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Life-threatening bleeding secondary to adenomyosis: a case report

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Adenomyosis is a common benign gynaecological disorder. However, it may lead to life-threatening bleeding and complications. We report one such case in a 26-year-old woman who was complicated with disseminated intravascular coagulopathy, shock, and renal failure. She underwent emergency hysterectomy as a life-saving procedure.

Keywords: Adenomyosis; dysmenorrhoea; disseminated intravascular coagulopathy; shock

Case Presentation

In September 2017, a nulliparous 26-year-old woman presented to the accident and emergency department with severe dysmenorrhoea on the first day of her period. She had known history of adenomyosis complicated with menorrhagia and anaemia. At age 23 years, she had been admitted with menorrhagia and anaemia (haemoglobin level of 5.4 g/dL) and had undergone blood transfusion. Ultrasonography showed an enlarged uterus to 14 weeks' size with thickened posterior myometrial wall and cystic spaces in the endometrial cavity. Hysteroscopy showed a 4-cm endometrial polyp, which was confirmed to be benign after polypectomy. She was prescribed cyclical norethisterone but achieved suboptimal control, with multiple admissions for blood transfusions the same year. Injectable medroxyprogesterone acetate also failed to control the severe menorrhagia. In September 2015, she switched to combined oral contraceptive pills (ethinylestradiol and levonorgestrel).

On presentation, her menstrual flow was not heavy and there were no urinary or bowel symptoms. However, the patient was in shock a few hours after admission, with blood pressure decreased to 86/65 mm Hg, tachycardia (119 bpm), tachypnoea, and desaturation requiring 4L oxygen. There were diffuse rhonchi on respiratory examination. Abdominal examination revealed an enlarged uterus to 18 weeks' size. Chest radiograph showed bilateral diffused haziness (Figure 1). Transabdominal ultrasound showed the uterus enlarged to 18 weeks' size with cavity distended with an echogenic shadow measuring 7.92 cm × 8.36 cm, with negative Doppler flow. Both ovaries were not well seen, and there was no adnexal mass or free fluid. The initial diagnosis was adenomyosis with dysmenorrhea and shock with acute respiratory distress syndrome (ARDS).

The amount of vaginal bleeding was not accountable for her haemodynamic instability. Haemoglobin level decreased from 9.1 g/dL to 5.4 g/dL within a few hours. There were features of disseminated intravascular coagulopathy (DIC) with thrombocytopaenia (47 $\times 10^9$/L), and the clotting profile was markedly deranged to an international normalised ratio of 2.8, and the fibrinogen level was 0.3 g/L. She was given double inotropic support with dopamine and noradrenaline and was intubated in the intensive care unit. Transfusion with packed cells and platelet concentrates was started and fresh frozen plasma and cryoprecipitate given. After resuscitation, computer tomographic scanning showed an enlarged uterus of 20 weeks' size with a large haematoma measuring 11.9 × 8.8 × 10.7 cm in the endometrial cavity. There was intra-abdominal fluid with mild dependent density in the pelvis, which could be due to haemoperitoneum or infection. There were bilateral renal cortical necrosis and bilateral lung changes suggestive of ARDS (Figure 2).

In view of continuous drop of haemoglobin level and unstable haemodynamics, emergency hysterectomy and bilateral salpingectomy was performed. Intra-operatively the uterus was distended to 24 weeks in size, bluish, and oedematous; both fallopian tubes were oedematous. Blood loss was 2.85 L, and the patient required intra-operative transfusion of 7 units of packed cells, 8 units of platelet concentrate, 8 units of fresh frozen plasma, and 4 units of cryoprecipitate.

Pathology report showed the uterus weighing 1005 g. Section of the uterus showed foci of haemorrhage

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and congested blood vessels in myometrium. Foci of endometrial glands and stroma in myometrium consistent with adenomyosis.

Postoperatively, the patient was hospitalised for nearly 2 months. Shortly after operation, she developed acute renal failure (with serum creatinine level of 417 μmol/L) and required haemodialysis. She also developed persistent swinging fever. Computed tomographic scans showed an irregular pelvic collection with rim enhancement suggestive of infection. Fever was subsequently resolved after intravenous piperacillin/tazobactam and metronidazole. At postoperative 3 months, her renal function was on gradual recovery and no longer required haemodialysis. She was followed up half-yearly by renal physicians and her renal function was stable.

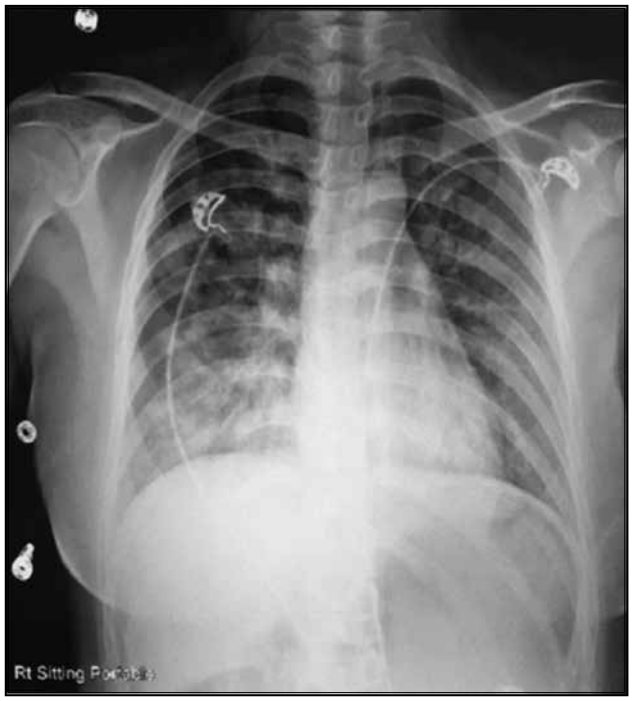

Figure 1. Chest radiograph showing bilateral diffuse haziness suggestive of acute respiratory distress syndrome.

Discussion

Adenomyosis is a benign condition characterised by the presence of ectopic endometrial tissues in the myometrium[1,2]. Patients usually present with dysmenorrhoea, menorrhagia, and anaemia. The diagnosis of adenomyosis requires histology confirmation. Ultrasonographic features suggestive of adenomyosis include echogenic nodules in the myometrium, myometrial thickening, enlarged globular uterus, and increased myometrial vascularity. The management strategy for adenomyosis is mainly symptomatic control. Definite treatment is hysterectomy.

The presentation of our patient is rare. There are few case reports in the literature on life-threatening presentation of adenomyosis. A case report from Taiwan described a patient who presented with exacerbation of menorrhagia and dysmenorrhoea and was later found to have adenomyosis-induced uterine rupture[3]. A case report from Japan described a patient with known adenomyosis who presented with menorrhagia and dysmenorrhea with blood picture showing DIC; she underwent anticoagulant therapy, and magnetic resonance imaging revealed spotty

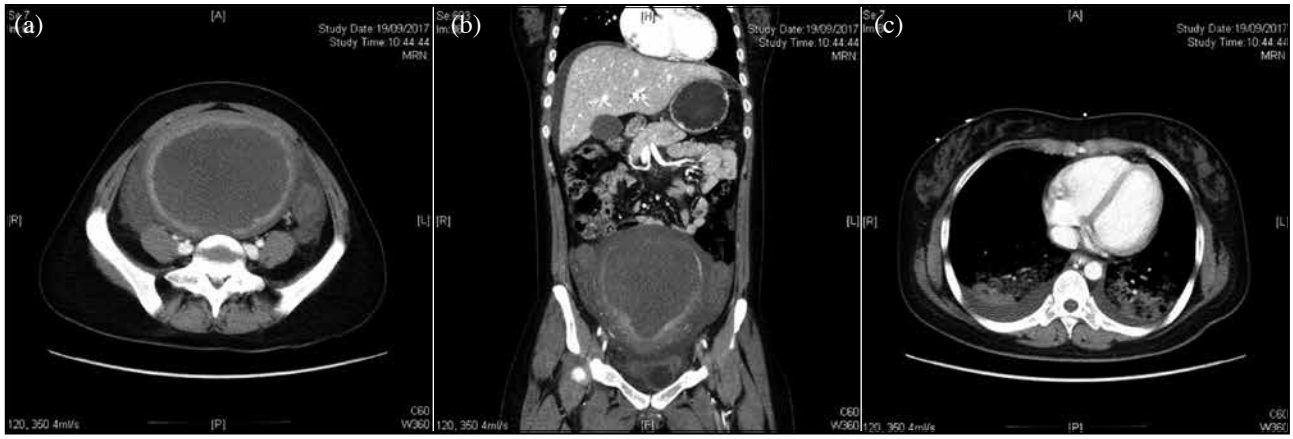

Figure 2. Contrast computed tomographic scans showing (a) an enlarged uterus with grossly distended endometrial cavity, (b) an enlarged uterus with distended cavity, and fluid in the peritoneal cavity in coronal view, and (c) bilateral lung bases with acute respiratory distress syndrome changes.

haemorrhagic lesions in the uterus[4]. A case report in Korea described a patient with known adenomyosis who presented with severe menorrhagia, dyspnoea, anaemia with DIC, and renal impairment; she was treated with hysterectomy and blood product transfusion[5].

It is postulated that ectopic endometrial tissues during menstruation may induce chronic inflammation, intramural haemorrhage, and tissue necrosis[1-3]. The coagulation cascade is activated with consumption of coagulation factors and leads to DIC. Haemorrhage associated with DIC further triggers the consumption of coagulation factors and leads to uncontrolled life-threatening bleeding. Bleeding occurs mainly within the myometrium, leading to the scenario similar to placental abruption[6]. Therefore, the uterus of our patient appeared grossly distended and bluish, like a Couvelaire uterus in placental abruption, but not much vaginal bleeding was observed.

Our case is unique in that the patient also developed ARDS, which was not reported in other case series. The underlying mechanism can be due to haemorrhagic shock, which rapidly induces pulmonary cytokine expression through an oxygen-radical dependent mechanism, leading to the development of widespread inflammation of the lungs and ARDS[7]. The present case is the extreme form of presentation of adenomyosis, and hopefully it can alert physicians that even benign condition can be life threatening.

Declaration

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