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CASE REPORT

Second Look of Endosalpingiosis: A Rare Entity

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Introduction

Endosalpingiosis is a rare condition characterized by the presence of tubal epithelium outside the fallopian tube [1]. The most accepted pathogenesis for this condition is metaplastic change of the coelomic epithelium into tubal-like epithelium [1]. Like endometriosis, endosalpingiosis is also classified as a lesion of secondary Mullerian system. It is a rare gynecological condition usually seen in females in reproductive age [2].

It is not unusual to find endosalpingiosis associated with endometriosis or endocervicosis, although it often appears alone. One retrospective study showed concurrent presence of endometriosis in 34.5% patients of endosalpingiosis [3]. The same study also quoted a strong correlation between endosalpingiosis and gynecological malignancy in pre-menopausal females (p value <0.0001) with no significant

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correlation in postmenopausal females. We published a case report on endosalpingiosis in the British Medical Journal of case reports in the year 2014. The present report is a follow-up of the same patient after 2 years and 6 months.

Case Report

A patient presented to our outpatient department with dysmenorrhea and heavy menstrual bleeding in January 2013. She was then 31 years old with two living issues born by lower segment cesarean section. An ultrasound examination showed a cyst measuring 4.3 × 3.2 cm. Medical management with continuous progesterone therapy was tried but without any improvement in symptoms; moreover, the size of the ovarian cyst increased to 6.8 × 5.5 cm on ultrasound in a month's period. She underwent laparoscopic left oophorectomy and fulguration of the vesicular deposits on the pelvic peritoneum and uterus in February 2013. A laparoscopic view of the deposits is shown in Fig. 1. Histopathological examination was suggestive of endosalpingiosis. She was put on continuous oral progesterone therapy for 3 months and was asked to follow up. The patient did not come for further follow-up after one visit during which she was fine. She had stopped oral progesterone after 6 months. After 2 years of the last visit, she came to us with 7-week pregnancy. She wanted to continue with the pregnancy. Regular antenatal care was provided. The patient did fine throughout the course of her visits. An elective lower segment cesarean section was done at 38 weeks of gestation. Preoperatively, the uterus was studded with multiple florid deposits (Fig. 2). Both tubes and the right ovary were buried under the deposits and adhered to the uterine surface. The right ovary was normal in size. The pelvic peritoneum was palpated thoroughly, and deposits were present all over it. A 2 × 2 cm nodule was also present on the omentum. Multiple biopsies were taken from the deposits on the uterus, from the deposits in the pelvic peritoneum, and omental nodule was also removed. Tubal sterilization was also performed along with the procedure. Closure was done uneventfully. Histopathology report showed endosalpingiosis in all the specimens.

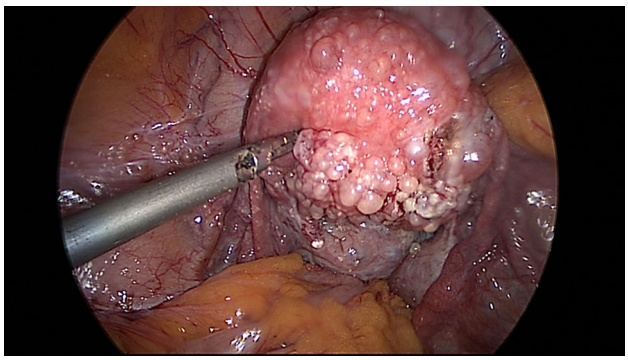

Fig. 1 Laparoscopic view of the deposits of endosalpingiosis

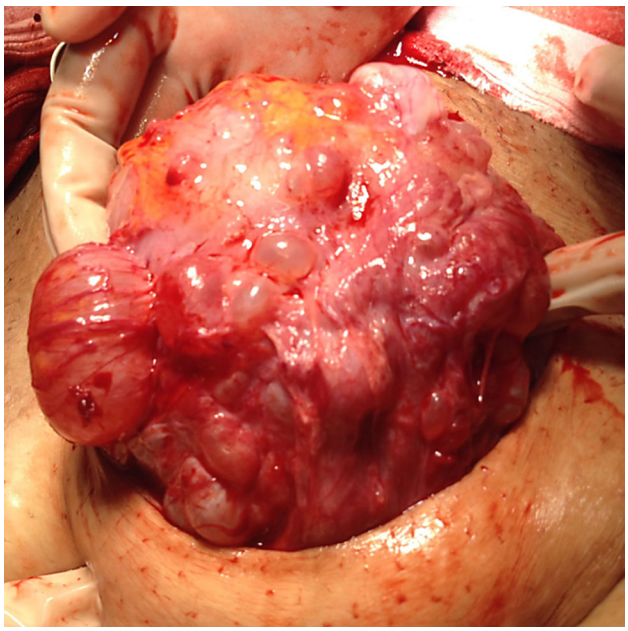

Fig. 2 Uterus studded with florid deposits with tubes and right ovary buried under them

Discussion

Sampson first described endosalpingiosis in 1930; he found epithelium resembling the fallopian tube in ectopic locations in women who had undergone previous salpingectomies or tubal sterilization. Endosalpingiosis can present with chronic pelvic pain, dysmenorrhea, menorrhagia, and infertility or can be asymptomatic. A 5-year retrospective study published in the journal of Fertility and Sterility showed that no significant association was found between endosalpingiosis and chronic pelvic pain, neither between endosalpingiosis and infertility, as against endometriosis, which is proven to be associated with these presentations [3]. This is the only study, which included 72 patients with endosalpingiosis without concomitant endometriosis, considering the rarity of the condition. The same study also noted that premenopausal women with endosalpingiosis were more than ten times more likely to have a gynecologic malignancy than women without endosalpingiosis [3]. Association between endosalpingiosis and serous borderline tumors has also been mentioned. The patient in our case study did not have any clinical symptom after the previous surgery, and she had conceived immediately on stopping the progesterone pills.

Prentice et al. [3] observed that the rate of previous gynecologic and abdominal surgery was 59.1% and a

history of tubal disease was documented in 33.6% of cases in their retrospective study, which raises the possibility that peritoneal implantation may be a factor in the etiology of endosalpingiosis. The patient in our study also had previous 2 cesarean sections before the laparoscopies.

Prentice et al. [3] also state that the hormone dependence theory of endosalpingiosis should be re-evaluated, as 40% women with endosalpingiosis in their study were postmenopausal.

Gross appearance of endosalpingiosis may mimic a primary peritoneal tumor or papillary carcinoma of ovary, but the absence of mitotic activity or atypia on histopathology contradicts the diagnosis of carcinoma [1]. Extensive endosalpingiosis involving the vaginal cuff, pelvic peritoneum, anterior abdominal wall and serosa of the sigmoid colon has been reported in a 68-year-old postmenopausal woman who was operated for Stage 1A carcinoma of ovary in the past. She had hysterectomy for abnormal uterine bleeding 31 years ago and bilateral salpingo-oophorectomy for FIGO stage 1A carcinoma of ovary 9 years back. This time, she had presented with nausea and vomiting. Examination showed a mass in the pelvis, and imaging showed the presence of 8-cm bilobed mass in the pelvis encasing the rectum. The patient was taken up for surgery with a preoperative diagnosis of recurrent ovarian malignancy. An extensive surgery with upper vaginectomy, complete pelvic peritonectomy, excision of the bladder peritoneum and the lower anterior abdominal wall peritoneum and cystic lesions of the sigmoid and infracolic omentectomy was also performed with recurrent ovarian cancer in mind; the final histopathology report came as endosalpingiosis. Although frozen section was done and its report was suggestive of endosalpingiosis, the appearance was so much in favor of malignancy that the surgeons preferred going for a radical surgery [4]. Endosalpingiosis is mostly asymptomatic [3]. Asymptomatic endosalpingiosis does not require any treatment. It may become symptomatic by mechanical irritation of abdominal organs. Surgical removal of the cystic structures may effectively abolish the symptoms.

In our case, we put the patient on continuous oral progesterone for 6 months after the previous surgery, although she was symptom-free, but the lesions were as furious as before, even pregnancy, which is a high progesterone state could not melt the lesions. From this we can conclude that unlike endometriosis, the lesions in endosalpingiosis do not respond to hormonal therapy.

Conclusion

Endosalpingiosis is a rare benign entity, masquerading peritoneal or ovarian malignancy. In spite of the benign nature, the patients need to be kept on follow-up because of significant association between endosalpingiosis and gynecological malignancies, specially the premenopausal patients.

Compliance with Ethical Standards

Conflict of interest Author Rinchen Zangmo declares that she has no conflict of interest. Author Neeta Singh declares that she has no conflict of interest. Author Sunesh Kumar declares that he has no conflict of interest. Author Richa Vatsa declares that she has no conflict of interest.

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

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