

## CASE REPORT

# Endometriosis coexisting with mature cystic teratoma in the same ovary and ectopic pregnancy of left fallopian tube: a rare coexistence

Heesuk Chae<sup>1,2</sup> & Chulhee Rheu<sup>1,2</sup>

<sup>1</sup>Department of Obstetrics and Gynecology, Chonbuk National University Medical School, Jeonju, South Korea

<sup>2</sup>Research Institute of Clinical Medicine, Chonbuk National University Medical School, Jeonju, South Korea

### Correspondence

Department of Obstetrics and Gynecology, Chonbuk National University Medical School, 20, Geonji-ro, Deokjin-gu, Jeonju, Jeollabuk-do 561-712, South Korea. Tel: +82-63-250-1373; Fax: +82-63-254-4833; E-mail: Rheu@jbnu.ac.kr

### Funding Information

No sources of funding were declared for this study.

Received: 27 September 2013; Accepted: 6 July 2014

*Clinical Case Reports* 2015; 3(5): 315–318

doi: 10.1002/ccr3.118

## Introduction

Mature cystic teratoma, the most common type of ovarian teratoma, is also the most common ovarian germ cell-derived neoplasm and it account for ~20% of all ovarian tumors [1]. Endometriosis coexisting with mature cystic teratoma in the same ovary is extremely rare, although both benign conditions are said to be common in women in the reproductive age group. We report a rare case of coexistence of endometriosis and mature cystic teratoma in the same ovary combined with fimbrial ectopic pregnancy.

## Case

A 28-year-old (G1P0) woman was referred to our center with a history of vaginal bleeding of 2 days duration and mild abdominal discomfort. Blood pressure was 112/66 mmHg and pulse rate 80 beats/min. Her abdomen was soft but mild tenderness of the lower abdomen. Serum  $\beta$ -hCG level was 5302.8 mIU/mL. Transvaginal ultrasonography showed an enlarged uterus containing an amorphous thickened endometrium and absence of fetal

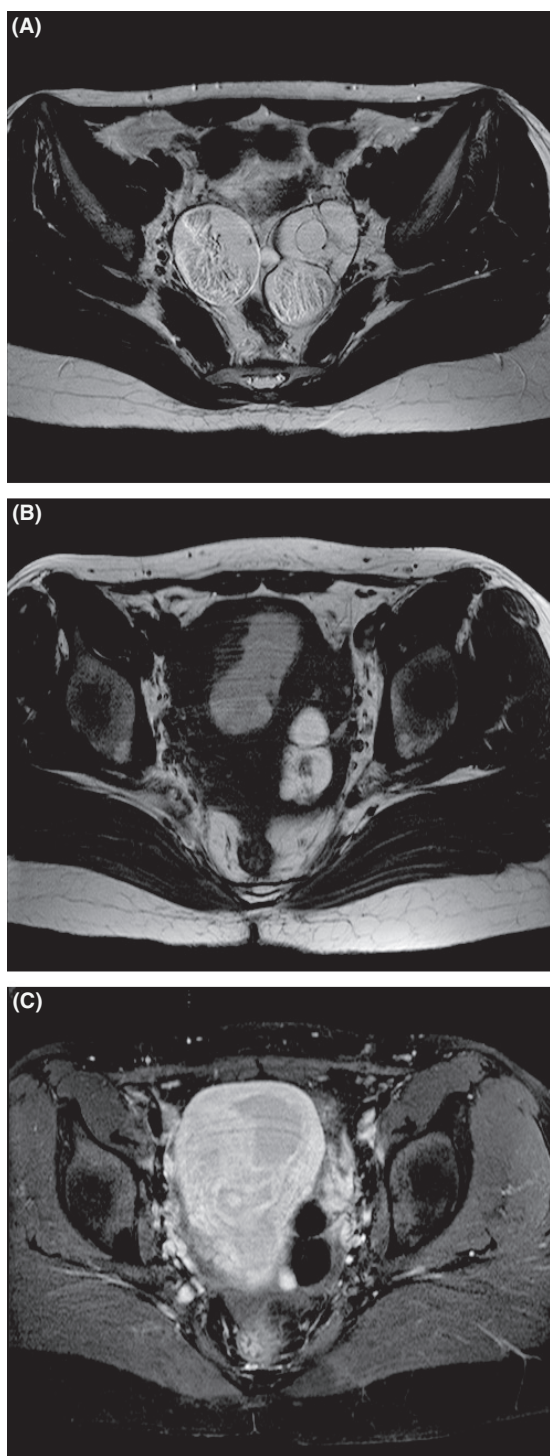
### Key Clinical Message

A coexistence of endometriosis and mature cystic teratoma in the same ovary is a rare occurrence although such tumors of ovaries are said to be common in the reproductive age group. We report a case of fimbrial ectopic pregnancy combined with simultaneous ipsilateral ovarian presentation of endometriosis and mature teratoma.

### Keywords

Endometriosis, fimbrial ectopic pregnancy, ovary, teratoma.

parts. Ultrasound revealed bilateral ovarian masses of ~6 cm, respectively, and a moderate amount of intraperitoneal fluid. Further evaluation with magnetic resonance imaging (MRI) revealed bilateral mature cystic teratomas and irregular nodular thickened endometrium (Fig. 1). After discussion, the patient underwent a laparoscopy. Intraoperative findings include a moderate amount of hemoperitoneum and enlarged ovaries bilaterally. Fibrinous material and clot adherent to the fimbrial end of the left fallopian tube were noted. After careful evacuation of fibrinous material and clot, the bleeding focus was identified in the fimbrial end of the left fallopian tube. As the bilateral tubes appeared to be unaffected by the ectopic pregnancy except for bleeding site of the left fimbrial end, the bleeding site was sutured. The ovarian cysts were then enucleated. Teeth, skin, and hair were found, characteristics of teratoma. The removed fibrinous material and clot were sent for pathological examination and the final histology confirmed the presence of the chorionic villi, consistent with ectopic pregnancy. In addition, on histologic analysis of the bilateral ovarian masses, the diagnosis of bilateral ovarian mature cystic teratomas was confirmed and the simultaneous presence of ovarian endometriosis



**Figure 1.** Bilateral mature cystic teratoma. Axial T2-weighted image without fat suppression (A) shows high signal intensity mass in both ovaries with well-defined margin. Axial T1-weighted (B) images show distended endometrial cavity filled with high signal intensity fluid. On axial gadolinium-enhanced fat saturated T1-weighted image. (C) homogeneous enhancement of the uterus with some enhancing endometrial nodularity is seen.

and mature teratoma in the left ovary was also noted (Fig. 2). To reduce the risk of persistent ectopic pregnancy, systemic administration of methotrexate ( $50 \text{ mg/m}^2$ ) was performed intramuscularly. Three weeks postoperatively, postoperative course was uneventful and the  $\beta$ -hCG decreased to  $32.83 \text{ mIU/mL}$ . But the patient had been lost to follow-up since then.

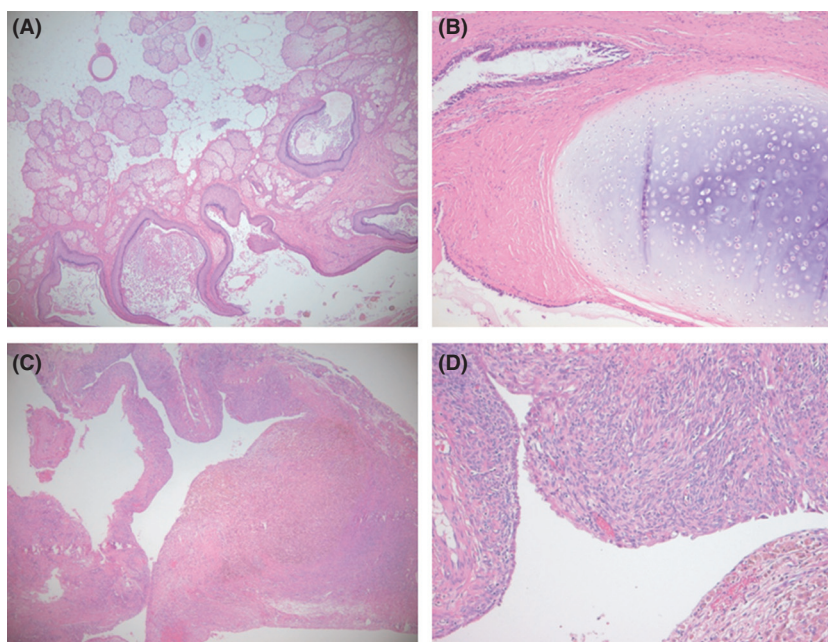
## Discussion

Mature cystic teratoma is a benign, cystic lesion containing tissue from all three embryonic layers; endoderm, mesoderm and ectoderm. Mature cystic teratomas tend to occur at relatively young age in the reproductive years [2]. These tumors are usually asymptomatic until they reach a considerable size. Mature cystic teratomas of the ovary are often discovered by routine physical examination, during radiographic examinations, or during abdominal surgery performed for other indications. When complications such as torsion, rupture, infection, and malignant transformation occur, these mature cystic teratomas present with discomfort, or pain [3].

Endometriosis is a chronic and recurrent disease characterized by the abnormal growth of the functional endometrial glands and stroma outside the uterine cavity, which occurs in  $\sim 10\%$  of women of reproductive age [4]. Although the most common symptoms of endometriosis are dysmenorrhea, dyspareunia, and chronic pelvic pain, many of its symptoms are of non-specific nature.

There are some important aspects to consider when dealing with endometriosis and mature cystic teratoma of the ovary. Although endometriosis is one of the most frequent causes of secondary infertility in young women, the association of teratoma with infertility is considered rare. Microscopically, endometriosis is associated with stromal implants and reduced follicular number and activity, whereas residual ovarian cortex containing several germinal follicles is frequently found even if teratoma is bilateral and very large in size [5]. In addition, the management of ovarian tumors can be challenging because of the risk of a possible malignancy. The frequency of endometriosis in malignant tumors was  $11.2\%$  and endometriosis may be closely related to ovarian tumors such as endometrioid adenocarcinoma, while mature cystic teratomas had a low risk of malignancy ( $0.17\text{--}3\%$ ) [2, 6]. Therefore, conservative ovarian surgery in patients who desire to preserve fertility should be carefully selected based on preoperative evaluation.

The coexistence of mature cystic teratoma and endometriosis in the same ovary is rare. Our literature review identifies only two of these cases, and the primary reports for one of these cases could not be retrieved from the available English literature. This one case is known only



**Figure 2.** Histologic slides of the left ovary. Components of benign cystic teratoma. (A) Tissue types present in this slide include skin, skin appendages, and adipose tissue. (B) respiratory epithelium and underlying cartilage (H&E, 100 $\times$ ). Endometriosis in ovary. (C and D) Endometriotic cyst associated with mature cystic teratoma. (H&E, D: 40 $\times$ , E: 200 $\times$ ).

**Table 1.** Summary of cases of simultaneous occurrence of endometriosis and teratoma in a single ovary reported, including the present case.

|                          | Age (years) | Parity | Symptoms and signs                 | Side      | Treatment                                | Histology  |
|--------------------------|-------------|--------|------------------------------------|-----------|--|--|
| Caruso and Pirrelli [8]  | 28          | 0      | Abdominal pain                     | Lt. ovary | Rt. oophorectomy<br>Lt. cyst enucleation | Teratoma and endometriosis                       |
| van der Merwe et al. [1] | 30          | 0      | Bloatness<br>Stomach swelling up   | Rt. ovary | Rt. oophorectomy                         | Teratoma, endometriosis,<br>mucinous cystadenoma |
| This study               | 28          | 1      | Abdominal pain<br>Vaginal bleeding | Lt. ovary | Both cyst enucleation                    | Teratoma and endometriosis                       |

through its inclusion in a subsequent literature review and is not included in our table [1]. Table 1 summarizes the cases of coexisting endometriosis and teratoma in a single ovary reported, including the present case. Ferrario [7] described the first case report of this coexistence of mature teratomas and endometriosis in a single ovary, which was a coincidental finding in a 23-year-old woman who underwent a bilateral salpingo-oophorectomy for a pelvic mass. Caruso and Pirrelli [8] confirmed histologically the separate endometrial and teratomatous lesions in the left ovary. Also, they stated that this coexistence of teratoma and endometriosis had clinical relevance because an endometriotic pathology could reveal a silent teratoma with bilateral localization [1, 8]. Furthermore, van der Merwe et al. [1] described a case of a large ovarian tumor composed of both endometriosis and mature cystic teratoma in a 30-year-old woman. In the patients with coex-

istence of varied pathology in a single organ, they mentioned that emphasis must be placed on accurate clinical assessment and decisive histologic verification.

The presented case showed co-occurrence of two types of problems: the site of implantation of the ectopic pregnancy in the tubal fimbria and endometriosis coexisting with mature cystic teratoma of the same ovary.

Ectopic pregnancies are mostly located in the fallopian tubes, accounting for 98.3% of all cases. Incidence of implantation in the ampulla, isthmus, fimbrial end, and interstitial (corneal) region is 79.6%, 12.3%, 6.2%, and 1.9%, respectively. Also, ectopic pregnancy outside the fallopian tube; only 1.4% of ectopic pregnancies are abdominal pregnancies, 0.15% ovary or 0.15% cervical [9]. Early detection of fimbrial ectopic pregnancy is often difficult because of the rarity and the asymptomatic nature before intra-abdominal hemorrhage from the fimbria

occurs. Transvaginal ultrasonography is the initial modality of choice to diagnose ectopic pregnancy. However, when transvaginal sonographic findings are indeterminate, MRI is potentially useful [10]. However, unusual types of ectopic pregnancy such as fimbrial ectopic pregnancy are difficult to ascertain, especially with the presence of ovarian cysts as in this case. The laparoscopic approach is preferred as a safe and feasible surgery offers a short hospital stay, little or no postoperative care, and allows a swifter recovery time and the majority of ectopic pregnancies can be managed this way. The management for fimbrial ectopic pregnancy must be individualized, according to the state of the affected fallopian tube and desire for future fertility. Our patient was thought that the tubal damage of fimbrial ectopic pregnancy was minimal and not significant enough to be resulted in loss of tubal function. Therefore, laparoscopic suturing after removal of an ectopic pregnancy rather than salpingectomy was performed. The incidence of persistent ectopic pregnancy can be significantly reduced after a single prophylactic dose of systemic methotrexate administered postoperatively [11].

In conclusion, this is the unique case, given the non-specific nature of symptoms, found to have a fimbrial ectopic pregnancy as well as a coexistence of endometriosis and mature cystic teratoma of the same ovary. Because endometriosis is a chronic disease with high recurrence rates, the management of this rare case documented the simultaneous presence of endometriosis and mature cystic teratoma in the same ovary is faced by unresolved questions such as whether intervention is surgery alone or in combination with medical therapy. Coexistence of varied pathology in a single organ makes it even more complex to justify one approach over another, especially for further follow-up. Close follow-up of this patient is imperative, given the risk of persistent ectopic pregnancy and the risk of future recurrence of endometriosis.

## Conflict of Interest

None declared.

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