic necrosis and cutaneous fistulation. A brief review of literature was done.

Patient and observation

Patient information

A 41-year-old female presented with a 4-month history of pelvic pain and purulent discharge from the supraumbilical region. The patient had no relevant medical, family, or psychosocial history. Menarche occurred at 13 years of age and the patient reported a regular 28-day menstrual cycle with a 5-day flow, without associated symptoms. She delivered by cesarean section 4 months earlier at 30 weeks of gestation due to a pathological fetal heart rate. During the procedure, a giant 30 cm intramural uterine fibroid with aseptic necrobiosis was identified but not resected.

Clinical findings and timeline of current episode

Four months later, the patient appeared to be in good general health on physical examination. However, abdominal examination revealed an increase in abdominal volume and a 3-cm supraumbilical parietal mass with fistulation to the skin, discharging pus (Figure 1). The remainder of the physical examination was unremarkable.

Figure 1. A 3-cm supraumbilical cutaneous fistulation of a uterine fibroid.

Diagnostic evaluation

Pelvic ultrasonography revealed a 30-cm intramural uterine fibroid undergoing necrobiosis, with a 3-cm fistulized collection that extended to the skin. Given the size of the pelvic mass, which exceeded the limits of ultrasound imaging, an abdominopelvic computed tomography (CT) scan was performed. The CT scan confirmed the ultrasonographic findings (Figure 2).
The biochemical tests, including a complete blood count with a hemoglobin level of 11 g/dL (normal range: 12.0–15.5 g/dL) and a platelet count of 220000/µL (normal range: 150000–450000/µL), as well as C-reactive protein < 10 mg/L (normal range: < 10 mg/L), all yielded normal results.

Figure 2. A 30-cm uterine fibroid complicated with aseptic necrobiosis and skin fistulation is shown by a CT scan of the abdominal cavity and pelvis.
(**a**: Axial view; **b**: Sagittal view; **c**: Coronal view; **d**: Axial view of a smaller region)

Diagnosis

A 30-cm intramural uterine fibroid in necrobiosis, with a 3-cm fistulized collection that extended to the skin, was identified.

Therapeutic interventions

The patient was admitted to our department of Obstetric gynecology of Sousse (Tunisia) and both diagnosis and treatment were discussed with the patient and a multidisciplinary team. Initial management involved systemic and local antibiotic therapy, followed by surgical intervention by laparotomy. A subumbilical midline incision was made, and a myomectomy was performed by enucleating the 30 cm posterior intramural uterine fibroid, along with resection of the fistulous tract (Figure 3).

Follow-up and outcome of interventions

Histopathology revealed a benign fibroid in necrobiosis that confirms the established diagnosis. The patient's recovery was uneventful and she was discharged 48 hours after the operation. However, long-term follow-up of the patient revealed no anomalies, with no evidence

of recurrence or postoperative complications. Notably, the patient's fertility was preserved, highlighting the success of the surgical intervention and postoperative management in addressing the condition without compromising reproductive potential.

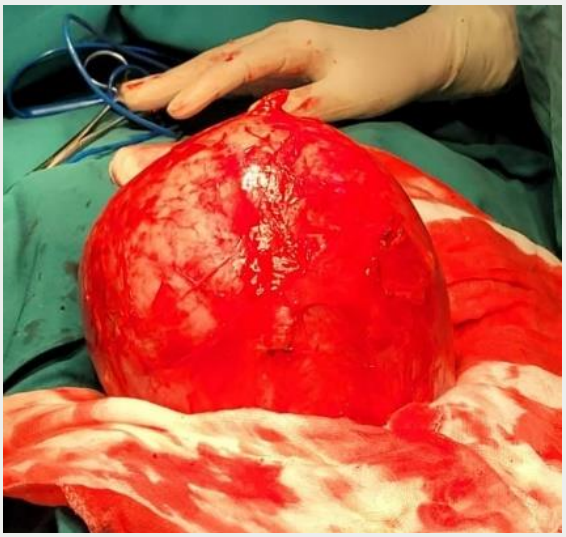

Figure 3. A 30-cm intramural posterior uterine fibroid in necrobiosis.

Patient perspective

From the patient's perspective, the experience of managing a giant uterine fibroid complicated by aseptic necrobiosis and cutaneous fistulation was both physically and emotionally challenging. Initially, the diagnosis and the severity of the condition were overwhelming, particularly given the rare nature and potential for long-term complications. However, the patient expressed relief and gratitude for the clear communication provided by the medical team, which helped her understand the treatment options and the surgical plan. The preservation of fertility was a significant concern, and the successful outcome was a source of immense reassurance. Postoperatively, the patient emphasized her satisfaction with the care received and the absence of recurrence or complications during follow-up. Despite the initial distress, the experience strengthened her trust in the healthcare system and highlighted the importance of early diagnosis and comprehensive care in managing such complex cases.

Discussion

This case highlights the rare and challenging complications of a giant uterine fibroid, including aseptic necrobiosis and cutaneous fistulation, underscoring the importance of early diagnosis and appropriate surgical intervention.

Uterine fibroids are the most common gynecologic tumors, affecting approximately 20 to 50% of females worldwide [1]. These tumors generally progress slowly and are often asymptomatic, although complications may arise [3]. Aseptic necrobiosis, a leading cause of pain in fibroids, occurs secondary to ischemia, which results from poor vascularization due to increased size [3]. This condition is more frequently observed during pregnancy when estrogen promotes fibroid growth [5].

On clinical examination, patients typically report intense paroxysmal pelvic pain, often accompanied by nausea, vomiting, and bowel transit disorders (reflex ileus) [5]. Fever (38-38.5°C) and mild systemic symptoms may also be present [5]. Vaginal examination commonly reveals a rounded and tender mass attached to the uterus [5]. Depending on the location of the myoma, both vaginal palpation and sometimes abdominal palpation detect a soft and painful tumor with regular contours [5].

Ultrasonography helps rule out adnexal torsion and reveals a characteristic "cocoon-like" myoma with central hyperechogenicity, which is painful during the passage of the probe [5]. Doppler studies confirm reduced or absent vascular flow to the fibroid [5]. A mild inflammatory syndrome can also be detected through elevated plasma C-reactive protein [5].

Giant uterine fibroids are rare, often exceeding 15 kg in weight, and can cause abdominal distension and pain [6]. Differential diagnosis of a giant uterine fibroid can be challenging, as it may mimic ovarian tumors due to overlapping clinical and radiological features [7]. Uterine fibroids are typically benign and composed of smooth muscle cells influenced by hormonal interactions [7]. While most fibroids are asymptomatic, larger ones may present with nonspecific symptoms such as abdominal pain, bloating, or pressure effects on adjacent organs [6]. In some cases, imaging modalities like ultrasound and CT may suggest an ovarian mass, as seen in reports where giant fibroids were initially misdiagnosed as mucinous ovarian tumors [7]. Even advanced imaging techniques, including magnetic resonance imaging, may not always distinguish fibroids from ovarian fibroma or Brenner's tumor due to their fibrous composition and signal similarities [7]. This was exemplified in the case described by Chowdhury et al. [7], where a large uterine fibroid was initially mistaken for an ovarian tumor based on clinical and radiological findings. This highlights the importance of considering uterine fibroids in the differential diagnosis of large abdominopelvic masses and underscores the role of laparotomy and histopathological examination for accurate diagnosis, especially when preoperative investigations are inconclusive [6].

Giant uterine fibroids may lead to malnutrition due to compromised blood supply, complicating their management [8]. Aseptic necrobiosis occurs when fibroids undergo ischemic changes, which can be particularly problematic during pregnancy [8]. In one case, a pregnant female presented with symptoms resembling a strangulated hernia, ultimately revealing necrobiosis of a fibroid during an emergency cesarean section [9].

Utero-cutaneous fistulas, although rare, can develop post-surgery or due to necrotic fibroids [10]. A case study highlighted a patient with a fistula resulting from multiple myomectomies, necessitating surgical intervention [10]. While giant fibroids can lead to severe complications, some cases may resolve with conservative management or surgical intervention. However, the potential for significant morbidity underscores the need for careful monitoring and timely treatment [10].

The uniqueness of this case lies in the rare complication of cutaneous fistulation. Most reported cases of uterocutaneous fistulas have occurred following surgical procedures, such as cesarean delivery or miscarriage management [11]. Sönmezer et al. [11] reported a case of a 30-year-old female who developed a uterocutaneous fistula following surgical treatment for an incomplete abortion. Similarly, Seyhan et al. [12] described a 25-year-old female who developed a fistula after her fourth cesarean delivery. Only one case of utero-cutaneous fistula resulting from red degeneration of an intramural fibroid has been previously documented [13].

In our case, we hypothesize that postpartum adhesions and fibroid degeneration caused direct contact between the uterus and the thin abdominal wall, leading to the formation of the uterocutaneous fistula. Indeed, ischemic necrosis of the fibroid, often triggered by compromised vascular supply during the postpartum period, creates a pro-inflammatory microenvironment rich in necrotic debris, which serves as a nidus for secondary bacterial infection. Postpartum adhesions, formed due to uterine trauma and inflammation, tether the necrotic fibroid to adjacent structures, such as the anterior abdominal wall, providing an anatomical pathway for infection to spread [3]. Persistent inflammation weakens tissue integrity, while enzymatic activity from bacteria and host immune cells facilitates progressive tissue erosion [3]. The formation of an abscess further increases local pressure, promoting the extension of necrotic and infected material through fascial planes and eventually to the skin surface, forming a fistulous tract [3].

The management of giant uterine fibroids involves a range of strategies, including medical, minimally invasive, and surgical interventions, tailored to the individual patient's needs, particularly their reproductive goals and overall health status [5]. Treatment is primarily medical, including analgesics, bed rest, and the application of ice to the abdomen. Non-steroidal anti-inflammatory drugs should be avoided during pregnancy, especially after 24 weeks' gestation [5]. Antibiotics are generally not required [5]. Surgery is typically delayed, except in cases involving large, symptomatic fibroids, where it may be considered after delivery [5]. Treatment options vary, with surgical interventions like myomectomy being preferred for fertility preservation, while hysterectomy may be necessary for definitive treatment, particularly in symptomatic cases [14,15]. Minimally invasive techniques, such as uterine artery embolization, can help reduce fibroid size and alleviate symptoms, either as a preoperative measure or as a standalone treatment [14]. Effective management typically requires a multidisciplinary approach, involving gynecologists, radiologists, and surgeons to ensure optimal outcomes [14,15]. While surgical intervention is often necessary for giant fibroids, conservative management may be an option for asymptomatic patients, emphasizing the importance of personalized care and shared decision-making in treatment planning [14]. Several treatment approaches for utero-cutaneous fistulas have been reported [12]. Seyhan et al. [12] demonstrated a nonsurgical approach using gonadotropin-releasing hormone agonists (GnRHa), which reduced surgical risks. GnRHa induced atrophy of the endometrial-like lining of the fistula tract, leading to its spontaneous closure by inhibiting the menstrual cycle [7]. In the present case, given the age of the patient, fertility concerns, and the presence of a large fibroid, we opted for surgical intervention. Following adhesiolysis, the fistula tract was excised, and a myomectomy was performed.

In our case, the cesarean section was indicated primarily due to acute fetal distress. The fibroid was not resected during the procedure. In the postpartum period, the patient presented abdominal and pelvic pain, increased abdominal size, and skin fistulation, as well as suggestive ultrasound findings. Based on these observations, the patient was started on antibiotics, and a scheduled myomectomy was performed.

Table 1 summarizes key aspects of similar case reports and studies. This table provides an overview of the clinical context in which rare complications, such as aseptic necrobiosis and cutaneous fistulation, occur and highlights the various management approaches used in these cases.

Table 1. Summary of similar case reports and studies on giant uterine fibroids with rare complications.

Study	Year	Fibroid characteristics	Complications	Management/Treatment	Outcome
Chowdhury et al. [7]	2015	Giant uterine fibroid	Aseptic necrobiosis cutaneous fistulation	Myomectomy	Uneventful recovery Fertility preserved
Rout et al. [14]	2023	Large fibroid with necrosis	Necrobiosis Pelvic pain	Myomectomy Post-operative follow-up++	Successful removal No recurrence
Timofeeva et al. [8]	2024	Giant uterine fibroid	Severe abdominal pain Mass effect	Hysterectomy	Postoperative recovery without complications
Kamińska et al. [15]	2023	Subserosal fibroid	Skin fistulation Abdominal distension	Myomectomy	Patient improved No recurrence
Irana et al. [16]	2023	Large uterine fibroid	Degeneration Infection	Uterine artery embolization Myomectomy	No further complications Fertility preserved

Conclusion

This clinical case highlights the diagnostic challenges frequently encountered in the management of aseptic necrobiosis of uterine fibroids. Aseptic necrobiosis of leiomyomas can present with complications such as skin fistulation. Effective management requires careful planning and a multidisciplinary approach to surgical decision making.

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