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Cutaneous Fistulization of a Necrobiotic Uterine Leiomyoma after Prophylactic Cesarean Delivery at 34 Weeks of Gestation in a Polymyomatous Uterus: A Case Report

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Abstract

Introduction: Aseptic necrobiosis is the most common complication of uterine fibroids during pregnancy. Progression to cutaneous fistulization is exceptional. We report a case of cutaneous fistulization of a necrobiotic uterine leiomyoma occurring after cesarean delivery with myomectomy. **Case Presentation:** A 30-year-old patient with a polymyomatous uterus experienced several painful episodes during pregnancy, requiring repeated hospitalizations. A therapeutic cervical cerclage was performed at 24 weeks of gestation because of regular uterine contractions and cervical changes. At 34 weeks and 4 days of gestation, after fetal lung maturation, cesarean delivery with myomectomy of an obstructive anterior previa fibroid was performed through a subumbilical laparotomy because of persistent, unbearable pain. The postoperative course was marked by delayed wound healing and chronic suppuration. Ten months later, the patient was admitted with an abdominopelvic mass fistulizing to the skin. Computed tomography revealed a 162 × 98 × 130 mm lesion containing air bubbles and communicating with the skin, without an intestinal fistula. Surgical exploration identified the fistulized mass as a pedunculated subserosal FIGO type VI fibroid, distinct from the anterior previa fibroid re-

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moved during cesarean delivery. En bloc excision of the mass with resection of the cutaneous fistulous tract and additional myomectomies was performed. Histopathological examination concluded that it was a uterine leiomyoma with aseptic necrobiotic changes and a fistulous tract, without signs of malignancy. Postoperative follow-up on days 7, 30, and 60 showed complete healing, with no superinfection or fistula recurrence. **Conclusion:** Cutaneous fistulization of a necrobiotic uterine leiomyoma is an exceptional complication. It may occur long after cesarean delivery in a patient with a polymyomatous uterus. Complete surgical excision led to a favorable outcome, with complete healing and no recurrence after two months of follow-up.

Keywords

Uterine Fibroid, Necrobiosis, Pregnancy, Cutaneous Fistula, Cesarean Delivery, Myomectomy

1. Introduction

Uterine leiomyomas are the most common benign tumors of the female genital tract. Their prevalence during pregnancy ranges from 1% to 10% [1]. Aseptic necrobiosis is their most common complication during pregnancy because of gestational vascular and hormonal changes [1]. Although abdominal pain, uterine contractions, and threatened preterm labor are classic manifestations, progression to cutaneous fistulization remains exceptional. We report the case of a patient admitted to our general surgery department for management of a chronic abdominal mass with a fistula following cesarean delivery and myomectomy, which revealed a necrobiotic uterine leiomyoma.

2. Case Report

A 30-year-old patient was referred to the surgery department of the Abomey-Calavi Zone University Hospital Center (CHUZ) for an anterior abdominal mass associated with chronic suppuration following a midline laparotomy for cesarean delivery with myomectomy performed 10 months earlier.

She had no significant medical history. Her gynecological history included menarche at 16 years and sexual debut at 29 years. Menstrual cycles were regular, without dysmenorrhea or dyspareunia. There was no history of hormonal contraception, alcohol or tobacco use, or family history of uterine fibroids.

The illness began during an ongoing singleton pregnancy in a polymyomatous uterus. Obstetric ultrasound examinations identified several uterine fibroids, including the largest, an anterior corporal FIGO type VI fibroid measuring 159 × 92 mm, with signs of necrobiosis (anechoic areas with hyperechoic margins). An anterior previa fibroid, distinct from the subserosal FIGO type VI fibroid, was also identified (**Figure 1**).

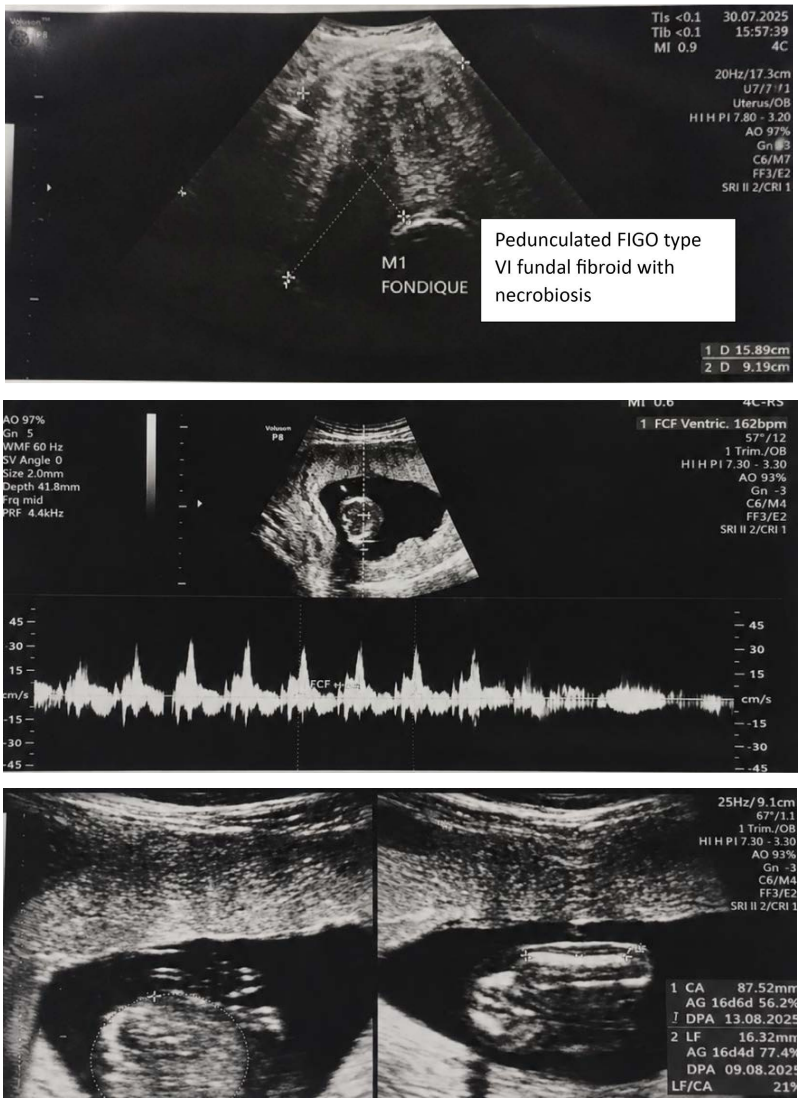


Figure 1. Obstetric ultrasound performed at 16 weeks of gestation showing the pedunculated FIGO type VI fundal fibroid.

The pregnancy was marked by several painful episodes requiring hospitalization. From 24 weeks of gestation, she experienced regular uterine contractions and cervical changes, with a mid-position, soft cervix admitting two fingers at the internal and external os. The membranes were intact, and vaginal examination revealed blood-streaked mucus, suggesting threatened preterm labor complicating aseptic necrobiosis in an ongoing singleton pregnancy at 24 weeks of gestation. She was managed with strict bed rest during hospitalization, nifedipine, analgesics, and therapeutic cerclage. Pain and uterine contractions improved. At 34 weeks and 4 days of gestation, after fetal lung maturation, cesarean delivery was performed through a midline subumbilical laparotomy, following myomectomy of the obstructive previa fibroid, because the painful episodes had recurred and become unbearable. A male infant weighing 2,430 g was delivered, with Apgar scores of 8/9/10. Histopathological examination of the surgical specimen showed

a leiomyoma with necrobiotic changes and no signs of malignancy. The postoperative course was marked by chronic suppuration and delayed healing of the surgical wound. A postpartum ultrasound showed a polymyomatous uterus, including a 131 × 92 mm pedunculated FIGO type VII fibroid, a 66 × 31 mm FIGO type VI fundal fibroid, and another FIGO type III fibroid measuring 42 × 38 mm **(**Figure 2**).**

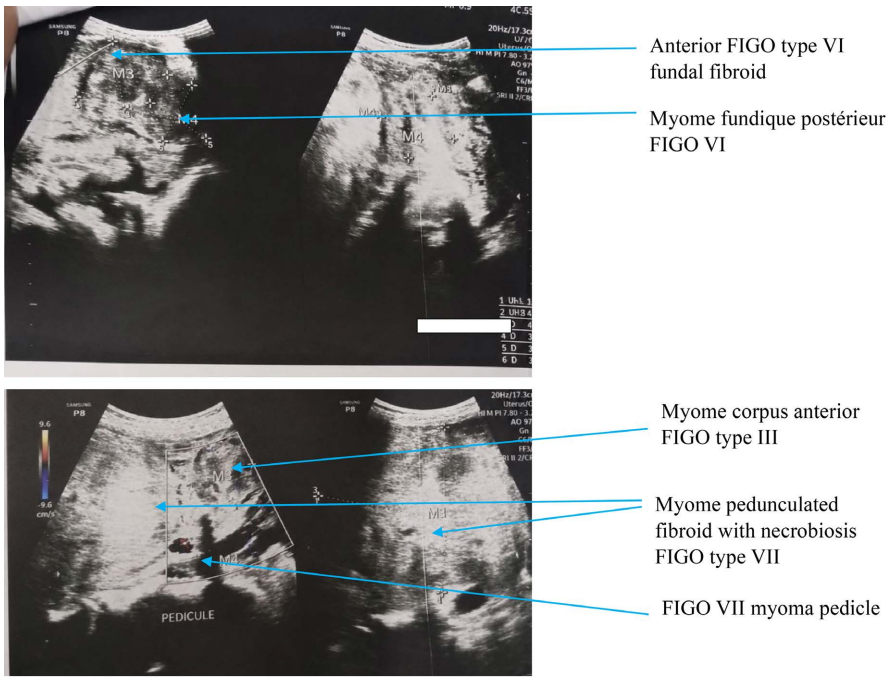


Figure 2. Post-cesarean pelvic ultrasound.

Because symptoms persisted, the patient was admitted to our surgery department for further management.

On admission, the patient's general condition was good, with a WHO performance status of 0. Abdominal examination revealed a large abdominopelvic mass associated with purulent discharge from a skin opening approximately 2 cm in diameter, located on the midline laparotomy scar, 3 cm below the umbilicus (Figure 3).

Palpation revealed a firm, regular, well-circumscribed mass measuring approximately 20 cm along its largest axis. The mass was adherent to the anterior abdominal wall, with limited mobility relative to the deep planes; pressure on it caused pus to discharge through the cutaneous opening. It occupied the pelvic region and extended to the umbilical region.

Gynecological examination revealed a normal-appearing vulva without abnormal discharge. On speculum examination, the vaginal walls and cervix showed no visible abnormalities. Vaginal examination did not identify a distinct pelvic mass. Assessment of uterine size was difficult because of the large abdominopelvic mass, whose boundaries could not be clearly distinguished from the uterus. A diagnosis



Figure 3. Ventral view of the patient's abdomen showing the protrusion of the abdominopelvic mass and the cutaneous fistulous opening.

of an abdominopelvic mass fistulizing to the skin, of undetermined nature, was retained, prompting further imaging investigations.

Abdominopelvic computed tomography showed a large oval abdominal lesion measuring 162 × 98 × 130 mm. The lesion contained dense, non-tissue material with several air bubbles and was surrounded by an 8-mm-thick wall. It communicated with the skin through a 29 × 16 mm cutaneous defect, with no communication with the digestive tract identified (**Figure 4**).

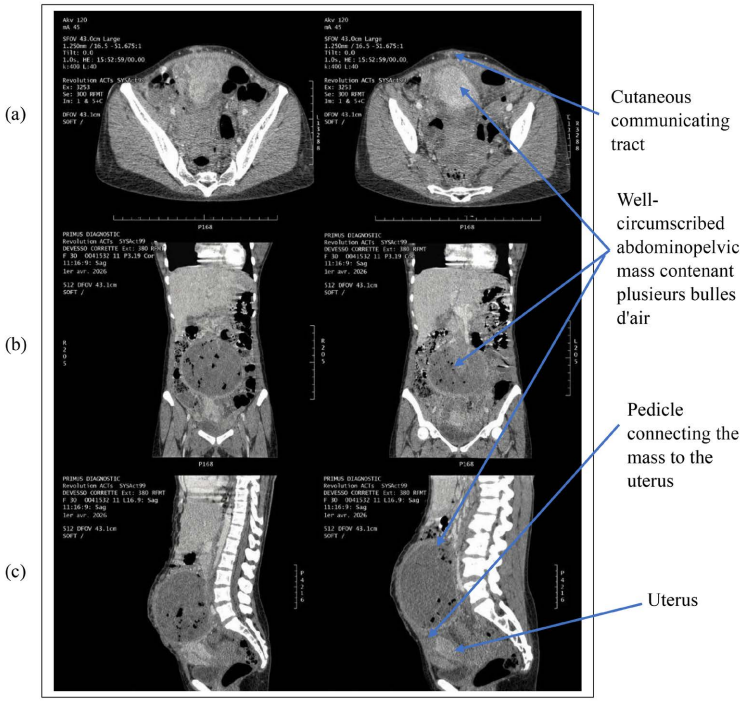


Figure 4. Abdominopelvic CT scan showing the mass and the cutaneous communicating tract. (a): Axial section of the abdomen through the abdominopelvic mass; (b): Coronal section of the abdomen; (c): Sagittal section of the abdomen and pelvis showing the abdominopelvic mass.

Exploratory laparotomy was indicated. Under general anesthesia, a midline incision was made, extending from below to above the umbilicus and bypassing the cutaneous fistulous opening, which had first been closed with a 2-0 nylon running suture. The rectus muscles adherent to the mass were resected. At laparotomy, multiple adhesions were observed between the greater omentum and the upper pole and posterior surface of the mass, on the one hand, and between the small bowel and the lateral surfaces of the tumor, on the other (**Figure 5(a)**). Careful ligation and division of the greater omentum, followed by adhesiolysis between the small bowel and the tumor, was performed. The fistulized mass was a pedunculated subserosal FIGO type VII fibroid that had remained in place after cesarean delivery; it was distinct from the anterior previa fibroid that had been removed during cesarean delivery. This mass arose from the uterine fundus. Two juxtaposed sessile subserosal FIGO type VI uterine fibroids, each measuring approximately 3 cm along its largest axis, were also identified on the posterior surface of the uterine body (**Figure 5(b)**).

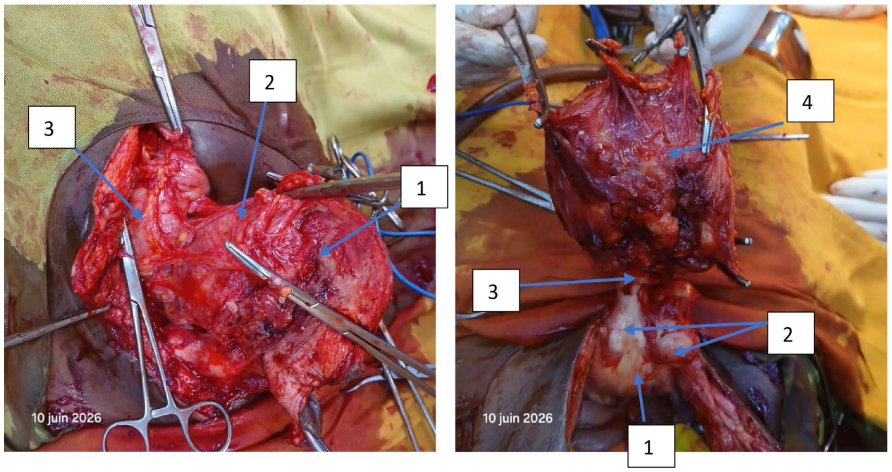
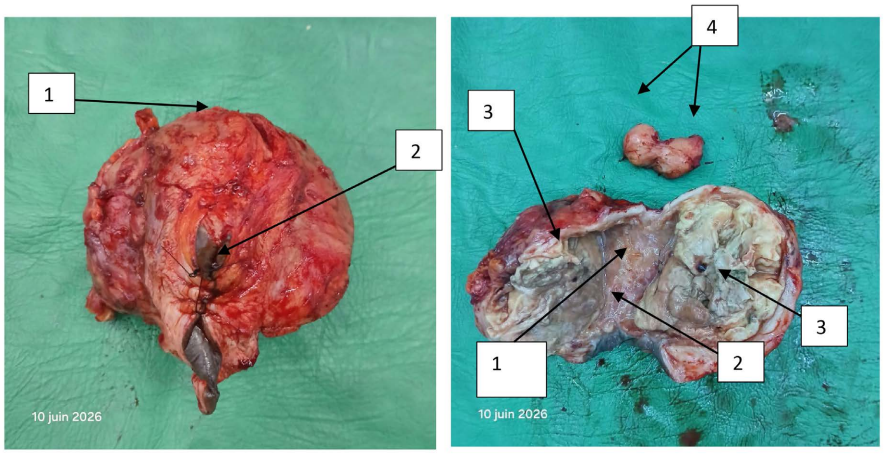


Figure 5. Intraoperative exploration of the abdominopelvic mass. (a): Abdominopelvic mass with adhesions between the greater omentum and the upper pole and posterior surface of the mass. 1. Abdominopelvic mass; 2. Greater omentum; 3. Large bowel; (b): Ventral view showing the mass arising from the uterine fundus. 1. Uterus; 2. juxtaposed FIGO type VI fundal fibroids; 3. Pedicle connecting the mass to the uterus; 4. Abdominopelvic mass.

En bloc excision of the mass, including the skin overlying the fistulous opening, was performed, along with enucleation of the two myomatous nodules (**Figure 6(a)** and **Figure 6(b)**).

The postoperative course was uncomplicated. The hospital stay was three days. The wound healed well (**Figure 7(a)** and **Figure 7(b)**). The patient was reviewed on postoperative days 7, 30, and 60, corresponding to two months of follow-up. By day 60, the lesion had completely healed, with no superinfection or fistula recurrence.



(a) (b)

Figure 6. En bloc excision specimen of the abdominopelvic mass. (a): En bloc excision specimen of the abdominopelvic mass. 1. Abdominopelvic mass; 2. Cutaneous opening closed with sutures; (b): Sagittal section of the abdominopelvic mass showing the myomatous nodules with aseptic necrobiosis. 1. Fistulous tract; 2. Cutaneous opening; 3. Myomatous nodules with necrobiosis; 4. Excision specimen of the juxtaposed FIGO type VI fundal fibroids.



(a) (b)

Figure 7. Ventral view of the patient's abdomen showing the clinical evolution of the postoperative laparotomy scar. (a): Wound status on postoperative day 7e postoperative day; (b): Complete wound healing on postoperative day 30.

Histopathological examination of the surgical specimen concluded that it was a uterine leiomyofibroma with aseptic necrobiotic changes and a fistulous tract.

3. Discussion

Uterine fibroids are the most common benign tumors of the female genital tract in women of reproductive age. Their prevalence during pregnancy is estimated at

1% to 10%, depending on the series [1] [2], owing to improved obstetric ultrasound performance.

In our case, the pregnancy occurred in a polymyomatous uterus containing a large anterior corporal fibroid measuring 159 × 92 mm with signs of necrobiosis. The course was marked by several painful episodes associated with threats of preterm delivery requiring repeated hospitalizations. This presentation is consistent with aseptic necrobiosis (or red degeneration), the most common complication of fibroids during pregnancy [2]-[4]. It results from a mismatch between tumor growth and its blood supply, leading to ischemia and necrosis of the myomatous tissue.

Clinically, it presents with sometimes severe abdominal pain that may mimic other abdominal or obstetric emergencies. Diagnosing myomatous necrobiosis during pregnancy can be difficult, particularly in resource-limited settings. Dohbit *et al*. [4] reported a case of myomatous necrobiosis discovered incidentally during cesarean delivery performed for suspected strangulated hernia in a term parturient, illustrating the diagnostic challenges of this complication.

In our case, the presence of an obstructive previa fibroid led to prophylactic cesarean delivery at 34 weeks + 4 days, combined with myomectomy. Myomectomy during cesarean delivery was long considered contraindicated because of the risk of hemorrhage. However, several recent studies indicate that it can be performed with acceptable morbidity in carefully selected patients, particularly when fibroids are obstructive or readily accessible [5]-[8]. Several authors have also reported potentially severe complications associated with performing cesarean delivery and myomectomy concurrently. These include a significant increase in operative time, increased intraoperative and postoperative blood loss that may require transfusion, and prolonged hospital stay. Higher rates of surgical-site infection, abdominal distension, and postoperative bowel obstruction have also been observed in some studies [6] [7]. Neonatal outcomes reported include a higher incidence of insufficient maternal lactation and significantly lower Apgar scores at various assessment times among infants delivered during these combined procedures [6] [7].

The originality of our case lies in the progression of a necrobiotic fibroid to cutaneous fistulization. This complication appears to be exceptional. A review of the literature shows that fistulas secondary to leiomyomas mainly involve communications with digestive or urinary organs, whereas cutaneous fistulas remain anecdotal and are reported primarily as isolated case reports [9] [10]. In our case, several pathophysiological mechanisms may be proposed. The initial myomatous necrobiosis probably favored secondary infection following prophylactic cesarean delivery. Chronic inflammatory processes led to tight adhesions between the fibroid, greater omentum, small-bowel loops, and anterior abdominal wall. The midline laparotomy performed during cesarean delivery created an area of reduced resistance, allowing the suppurative process to gradually extend through the surgical scar. The CT appearance of a heterogeneous mass containing gas bub-

bles and communicating with the skin strongly suggested this progression. The literature describes this possibility [9] [10], although cases of fibroids with spontaneous fistulization without prior laparotomy or uterine artery embolization have been reported by Anderson KB et al. in Denmark in 2020 [9].

In this case, the initial necrobiosis was described as aseptic, whereas the postoperative course involved chronic suppuration suggestive of secondary infection. The fistulized mass was a pedunculated subserosal FIGO type VII fibroid that had been left in place, distinct from the anterior previa fibroid removed during cesarean delivery. The FIGO classifications mentioned in this report were derived from successive ultrasound examinations performed during pregnancy and postpartum and should be interpreted according to the date of each examination.

Preoperative diagnosis may be difficult in the setting of a chronic postoperative abdominal mass. The main differential diagnoses include chronic abdominal wall abscess, foreign-body granuloma, complicated incisional hernia, and soft-tissue tumor [11]. In this situation, CT and MRI are essential for defining anatomical relationships, looking for bowel communication, and planning the surgical strategy.

Treatment consists of complete surgical excision of the mass, resection of fistulous tracts, and release of associated adhesions [12]. In our patient, this approach resulted in a favorable postoperative course, a short hospital stay, and good abdominal wall healing. Histopathological examination of the surgical specimen concluded that it was a uterine leiomyofibroma with aseptic necrobiotic changes and a fistulous tract, without signs of malignancy.

4. Conclusion

Cutaneous fistulization of a necrobiotic uterine leiomyoma after pregnancy and cesarean delivery is an exceptional complication. It should be considered in any patient with a polymyomatous uterus who presents with delayed postoperative wound healing and chronic discharge. Imaging plays an essential role in the preoperative assessment. Treatment consists of complete surgical excision, which can provide definitive healing.

Consent for Publication

Written informed consent, signed by the patient, was obtained for publication of this case report and the associated images.

Ethical Considerations

The ethical considerations applicable to the management and publication of this case report were observed.

Author Contributions

All authors read and approved the final version of the manuscript.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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