Ovarian endometriosis with co-existing granulomatous inflammation—a rare association

Lt Col Tanushri Mukherjee*, Bharat Rekhi†

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INTRODUCTION

Endometriosis is a benign, oestrogen-dependent, chronic gynaecological disorder, commonly associated with pelvic pain and infertility. It occurs at various sites, including the ovaries, where it is described as a 'chocolate cyst'. On histopathology, it reveals variable amount of endometrial stroma, pigmented macrophages, and endometrial glands.¹

Granulomatous inflammation of the ovary is commonly associated with tuberculosis, including tubercular salpingitis that clinically present with pelvic pain and/or infertility. Ovarian endometriosis has been uncommonly documented with a co-existing granulomatous inflammation.^{2–5} Here, we present a rare case of a young lady, who presented with a unilateral ovarian mass, which on biopsy unravelled endometriosis with co-existing epithelioid granulomas. This case report is the third such documented case, to the best of our knowledge.

CASE REPORT

A 35-year-old averagely built lady, para 2, presented with complaints of acyclic pelvic pain. She acquired menarche at the age of 13 years. She denied history of intake of hormonal therapy or a prior history of pelvic pain and/or dysmenorrhoea. There was no associated history of fever or cough. She had normal deliveries and had no history of any prior surgical intervention. She had no familial history of endometriosis. On her physical examination, a uniformly enlarged, left-sided adnexal mass was noted. A pelvic ultrasonography scan revealed a left ovarian cystic, homogenous mass that measured $4.4 \times 2.7 \times 2.7 \text{ cm}^3$ in size. The Doppler blood flow study suggested this as a benign ovarian mass. A clinical diagnosis of a left ovarian endometriosis was considered.

The tumour marker levels for cancer antigen-125, alpha-fetoprotein (AFP), and carcino-embryonic antigen were within normal limits. The patient underwent a laparoscopy that revealed a left-sided, cystic adnexal mass, without adhesions, or other pelvic endometriotic lesions. She was subjected to a left salpingo-oophorectomy, elsewhere that was subsequently submitted to us for histopathological analysis.

Grossly, the ovarian mass measured $6.0 \times 5.5 \times 3.5 \text{ cm}^3$. Cut surface showed multiple cysts containing thick, 'chocolate brown' material. The cyst wall thickness varied from 0.1 cm to 0.3 cm (Figure 1). Microscopic examination revealed cystic areas lined by benign cuboidal epithelium along with endometrial glands and stroma, the latter exhibiting peri-arteriolar arrangements of oval to short spindly cells. Besides, there were sheets of pigmented macrophages. In addition, well-defined epithelioid cell granulomas containing Langhan's type of giant cells and focal punctuate necrosis were noted (Figure 2). Acid fast staining did not reveal any mycobacteria. On immunohistochemistry, CD10 was positive within the ovarian cyst wall, reinforcing endometrial stroma. Diagnosis of an ovarian endometriotic cyst with co-existing granulomatous inflammation was formed. Attached fallopian tube was unremarkable. Further treatment details were inaccessible as this being a referral patient.

Figure 1 Gross findings of ovarian endometriosis. Ovarian mass with cut surface showing multiple cysts containing thick, 'chocolate brown' material.

*Graded Specialist (Pathology & Oncopathology), 166 Military Hospital, C/o 56 APO, †Associate Professor (Pathology), Tata Memorial Hospital, Parel, Mumbai.

Correspondence: Lt Col Tanushri Mukherjee, Graded Specialist (Pathology & Oncopathology), 166 Military Hospital, C/o 56 APO.
E-mail: tanujamukherjee@yahoo.com

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Figure 2 Microscopic findings. (A) A cyst lined by cuboidal epithelium with endometrial stromal component within the cyst wall; dilated blood vessels and well-defined epithelioid granulomas containing Langhans type of giant cells (200 $\times$, H&E), (B) cyst wall showing endometrial glands with co-existing epithelioid granulomas (200 $\times$, H&E), (C) areas showing sheets of pigmented macrophages within the cyst wall (200 $\times$, H&E), and (D) CD10 highlighting the endometrial stroma within the ovarian cyst wall. Negative staining in the cuboidal epithelium of the cyst (200 $\times$, DAB). DAB: 3'-3'-diaminobenzidine tetrahydrochloride, H&E: haematoxylin and eosin.

DISCUSSION

The present case is an extremely uncommon case of ovarian endometriosis with co-existing granulomatous inflammation, occurring in a young, para 2, lady, who presented with acyclic pelvic pain. While she was clinicoradiologically suspected with ovarian endometriosis, a co-existing granulomatous inflammation, in addition, was noted on histopathological examination of the salpingo-oophorectomy that she underwent.

In a seminal paper, Cluggage et al identified foreign-body reaction to suture, Crohn's disease, previous diathermy, tuberculosis and postoperative necrotizing reaction, in decreasing order of frequency, as identifiable causes of ovarian granulomas. Only one of their 32 cases of ovarian granulomas showed co-existing endometriosis.³ Thereafter, Ghosh et al have documented a single case of co-existing genital tuberculosis and endometriosis in an infertile patient.⁵

Diathermy-induced tubo-ovarian granulomas in form of amorphous coagulated tissue surrounded by a granulomatous reaction containing foreign-body giant cells, can occur in patients who have had laparoscopic cauterisation for conditions such as endometriosis. A histiocytic reaction in the ovary is

known to occur in endometriosis. However, such reactions are not granulomatous, but consist of histiocytes which may contain lipofuscin and haemosiderin pigments, the latter forming a part of endometriosis.³ In the present case, apart from pigmented histiocytes that formed the endometriotic component, discrete epithelioid granulomas containing Langhan's giant cells with focal necrosis, similar to tubercular granulomas, were noted. However, special stain did not reveal mycobacterial organisms and culture was not performed. Besides, peritoneal necrotic pseudoxanthomatous nodules have been observed in patients with endometriosis.⁴ According to a western study by Cluggage et al, the most common cause of ovarian granulomas is a reaction to the suture material introduced during a previous operation.³ With the increasing emphasis on conservative surgery including ovarian preservation, in form of a cystectomy and wedge resection, granulomas are being encountered more frequently. Talc and starch granulomas are also known to occur postoperatively. These are usually typical foreign-body type. In the present case there was no history of surgical intervention and no foreign-body material within the giant cells. Postoperative necrobiotic ovarian granulomas are composed of central fibrinoid necrosis surrounded by palisaded histiocytes and giant

cells, raising other possibilities such as rheumatoid disease. Earlier, Herbold et al described apparently four cases of idiopathic ovarian granulomas in premenopausal women, none of whom had a history of systemic granulomatous disease.² Ovarian involvement has been uncommonly reported in sarcoidosis with a co-existing mucinous cystadenoma.⁶

Granulomatous inflammation in the present case could possibly have been as a result of associated endometriosis, co-existing mycobacterial infection (despite negative mycobacterial staining), sarcoidal reaction or idiopathic. Nonetheless, it is an intriguing and an uncommon association. Even though the patient did not present with any pulmonary symptoms, chest radiography was recommended.

CONFLICTS OF INTEREST

None identified.

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