

## An Unexpected Presentation of Endometriosis – A “Parasitic” Cyst of the Bowel in a Menopausal Woman on Hormone Therapy

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### Abstract

**Introduction:** This report describes a “parasitic” endometriotic cyst of the small bowel. **Clinical Picture:** A menopausal woman with a pelvic mass presenting years after commencing hormone therapy. **Treatment:** We performed laparoscopic excision of a cystic tumour attached to the small bowel with a solitary vascular pedicle. **Outcome:** Histology confirmed it to be an endometriotic cyst of ovarian origin, probably resulting from spillage during previous surgery and reactivation with hormone therapy. **Conclusions:** We discuss the possible aetiology of this unusual presentation of endometriosis and review the literature on parasitic gynaecological tumours.

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**Key words:** Menopause

### Introduction

Endometriosis commonly affects women during their reproductive years and may involve any organ. Although rare, endometriosis has been reported in postmenopausal women and is often associated with hormone replacement<sup>1</sup> or tamoxifen.<sup>2</sup> Extra-genital endometriosis following pelvic surgery has also been described.<sup>3,4</sup> This case illustrates an unusual presentation of an endometriotic cyst which first appeared as a “parasitic” tumour affecting the small bowel in a postmenopausal woman on hormone therapy. To our knowledge, there is no previously published account of an endometriotic cyst completely detached from the ovary. The theories of its origin may be debated but will be difficult to confirm.

### Case Report

The woman sought treatment for primary infertility in 1984, aged 31 years. Laparotomy, cystectomy and adhesiolysis were performed twice for pelvic endometriosis in 1984 and 1986 but she failed to conceive. She was first seen at our institute in 1988 aged 35 years, seeking further infertility treatment. She had no symptoms of endometriosis. Diagnostic laparoscopy demonstrated adhesions in the left

ovarian fossa obscuring the left ovary and tube, which was obstructed. Multiple endometriotic deposits were noted in the pelvis along with adhesions in the cul-de-sac. Adhesiolysis, ablation of endometriotic deposits and bilateral ovarian biopsies were performed. Histological examination revealed the absence of primordial follicles in the right ovary. Subsequent fertility treatments were unsuccessful.

She reported climacteric symptoms and oligomenorrhoea at age 43 years. With an elevated serum follicle stimulating hormone level (FSH) of 45.2 IU/L, ovarian failure was diagnosed. Pelvic examination and ultrasonography were normal and both ovaries were identified. She was started on hormone therapy in 1996 (conjugated equine oestrogens 625 mcg, norethisterone 150 mcg cyclically) and continued this for the next 7 years for persistent symptoms. Menstruation ceased during the first year of use. Annual pelvic examinations were unremarkable.

During routine examination in 2002, a pelvic mass was detected. Sonography showed a 67 mm left ovarian endometrioma. Serum glycoprotein cancer antigen CA 125 was normal at 13.1 U/mL. Though the clinical suspicion

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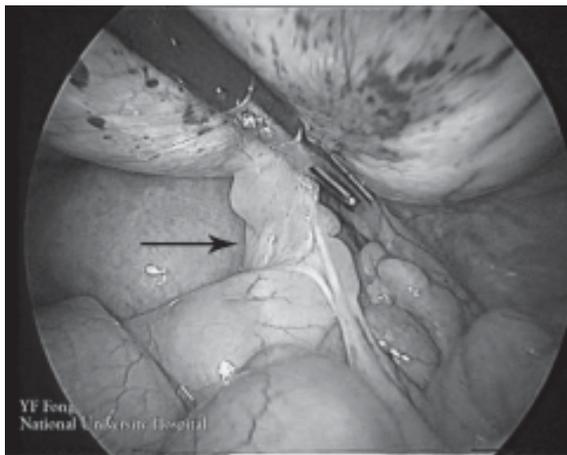


Fig. 1. Large endometriotic cyst attached to a loop of small bowel by a vascular pedicle. The cyst initially lay in the cul-de-sac but was easily mobilised, having no other points of attachment.

of malignancy was low she was advised to stop hormone therapy and to undergo surgery, but she resisted any change in management as she felt well.

Regular surveillance with ultrasound and CA 125 levels showed no appreciable change in the cyst. In 2003, she started tibolone and remained asymptomatic for the pelvic mass. However, repeat sonography in 2005 demonstrated an increase in the size of the lesion to 78 x 70 mm (CA 125 was 7.2 U/mL), and she agreed to surgical management.

### Treatment

Diagnostic laparoscopy revealed a pedunculated cystic tumour measuring 7 cm arising not from either adnexa but, unexpectedly, solely connected by a single vascular pedicle to the small bowel surface (Fig. 1). This smooth-walled mass was similar in appearance to an ovary containing an endometriotic cyst. The cyst was delivered after ligation of the pedicle and controlled aspiration in an endoscopic pouch. The contents were a characteristic chocolate material. The right ovary was atrophic; the left ovary was not visualised. No other abnormalities were noted.

### Outcome

Histological examination of the excised mass showed a unilocular, thin-walled cyst lined by unilayered cuboidal epithelium (Fig. 2). The subepithelial endometrial type stroma contained haemosiderin-laden macrophages (Fig. 3). This triad established the diagnosis of an endometriotic cyst. With clinical correlation, in particular the appearance of the cyst, the solitary vascular attachment to the bowel and the patient's history, the final diagnosis of a parasitic endometriotic cyst was made.

### Conclusions

This case adds to the literature describing the diverse

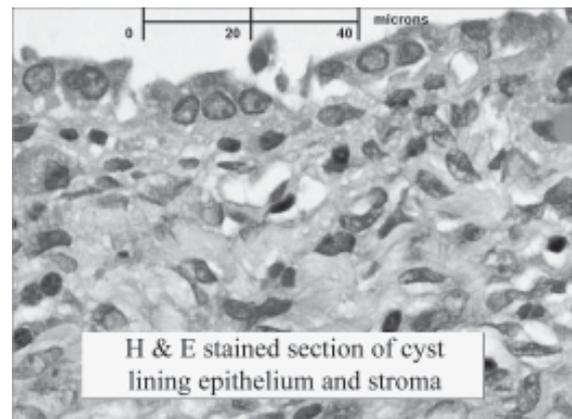


Fig. 2. Cyst wall lined by unilayered cuboidal epithelium.

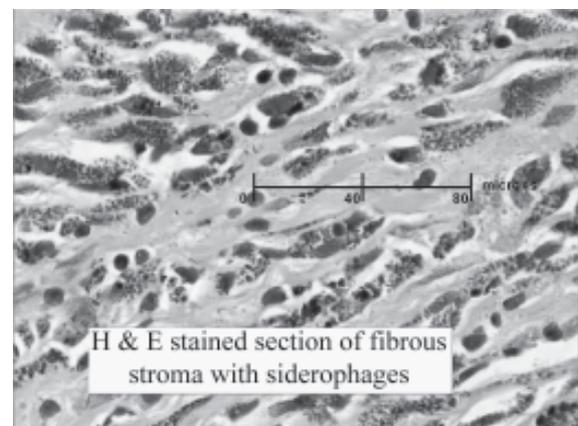


Fig. 3. Subepithelial endometrial type stroma with haemosiderin-laden macrophages.

clinical manifestations of endometriosis, and is believed to be the first report of intestinal endometriosis presenting as a pedunculated endometrioma. Intestinal endometriosis commonly appears as infiltrating lesions that can extend to various depths from the bowel serosa down to the mucosa.<sup>5</sup> Thus, the lesion did not at first appear to be a variant of intestinal endometriosis. Debating the origin of this cyst raises the intriguing possibility of a parasitic tumour. The instigating factor is thought to be spillage of viable ovarian or endometriotic tissue during previous surgery, with transplantation and formation of an alternate blood supply from the bowel. Supported by a parasitised blood supply, proliferation of the endometriotic tissue continued. The process may have started prior to menopause and commencement of hormone therapy, but was very likely sustained by exogenous hormones as the cyst was detected after many years of hormone use.

Parasitic tumours are a rare medical entity. Parasitic leiomyomata have been reported.<sup>6</sup> These originate as subserosal fibroids which develop a secondary "parasitised"

blood supply from an adjacent organ eventually becoming detached from the uterus. There are published reports of parasitic teratomata originating from ovarian dermoid tumours and found reattached to the omentum as a result of torsion, autoamputation and reimplantation.<sup>7</sup> To our knowledge, this is the first such report of a parasitic ovarian endometriotic cyst. A search on PubMed using key terms including, but not limited to, “parasitic”, “ovary”, “cyst” and “endometrioma” did not reveal any other published account.

Either of the main theories regarding the aetiology of endometriosis could also be used to explain the origin of this lesion.<sup>8</sup> It may have started with implantation prior to menopause, direct transplantation onto the bowel serosa or surrounding scar tissue during surgery, or resulted from coelomic metaplasia under the influence of exogenous hormonal stimulation.

This hypothesis is difficult to prove. Although at histological examination the finding of cuboidal epithelium was compatible with ovarian tissue, a metaplastic phenomenon arising in a mesenteric cyst could also look histologically identical. Histochemical markers would be identical in both. Thus while endometrioid differentiation is demonstrated, the ability to prove ovarian histogenesis is limited.

Reports of postmenopausal endometriosis often feature exogenous hormones.<sup>1</sup> There is little evidence to suggest

whether cyclical or continuous regimes would be better for a woman with endometriosis. This case underlines the need to be prudent when using hormone therapy in women with endometriosis and the need to regularly screen for recurrence.

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