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CASE REPORT

- 8 Association of endometrioma with ovarian teratoma and mucinous cystadenoma in a patient diagnosed with endometriosis: A case report
Rokhgireh S, Mehdizadehkashi A, Tahermanesh K, Gorginzadeh M

REVIEW

- 13 Primary extragenital mixed malignant Mullerian tumour presenting as a painful splenic mass: A case report and review of the literature
Kwok AMF

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Association of endometrioma with ovarian teratoma and mucinous cystadenoma in a patient diagnosed with endometriosis: A case report

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Abstract

BACKGROUND

Co-occurrence of different tumor types in a same patient's ovaries diagnosed with endometriosis is a rare phenomenon.

CASE SUMMARY

In this article we present an uncommon association of three distinct ovarian pathologies in a same woman presenting with adnexal mass. A 31-year-old nulliparous woman with a large persistent adnexal mass underwent laparoscopic surgery. Imaging demonstrated a multi-cystic mass with internal echoes. Tumor markers were within normal range. Based on histopathologic assessment, benign mucinous cystadenoma and mature cystic teratoma of the right ovary together with endometrioma of left ovary were revealed.

CONCLUSION

In cases of large adnexal mass, the of existence of more than one tumor type and the involvement of the contralateral ovary is possible. Also, the possibility of concurrent underlying malignancy or diminished ovarian reserve should be kept in mind.

Key words: Endometriosis; Ovary; Teratoma; Mucinous cystadenoma; Laparoscopy; Case report

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Core tip: Existence of three distinct tumor types in the ovary is a rare phenomenon. This manuscript presents scarce association of endometrioma with teratoma and mucinous cystadenoma which was confirmed by histologic assessment.

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INTRODUCTION

Though its true incidence is not defined yet, endometriosis affects approximately 10% of women in their reproductive life^[1,2]. Endometrioma is considered as a common manifestation with the incidence of more than 40% in women diagnosed with endometriosis^[1-3]. According to a large study by Grammatikakis *et al*^[3], endometriosis was the most common diagnosis amongst 1522 women with adnexal mass in whom laparoscopy was performed. Coincidence of endometrioma which is of epithelial origin with other ovarian tumors and particularly those with a different cellular origin, however, is not frequent. Dermoid cysts or mature cystic teratomas are regarded as the most frequent germ cell tumors of the ovary and mucinous cysts are the second most common masses of epithelial origin^[4,5]. Only few cases of the endometrioma concurrence with other epithelial masses like mucinous cystadenoma or germ cell tumors have been reported in the literature^[4-7]. It is well established that the pathogenesis of ovarian endometriosis could harbor malignant transformation^[8]. Although endometriosis is not considered a premalignant condition, based on epidemiologic, histopathologic, and molecular data, it may lead to some specific subtypes of ovarian cancer particularly for atypical endometrioma or lesions larger than 10 cm^[6,8]. Generally, there is a 2 to 3 fold increase in the risk of ovarian malignancy in patients suffering from endometriosis with the endometrioid and clear cell carcinoma as the most common subtypes^[8,9]. According to a study by Pearce *et al*^[9], there is also a significant association between the pre-existing endometriosis and low-grade serous ovarian malignancy. This study presents the coincidence of three different ovarian cellular growths in a same woman who underwent laparoscopic surgery for management of a large adnexal mass.

CASE PRESENTATION

Chief complaints

A 31-year-old woman presented with progressive dysmenorrhea and abdominal fullness.

Laboratory examinations

Laboratory examination indicated serum tumor markers including CA-125, HE4, CA19-9, AFP, CEA and HCG within normal range. Due to the reason that she had no children and had a low ovarian reserve, she was referred to the infertility clinic for oocyte preservation first.

Imaging examinations

Trans rectal ultrasound reported a tubular multicystic lesion with a size of 9 cm suggestive of hydrosalpinx in the right adnexa accompanied by a small endometrioma in the left ovary (Figure 1).

FINAL DIAGNOSIS

According to pelvic MRI which was requested three months later, a 95 mm × 90 mm cystic area was revealed in the midline with internal septae. The second trans rectal ultrasound revealed a multi cystic mass with internal echoes and the size of 11 cm suggestive of mucinous cystadenoma. Finally, she was scheduled for laparoscopic surgery due to the increase in the size of the mass. During operation, a huge cystic mass was uncovered occupying the whole pelvic cavity.

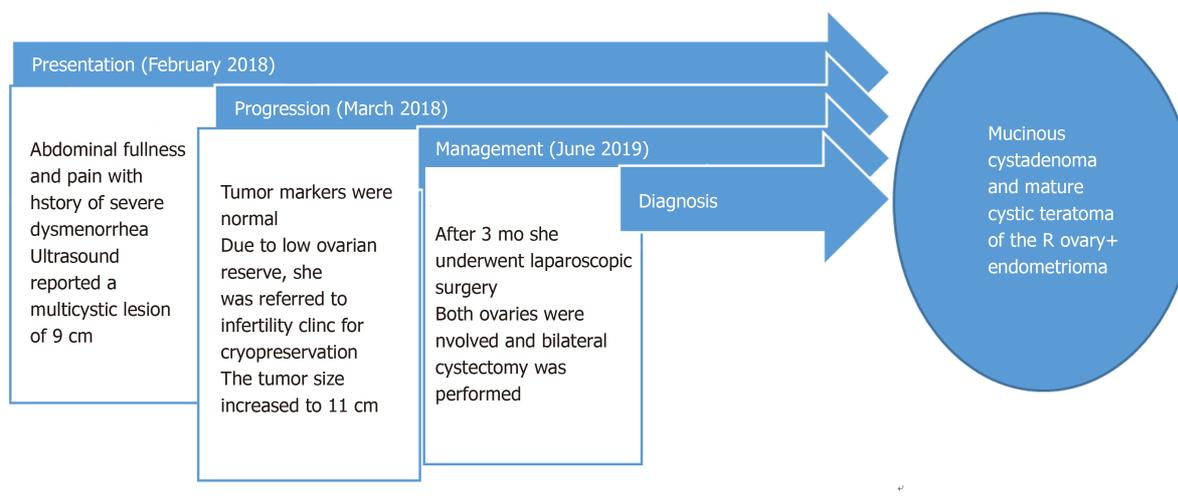


Figure 1 Timeline of the patient's progression of her disease.

TREATMENT

After obtaining the fluid sample for cytological assessment, the cyst was aspirated through the suprapubic port without any spillage of its content (Figure 2). Then, the ovaries were appeared. Another lesion was found which was adhered to the first cyst and originated from the right ovary (Figure 3A). A small endometrioma was also detected in the left ovary (Figure 3B).

OUTCOME AND FOLLOW-UP

Cystectomy was performed and all these lesions were sent for pathologic evaluation. Moreover, the uterosacral endometriotic nodule (histologically confirmed) was excised. Microscopically, 3 tumor types were revealed: Mucinous cystadenoma together with mature cystic teratoma of the right ovary and endometrioma of the left ovary.

DISCUSSION

This study presented a case of the simultaneous mucinous tumor associated with mature cystic teratoma and endometriosis in the same patient ovaries in her reproductive age. Although each of these cystic pathologies are commonly managed by laparoscopic approach, their co-existence together is rare especially when there are more than two tumor types^[4-7]. There are a few studies in the literature on the simultaneous occurrence of two ovarian histopathologic findings in a same ovary of a patient or the ovaries of a same patient^[4-7]. This study was a collision tumor case (consisting of mucinous cystadenoma and dermoid) of the right ovary accompanied by an endometrioma of the left ovary. Collision tumors are uncommon tumors composed of two or more histologically distinct neoplasms coinciding at the same organ^[7]. However, the specific feature of this case is the coexistence of three different ovarian pathologic findings in a patient suffering from endometriosis. There was also a similar study by Joo *et al*^[4] in which three different tumor types including borderline mucinous tumor, mature cystic teratoma, and endometriosis in the same ovary were revealed. Their case was a 42-year-old woman with severe dysmenorrhea and a huge unilateral ovarian mass^[4]. For our case, endometrioma was derived from the left ovary and mature cystic teratoma adhered to a large mucinous cystadenoma was related to the right ovary. Chae *et al*^[5] in another study described a 28-year-old Gravida 1 woman with bilateral ovarian cysts who underwent laparoscopic surgery due to her persistent vaginal bleeding and positive serum β -hCG (more than 5000 IU) suspicious of ectopic pregnancy. Imaging evaluation was in favor of bilateral mature cystic teratomas which was confirmed by final histologic assessment but involvement of the left ovary by endometriosis was also reported^[5]. For all these cases, simultaneous presence of different tumor types was affirmed only after histopathologic examination. For the Chae *et al*^[5] case, the surgeons did not expect to be encountered

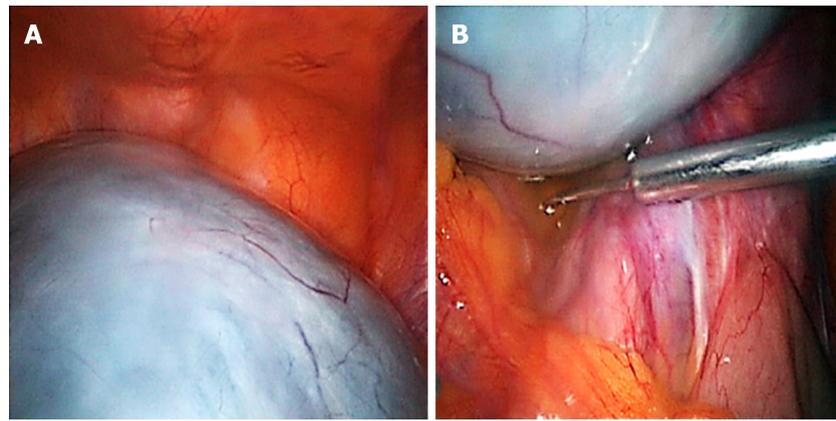


Figure 2 Large mucinous cystadenoma of the right ovary. A: Laparoscopic view following entry; B: Cytological sample was obtained from pelvic fluid.

with more than one ovarian pathology based on imaging. The type of ovarian tumor or cyst is of paramount importance concerning the ovarian reserve. While large or bilateral dermoid cyst do not significantly alter the ovarian reservoir, even a small endometriotic implant could jeopardize the future fertility of a woman^[10,11]. Our patient declared a history of aggravating dysmenorrhea. In addition, her anti-müllerian hormone level was only 1.05 ng/mL, both of which were compatible with the diagnosis of endometriosis. Hence, some measures were taken before she was scheduled for the surgery. First, she was referred to the infertility clinic for oocyte cryopreservation. Furthermore, with respect to the mass size (> 10 cm) which was suspicious of malignancy, written informed consent was obtained from the patient including the possibility of salpingo-oophorectomy. Endometriosis concurrence with other gynecological diseases like adenomyosis has been noted in the literature^[4,5,11]. Matalliotaki *et al*^[11] evaluated 1000 women with endometriosis during a 10-year period. More than 90% of their patients were also diagnosed with other benign conditions with adenomyosis, leiomyoma and benign ovarian cysts at the top of the list. Under the influence of genetic, environmental and inflammatory factors, benign tumors can occur in a same woman^[7,11,12]. However, the perplexing nature of endometriosis with various underlying mechanisms further could complicate the process of disease in women^[8-10]. Hence, as Matalliotaki *et al*^[11] reported, women suffering from endometriosis should be informed of the possibility of being diagnosed with other benign conditions. Another finding in their study was the left ovary predominance for developing endometrioma which was also consistent with our case^[12].

Each constituent in a collision tumor has its own biologic behavior which could harbor malignancy^[7]. Since the appropriate management could differ depending on the most aggressive component, the exact diagnosis is quite important and should be reaffirmed^[4-7]. Patients diagnosed with these tumors should be followed strictly due to the added potential of malignant transformation of different cell types^[6,7]. Also, more emphasis should be given to those who are suffering from endometriosis as they are at risk for recurrence and further progression of disease like this case^[2,6,10].

CONCLUSION

In conclusion, considering the scarcity of coincidence of more than two histopathologic findings related to the ovary, special attention should be given to appropriate diagnosis, management and long-term follow-up of patients suffering from these types of tumors in order to increase their quality of life.

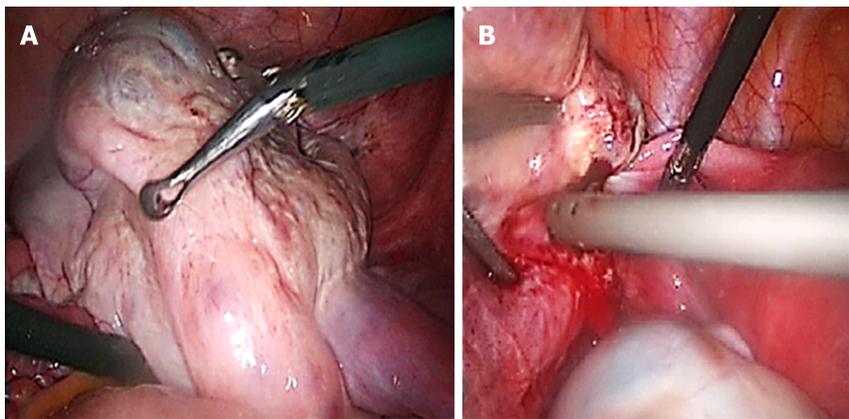


Figure 3 Laparoscopic views of ovaries after aspiration of the cyst. A: Dermoid cyst adhered to the mucinous cystadenoma of the right ovary; B: Endometrioma of the left ovary.

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