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COMMUNICATION

Unsuspected unusual endometriotic ovarian cancer

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Abstract In recent years, laparoscopic surgery has become the gold standard in the management of presumed benign adnexal masses. As preoperative selection is not perfect, some cases of ovarian malignancy are encountered at laparoscopy, during the management of adnexal masses presumed to be benign. We report the case of a 45-year-old lady with endometriosis, who presented with a pelvic abscess 6 months after the treatment of an endometrioma managed at another unit. She subsequently underwent a laparoscopic salpingo-oophorectomy. The histology returned showing an endometrioid adenocarcinoma of the ovary. We review the literature regarding the use of laparoscopy for ovarian masses, especially in the case of ovarian endometrioma in the perimenopause. Though conservative surgical management of endometriomas in young women is advocated by some authors, this article highlights the possible pitfalls in the management of endometriomas in women over the age of 40 years, as endometriomas can be associated with an increased risk of endometrioid and clear-cell adenocarcinoma of the ovary.

Keywords Endometrioma · Ovarian cancer · Pelvic abscess

Introduction

Laparoscopic surgery in the management of adnexal masses has enormously increased in popularity since the late 1980s, and has mostly replaced the abdominal approach. Several prospective randomised studies have demonstrated the value of laparoscopic surgery in the management of adnexal masses [1].

Most authors consider operative laparoscopy superior to laparotomy, especially in the management of endometriomas [2]. Others consider this issue controversial, since the preoperative assessment for the diagnosis of malignancy in adnexal masses cannot be established in all cases, especially before the menopause [3].

As a consequence, adnexal masses expected to be benign may be later found to be malignant and possible spillage at the time of laparoscopic surgery can lead a worsening of the prognosis [4].

Case report

A 45-year-old nulliparous lady presented to our gynaecological emergency service complaining of abdominal and pelvic pain of acute onset. The pain was aggravated by movement and was radiated to the shoulder.

She was pale, tachycardic and with a temperature of 38.5°C. On examination, there was rebound tenderness and guarding, especially in the left iliac fossa. Vaginally, there was left adnexal tenderness. Her pregnancy test was negative, urinalysis and blood profile was normal, except for the Ca125, which was slightly elevated.

Ultrasound of the pelvis revealed a 7–8-cm left ovarian cyst, suggestive of an "endometrioma," with some free fluid in the pouch of Douglas.

Six months earlier, she had presented to another unit with lower abdominal pain and dyspareunia. A scan at the time had shown a 3-cm endometrioma, for which she had undergone laparoscopic management; however, no tissue biopsy was taken. At her 2-month follow-up, the cyst had

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recurred. As her symptoms had not changed, it was decided to manage her conservatively with a progestagen and 6-weekly scans. The Ca125 estimations were elevated during her conservative management (209.2 kU/L).

During her current presentation, despite analgesia and antibiotics, her pain became uncontrollable. She was, thus, offered an emergency laparoscopy after counselling regarding the possible loss of the left adnexa and that further operation may be necessary, depending on the findings.

The intraoperative findings were: a ruptured left tubo-ovarian abscess of 8 cm which was adherent to the pelvic sidewall and left uterosacral ligament, and 300 ml of purulent fluid in the pouch of Douglas and under the diaphragm (Fig. 1).

There was endometriosis on both uterosacral ligaments, the anterior pouch and, on the right ovary, there was grade II–III endometriosis, as per rAFS classification.

Peritoneal fluid was aspirated and sent for microbiology and cytology.

Once the ovary was drained, the interior looked suspicious, thus, a left salpingo-oophorectomy with excision of the peritoneum of the sidewall where the ovary was attached was performed. The specimen was removed through a 10-mm port with an endobag and was sent for urgent histology.

The patient recovered from the surgery uneventfully and was discharged from hospital 2 days post-operatively.

Pathology analysis of the specimen revealed an endometrioid neoplasm with high-grade proliferation, showing prominent foci of squamous metaplasia (Fig. 2).

Upon receipt of the pathology report (stage IIb), referral was made to a gynaecologic oncologist at the local cancer centre.

The patient underwent completion surgery, at which there was no upstaging of disease, followed by chemotherapy.

Discussion

The ovary is a common site for endometriosis [5]. The exact incidence and prevalence of malignant transformation in endometriosis is unknown. However, it is thought to occur in 0.7–5% of all cases with ovarian endometriosis [6].

Fig. 1 Endometrioma at the second laparoscopy

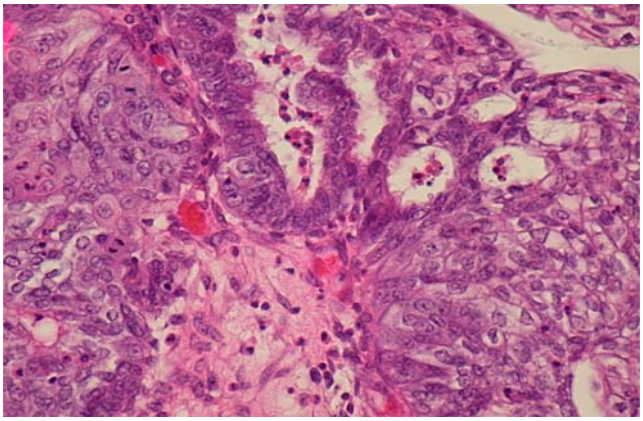

Fig. 2 High-grade endometrioid ovarian adenocarcinoma showing prominent focal squamous differentiation

When it occurs, carcinoma of the ovary is seen in women with endometriosis 10 to 20 years younger than those who develop endometrial or ovarian cancer. In Heaps et al.'s review of 195 cases of endometriosis associated with ovarian cancer, the average age of the patients was 46 years and 50% of the patients were nulliparous [7].

In patients with long-standing endometriomas, the relative risk of ovarian cancer has been shown to be increased (standardised incidence ratio of 4.2) [8], which is in keeping with the follow-up study by Melin et al., which had a longer follow-up [6].

The working diagnosis in our patient was that of an endometrioma with a pelvic infection. She thus underwent laparoscopy, as it is known that pelvic abscesses can result after the treatment of endometriomas and that laparoscopy, apart from being the best way to manage endometriomas, also has a place in the management of pelvic abscess [9].

It was also kept in mind, however, that she was a 45-year-old nulliparous woman with an elevated Ca125.

At the time of the surgery, laparoscopic drainage of the abscess may have been adequate, as suggested by some authors [10]. It is, however, important with endometriomas to perform a "cystoscopy" to visualise the interior of the cyst. All of the fluid should be washed out and a biopsy taken from the cyst. This approach may not be appropriate for women over the age of 40 years because of the association of endometriosis and cancer [8]. The interpretation may, however, be difficult in the presence of infection. In our case, suspicious features were noticed and, on further dissection, it was difficult to separate the ovary from the pelvic sidewall. This difficulty of cleavage is considered by some authors as a pointer to malignancy [1].

The decision was, thus, between performing a frozen section or the removal of the adnexa with urgent histology followed by completion surgery.

It was decided not to perform a frozen section, as success of frozen sections in the presence of infection was doubtful [11] and the suspicious appearance in itself may have been due to the presence of acute on chronic inflammation. In addition, there are reports of tubo-ovarian abscess resembling ovarian malignancy and cases of ovarian abscess arising within an endometrioma have been described [12].

The patient underwent salpingo-oophorectomy with excision of the site of adherence of the ovary, which is important in staging, should the mass turn out to be malignant, as in this case.

If a frozen section is not carried out, it is important that the specimen be sent off for histology urgently, as a delay beyond 17 days has been reported to increase the stage of disease [13]. The specimen, however, should also be removed in an endoscopic bag, as this, theoretically, would reduce the occurrence of port site metastasis.

The choices for the surgical management of endometriomas include: laparoscopic cyst aspiration, fenestration and ablation or excision of the cyst. Women undergoing endometrioma coagulation or vaporisation are three times as likely as women undergoing cyst excision to have a recurrence, with recurrence rates ranging from 6–30% [14].

Some investigators have also suggested a two-stage procedure, where the cyst is aspirated, irrigated and inspected, followed by GnRH analogues. This procedure may be appropriate for large cysts [15].

Excision should be the treatment of choice for endometriomas, such as that for our 45-year-old nulliparous patient in this report.

Another issue is the follow-up of women after the treatment of endometriomas. In this patient, there was a recurrence with minimal symptoms; thus, she was followed up with serial scans instead of re-operation. The possibility of malignancy should be kept in mind in older women. Conservative management without tissue diagnosis should be reserved for women with ultrasonographically suspected simple ovarian cysts [16].

The patient in this study received the most appropriate operation for the presenting symptoms, but at her original treatment, her background history was overlooked and tissue diagnosis was not made.

Though laparoscopy is the best management for women with ovarian cysts, it is important that the background history should always be taken into consideration, as malignancy is a possibility, especially in older women with an elevated Ca125 and an “endometrioma,” as demonstrated by this case.

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