Singapore Med J 2012; 53(6) : e114

Spontaneous uterine rupture secondary to recurrent haematometra from cervical stenosis

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ABSTRACT Cervical stenosis is a challenging condition that often recurs despite intervention. Multiple therapeutic options have been described, but a clearly effective and reliable treatment method has yet to be identified. Patients with recurrent stenosis are at risk of developing severe complications such as chronic pelvic pain and infertility. We describe a case of congenital cervical stenosis with secondary haematometra in which repeated cervical dilatation, hysteroscopic canalisation and administration of medications to retard endometrial development were unsuccessful in relieving the obstruction and preventing re-accumulation of menstrual blood. Total hysterectomy was eventually mandated by spontaneous rupture of the haematometra.

Keywords: cervical dilatation, cervical stenosis, haematometra, uterine rupture
Singapore Med J 2012; 53(6): e114–e116

INTRODUCTION

Cervical stenosis may be congenital or could result from cervical trauma, carcinoma or atrophy. Patients typically present with dysmenorrhoea, reduced or absent menstrual flow or abdominal distension from haematometra. Treatment is challenging, as stenosis often recurs despite intervention and may give rise to complications such as infection and infertility. We report a case of congenital cervical stenosis with secondary haematometra in which repeated surgery was unsuccessful. Total hysterectomy was eventually mandated by rupture of the haematometra.

CASE REPORT

A 24-year-old single and nulliparous Chinese woman presented with dysmenorrhoea and abdominal distension, as well as episodes of light inter-menstrual bleeding, particularly during exercise. Her menstrual cycles had otherwise been regular with normal flow since menarche. She had undergone an open adenomyomectomy and endometriotic cystectomy six years ago, after complaining of painful menses, but no other abnormalities had been noted intra-operatively. She had been asymptomatic till several months before this presentation. Clinical examination revealed a 20-week-sized pelvic mass (Figs. 1a & b) that appeared to be a large fundal fibroid with cystic degeneration on imaging.

The patient was planned for an open myomectomy. Intra-operatively, however, her uterus was densely adherent to the Pouch of Douglas and globularly enlarged with a thin-walled fundus. The deficient uterine myometrium was inadvertently breeched during adhesiolysis to reveal a large haematometra. This was drained and the uterine cavity was explored to reveal a stenotic internal os, which was progressively widened from

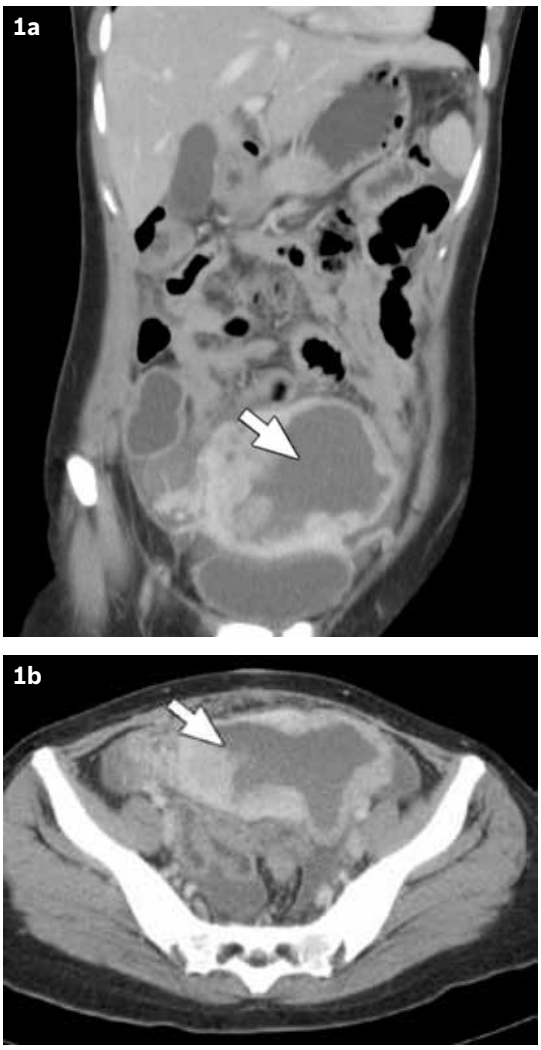

Fig. 1 (a) Axial and (b) coronal CT images show a large haematometra (arrowhead) with a thin uterine fundus at first presentation.

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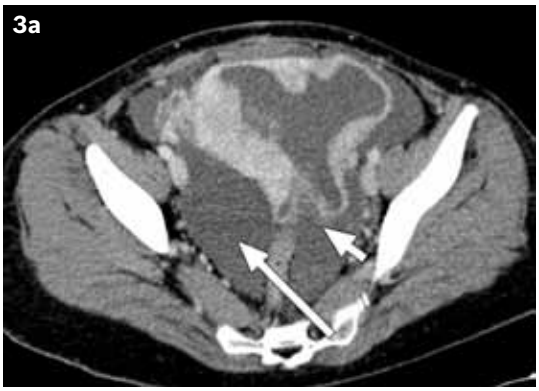
Fig. 2 Hysteroscopic view of the cervical canal during resection.



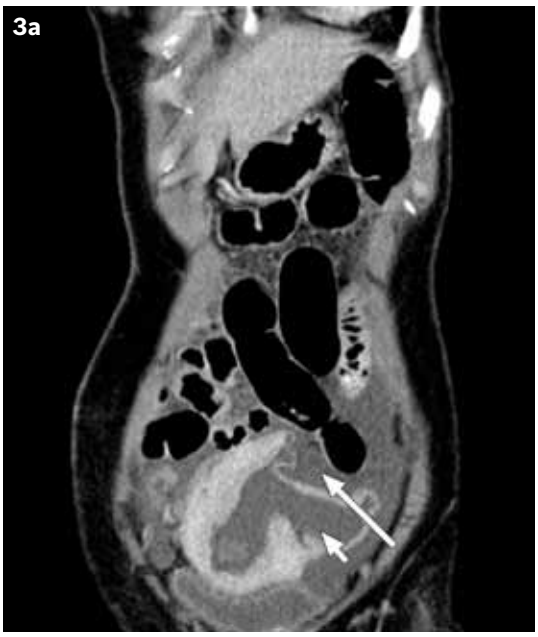


Fig. 3 (a) Axial and (b) coronal CT images show uterine rupture (arrowhead) and the resultant haemoperitoneum (arrow).

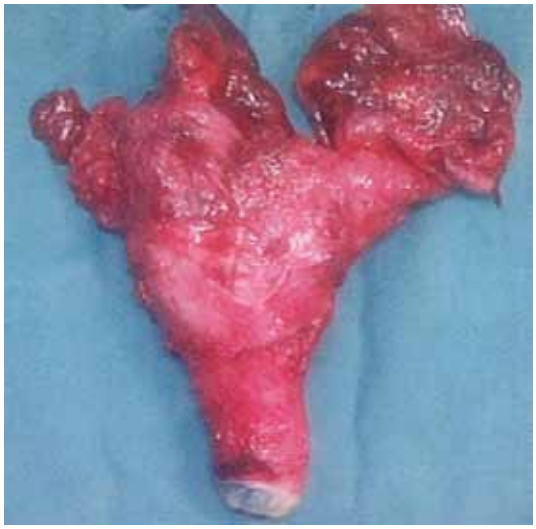


Fig. 4 Photograph shows the hysterectomy specimen, which demonstrated uterine rupture.

above using Hegar dilatators of up to size 12. The uterus was then repaired in two layers.

The patient subsequently remained asymptomatic. However, the stenosis recurred, and follow-up ultrasonography six months later demonstrated significant re-accumulation of blood in the uterine cavity. Cervical dilatation under general anaesthesia was performed vaginally using Hegar dilators of up to size 12 in order to enable passive drainage of the retained blood. A large 20-week-sized haematometra was again evident five months after this procedure. Cervical dilatation was repeated, this time with Hegar dilators of up to size 16. The patient was also given a dose of 11.25 mg leuprorelin acetate to induce amenorrhoea, and a Mirena intra-uterine system (IUS) was inserted to maintain an atrophic endometrium. These measures were undertaken as it was felt that the obstruction was not complete and that effective reduction of menstrual loss might prevent re-accumulation of the haematometra. Unfortunately, they only succeeded in slowing the re-accumulation of blood, and the haematometra recurred, with the patient eventually requiring re-drainage one year later.

During this third attempt, hysteroscopic resection of the upper cervical canal (Fig. 2) was also performed in an attempt to create a patent channel for menstrual flow. The patient was additionally given 150 mg of intramuscular medroxyprogesterone acetate after surgery and another dose three months later. However, a re-accumulation of the haematometra to 16-weeks size was again observed after six months.

The patient opted for conservative management with another dose of medroxyprogesterone acetate instead of repeat dilatation, as she remained comfortable and asymptomatic. However, she presented the following month with acute central abdominal pain that was preceded by a 'popping' sound. Although her abdomen was tender and guarded, she remained haemodynamically stable. Ultrasonography and computed tomography (Figs. 3a & b) demonstrated a large haemoperitoneum secondary to uterine rupture.

The extent of rupture and the severely deficient myometrium meant that achieving adequate repair to enable future reproductive success was unlikely, while the failure of multiple previous attempts at conservative surgery indicated that the risk of recurrent haematometra with repeat uterine rupture eventually necessitating hysterectomy was high and also carried with it the possibility of life-threatening haemorrhage. In addition, the patient's fertility prospects were anticipated to be poor due to endometrial and tubal damage resulting from back pressure

from a chronically distended uterine cavity, while the extremely thin endometrium put the patient at risk of uterine rupture in the event that a successful pregnancy was achieved. In view of these considerations and also because the patient was keen for a definitive cure as her lifestyle had been significantly disrupted by repeated hospital admissions and surgical procedures, we discussed with the patient and her family the option of a total hysterectomy, and they agreed to the procedure.

During the laparotomy, the patient's uterine fundus was found to have ruptured, resulting in the release of altered blood into the peritoneal cavity (Fig. 4). A total hysterectomy and adhesiolysis was done; histology of the uterine specimen showed extensive adenomyosis. The patient recovered well postoperatively and was coping well psychologically at the time of this writing.

DISCUSSION

Cervical stenosis can be congenital or acquired and complete or partial. Patients with incomplete cervical stenosis may initially be asymptomatic, as the cervical canal still allows partial drainage of menstrual blood. Our patient had regular cycles with seemingly normal menstrual flow from menarche till she presented in her early twenties with dysmenorrhoea, abdominal distension and intermenstrual bleeding, and she had by then already developed a sizeable haematometra. This collection was likely to have gradually accumulated since puberty and subsequently worsened with the development of uterine adenomyosis. Patients with cervical stenosis are also more prone to endometriosis due to retrograde flow of menstrual blood into the pelvic cavity:[1] our patient had an endometriotic cyst and severe endometriotic adhesions, with the uterus densely adherent to the bowel loops and the Pouch of Douglas.

The management of cervical stenosis is challenging, and no clearly effective and reliable treatment is currently available. The methods previously described in the literature include cervical dilatation, stent insertion to maintain cervical patency, hysteroscopic canalisation and administration of medications to retard endometrial development.[2-4] Cervical dilatation is performed using Hegar dilatators under antibiotic cover in order to reduce the risk of endometritis. The cervix may be primed with misoprostol or laminaria, and dilatation is done under ultrasonography guidance to reduce the risk of perforation. However, as illustrated by our case, recurrence after dilatation is common and may occur as soon as a few months after the procedure. This has prompted several authors to propose the insertion of stents,[2,3] pessaries[5] and catheters[6] to maintain a continuous expansive force on the cervix. These devices have been reported to achieve good long-term outcomes, but may get dislocated or increase the risk of uterine infection. Other authors have described the successful utilisation of Interceed,[7] an absorbable adhesion barrier, in preventing recurrent stenosis. Evidence for this method is, however, scanty and limited to case reports, and it may indeed be more effective when the recurrent stenosis is due to adhesions rather than a true narrowing.

Hysteroscopic canalisation involves using a hysteroscopic loop resector to refashion stenotic portions of the cervical canal. This has been reported in a small case series to be successful in enabling embryo transfer in women with cervical stenosis undergoing *in-vitro* fertilisation.[4] It did not, however, adequately relieve the obstruction or prevent haematometra reformation in our patient.

Hormonal medications can be used as an adjunct to the methods described above to reduce endometrial thickness and prolong the interval between cervical dilatations. The combined use of leuprorelin acetate and the Mirena IUS succeeded in delaying re-accumulation of haematometra in our case, but the therapeutic effect of the Mirena IUS was likely to have diminished as our patient's uterine cavity gradually distended with blood.

In conclusion, treatment of intractable cervical stenosis remains a therapeutic challenge, yet failure to relieve the obstruction can result in severe complications such as endometritis, chronic pelvic pain, infertility and spontaneous uterine rupture. Further studies are needed to determine the effective methods for treating this difficult condition.

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