## CASE REPORT

# Non-puerperal uterine inversion associated with an atypical leiomyoma

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# CASE REPORT

A 27-year-old nulligravid 104 kg woman was referred from her general practitioner with a 2-year history of worsening menorrhagia, dysmenorrhoea, dyspareunia and difficulties with sexual penetration. She had not had a cervical smear for 4 years, but had tried a recent unsuccessful course of continuous progestogens for control of the increasing metrostaxis. An ultrasound performed 14 months earlier revealed a normal sized uterus with a 3 cm fundal fibroid. Examination was hampered by the patient's significant obesity, nulligravity and particularly long vagina.

A large, tender, firm haemorrhagic mass filled the vagina, the nature of which was not immediately obvious. It protruded to a level 2 cm above the hymenal ring, with the mass extending to a level equal to a uterus at 14 weeks gestation. The cervix could be neither seen nor palpated. Magnetic resonance imaging demonstrated a 6 cm heterogeneous uterine leiomyoma at the fundus of an inverted uterus, with T1 weighted images demonstrating fluid collections in the vaginal fornices (Figure 1). Normal ovaries were seen to be lying adjacent to inverted broad ligaments on the superior surface of the mass. Haemoglobin level was 6.8 gm/dl, so a 4-unit blood transfusion was organised prior to any further procedures.

An examination under anaesthetic, laparoscopy and biopsy was arranged in order further to delineate the lesion. The findings were of a beefy red mass entirely filling the vagina and attempts by palpation and vaginoscopy to identify a cervix were again unsuccessful. Laparoscopy revealed the doughnut appearance of an inverted fundus along with broad ligaments and round ligaments on stretch. No obvious vascular compromise to the ovaries or distal fallopian tubes was

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**Figure 1** T1-weighted sagittal image of an MRI through the uterus in the midline, demonstrating uterine inversion with fluid in the vaginal fornices, and an ovary on the cephalad surface of the uterus.



noticed. Simple efforts at pressure from below or traction upon the round ligaments made no significant impact upon returning the fundus to its appropriate position. The fibroid-like mass was transmural in position, and it was not possible to find discrete margins between the mass and normal myometrium. There was inadequate vaginal capacity in which to operate. Haemorrhage from the biopsy and tenaculum sites was a considerable problem, and subsequent vaginal packing and a further 3-unit blood transfusion were required at the end of the procedure which was followed by ongoing oral iron supplementation.

Biopsy findings demonstrated spindle-shaped cells with moderate nuclear enlargement and pleomorphism. The number of mitoses seen per high power field was still within the limits of a benign category. Patches of degeneration were noted. Immunohistochemistry confirmed smooth muscle phenotype, suggesting an atypical leiomyoma with bizarre nuclei.

The woman was committed to retaining her fertility. A decision to give gonadotrophin-releasing

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hormone (GnRH) analogue for 3 months was made on the basis of reducing blood flow to the inverted uterus and reducing the size of the fundal fibroid. Following this, a repeat attempt at removing the fibroid and restoring the normal fundus was planned. The woman had little in the way of vasomotor symptoms but had significant reductions in blood loss per vaginam while on GnRH analogue. Discussions regarding management issues should it be impossible to correct the inversion included hysterectomy, and the chance that there may be more sinister pathology within the body of the fibroid.

Following the 3 months of GnRH analogues, a repeat laparoscopy, examination and pervaginal transmural myomectomy was planned prior to correction of the inversion. During the procedure, the fibroid was found to be 3 cm in diameter, and the endometrium on the external surface of the uterus was markedly atrophic. An electrosurgical loop, similar to those used for excision of the cervical transformation zone, was utilised to debulk the fibroid. Despite removing the bulk of the fibroid, attempts at restoring the position of the fundus through the cervicovaginal junction were unsuccessful. The tightness of this junction proved to be the limiting feature. Laparotomy was performed, which did not assist in repositioning the fundus, despite traction on round ligaments. A total abdominal hysterectomy was performed, and the woman made an uneventful postoperative recovery.

Final pathological examination confirmed the presence of an atypical leiomyoma, with patchy hyalinisation, and atrophy of the endometrium. No evidence of malignant changes was seen.

### DISCUSSION

Non-puerperal uterine inversion is a very unusual condition, which most gynaecologists will never come across, and thus it has to be managed based upon little or no previous experience. There are few case reports in the English literature, and only one review.<sup>1</sup> This case highlights the importance of full anatomical assessment and the difficulty encountered in restoring the position of the fundus in the situation of chronic inversion. Initial assessment appeared to be that of a chronically prolapsed enlarged leiomyoma, and laparoscopy and magnetic resonance imaging (MRI) were utilised to accurately determine the gross

anatomical abnormality. The round ligaments and fundal position reveal if true inversion has occurred. Compromise to ovarian blood supply is uncommon due to the accommodation afforded on chronically stretched infundibulopelvic ligaments. Uterine ischaemia with subsequent necrosis and infection has been reported and requires more immediate action.<sup>2</sup>

Mwinyoglee et al summarised 77 reported cases in a review of in the literature; 75 (97.4%) of these were associated with uterine tumours, of which 20% were malignant.<sup>1</sup> This emphasises the need to perform biopsies prior to definitive treatment.

A classification of genital inversion has been described:  $^{2} \ \ \,$ 

- Stage 1: Inversion of the uterus is intrauterine or incomplete. The fundus remains within the cavity.
- Stage 2: Complete inversion of the uterine fundus through the fibromuscular cervix.
- Stage 3: Total inversion, whereby the fundus protrudes through the vulva.
- Stage 4: The vagina is also involved with complete eversion through the vulva along with an inverted uterus.

Treatment of the inversion depends on the stage and associated pathology. While stage 1 will often afford easy repositioning of the fundus, stages 2–4 will more likely lead to hysterectomy.<sup>2</sup> In our case, the repositioning of the fundus was physically hampered by the tightness of the fibromuscular cervix.

Cervical resistance to dilatation has been found to be markedly increased by pre-treatment with GnRH analogue prior to endometrial ablation.<sup>3</sup> While GnRH analogue was at the time expected to be beneficial by reducing vascularity and size of the leiomyoma, the re-positioning of the fundus might have been exacerbated by pre-treatment with GnRH analogue.

### REFERENCES

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