Case report

Perioperative transcatheter embolisation of a locally recurrent metastatic uterine leiomyoma

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Abstract

Introduction
We present an unusual case of large recurrent invasive pelvic leiomyoma 14 years after hysterectomy and 12 years after treatment of pelvic benign recurrence. Embolisation immediately after pelvic resection was performed, which we believe is the first reported case of such treatment.

Case report
A 55-year-old black female presents with recent development of obstruction and decreased calibre of stool. On current admission, physical examination showed a soft and non-tender abdomen, fullness in the left lower quadrant and clear lungs. Emergent angiography and embolisation was performed. Post-operative course was unremarkable.

Conclusion
To our knowledge, this is the first reported case of angiography and embolisation involving extra-uterine and recurrent metastasizing leiomyomas.

The rarity of these lesions would obviously preclude attempting a prospective clinical trial of pre-operative embolisation for the purposes of either reducing intra-operative blood loss or entirely avoiding surgical resection.

Introduction
Benign metastasizing uterine leiomyoma (BML) is a rare entity representing extra-uterine foci of leiomyomatous lesions of variable size and locations and variable delay in presentation after initial diagnosis of uterine fibroids. The nature of benign metastasizing leiomyoma has been debated since it was first reported in 1939, and only a few dozen cases have been reported in the English literature. Recent findings at the proteomic and genomic level demonstrate that the metastatic lesions are clonally related to the primary uterine fibroids and suggest that their pathogenesis relies on metastatic spread and not on simultaneous multifocal appearance. Therefore, these tumours could be regarded as borderline tumours or tumours with low malignant potential. Due to the rarity of these cases, there is currently no consensus on their management. The high expression of oestrogen (ER) and progesterone (PR) receptors supports the use of hormonal therapy.

Surgical intervention can be challenging due to the recurrent, often disseminated nature of these tumours and their aberrant vascularity. Over the last few decades, uterine artery embolisation has emerged as an established treatment for primary uterine fibroids. This case suggests that angiography and embolisation should be considered in the management of the patients with BML in an attempt to avoid surgical intervention or decrease the risk for perioperative bleeding.

Case report
A 55-year-old black female, Gravida 2, Para 0, presents with a recent development of obstruction and decreased calibre of stool. There is a history of total abdominal hysterectomy in 1997 for benign uterine fibroids. Repeat surgery in 1999 was performed for local pelvic recurrence.

Surgery involved resection of several pelvic and mesenteric masses, the largest measuring 11 cm, uninvolved bilateral ovaries and fallopian tubes, a 10 cm segment of adherent but otherwise normal colon and several pelvic lymph nodes, one of which was involved pathologically. Pathology of all lesions was consistent with leiomyoma, all of which showed cystic degeneration. ERs and PRs were both highly expressed (>90%). There was very minimal focal cytologic atypia and no evidence for leiomyosarcoma (Figure 1). The diagnosis of benign metastasizing leiomyomas was made in 1999. On current admission, physical examination showed a soft and non-tender abdomen, fullness in the left lower quadrant and clear lungs. Relevant laboratory findings include haemoglobin 10.7 g/dl, haematocrit 33.1, platelets 266,000/ml, WBC count 3340/ml, BUN 12 mg/dl and Cr 1.2 mg/dl. MRI showed two large pelvic masses (Figure 2). A 5 × 8 cm mass adjacent to the aortic bifurcation elevated the right common iliac artery (Figure 2a) and a 9 × 15 cm mass in the mid-to-left anterior lower pelvis displaced the bladder and rectosigmoid with no sign of invasion (Figure 2b). There was mild right hydrourerter with normal renal size and cortical thickness and severe left hydrourerter with thinned parenchyma due to unilateral chronic obstruction. Chest radiograph was unremarkable. The patient was taken for surgical resection that included lysis of adhesions, debulking of the larger inferior mass and repair of small bowel injury. There was significant haemorrhage from the larger...
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Table 1: Magnetic resonance imaging (MRI) finding of the left pelvic area.

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<th>MRI Findings</th>
<th>Description</th>
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<td>Lesion 1</td>
<td>A 9×15 cm pelvic mass in the mid-to-left anterior lower pelvis displacing the bladder and rectosigmoid.</td>
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<tr>
<td>Lesion 2</td>
<td>A 5×8 cm pelvic mass adjacent to the aortic bifurcation.</td>
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Discussion

Extra-uterine leiomyomas are rare lesions. The clinical spectrum includes benign metastasizing leiomyomas, disseminated peritoneal leiomyomatosis and intravenous leiomyomatosis.

Collaterals from the left external pudendal artery off the deep femoral artery which course medially and superiorly. The right internal iliac artery does not appear to feed either of the two lesions. The middle sacral, left internal iliac and medial segment of the left external pudendal artery were all embolised to proximal occlusion with embospheres (tris-acryl gelatin microspheres, Merit Medical, S. Jordan, Utah) 300–500 micron followed by 500–700 micron size. Post-embolisation angiogram (Figure 3b) shows complete occlusion of the distal middle sacral, proximal left internal iliac and distal aspect of the medial segment of the left external pudendal arteries; absence of flow to the bifurcation lesion and almost complete resolution of the left pelvic region of hypervascularity. The patient was returned the following day for removal of abdominal packing, which was uneventful, with no sign of further haemorrhage. Post-operative course was unremarkable.

Figure 1: Benign metastatic leiomyoma without evidence of leiomyosarcoma. Only very minimal focal atypia is seen. Mitotic figures are rare, no more than 1/10 HPF. No coagulative necrosis (haematoxylin and eosin, 400×).

Figure 2: MRI pelvis. (a) A 9×15 cm pelvic mass in the mid-to-left anterior lower pelvis displacing the bladder and rectosigmoid. (b) A 5×8 cm pelvic mass adjacent to the aortic bifurcation.

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Benign metastasizing leiomyomas involve numerous tumours, usually in patients post-hysterectomy with identical pathology to the original uterine fibroid. The most common location is the lungs, and the time from hysterectomy to the presence of lung nodules has been reported to be from 3 months to 20 years. Involvement of the heart, brain, bones or lymph nodes has also been described.

A wide spectrum of clinical manifestations has been reported depending on the location of the lesions. While some metastatic myomas are discovered incidentally, symptoms such as dyspnea and cough from lung lesions and constipation, abdominal pain and urinary retention from pressure to adjacent abdominal or pelvic organs have been reported.

To our knowledge, this patient never had lung involvement. The diagnosis of benign metastasizing leiomyomas was made at the second operation with the presence of lesions in the pelvis, mesentery and regional lymph nodes. There was no evidence of peritoneal implants or leiomyomatosis and likewise no venous invasion. Several theories have been proposed to explain the pathogenesis of these lesions. Possible mechanisms for the development of metastatic leiomyomas include overgrowth of smooth muscle tissue under the effect of hormones or growth factors and embolisation of mesenchymal cells and growth in distant sites. Some authors consider the metastatic myomas low-grade leiomyosarcomas with malignant potential.

Given the unclear pathophysiology of this pathologic entity and the multiple factors that are implicated, several treatment modalities have been proposed and an optimal management strategy has not yet been defined. Several authors discuss the use of drug therapy, in the form of selective ER modulators (Tamoxifen, Raloxifen), aromatase inhibitors (Anastrozole), GnRH agonists and hormones (progesterone), as treatment for BML. However, these have usually been performed in the presence of lung lesions and pelvic recurrence within a short time interval post-hysterectomy and were not considered in this case. The presence of bowel symptomatology, the large size of the lesions and the need to obtain pathologic diagnosis were the reasons to proceed directly to surgical treatment.

Transcatheter embolisation is a well-established treatment for the presence of symptomatic uterine fibroids. Uterine fibroid embolisation results in significant decrease in the size of lesions and improvement in symptomatology in the vast majority of patients.

**Conclusion**

To our knowledge, this is the first reported case of angiography and embolisation involving extra-uterine and recurrent metastasizing leiomyomas.

The rarity of these lesions would obviously preclude attempting a pro-

**Figure 3:** (a) Initial pelvic aortogram demonstrates the well-delineated hypervascular mass at the aortic bifurcation and an area of ill-defined hypervascularity at the left inferior lateral pelvis. (b) Post-embolization angiogram shows complete occlusion of proximal left internal iliac, distal middle sacral and absence of flow to the bifurcation lesion and decreased vascularity in the left pelvis.

Broad ligament leiomyoma is a separate entity where serosal uterine fibroids adhere to the broad ligament, detach and parasitise an auxiliary blood supply.

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spective clinical trial of pre-operative embolisation for the purposes of either reducing intra-operative blood loss or entirely avoiding surgical resection. However, based on the impressive angiographic results, this option may be considered in patients subsequently presenting with similar findings. There also may be a consideration in selected future cases for a combination of embolisation and drug therapy for the purpose of causing shrinkage of recurrent pelvic lesions in patients that do not need require urgent surgical resection.

Abbreviations list
BML, benign metastasizing uterine leiomyoma; ER, oestrogen receptor; PR, progesterone receptor

References