CASE REPORT

Endometriosis: a rare and interesting cause of recurrent haemorrhagic ascites

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SUMMARY

Recurrent haemorrhagic ascites as a cause of endometriosis is rare. We report the case of a 36-year-old woman presenting acutely with abdominal distension, ascites and an elevated CA-125 raising the suspicion of ovarian malignancy. Tissue biopsies retrieved during laparoscopy confirmed the diagnosis of endometriosis associated with haemorrhagic ascites. Gonadotropin-releasing hormone (GnRH) analogues were started to manage symptoms, with good effect. Subsequently, *in vitro* fertilisation resulted in a successful singleton pregnancy and by the second trimester, there was full resolution in symptoms. During the early puerperal period, the development of massive ascites recurred, requiring symptomatic relief through repeated ascitic drainage and GnRH analogues. Long-term follow-up is planned with the hope of continuing with medical management at least until the patient's family is complete when the surgical option of bilateral salpingo-oophorectomy with or without hysterectomy will be discussed.

BACKGROUND

Endometriosis is a common cause of morbidity in young women. This case highlights a rare but interesting presentation of endometriosis and the obstacles faced with managing this poorly understood disease.

CASE PRESENTATION

A 36-year-old Afro-Caribbean nulliparous woman presented to the emergency department with acute abdominal pain and bloating. Further questioning revealed a 7 month history of worsening dysmenorrhoea, deep dyspareunia and constipation. Her menstrual cycles were regular and smear history was normal. The patient's medical history included a long history of recurrent urinary tract infections. There was no significant family history and she was not on any regular medication; she neither smoked nor drank alcohol. Abdominal examination revealed generalised tenderness and distension, however, gynaecological examination was unremarkable.

INVESTIGATIONS

Blood analysis showed a microcytic anaemia with a haemoglobin of 10.8 g/dL and a lactate dehydrogenase of 405 IU/L. An ultrasound scan of the abdomen and pelvis revealed extensive ascites with no evidence of adnexal masses. MRI of the pelvis was also performed confirming a large amount of fluid in the pelvis with a faint fluid level suggesting a collection. The uterus was normal and both ovaries appeared

polycystic. Serum tumour markers demonstrated a markedly raised CA-125 of 1123 U/mL. A sample of ascitic fluid was obtained through paracentesis. Cytological analysis of the ascitic fluid failed to detect any evidence of malignant cells. Owing to the extensive volume of ascites, an ascitic drain was inserted under ultrasound guidance. In total, 3.5 L of blood stained fluid was drained within the first 24 h. Cultures were negative and no acid-fast bacilli were detected.

CT of the abdomen and pelvis (figure 1) further demonstrated ascites and suggested evidence of disease affecting the transverse colon and small bowel with no evidence of pelvic pathology.

DIFFERENTIAL DIAGNOSIS

After discussion at a gynaecology oncology multidisciplinary team meeting, the decision was made to perform an exploratory laparoscopy to obtain a tissue biopsy as it was thought that an imaging-guided attempt for biopsy carried a higher risk of bowel injury.

Findings at laparoscopy included: 1.6 L of haemorrhagic ascites, large nodules at both uterosacral ligaments and multiple nodules affecting the

Figure 1 CT study showing gross ascites at the time of presentation.

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Figure 2 Haemorrhagic ascites.

Figure 4 Abdominal wall bowel adhesion.

ascending colon, transverse colon, small bowel and peritoneum. The sigmoid colon was densely adherent to the left pelvic side wall and posterior uterine wall obstructing the view of the left adnexum (figures 2–4). Furthermore, a dilated oedematous right tube was found with the right ovary adherent to the pelvic side wall. Multiple biopsies were obtained and histology was consistent with cyclical endometrium in the proliferative phase. A diagnosis of stage IV endometriosis¹ with associated recurrent haemorrhagic ascites was made.

TREATMENT

Owing to the widespread nature of her disease extending beyond the pelvis, the patient was started on gonadotropin-releasing hormone (GnRH) analogues during which time her symptoms gradually improved. Fertility implications were discussed and the couple expressed the desire to start a family. After 5 months of GnRH analogue injections, during which National Health Service funding was secured, the couple was referred to a fertility clinic and underwent a cycle of *in vitro* fertilisation (IVF) treatment. Fourteen eggs were retrieved and 2.5 L of haemorrhagic ascitic fluid was drained at the time of egg collection. Six embryos developed to the blastocyst stage, five were frozen and the patient underwent a single embryo transfer. She had a positive pregnancy test 2 weeks later. Episodes of recurrent ascites persisted during the first trimester; however, by the second trimester of pregnancy, her symptoms had settled.

OUTCOME AND FOLLOW-UP

At 32 weeks gestation she presented in preterm labour and underwent an emergency caesarean section for fetal distress.

Figure 3 Multiple active endometriotic nodules.

At the time of caesarean section a small amount of haemorrhagic ascites was found and endometriotic implants were found at the bladder peritoneum. The lower uterine segment was thick and vascular, and the pouch of Douglas was obliterated with sigmoid colon adherent to the back of the uterus. A live female infant was delivered in good condition and admitted to the special care baby unit. The patient's postoperative recovery was slow but she was eventually discharged home with advice to continue regular expression of breast milk to induce lactational amenorrhoea and prolong ovarian suppression.

Two weeks postnatally, the patient re-presented to the emergency department with acute abdominal pain and distension. Imaging confirmed a recurrence of ascites, which was drained. She was unable to continue with breast feeding in view of the severe pain. She was restarted on GnRH analogues and required a further three ascitic drains during the puerperium. After the fifth GnRH injection, an ultrasound scan showed no evidence of ascites, and add-back tibolone was started. She is currently also under the care of the pain team and her symptoms are well controlled on pregabalin and non-steroidal anti-inflammatory medication. The surgical option of bilateral salpingo-oophorectomy with or without hysterectomy remains an option and will be considered further when her family is complete.

DISCUSSION

Endometriosis is defined as the presence of endometrial tissue located outside of the uterine cavity. In cases of pelvic endometriosis, symptoms often include chronic pelvic pain, dysmenorrhoea, deep dyspareunia and subfertility. Recurrent haemorrhagic ascites secondary to endometriosis is a very rare complication of the condition^{2–3} and is often initially mistaken for ovarian/primary peritoneal malignancy. The first case of endometriosis-related ascites was described by Brews *et al* in 1954,⁴ and since then a further 63 cases have been reported worldwide with a predominantly higher occurrence in nulliparous women of African descent.⁵ Most cases of endometriosis-related ascites present with a history of dysmenorrhoea and chronic pelvic pain alongside progressive abdominal distension.^{2–5–7} The pathophysiology of endometriosis is not completely understood, however, various theories exist. Furthermore, the exact cause of the development of recurrent ascites is not known, however, Bernstein *et al*⁸ described a theory proposing the formation of ascites due to irritation of serosal surfaces by free blood released from ruptured chocolate cysts.

Elevated CA-125 levels may be associated with ovarian endometrioma and advanced endometriosis, as demonstrated in our

case. This is often a cause for concern at initial presentation, often raising the suspicion of ovarian malignancy.

The long-term management of severe endometriosis associated with recurrent ascites is difficult. Consideration of the patient's age, surgical history and desire for preservation of fertility is important in choosing between medical and surgical management. Currently, there are no controlled studies to determine whether surgical excision of moderate-to-severe endometriosis enhances fecundity rates and thus surgery is generally not recommended unless performed for other indications such as management of chronic pain. In our case, with multifocal disease and extensive bowel involvement, it was felt that the aim of any surgical intervention would be to excise the large nodules rather than aim to treat all of the endometriotic implants. This would involve extensive bowel surgery with a high risk of resection with no good evidence of improvement in fertility and no guaranteed resolution of ascites. In a Cochrane review of three randomised controlled trials, 3–6 months of ovarian suppression with GnRH agonists before IVF in women with stage III–IV endometriosis led to a fourfold increase in clinical pregnancy rates.⁹

Management in previously reported cases has included medical management with GnRH analogues and surgical management in the form of unilateral/bilateral salpingo-oophorectomy, with or without hysterectomy in women who have completed their family.

GnRH analogue treatment is considered an effective treatment option particularly where preservation of fertility is necessary, with evidence in some cases of full remission after 2 years.¹⁰ Progestogens are also used in the medical management of endometriosis and have been shown to induce regression and apoptosis of endometriotic lesions. Potential side effects include acne, hirsutism, weight gain and unscheduled vaginal bleeding, which may render them unfavourable in some women. Side effects of GnRH analogues include hot flushes, vaginal dryness and sexual dysfunction. In our case, GnRH analogues were used prior to IVF and, after discussion about the options, were chosen again in the puerperium.

The effectiveness of medical and/or surgical ovarian suppression supports the hypothesis of the ovary being a key element in the pathogenesis of the disease.

It has been reported that endometriosis-related ascites has a high risk of recurrence^{5,6} with a 50% recurrence risk reported after unilateral oophorectomy or cystectomy. As a result, long-term follow-up care is important, particularly in patients undergoing medical management due to the high recurrence rates.⁶

Although rare, this case highlights the importance of considering endometriosis in the differential diagnosis of women of reproductive age and African descent presenting with massive haemorrhagic ascites. Where possible, radical surgery consisting

of hysterectomy and bilateral oophorectomy should be avoided as evidence has demonstrated successful resolution in symptoms with medical therapy alone.

Learning points

- ▶ Consider endometriosis as a differential diagnosis in women of reproductive age and African descent presenting with chronic pelvic/abdominal pain, anaemia and haemorrhagic ascites.
- ▶ The long-term management of severe endometriosis associated with recurrent ascites is difficult. Consideration of the patient's age, surgical history and desire for preservation of fertility is important in choosing between medical and surgical management.
- ▶ Endometriosis-related ascites has a high risk of recurrence.
- ▶ Multidisciplinary input including the pain team is essential in providing holistic care.

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Competing interests None.

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