CASE REPORT

Postnatal pyomyoma: a diagnostic dilemma

Ravneet Sirha,¹ Aditi Miskin,² Amin Abdelmagied³¹Department of Obstetrics and Gynaecology, St Mary's Hospital, London, UK²Department of Obstetrics and Gynaecology, Princess of Wales Hospital, Wales, UK³Department of Obstetrics and Gynaecology, Royal Gwent Hospital, South Wales, UK**Correspondence to**Dr Ravneet Sirha,
r_sirha@hotmail.com**SUMMARY**

Pyomyoma is a rare, yet potentially fatal complication of uterine leiomyoma. Clinically difficult to diagnose as a result of non-specific symptoms, its presentation is commonly confused with fibroid degeneration. Late diagnosis has severe implications, with the mortality of the condition remaining high. Despite the availability of powerful antibiotics, surgical intervention is frequently required for the curative treatment of the critically ill patient. Here, we report a case of postpartum pyomyoma developing after a complicated antenatal course of placenta praevia resulting in recurrent antepartum haemorrhage, preterm prelabour rupture of membranes and eventual emergency caesarean section for cord prolapse. We highlight the diagnostic difficulty and delay in definitive surgical intervention. Using this case, we have emphasised the importance of strong clinical suspicion when faced with a triad of pain, sepsis without an obvious source and a known diagnosis of leiomyoma to prevent fatalities.

BACKGROUND

Pyomyoma is a rare but serious complication of common leiomyoma.¹ Despite the widespread availability of potent antibiotics, the mortality of this condition has not changed from 20–30%.² This may be explained by the rarity of pyomyoma resulting in inexperience in making the diagnosis. The aim of this communication is to increase awareness of the condition and its potential as a differential in cases of unexplained sepsis, pain and known leiomyoma. It highlights the importance of considering pyomyoma in pregnant and postpartum women who often have a suppressed immune system and can clinically deteriorate and become critically septic very quickly. We discuss progression from surgery as the sole curative treatment to the use of interventional radiology in managing the condition.

CASE PRESENTATION

The patient, a 37-year-old para 3 (all spontaneous vaginal deliveries) with a known 9×8 cm posterior leiomyoma presented to hospital several times antenatally. At 20 weeks gestation ultrasound scan (USS) revealed a low-lying placenta completely covering the cervical os and echogenic fetal bowel. The patient subsequently experienced recurrent episodes of vaginal bleeding. She presented at 23 weeks gestation after a further unprovoked bleed and good history of spontaneous rupture of membranes. This was confirmed on speculum examination and USS measured a reduced amniotic fluid index of 4 cm. Risks of preterm prelabour rupture of membranes (PPROMs) were discussed

with the patient, including the significant risk of chorioamnionitis, currently unviable fetus, future developmental problems including pulmonary hypoplasia and bone deformities should the pregnancy continue. Despite this, the patient opted to continue with the pregnancy. She was therefore started on erythromycin 250 mg four times a day and attended the day assessment unit as an out-patient for regular review. She remained afebrile and well, with normal inflammatory markers and clear, non-offensive liquor. Steroids were administered for fetal lung maturation. At 26+6 weeks gestation after a significant antepartum haemorrhage with clots, it was felt inpatient care was appropriate. After lengthy discussions with the neonatal team, and in line with the patient's wishes, a conservative approach was taken as a result of severe prematurity. Erythromycin was continued and she was monitored closely, with twice daily cardiotocography.

At 27+4 weeks gestation, the patient reported feeling a lump at the introitus. On parting the labia, a loop of cord was visualised and the patient was taken to the operating theatre immediately for an emergency caesarean section. A male infant weighing 1015 g was eventually delivered after a difficult surgery due to reduced liquor, requiring breech extraction and internal manipulation. The intraoperative blood loss was 2000 mL and the patient was transfused 2 units of red blood cells. As a result of the blood loss and traumatic delivery she was observed on a high-dependency unit. She became symptomatic of anaemia with a persistent tachycardia and reported feeling lightheaded so was transfused a further 2 units of red blood cells. Post-transfusion she remained stable and was stepped down to the postnatal ward the following day. Postoperatively, the patient spiked a single temperature of 38°C, which responded to antipyretics. Blood cultures were negative and urine contained leucocytes and nitrates. She was started on cefalexin for a presumed urinary infection and discharged home well 5 days postpartum.

Ten days postpartum, the patient reattended with vomiting and feeling unwell. On arrival she appeared pale, clammy and tearful. She was hypotensive with a blood pressure of 90/40, tachycardic at 100 bpm and experiencing rigours with a temperature of 39.1°C. A full examination revealed normal heart sounds, a clear chest and a soft abdomen with an involuted uterus and a clean healing caesarean wound. There was no offensive vaginal discharge and bleeding had settled. Breast examination showed no evidence of mastitis. She had no leg swelling and no obvious source of infection. She was admitted for a full septic workup,

CrossMark

To cite: Sirha R, Miskin A, Abdelmagied A. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2013-201137

which included full blood count (FBC)/C reactive protein (CRP), blood and urine cultures, wound swab, high vaginal swab and chest X-ray. USS of the pelvis was also performed for completeness, excluding a pelvic collection. It was difficult to comment on retained products of conception as the uterus was bulky postpartum, however, this was unlikely as she was not experiencing any vaginal bleeding or discharge. She was started on broad spectrum antibiotics, cefuroxime and metronidazole intravenously as per local protocol. Provisional diagnosis was one of urosepsis in light of a previous urinary tract infection, leucocytes on urine dipstick and suprapubic abdominal pain. She responded well to fluid resuscitation, although continued spiking temperatures. Repeat blood cultures were persistently negative for any bacterial growth, however, the CRP continued to rise to 300. Initial response to antipyretics and cooling was good and broad spectrum antibiotic therapy was continued in line with microbiology advice. With a known history of leiomyoma, significantly raised inflammatory markers despite a few days of antibiotics and a negative septic screen, it was initially thought fevers and symptoms of abdominal pain may be the result of postpartum fibroid degeneration. However, it soon became apparent that the temperatures were not settling and the peaks were becoming more frequent and severe. Microbiology was consulted for antibiotics advice.

On day 16 postpartum, the patient had rigours with a temperature reaching 40.1°C. Repeat ultrasound of the pelvis showed a bulky postpartum uterus with an area of mixed echogenicity within the uterus measuring 9.6×8.8×6.6 cm, consistent with the known fibroid. On day 18 postpartum, the patient reported chest pain, ECG showed normal sinus rhythm and oxygen saturations were satisfactory, however, given the patient's immobility, sepsis and recent surgery her risk of venous thromboembolism was high. A CT pulmonary angiogram was negative and the patient's symptoms resolved spontaneously. A CT of the abdomen and pelvis was performed to exclude a pelvic collection. It reported a bulky postpartum uterus with a multiloculated collection within the uterine cavity near the fundus with peripheral enhancement of the loculations. At this point, the patient showed signs of improvement clinically and numerically on her bloods. Microbiology was consulted and Tazocin was given.

On day 24 postpartum, she deteriorated further with the same symptoms. On examination, she was mildly tender over the lower abdomen with no guarding, rebound or palpable masses. Vaginal examination revealed a boggy collection to the left of the uterus. The case was discussed by the multidisciplinary team (MDT) and the imaging reviewed by our radiology colleagues. The need for MRI was reviewed, however, the consultant radiologist recommended repeat CT scan. CT pelvis showed an enlarged uterus with a left lateral bulge representing the previous fibroid. There were low-density changes within the uterus, particularly at the fundus. The radiologist reported haematoma/pus or even retained products of conception could not be excluded. After further MDT discussion the consensus was that the images suggested very little free pus within the uterine cavity itself, but rather the possibility of a loculated enclosed area of infection in the region of the fibroid. The treatment options of conservative management, dilation and curettage or laparotomy ± total abdominal hysterectomy (TAH) were discussed with the patient. She was able to appreciate that the conservative approach was not a suitable option in light of her ongoing sepsis. Dilation and curettage was offered but it was highlighted it would be of minimal use if the infection was walled off in a capsule. As the patient emphasised she had

completed her family and fertility was no longer required, a decision was made to treat her surgically and promptly as per her wishes. TAH with conservation of ovaries was performed the next day and the patient's condition significantly improved. Microbiology recommended 72 h of imipenem postoperatively, followed by 1 week of oral cefalexin and metronidazole. Her postoperative course was uncomplicated and she was discharged home 5 days later.

Intraoperatively, a soft bulky uterus containing a large soft fibroid was removed. The specimen was opened to reveal a healthy endometrial cavity with a collection of pus within a degenerating fibroid towards the fundus. Swabs were not taken of the pus as they would not have been appropriately representative given the patient's previous antibiotic load.

The histology of the pathology specimen confirmed a uterus with benign proliferative type endometrium containing a partially infarcted fibroid measuring 14 cm in diameter at the uterine fundus. The fibroid showed large areas of infarction type necrosis associated with acute inflammation (figures 1–4).

INVESTIGATIONS

- ▶ Septic screen—FBC/CRP, blood cultures, urine cultures, wound swab, high vaginal swab, chest X-ray
- ▶ USS of the pelvis.
- ▶ CT scan of the abdomen/pelvis.

OUTCOME AND FOLLOW-UP

The patient's findings were discussed with the patient after surgery. She remained well on discharge and was seen in clinic 6 weeks posthysterectomy and fully debriefed.

Neonatal summary

The male infant was delivered by emergency caesarean section weighing 1015 g. He was resuscitated and transferred to Bristol special care baby unit (SCBU) 3 days postdelivery. Problems included respiratory distress syndrome, bronchopulmonary dysplasia, bacterial sepsis and contractures of all the four limbs. Bowel obstruction secondary to meconium ileus resulted in a laparotomy during which the small bowel was perforated. A bowel resection was performed and stoma fashioned. He was transferred to the SCBU at the University Hospital of Wales, Cardiff at 1 month postdelivery for continuation of care.

DISCUSSION

Leiomyomas are a common occurrence in the general female population, inevitably leading to their presence in 1–2% of pregnancies.³ Complications of leiomyoma in pregnancy are

Figure 1 Bulky uterus at abdominal hysterectomy.

Figure 2 Uterus opened after hysterectomy, instruments illustrating cervix. Partially necrotic pyomyoma seen at uterine fundus.

Figure 4 Uterus opened after hysterectomy. Partially necrotic pyomyoma seen at uterine fundus.

spontaneous abortion, PPROMs, preterm labour, obstructed labour, postpartum haemorrhage and degeneration.⁴ Infection of leiomyoma with the resultant development of a pyomyoma can be fatal.

More common in the preantibiotic era, the occurrence of pyomyoma is now rare. Miller *et al*'s literature review in 1945⁵ found only 75 cases of pyomyoma reported between 1871 and 1945, and mortality was high. A series published in 1928 by Ruch⁶ concluded the patient died in 29% of cases, surgical intervention being the only hope of controlling the infection and enabling curative treatment. Fewer than 20 cases have been reported since 1945. Interestingly, despite more potent antibiotics, the mortality remains around 21%^{1 2} with surgical intervention being the eventual treatment. Of these cases, six were pregnancy related, several were postmenopausal, a couple were related to diabetics and one was associated with intravenous drug use.²

The postulated mechanism of pyomyoma development is (a) direct spread of infection from the uterine cavity, (b) extension of infection from the surrounding structures, such as the bladder or bowel, (c) vascular insufficiency in diabetics/women with atherosclerotic disease or (d) haematogenous/lymphatic spread.⁵

In our case, there were several potential routes of transmission of infection. It is very possible that bacterial seeding of the fibroid may have begun antenatally as a result of PPROMs and ascending infection. Increased uterine blood flow in pregnancy could cause haematological spread of infection from elsewhere in the body although as blood cultures had been persistently

negative this is unlikely. Instrumentation of the uterus during emergency caesarean section is another potential. The fibroid was noted intraoperatively and appeared normal. Postpartum acute degeneration of the fibroid resulting in bacterial seeding in a necrotic centre is another explanation.

The differentiation between postpartum fibroid degeneration and pyomyoma presents a diagnostic challenge due to the similarities in signs and symptoms.⁷ It is evident from reviewing the literature and indeed our case, that much of the delay in making the diagnosis is the assumption that the clinical picture is due to more commonly occurring degeneration. This case highlights that despite the rarity of pyomyoma, maintaining high clinical suspicion in a postpartum woman with a triad of pain, sepsis with no obvious source and history of uterine leiomyoma, is important.

Although TAH was an acceptable option for our patient, often this will not be the case in lower parity, younger women. Recent studies have used imaging to optimise and plan surgical intervention, so as to treat the pyomyoma but conserve the uterus and therefore fertility. Shona *et al* illustrated the role of dynamic MRI to clarify the location of the fibroid along with its vascular pedicles and extent of myometrial involvement.⁸ MRI has also successfully differentiated between necrotic non-liquefied leiomyomas and degenerating fibroids. MRI has allowed successful myomectomy of a pyomyoma maintaining the uterus.⁸

Laubach *et al*⁹ have recently shown the use of CT-guided drainage and lavage of pyomyoma related to pregnancy in two out of three cases successfully. They suggest that image-guided drainage is a safer, minimally invasive option for these patients who can be critically ill and septic, exposing them to less risks of extensive surgery and also preserving fertility.⁹ In the absence of complete recovery, however, the option of TAH still remains.

Figure 3 Uterus opened after hysterectomy. Partially necrotic pyomyoma seen at the uterine fundus.

Learning points

- ▶ Presentation is clinically similar to degenerating leiomyoma.
- ▶ Mortality from pyomyoma remains high.
- ▶ Sepsis can progress rapidly.
- ▶ A high index of suspicion with a triad of pain, sepsis without a clear focus and a known history of leiomyoma will facilitate prompt treatment.
- ▶ Radiologically directed surgery/drainage of pyomyoma can preserve fertility.

Contributors The authors have reviewed and approved the final version of this manuscript.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Genta PR, Dias ML, Janiszewski TA, *et al.* Strep agalactiae endocarditis and giant pyomyoma simulating ovarian cancer. *South Med J* 2001;94:508–11.
- 2 Greenspoon JS, Ault M, James BA, *et al.* Pyomyoma associated with polymicrobial bacteraemia and fatal septic shock: case report and review of the literature. *Obstet Gynecol Surv* 1990;9:563–9.
- 3 Cunningham CF, Grant N, Leveno K, *et al.* *Williams obstetrics*. 21st edn. New York: McGraw-Hill, 2001:927.
- 4 Coronado GD, Marshall LM, Schwartz SM. Complications in pregnancy, labour and delivery with uterine leiomyomas: a population-based study. *Obstet Gynecol* 2000;95:764–9.
- 5 Miller I. Suppurating fibromyomas: report of case with a review of literature. *Am J Obstet Gynecol* 1945;50:522–6.
- 6 Ruch UA. Intrapartum rupture of a suppurative myoma, with purulent peritonitis: report of a case. *Obstet Gynecol* 1963;21:593–6.
- 7 Patwardhan A, Bulmer P. Pyomyoma as a complication of uterine fibroids. *J Obstet Gynaecol* 2007;27:444–5.
- 8 Torrance S, Muhn N, Ellis S, *et al.* Role of dynamic MRI in surgical decision-making for postpartum woman with a prolapsed degenerating uterine leiomyoma. *J Obstet Gynaecol Can* 2009;31:446–51.
- 9 Laubach M, Breugelmans M, Leyder M, *et al.* Non surgical treatment of pyomyoma in the postpartum period. *Surg Infect* 2011;12:65–8.

Copyright 2013 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow