

CASE REPORT

An Unusual Case of Leiomyoma - Controversies in Management

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ABSTRACT

A 48-year-old nulliparous woman was referred to Gynaecology for a suspected gynaecological cancer. She presented with unexplained abdominal symptoms, iron deficiency anaemia and a large pelvic mass. Her menstrual cycle was described as heavy and regular. She had a normal cervical smear history and was in the perimenopausal stage of her life. There was no reported history of weight loss. Her past gynaecological and surgical history included a previous ovarian cystectomy and appendicectomy many years ago. There was no other significant medical or family history. On examination she had a large fibroid uterus. Ultrasound scan was suggestive of a large multi-loculated pelvic mass, but Ca125 was in the normal range. The pelvic mass was considered benign and she underwent a surgical treatment with total abdominal hysterectomy and bilateral salpingo-oophorectomy. During the surgery a distended fluid filled uterus was noted, with an appearance typical of a pregnant uterus. Histological examination confirmed a bizarre, symplastic leiomyoma of the uterus.

Keywords: Pelvic mass, Symplastic uterine fibroids, Malignant transformation

INTRODUCTION

Leiomyomas are common, benign smooth muscle tumours (fibroids) of the female genital tract. They are rare before the age of 20, and regress after the menopause.¹ Leiomyoma usually grows slowly, and often are asymptomatic, however, large symptomatic leiomyomas may need to be removed surgically. Malignant transformations of leiomyomas are rare and those with low mitotic activity, and lacking nuclear atypia have little or no malignant potential.¹ A bizarre, symplastic leiomyoma is a rare histological variant of a leiomyoma.²

This case demonstrates an unusual case of leiomyoma, and illustrates the difficulties associated with establishing a clinical and radiological diagnosis, and the consequent impact on a

patient's journey from symptoms to diagnosis. It also highlights the importance of histological examination in reaching a final diagnosis of bizarre, symplastic leiomyoma.

Case Report

A 48-year-old nulliparous woman was referred to the gynaecological department for a suspected pelvic malignancy. She initially presented to her GP complaining of shortness of breath, iron deficiency anaemia and persistent unexplained abdominal symptoms. This led to a significant impact on her work, as she was experiencing difficulty in undertaking manual work related responsibilities, which resulted in her being absent from work. Her menstrual cycle was described as heavy and regular, but there was no report of intermenstrual or postcoital bleeding. She had a normal cervical smear history and was in the perimenopausal stage of her life. There was no reported history of weight loss. Her past gynaecological and surgical history included a previous ovarian cystectomy and appendicectomy many years ago. There was no other significant medical or family history. Clinical examination was suggestive of a large fibroid

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uterus/pelvic mass. An abdominal and pelvic ultrasound scan that was arranged by her GP, demonstrated a 13 cm midline complex cystic mass, (**Fig 1**).



Figure 1 Ultrasound image of complex cystic pelvic mass

The origin of that complex cystic mass, (**Fig 2**) was unclear, though it was likely to be either a uterine or ovarian tumour. The endometrium appeared thickened measuring 1.5 cm in diameter. The patient was clearly very anxious with the findings as expected. A repeat ultrasound scan in the gynaecology department was suggestive of a large multi-loculated ovarian cyst, (**Fig 3**), nonetheless, a Ca125 was within the normal range.



Figure 2 Ultrasound image of complex cystic pelvic mass



Figure 3 Ultrasound image of a multi-loculated suspected ovarian cyst

Clinically and radiologically the impression was that of a benign mass, and therefore the patient was counselled and reassured accordingly. A total abdominal hysterectomy and bilateral salpingo-oophorectomy was discussed in view of her symptoms and provisional diagnosis. Thereafter the patient sought a second opinion from her private gynaecologist who also recommended the same surgical treatment. Although the clinical examination and ultrasound scans suggested either a uterine or pelvic mass, surgery was necessary to confirm the origin of that mass, and histology was of paramount importance in reaching the final diagnosis.

The patient returned to the gynaecological department to proceed with the recommended treatment. She had an uncomplicated total abdominal hysterectomy and bilateral salpingo-oophorectomy. During the surgery, a soft, distended, eighteen weeks' uterus, typical of a pregnancy was found. However, it was filled with a litre of clear fluid requiring drainage prior to hysterectomy. Following the drainage of the uterine fluid, the uterus measured approximately 11.5 cm size on gross histological examination. A small defect was noted on the serosal surface of the uterus. The endometrial cavity contained a small endometrial polyp, but there were no other lesions identified in the endometrial cavity. The endometrium was thin. There were several intra-mural yellowish – walled nodules measuring up to 1.3 cm. In the wall of the uterus was a large cystic area measuring 6 x 3 x 2 cm, but this did not communicate with the endometrial cavity. The cervix and both fallopian tubes and ovaries were normal.

Microscopically, there was no evidence of endometrial hyperplasia or malignancy. Similarly, ovaries, fallopian tubes, parametria and cervix were unremarkable. There were several unremarkable leiomyomas composed of fascicles of bland spindle cells without conspicuous mitotic activity. Sections from the cystic mass in the uterus showed highly pleomorphic spindle cells arranged into loose fascicles, but there were no signs of necrosis and mitotic activity was inconspicuous. The features were in keeping with a symplastic /bizarre leiomyoma that had undergone cystic degeneration. In some sections the atypical cells appeared to be adjacent to areas of more typical leiomyoma. The slides were sent to a tertiary centre for a second opinion and a final diagnosis confirmed bizarre, symplastic leiomyoma of the uterus.

The patient made a good post-operative recovery. The final histological diagnosis was discussed with the patient in writing and during a face-to-face consultation at 6 weeks' follow up clinic. According to multidisciplinary team no further follow up was deemed necessary.

Discussion

Smooth muscle tumors (leiomyoma) represent the most common group of uterine mesenchymal neoplasms. Although leiomyoma does not usually cause a diagnostic challenge for the clinician, yet its histological variations must be understood in order to reach a final diagnosis, and to differentiate it from its malignant counterpart, leiomyosarcoma.³ This knowledge is clearly essential, in providing adequate patient counselling and

alleviating patient anxiety associated with the diagnosis, treatment, and follow up.

Symplastic leiomyoma is a rare histological variant of uterine leiomyoma² characterized by nuclear atypical tumour cells with low mitotic counts and no coagulative tumour cell necrosis on microscopic examination.^{4,5,6} Atypical leiomyoma is differentiated from leiomyosarcoma by a lack of necrotizing tumour cells and a mitotic count <7 per 10 high power fields. Nuclear atypia makes the difference with mitotically active leiomyoma.⁷

Although symplastic leiomyomas are benign,⁸ it is more likely to undergo malignant transformation compared to uterine leiomyomas; as a result, hysterectomy is often the recommended treatment.⁹ Although our patient was nulliparous, fertility was not her main priority. Also she was in the perimenopausal stage of her life. A diagnosis of symplastic leiomyomas in a younger premenopausal nulliparous woman may cause a therapeutic dilemma, particularly in those wanting a more conservative approach. Montgomery et al described 3 cases of conservative treatment where a more conservative approach was supported; two patients had hysteroscopic resection of fibroid and one had a laparoscopic myomectomy. Diagnosis of symplastic leiomyoma was made later on histological specimens. All patients were followed up with imaging but no recurrence was suspected.¹⁰ However, the best management in women wanting to preserve their fertility is controversial as the exact risk of malignancy and recurrence risk remains unclear in literature.

Conclusion

Although Leiomyoma is a common presentation to the gynaecologist, a bizarre, symplastic leiomyoma is a rare histological diagnosis. Symplastic leiomyomas are benign and malignant transformation is rare, therefore the final treatment must be individualised, taking into account a patient's desire for fertility, co-morbidities and menopausal status. Multidisciplinary input is an important aspect of decision-making.

Consent of the patient: Obtained

Conflict of interest: None

Contribution of Authors: The 1st author wrote the case report and the co-authors reviewed it.

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