CASE REPORT

Florid cystic endosalpingiosis, masquerading as malignancy in a young patient: a brief review

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SUMMARY

Endosalpingiosis is a rare condition characterised by the presence of tubal epithelium outside the fallopian tube. Most of the previous case reports have described this condition in women in their fifth decade or older. We report a case of a woman presenting at 31 years with a history of heavy bleeding during menses and pain in the lower abdomen for the past 2 years. An ultrasound examination showed a left ovarian cyst of 4.3×3.2 cm with multiple septations. CA 125 was within normal range. Laparoscopy was performed with the plan of ovarian cystectomy, peroperatively; there were papillary projections all over the uterus and the peritoneal surface. The left ovary was enlarged with papillary projections on the surface. Multiple biopsies were taken from the surface of the uterus, ovary and the peritoneum as the picture was quite suspicious of malignancy. Histopathological examination gave the picture of endosalpingiosis. The patient underwent laparoscopic ovariectomy with fulguration of lesions 4 months later, in view of persistent pain and discomfort, and increase in the size of the cyst on ultrasound monitoring. Presently, she is free of symptoms.

BACKGROUND

Endosalpingiosis is a rare benign condition of the Mullerian system characterised by the presence of glands lined by ciliated tubal-type epithelium and involves the peritoneum, subperitoneal tissues, omentum, retroperitoneal nodes, etc. It was first described by Sampson,¹ who found epithelium resembling the fallopian tube in the ectopic locations. The most accepted pathogenesis is metaplastic change of the coelomic epithelium into tubal-like epithelium. Endosalpingiosis is associated with dysmenorrhoea and chronic pelvic pain. It is important to differentiate it with papillary serous adenocarcinoma due to presence of papillary and grapes like excrescences on the pelvic peritoneum, ovary and uterus as the management of this condition is primarily conservative. The other differential diagnosis includes benign cystic mesothelioma and multiple peritoneal inclusion cysts.

We have encountered this case for the first time in our practice. Proper histopathological diagnosis of the condition is crucial because its gross appearance mimics ovarian or primary peritoneal malignancies in most situations. The previous case studies have reported the condition in relatively older women. Our patient was different in this respect as she was 31 years old.

CASE PRESENTATION

A 31-year-old woman presented to the outpatient department of our hospital in June 2012 with a history of heavy bleeding during menses and pain in the lower abdomen for 2 years. She had two living issues, both were born by lower segment caesarian sections; postoperative period was uneventful, and she was not on any contraceptives. The last childbirth was 3 years ago. General examination was normal; on pelvic examination, her uterus was normal in size, the right fornix was free but the left fornix was full and tender.

INVESTIGATIONS

An ultrasound of the pelvis was performed in which the uterus and the right ovary were normal and the left ovary had a cyst of 4.3×3.2 cm with multiple septations. CA 125 was 25 U/mL.

Hysteroscopy and endometrial aspiration were performed in view of her symptoms of heavy menstrual bleeding; the results were normal with secretory pattern of the endometrium on histology. Transvaginal ultrasound was repeated after 1 month and it was found that the size of the left ovarian cyst has increased to 6.8 cm×5.5 cm with multiple septa mimicking hyperstimulated ovary. Doppler test was normal. CA 125 was repeated and the value was 25 IU/mL.

On laparoscopic examination, vesicular deposits of varying sizes were present all over the surface of the uterus and the pelvic peritoneum, and the left ovary was multicystic with papillary projections.

Biopsy from the deposits and the surface of the left ovary was suggestive of endosalpingiosis.

DIFFERENTIAL DIAGNOSIS

The term mullerianosis include lesions with tubal or serous differentiation called endosalpingiosis and the homologous lesions of endometriosis and endocervicosis.²

Mixed forms of mullerianosis also occur.³ It has varied clinical presentation, and sometimes it can be asymptomatic. It may resemble a gynaecological malignancy on imaging findings, therefore clinicians and radiologists should be aware of this benign entity to render a correct diagnosis and to avoid overtreatment.⁴ The gross appearance of endosalpingiosis may mimic a primary peritoneal malignancy as well.

TREATMENT

She was given a course of antibiotics and started on norethisterone 5 mg twice daily for menorrhagia. The patient did not respond to the treatment and

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reported again in December 2012 with acute pain in the lower abdomen. In view of her persistent symptoms and increase in size of the ovarian cyst, laparoscopy was planned. On laparoscopic examination, the uterus was found covered with vesicular deposits; deposits were also seen on the pelvic peritoneum and the left ovary was multicystic with papillary projections. However, the upper abdomen, undersurface of the diaphragm and surface of the liver were normal with no deposits. Multiple biopsies were taken from the surface of the uterus, ovary and peritoneum as the picture was quite suspicious of ovarian carcinoma (figure 1). Frozen section of the biopsied tissue was suggestive of some benign pathology; therefore, it was decided to wait for the final histopathology report, which suggested of endosalpingiosis (figure 2).

The patient was started on continuous progesterone tablet medroxyprogesterone acetate 10 mg twice daily. After 6 weeks, there was no improvement in pain and the pelvic discomfort had increased. The patient was offered laparoscopic ovariectomy in view of her pain and discomfort. All the superficial deposits were fulgurated. The peritoneal cavity was thoroughly lavaged.

OUTCOME AND FOLLOW-UP

The patient is off all medications at present and was free of symptoms during her last follow-up visit a week ago in August 2013.

DISCUSSION

The clinical presentation of endosalpingiosis is not specific. The common symptoms are pelvic pain, dysmenorrhoea, menorrhagia and infertility; some patients are asymptomatic, with an incidental finding of ovarian cysts on imaging or during laparotomy for other indications.^{5 6} Endosalpingiosis represents a diagnostic challenge that can only be made by histopathological examination.⁷ On rare occasions, it presents as a macroscopic mass that can be confused with a neoplasm.

Taneja *et al* reported a case of a 40-year-old woman who presented with chronic pelvic pain and dysfunctional uterine bleeding. Her CT scan revealed an irregular soft tissue mass in the pelvis indenting the posterolateral aspect of the uterus and cervix. Total abdominal hysterectomy with bilateral salpingo-oophorectomy was performed and histopathological examination revealed cystic structures lined by the tubal type of epithelium.⁴

Heatley and Russell⁸ reported a case of a 73-year-old woman who presented with a right-sided adnexal cystic mass, which at laparotomy appeared to be a benign serous ovarian cyst with an aggregation of thin-walled subserosal soft tissue cysts in the side wall of the uterus and adjacent parametrium. Total abdominal

Figure 1 Laparoscopic view of enlarged ovary with multiple papillary projections over ovary and uterus.

Figure 2 Histology of the ovarian biopsy: calretinin-positive polypoidal endosalpingiosis.

hysterectomy and bilateral salpingo-oophorectomy were performed. On histopathological examination, the features indicated multidirectional Mullerian differentiation which was consistent with florid cystic endosalpingiosis.⁸

Papavramidis *et al* reported a case of a 50-year-old Greek woman who presented with a pale brown nodule in her umbilicus, which was asymptomatic, with no cyclical discomfort or variation in size. A cystic lesion measuring 2.7×1.7×1 cm was removed. Histopathological examination was in favour of cutaneous endosalpingiosis.⁹

According to a retrospective study by Heinig *et al*,¹⁰ endosalpingiosis was diagnosed in 13 patients in a centre within a period of 6 years. Of those, nine patients had a history of some surgeries being performed in the past, 2 (15%) patients had tubal surgery, 2 (15%) had caesarean section and 5 (38%) had a history of more than two abdominal operations. Our patient too had a history of two full-term caesarean sections. Thus, history of surgeries may be one of the predisposing factors for endosalpingiosis. The usual age of diagnosis of endosalpingiosis is above 40 years according to most of the case reports; but our patient presented at a younger age as compared with the usual age of presentation.

Learning points

- ▶ Endosalpingiosis is a rare disorder of the Mullerian system characterised by the presence of glands lined by ciliated tubal-type epithelium on the parietal and visceral peritoneum, subperitoneal tissues, omentum, retroperitoneal nodes, etc.
- ▶ It may be associated with endometriosis or endocervicosis.
- ▶ Gross appearance of the lesions mimic peritoneal or ovarian malignancies.
- ▶ Confirmation of diagnosis by histopathological examination is very important, so as to impart proper line of management to the patient.

Contributors All the authors have contributed significantly in the management of the patient and preparation of the case report.

Competing interests None.

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