CASE REPORT

Novel management of a giant cervical myoma in a premenopausal patient

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SUMMARY

Cervical myomas are rare and account for <1% of uterine leiomyomas. Clinical complications include bulk-related symptoms of pelvic pain, abnormal bleeding and infertility. While hysterectomies may be readily performed for postmenopausal women; the management of women with cervical myomas of childbearing age remains a challenge.

Traditionally described fertility-preserving procedures such as myomectomy, endometrial ablation or myolysis may only be applied to leiomyomas within the uterine corpus. Little is known about the surgical management of its cervical counterpart.

Radical abdominal trachelectomy has been described as a potential fertility-preserving procedure in the management of women with early cervical cancer. As such, we present a case of a giant cervical myoma that was treated with an abdominal trachelectomy in an attempt to preserve fertility.

BACKGROUND

Uterine leiomyomas are the most common pelvic tumours in women. However, cervical myomas, which are leiomyomas arising from the cervix rather than the uterine corpus, are rare, with a reported incidence of <1%.¹ Risk factors for the development of cervical tumours are not dissimilar to their corpus counterparts and include a combination of genetic predisposition, environmental factors and exposure to steroid hormones. According to the International Federation of Gynaecology and Obstetrics (FIGO) classification, myomas are classified based on their location within the uterus: intramural myomas are FIGO types 3–5, submucosal are types 0–2, subserosal are types 6 and 7 while cervical myomas are type 8.

Surgery remains the mainstay of treatment for most leiomyomas. Indications for surgical intervention include tumour-related bulk symptoms, abnormal uterine bleeding or recurrent pregnancy loss and infertility. In postmenopausal women, hysterectomy is the procedure of choice as it eliminates both symptoms and the possibility of future recurrences.² In women of childbearing age who wish to preserve fertility, myomectomies may be performed; however, it is only applicable to patients with FIGO 1–7 myomas which originate from myometrial cells within the uterine corpus.³ The management of FIGO 8 cervical myomas in women who would like to preserve fertility remain a challenge.

Radical abdominal trachelectomy has been described in the management of selected premenopausal patients with early cervical cancer in an attempt to maintain reproductive potential.⁴ Reported rates of successful conception post trachelectomy range from 30% to 66%.^{4–6} Though potential obstetrics complications include early miscarriages and preterm labour, 65% of pregnancies result in a healthy term infant.

Here, we present a case of a giant symptomatic cervical myoma where abdominal trachelectomy was performed in an attempt to preserve fertility.

CASE PRESENTATION

Mdm CW is a 36-year-old, gravida 1, para 1, premenopausal Chinese woman with no medical nor surgical history. She is a non-smoker, non-drinker with body mass index of 22 and no family history of cancer.

Mdm CW presented with a 3-month history of increasing abdominal distension. She denied abdominal pain, symptoms of intestinal obstruction or constitutional symptoms. There was also no history of menstrual-related complaints such as dysmenorrhoea or menorrhagia; menarche occurred at the age of 12 years and menstrual cycle was described as regular with no prolonged bleeding. Her first pregnancy was at the age 32 years, and was non-eventful with a normal vaginal delivery at a fetal gestational age of 38 weeks.

On examination, Mdm CW was not cachectic, had no pallor and was well hydrated. There was a large 10 cm mass that arose from the pelvis and extended to the epigastric region. The mass was firm in consistency, had a smooth outline and was not ballotable. It did not appear to fix to the overlying skin. The abdomen was not tender and there was no ascites. Transvaginal examination did not reveal any cervical nor vaginal masses. Digital rectal examination was unremarkable.

INVESTIGATIONS

Transvaginal and transabdominal ultrasound studies performed found a 10×6×20 cm hypoechoic mass containing areas of cystic necrosis, internal vascularity and calcification. In view of its large size, MRI was performed to better characterise the mass as well as its anatomical relationship with surrounding structures.

MRI revealed a large 20×10 cm intra-abdominal tumour arising from the pelvis (figures 1 and 2). The mass was seen to be either arising from the

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Figure 1 Pelvic mass with possible origin from the uterus and displaced normal adnexa. LO, left ovary.

uterus or was invading into the uterus. The bilateral ovaries were displaced but appeared normal. In addition, the mass had a characteristic whorled appearance and appeared hypointense on T1-weighted images and hyperintense on T2-weighted image with variable enhancement. The bowel loops were displaced to the upper abdomen but there were no signs of invasion from the mass.

CT scan of the chest did not reveal any lung lesions.

Serum tumour markers were performed and her Ca125 was 10.6 U/mL (normal <35.1 U/mL), CEA was 1.9 UG/L (normal <5.2 UG/L) and Ca19-9 was 15.1 U/mL (normal <34.1 U/mL).

DIFFERENTIAL DIAGNOSIS

1. Giant leiomyoma

Figure 2 Coronal view of the pelvic mass.

Figure 3 Whorled appearance of mass with displaced bowel loops. SMA/SMV, superior mesenteric artery/vein; RO, right ovary.

2. Angiomyxoma: in view of whorled appearance on MRI imaging (figure 3).

TREATMENT

In view of the imaging findings, Mdm CW was counselled for surgery and possible hysterectomy. She underwent an exploratory laparotomy, and intraoperatively, a large tumour was seen arising from the pelvis with a stalk originating from the uterine cervix (figure 4). It was highly vascular and was adherent to but not invading the uterine corpus and bladder. There was no

Figure 4 Large pelvic mass 19×10 cm arising from the cervix.

Table 1 Summary of current literature on cervical myomas

Study	Year	Study type	Presenting complaint	Investigations	Management
1 Goel and Seth (India) ⁷	2016	Case report	40-year-old G2P2: abdominal distension	US 12×10 cm pelvic mass	Total hysterectomy, left salpingo-oophorectomy
2 Peng <i>et al</i> (Taiwan) ⁸	2016	Case report	42-year-old G2P2: urinary frequency	US & Computed topography (CT)10×7 cm pelvic mass	Cervical myomectomy
3 Keriakos and Maher (UK) ⁹	2013	Case report	29-year-old nulliparous: obstructed labour	US 10×12 cm polypoidal cervical lesion	Emergency caesarean section (LSCS) followed by vaginal cervical myomectomy
4 Kamra <i>et al</i> (India) ¹⁰	2013	Case report	28-year-old nulliparous during pregnancy: no symptoms	Antenatal US 5 cm cervical lesion	Vaginal myomectomy at 8-week fetal gestational age.
5 Ikechebelu <i>et al</i> (Africa) ¹¹	2013	Case report	37-year-old G3P3: presenting protruding vaginal mass	CT 24×15 cm mass with vaginal & intra-abdominal components	Vaginal myomectomy No treatment of intra-abdominal leiomyoma
6 Tian and Hu (China) ¹²	2012	Case series (n=17)	Mean age 32 (range 28–41 years) During antenatal screening: 75% obstructed labour	Antenatal US Mean size 10.4±7.2 cm	94.1% (n=16) emergency LSCS 1 Normal vaginal delivery 56.3% (n=9) Myomectomy 18.8% (n=3) hysterectomy 25% (n=4) No treatment
7 Chang <i>et al</i> (Taiwan) ¹³	2010	Case series (n=28)	Dysmenorrhoea/hypermenorrhoea (75%); urinary frequency (36%); tenesmus (29%); infertility (22%)	US No reported size	Laparoscopic cervical myomectomy Mean myoma weight 2.8 kg (range 0.3–12)
8 Matsuoka <i>et al</i> (Japan) ¹⁴	2010	Case series (n=16)	Not reported	US No reported size	Laparoscopic cervical myomectomy Mean myoma weight 2.08 kg (range 0.9–3)
9 Del Priore <i>et al</i> (UK) ¹⁵	2010	Case series (n=3)	All had lower abdominal pain	CT & MRI All had large pelvic mass cervix by hydroureters (1 reported 18 cm)	Radical abdominal trachelectomy

LSCS, lower segment caesarean section; US, ultrasound.

peritoneal disease and the ovaries were normal. Intraoperative frozen section was that of a spindle cell neoplasm with degenerative changes likely smooth muscle in origin, with no overt cytological atypia.

Excision of the tumour without transection of the cervix was not possible in view of its broad-based attachment to >70% circumference of the cervical neck. As such, resection of the tumour with trachelectomy was performed. A Foley's catheter was placed transvaginally through into the remnant uterus across the anastomosis.

OUTCOME AND FOLLOW-UP

Mdm CW had an uneventful postoperative recovery and was discharged on the fifth post-operative day (POD). The foley's placed across the anastomosis was remove on the POD4. The final histology of the pelvic tumour was that of a 20 cm leiomyoma arising from the uterine cervix. There was no excess mitotic activity, nuclear atypia or necrosis.

DISCUSSION

Leiomyomas of the uterine cervix are uncommon and present both a diagnostic and therapeutic challenge. The risk factors for its development are not dissimilar to its uterine corpus counterparts though little is known with regards to the pathogenesis of cervical myomas and the reason for its significantly lowered frequency.¹ In light of its rarity, studies published in this area have been limited to retrospective case reports and series (table 1). Presenting symptoms are often vague and non-specific, but are most commonly a result of mass effect on the surrounding pelvic organs. Common complaints include abdominal distension, lower abdominal pain, urinary frequency and tenesmus,^{7–15}

while menstrual-related complaints are rare. This differs from FIGO type 0–5 myomas where symptoms of abnormal uterine bleeding predominates.^{3 16} As such it was not surprising that our patient reported no menstrual-related symptoms and only had lower abdominal pain and distension.

In view of the lack of menstrual symptoms, it is not surprising that cervical myomas grow to large sizes before detection. Mean size in reported series was 13 cm (range 5–24 cm).^{7–15} Differentials for a large pelvic mass originating from the uterus in a premenopausal female patient will include:

1. Pregnancy
2. Uterine corpus lesions: benign (leiomyomas, adenomyosis and polyps) or malignant (leiomyosarcoma, endometrial stromal sarcomas, carcinosarcoma and endometrial carcinoma)
3. Uterine cervix lesions: benign (leiomyomas) or malignant (cervical carcinoma)

Ultrasound pelvis is the most commonly used imaging modality. However, once pregnancy has been excluded, a CT scan or MRI may then be performed to further characterise the mass. This is especially important when a sarcoma is suspected. Though the presence of mixed echogenicity, central necrosis and irregular vascular distribution on sonography may suggest a malignant lesion, up to 20% of large fibroids possess similar features. MRIs though unable to provide a definitive diagnosis of uterine sarcomas, have a higher specificity and positive predictive value than their sonographic counterparts.¹⁷ Cervical leiomyomas arise from the cervix and appear as hypointense masses that enhance homogeneously on T2-weighted MRI images.¹⁸ CT or MRI also provide further information with regards to the relation of the mass to the

surrounding pelvic organs; this is imperative for future surgical management.

Most women with cervical myomas are at a reproductive age. Mean age at diagnosis was 34 years (range 29–42).^{7–12} While hysterectomy may be the surgical procedure of choice for large symptomatic fibroids, it is not ideal in management of female patients who desire to preserve their fertility. Laparoscopic cervical or transvaginal/abdominal myomectomies have been described,^{8 9 12–14} however their utility are often limited to small myomas <10 cm in size or myomas which involve only part of the uterine cervix, for example, a predominantly anteriorly or posteriorly based lesion. Furthermore, myomectomies risk future recurrences in view of the remnant cervix left behind.

Radical trachelectomy in an attempt to preserve fertility has been described in the management of cervical carcinoma.^{5 6} Okugawa *et al* reported a successful pregnancy rate of 25% post-trachelectomy in a series of 151 patients.⁴ To date, this procedure has only been performed in benign symptomatic cervical lesions in three patients.¹⁵ In this series, Del Priore *et al* described three young patients with huge cervical leiomyomas which were complicated by symptoms of lower abdominal pain, distension and imaging findings of severe bilateral hydroureters. Though myomectomy was planned, due to the large size of the lesions, patients were all counselled for the possibility of hysterectomy. The trachelectomy that was performed was unplanned and was in fact a novel technique adopted intraoperatively as a salvage attempt to preserve fertility. Similarly, Mdm CW had a huge symptomatic 20 cm leiomyoma for which excision and possible hysterectomy was counselled. Intraoperatively, on transection of the cervix from the upper vaginal canal, decision was made for primary anastomosis of the remnant uterus to the upper vagina. Uterine vessels were preserved. We believe that this novel method may be widely adopted in benign cervical lesions in women of childbearing age and should be considered as an alternative to upfront hysterectomy.

In patients who successfully conceive post-trachelectomy, a short residual cervical stump predispose to higher rates of first and second trimester miscarriage as well as preterm delivery.¹⁹ Prophylactic cerclage performed at the time of vaginal trachelectomy has been described and is aimed at ‘strengthening’ the cervix to reduce the risks of future perinatal events.²⁰ In Mdm CW, cerclage was not performed at the primary surgery but should have been considered.

Learning points

- Cervical myomas represent a rare subtype of uterine leiomyoma.
- When symptoms develop, hysterectomy is the procedure of choice in postmenopausal women.
- Management of premenopausal women with cervical myomas is a challenge.
- Abdominal trachelectomy may have a role in the management of clinically significant/symptomatic cervical myomas as a fertility preserving procedure.

To conclude, cervical myomas are rare but often present in women of a reproductive age. Abdominal trachelectomy should be considered as a management strategy in premenopausal females who wish to preserve fertility.

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