Endometrioma Coexisting with Dermoid Tumor in a Single Ovary Presenting as Atypical Endometrioma

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ABSTRACT

Introduction: Ovarian endometrioma, the clinical phenotype of endometriosis, is found in 17% to 44% of patients with endometriosis and is related to the severe form of the condition. The observation of endometrioma and dermoid tumor in the same ovary can be evaluated as atypical endometrioma or ovarian cancer in ultrasound findings.

Case Description: The objective of this report is to present two cases in which an endometrioma and a dermoid tumor are present in the same ovary and to review the subject together with the results of 10 previously reported cases in the literature.

Discussion: The ultrasound findings in atypical endometriomas and endometriomas that mimic ovarian tumors are combinations of papillary projections, hyperechoic irregular walls, a distinct solid mass, and calcifications. Magnetic resonance imaging is the second option for patients who are ultrasonographically identified as having an indeterminate adnexal mass, and it can provide additional information in distinguishing between the two pathologies within the same ovary. Due to suspected malignancy, unnecessary tests and gynecologic oncology consultations are recommended in cases of atypical endometrioma. This may lead to an increase in the frequency of unnecessary midline laparotomy.

Key Words: Endometrioma; Dermoid cyst; Ultrasonography.

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INTRODUCTION

Ovarian endometrioma, the clinical phenotype of endometriosis, is found in 17% to 44% of patients with endometriosis and is related to the severe form of the condition. It is thought that endometriomas, evolve by the invagination of the cells over the ovarian serosa, which results in the remodeling of the ovarian cortex with successive ovulations.[1]

The typical endometrioma, which displays a ground-glass pattern, is defined as a unilocular or multilocular (fewer than 5 locules) cyst with low-level homogeneous echogenicity; only 50% to 65% of identified endometriomas are

"typical."[2] The diagnosis of an endometrioma with transvaginal ultrasonography is not generally problematic for typical endometriomas. The observation of a papillary projection inside the endometriotic cyst on ultrasonography is the most frequent reason for the diagnosis of atypical endometrioma. The absence of vascular flow in these papillary projections, observed by ultrasonography, differentiates an atypical endometrioma from an ovarian malignancy.[2] The common ultrasound findings in atypical endometriomas and endometriomas that mimic ovarian tumors are papillary projections, hyperechoic irregular walls, distinct solid masses, calcifications, or any combinations of these[3] (**Figure 1**). Magnetic resonance imaging (MRI) is the second option for patients who are ultrasonographically identified as having an indeterminate adnexal mass, and it can provide additional information in distinguishing between the two pathologies within the same ovary.[4] Although endometriomas and dermoid tumors are rarely observed in the same ovary, when seen, many of the ultrasonographic findings observed in atypical endometriomas and the endometriomas that mimic ovarian cancer are observed.[5]

The objective of this report is to present two cases evaluated as atypical endometriomas using transvaginal ultrasonography, wherein both endometriomas and dermoid tumors are present within the same ovary. The subject is also reviewed together with the results of 10 previously reported cases in the literature.

CASE DESCRIPTION

Case 1

A 25-year-old patient, with a 2-year history of primary infertility and who desired a pregnancy, presented to our clinic with pelvic pain and mild-to-moderate dysmenorrhea. The patient had no history of previous surgery or chronic illnesses. In the speculum examination, the cervix and vagina were normal and no nodules were detected in the rectovaginal septum. In the bimanual digital examination, a mass in the left adnexal and Douglas pouch was felt. In the transvaginal sonography, a cystic mass, 13 × 11 × 10 cm in size, containing bilocular, hypoechoic, and hyperechoic areas was observed in the left adnexal area (**Figure 2**). An MRI scan was conducted and the patient was evaluated as having an atypical endometrioma. In the T1-weighted (T1W) and the fat-suppressed T2-weighted (T2W) sequences, the 5-cm hyperechoic part of the mass was reported as a dermoid cyst and the 8-cm part as an endometrioma (**Figure 3**). In laboratory tests, the white blood cell count was 5600/mm^3, hemoglobin level was 11.2 g/dL, and CA-125 was 63 U/mL. The other tumor markers were within normal limits.

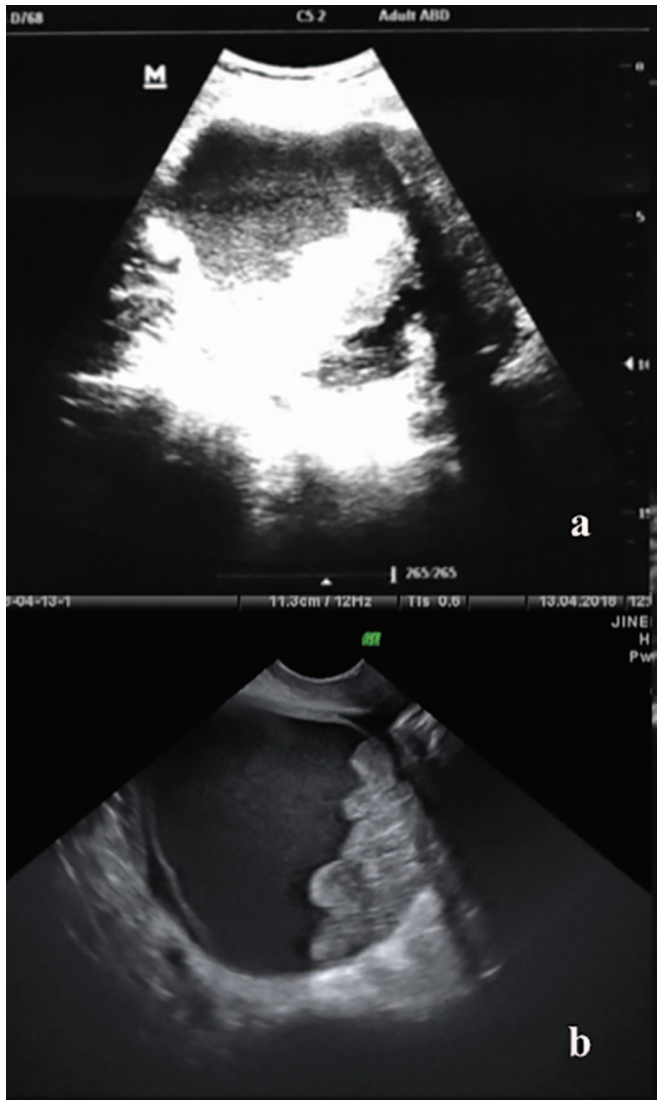

Figure 1. (**A**) Atypical endometrioma with papillary projection. (**B**) Images of ovarian cancer with papillary projections bearing resemblance to atypical endometrioma.

The decision was taken to perform a laparoscopy for this patient. In the intra-operative view, dense adhesions were present around the cyst. Adhesiolysis was performed. The dermoid cyst was removed from the abdomen using an endobag (Lagis Enterprise Co. Ltd, Taichung, China). There were pathologic structures compatible with teratoma in the removed material. Following the removal, the cyst was excised by peeling the endometrioma capsule. Upon histopathologic examination of the cystectomy material removed, macrophages infiltrating the endometrial

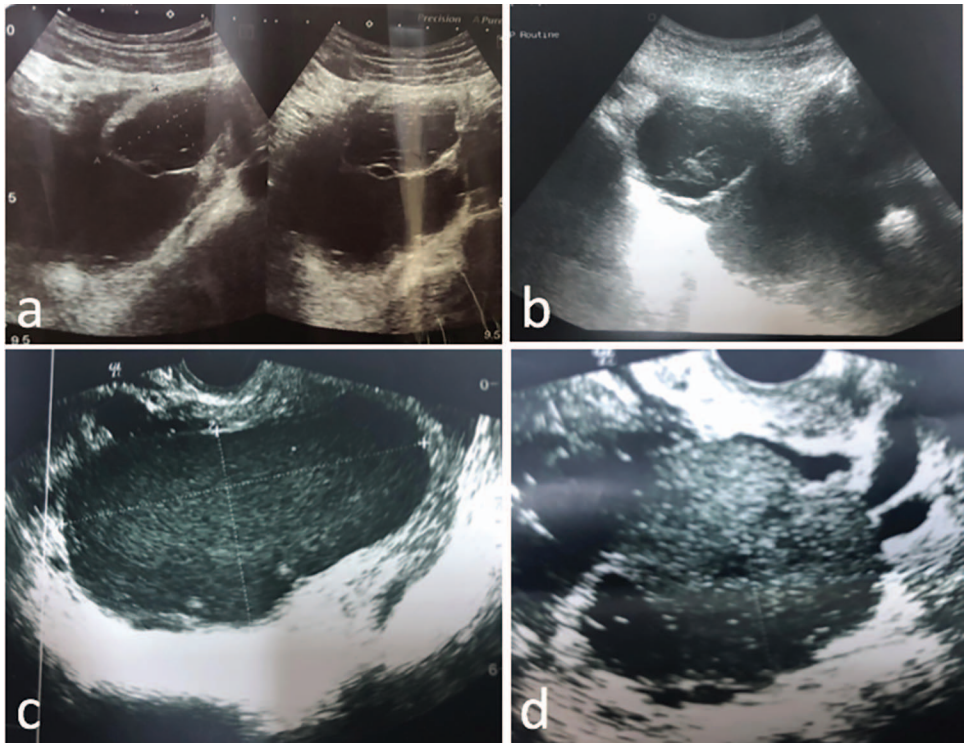

Figure 2. (**A**, **B**) Transabdominal, and (**C**, **D**) transvaginal sonographic images of Case 1. (**A**, **B**) Indeterminate adnexal mass; (**C**) endometrioma part of adnexal mass; (**D**) dermoid cyst.

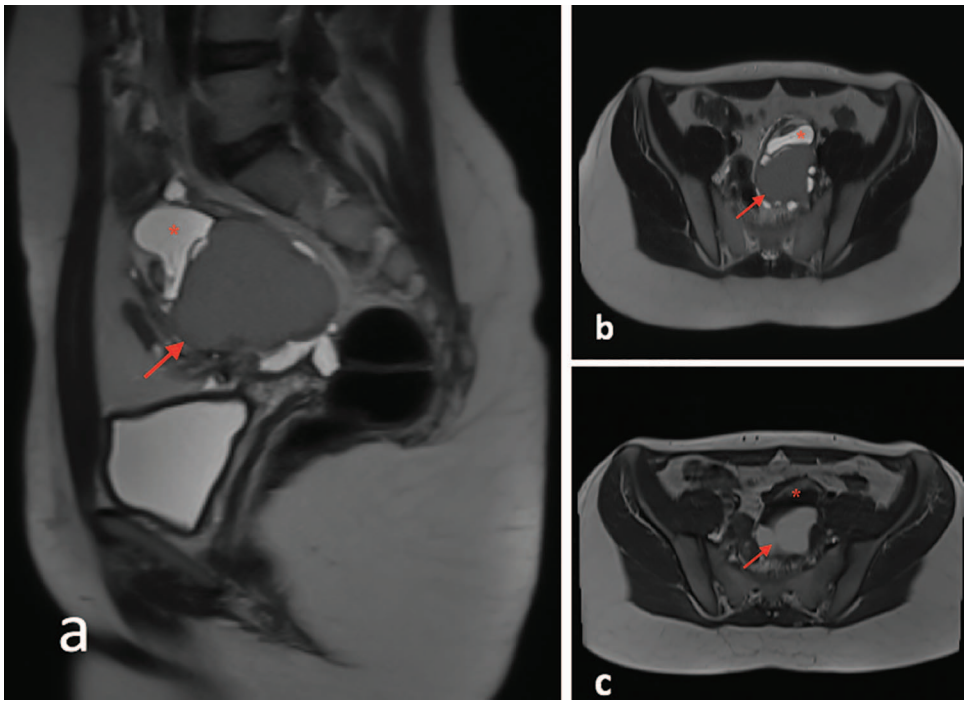

Figure 3. MR images of Case 1. (**A**) Sagittal and (**B**) axial T2-weighted, and axial (**C**) T1-weighted MR images show an endometrioma (arrow) and a dermoid cyst (*) within the same ovary.

epithelium, stromal cells, and hemosiderin, were detected, supporting a diagnosis of endometriosis. In the other part of the removed material, skin appendages, sebaceous glands, and cystic teratomas containing fat tissues were observed. No complications occurred during or after the surgery and the patient was discharged on the first postoperative day. The patient became pregnant spontaneously in the third postoperative month.

Case 2

A 22-year-old virgin patient consulted the outpatient clinic due to pelvic pain. She had dysmenorrhea irresponsive to nonsteroidal anti-inflammatory drug treatment. Eight years previously, ovarian torsion and necrosis induced by a dermoid tumor in the right ovary were observed and the right ovary was removed. By transabdominal pelvic ultrasonography, a cyst consisting of 6-cm hypoechogenic and hyperechogenic areas were observed in the left adnexal area (**Figure 4**). Although the first clinical diagnosis was compatible with endometrioma, hyperechogenic pathologic structures resembling a teratoma were noted inside the cyst. The ultrasonographic images of the patient were evaluated as an atypical endometrioma. An MRI was performed to provide diagnostic clarity and the patient was diagnosed as having an endometrioma and dermoid tumor in the same ovary (**Figure 5**). The white blood cell count was 4400/mm^3 and hemoglobin level was 13.1 g/L in the laboratory analysis. A laparoscopy was decided upon for the patient, revealing intra-operatively a 6-cm adnexal mass in the left adnexal area. The cyst contents, resembling liquid chocolate and compatible with a diagnosis of an endometrioma, were drained upon incision of the cyst. The 2-cm cystic structure was removed by peeling its capsule. Following this procedure, the components of the 4-cm dermoid cyst were removed from the abdomen using an endobag (Lagis Enterprise Co. Ltd, Taichung, China). The extracted material comprised pathologic structures (e.g., hair, teeth). In the material removed after cystectomy, the histopathologic examination showed macrophages infiltrating the endometrial epithelium, stromal cells, and hemosiderin, supporting the diagnosis of endometriosis. In the remaining extracted material, skin appendix, sebaceous glands, and cystic teratomas containing fat tissues were observed.

DISCUSSION

Endometriosis is a benign chronic inflammatory disease that usually manifests as pain and infertility. It is characterized by the presence of endometrium-like stroma and glands outside the uterine cavity.[6] The prevalence of endometriosis can reach up to 25% to 40% in infertile women, a 10-fold increase compared with that in the fertile population. Its prevalence is between 4% and 65% in women with chronic pelvic pain.[7] Among the locations where endometriosis is noted, the most frequent are the ovaries, peritoneum, and uterine ligaments, and less frequently the urinary bladder, vagina, and digestive system. The diagnosis is predominately confirmed based on histopathological findings, and often, the diagnosis is made based on visual assessment.[8,9] Endometriomas, also known as "chocolate cysts," are cystic masses arising from ectopic endometrial tissue over the ovarian cortex.

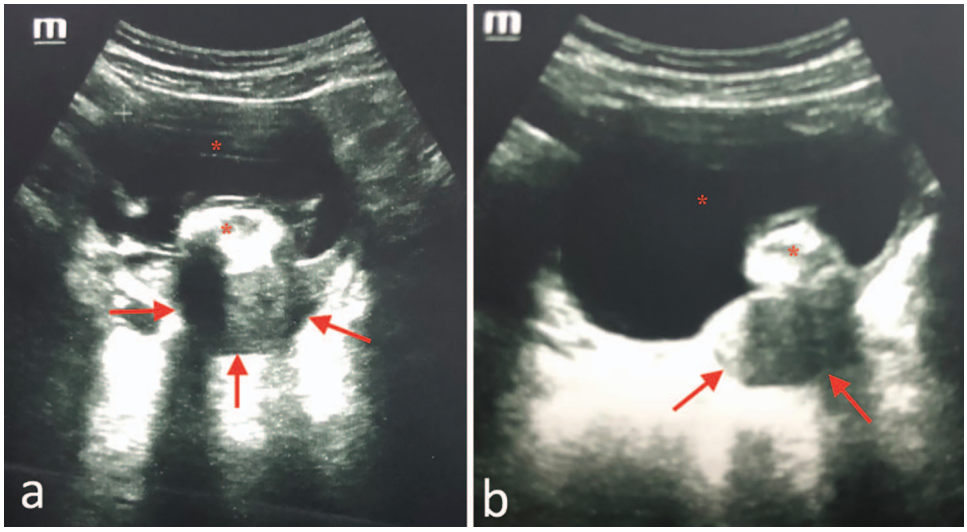

Figure 4. Sonographic image of the left adnexal mass of Case 2. Dermoid cyst (*); endometrioma (arrow).

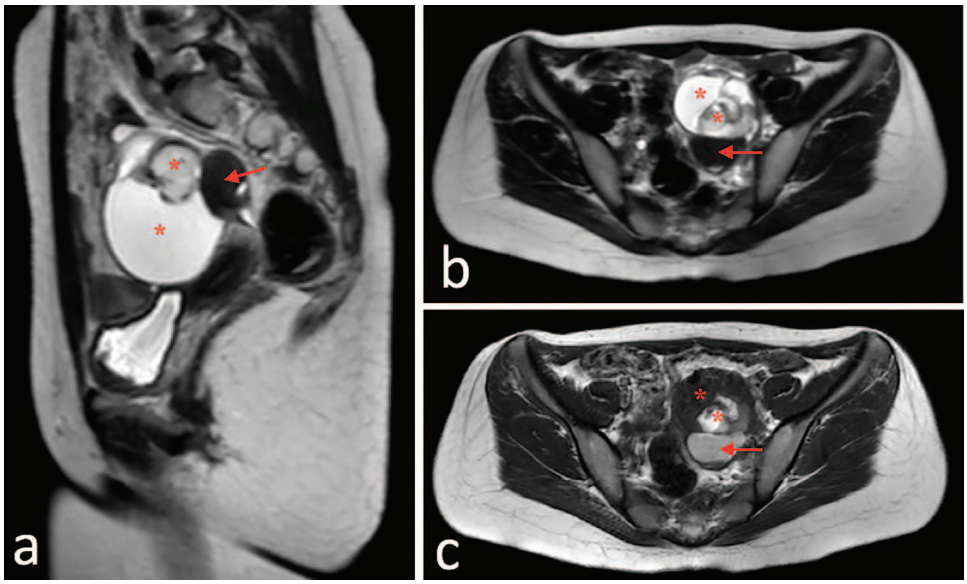

Figure 5. MR images of Case 2. (**A**) Sagittal and (**B**) axial T2-weighted MR images show an endometrioma (arrow) and a dermoid cyst (*) within the same ovary. Axial (**C**) T1-weighted MR image shows an endometrioma (arrow) and a dermoid cyst (*).

Mature cystic teratomas are benign cystic lesions consisting of all three of the embryonic layers (endoderm, mesoderm, and ectoderm). Teratomas are the most frequently observed germinomas and more than 80% of cases are noted in the reproductive period.[10,11] They constitute approximately 43%–70% of benign ovarian tumors and 15% of all ovarian tumors.[12] Mature cystic teratomas can be discovered during routine physical exams, radiographic exams, or incidentally during abdominal operations performed for other indications. These tumors are usually asymptomatic until they reach a certain size. They can become symptomatic upon ovarian torsion, rupture, infection, or malignant transformation.[13] These tumors are rarely seen in combination with other ovarian masses.

Studies show the effectiveness of ultrasonographic evaluation in diagnosing ovarian cysts.[9] The ultrasound examination findings of endometrioma can overlap with that of other adnexal masses, including hemorrhagic cysts, tuboovarian abscess, and dermoid and cystic ovarian neoplasm. Many studies have attempted to define the typical ultrasound characteristics of endometrioma. Sonographically, the most typical appearance of endometrioma is a hypoechoic ovarian mass that increases through transmission. In addition, homogenous low-to-moderate level echo is often observed within the ground-glass pattern. The ancillary findings of endometrioma are punctate echogenic foci located inside the walls, due to blood clotting and nodules that appear to be solid.[9] Many cases of endometrioma exhibit shadows of low-to-moderate intensity signal, depending on the different stages of blood products inside the cyst. The most frequent false diagnoses are hemorrhagic cysts and dermoid cysts. Echogenic areas related to posterior shadowing with focal or diffuse intensity are often present in dermoid cysts. A posterior acoustic shadow related to cystic echo pattern and an intense echogenic wall, hyperechoic bright lines and spots in the dark areas, tape-like echogenicities, and an intense echogenic area related to posterior acoustic shadowing with or without cystic components are the characteristics of a cystic teratoma.[9]

Laparoscopic surgery indications for adnexal masses depends on the patient's age, pelvic examination findings, imaging methods, and results of the tumor marker test.[14,7] The management of benign ovarian tumors can be challenging due to the risk of malignancy. Malignant transformation is a rare complication that is well defined but is estimated to be observed in approximately 1% of endometriomas.[15] Women with a history of endometriosis have a 4.2-fold increase in ovarian cancer risk compared with that in other women.[16] The most frequent histologic findings are clear-cell carcinoma and endometrioid carcinoma related to glandular elements. Less frequently, endometrial stromal sarcoma can be caused by stromal elements. The malignancy risk of mature cystic teratomas is approximately between 0.17% and 3%.[11,17] Therefore, the indications should be carefully considered when deciding whether to perform conservative surgery (i.e., cystectomy or cyst drainage) based on preoperative evaluation, espe-

cially when the woman has a desire for pregnancy in the future.

The observation of endometrioma and dermoid tumor in the same ovary can be mistaken for an atypical endometrioma or ovarian cancer with ultrasound examination. In the differential diagnosis, the absence of vascular flow in the papillary projection and MRI may be helpful. The latest MRI protocol for the endometriosis diagnosis includes T1 and fat-suppressed T2W sequences. In fat-suppressed MRI, the loss of signal intensity inside the T2W hyperintense adnexal mass is characteristic of mature cystic teratoma.[4] In addition, the saturation of the high-signal intensity of the fat increases the differences between the T2-hyperintense structures that do not contain fat and enables a more sensitive detection of smaller endometriomas.[18] Due to the strong suspicion of malignancy, unnecessary tests and gynecologic oncology consultations can occur in the case of atypical endometriomas.[3,5] This may lead to an increase in the frequency of unnecessary midline laparotomy.[3]

Although mature cystic teratomas and endometriomas are frequently seen pathologies in women of reproductive age, the number of cases reported in the literature where both pathologies are seen in the same ovary is fairly low[8,19] (**Table 1**). In 1960, both pathologies were incidentally detected in the same ovary of a 23-year-old female patient and a salpingo-oophorectomy was performed by Ferrario et al.[20] The other case was of a 28-year-old patient with a bilateral ovarian mass presented by Caruso et al.[21] In this case, the presence of endometrial and teratomatous lesions was shown in the histopathologic analysis.[21] A similar case, involving a patient with pelvic endometriosis and bilateral ovarian dermoid cysts, was presented by Frederick et al[11] in 2003. Van Der Merwe et al[8] reported a case of a 30-year-old patient with a large ovarian tumor with both endometriosis and mature cystic teratoma. In 2013, Prororic et al[22] documented a bilobular complex cyst detected in the left ovary of a 33-year-old infertile patient

Table 1.
Summary of Reported Cases of Endometrioma Coexisting with Dermoid Tumor in a Single Ovary

	Age, Years	Symptom and Sign	Side	Preoperative Diagnosis	Treatment	Histology
Wagners et al,[25] 2015	34	Pelvic pain dysmenorrhea	Bilateral	No	Laparoscopic cystectomy, hysterectomy	Dermoid tumor and endometriosis
Chen et al,[23] 2011	35	Abdominal pain	Right	No	Laparoscopic cystectomy	Dermoid tumor and endometriosis
Frederick et al,[11] 2003	NS*	Pelvic pain	Bilateral	No	Laparoscopic cystectomy	Dermoid tumor and endometriosis
Chae et al,[24] 2015	28	Abdominal discomfort	Bilateral	No	Laparoscopic bilateral cystectomy, salpingectomy	Dermoid tumor and endometriosis
Caruso et al,[21] 1997	28	Abdominal pain Acute abdomen	Bilateral	No	Left cystectomy, right oophorectomy	Dermoid tumor and endometriosis
Van der Merwe et al,[8] 2010	30	Abdominal bloating, swelling	Right	No	Midline incision, right oophorectomy	Dermoid tumor and endometriosis, mucinous cystadenoma
Prorocic et al,[22] 2013	33	Infertility	Left	No	Laparoscopic cystectomy	Dermoid tumor and endometriosis
Ferrario et al,[20] 1960	23	NS†	NS†	NS†	Bilateral salpingo-oophorectomy	Dermoid tumor and endometriosis
Hwang et al,[26] 2017	22	Flank pain	Bilateral	No	Laparoscopic cystectomy	Dermoid tumor, struma ovarii and endometriosis
Taylor et al, 2014	33	Attended-in ultrasonography pregnancy	Right	No	Caesarean section and Salpingo-oophorectomy	Dermoid tumor and endometriosis

*, Not specified.

†, non-English literature.

that was found during her preoperative imaging; the cyst comprised a dermoid cyst and endometriosis.

In 2011, Chen et al[23] performed a laparoscopic excision of bilateral dermoid tumor and a left endometrial cyst in a patient diagnosed with acute abdominal pain. In 2013, Chae et al[24] reported the findings of bilateral dermoid cyst and left endometrioma during laparoscopy performed for ectopic pregnancy. In 2015, Wagner et al[25] detected the coexistence of endometrioma and dermoid cyst in a 34-year-old patient with pelvic pain and performed a laparoscopic hysterectomy and cystectomy in the patient who had completed her family and had no desire of child further. In 2014, Taylor et al[5] showed the coexistence of endometrioma and dermoid cyst on ultrasonography in a 33-year-old patient who was 12 weeks pregnant. Gynecology and gynecological oncology departments were both involved in the followup and management of this patient and in the 38th week of her pregnancy, a C-section and right salpingo-oophorectomy were performed. The difficulty in establishing a final diagnosis preoperatively and the risk of ovarian malignancy made the management difficult. In 2018, Hwang et al[26] reported the coexistence of endometrioma and bilateral dermoid in a 22-year-old patient with flank pain in the histopathological evaluation when struma ovarii was detected. An accurate preoperative diagnosis was not possible in any of the previously reported cases.

Similar to our cases, the patients in previously published studies were of reproductive age and their ages ranged from 22 to 35 years. Our first case was a 25-year-old woman with infertility and the second was a 22-year-old virgin female with pelvic pain. Of the 10 patients in prior studies, 7 (70%) presented with pelvic pain. Cysts were detected incidentally in two of the remaining 3 patients.[5,22] The last patient complained of abdominal bloating.[8] In our first patient, the main complaint was pelvic pain. The other patient's cysts were discovered during the evaluation for infertility.

In the evaluation of adnexal structures, ultrasonography was used in 9 patients and MRI was additionally used in 3 patients.[5,24,26] Adnexal mass was considered to be indeterminate in 6 patients based on ultrasonography and malignancy was suspected. Among these patients, the case presented by Taylor et al[5] raised the suspicion of malignancy because, besides having an uncommon dyad of endometrioma and dermoid cyst, there was extensive vascularization in the decidua of the endometrioma due to pregnancy.

When the locations of ovarian cysts were examined, 6 (60%) patients in previous reports had bilateral dermoid tumor. Cystic masses were found on the left in 1 patient and the right in 3 patients (**Table 1**). In our case, cystic masses were observed in the left ovary.

In our first case, the mass in the left ovary could not be evaluated by transvaginal ultrasonography because it was approximately 13–14 cm in size and was present outside of the true pelvis. Since transabdominal ultrasonography revealed an atypical endometrioma, MRI was requested because of suspected malignancy. MRI showed a 66 × 81-mm hemorrhagic lesion, hyperintense on T1W and isointense on T2W images, probably consistent with an endometrioma. There was another cystic mass in the anterior proximity of the left ovary that was hypointense on T1W and had hypo-iso-hyperintense components on T2W images, with a diameter of 57 × 45- mm and consistent with dermoid cyst in T2 fat-suppressed sequences.

In our second case, only transabdominal ultrasonography could be performed because the patient was a virgin. The patient did not provide approval for a transrectal ultrasonography. An MRI was performed because of the suspicion of malignancy due to the findings of abdominal ultrasonography, and an adnexal mass of 84 × 76 mm and a solid mass with a thick septum were noted. MRI showed a cystic mass in the left ovary approximately 45 × 51 mm in size compatible with a diagnosis of a dermoid, with nodular signals consistent with hyperintense fat in T1W sequences and in T2W fat suppressed sequences. Inferior to the cyst, there was a cystic structure resembling an endometrioma 34 × 21 mm in size, with hypointense signals on T1W and hypo-isointense signal on T2W images. The cystic mass could be compatible with endometrioma with a hypo-isointense hemorrhagic component in the T2W images.

In the report by Hwang et al,[26] a 2.8-cm cystic mass, hyperintense on T1W and hypointense on T2W images, and a cystic mass with a high fat content in T1W images of 1.4 × 2.7 cm in size were reported. An operation was recommended on the grounds that these 2 masses gave rise to the suspicion of malignancy. In the case presented by Chae et al,[24] the patient had an ovarian mass approximately 6 cm in size and moderate intraperitoneal fluid was in the ultrasonography, and thus, MRI was requested with the suspicion of ovarian cancer. MRI revealed bilateral mature cystic teratoma and irregular nodular and thickened endometrium. In laparoscopy, the fluid was speculated to have been given rise to by the rupture of an ectopic pregnancy and a salpingectomy with bilateral cystectomy was performed. Taylor et al[5] reported the case of a pregnant woman with an endometrioma harboring projections in the lower, upper, and

posterior walls of the cyst having magnetic resonance (MR) findings of hyperintense signals on T1–T2-weighted images and unsuppressed fat-depleted sequences, who was operated on due to a high risk of malignancy. As in our cases, dermoid tumor and endometriomas were described separately in the first two MRI evaluations presented above,[24,26] and histopathological diagnosis was recommended because the 2 conditions were rarely seen together. In the third case, although the endometrioma and dermoid tumor were detected separately in the MRI of the pregnant patient, malignancy could not be excluded due to the decidualization of endometrioma after pregnancy.[5]

In the cases where endometrioma and dermoid tumors were observed in the same ovary, no definite diagnosis could be made, including in our cases. Hwang et al[26] reported that endometrioma and dermoid were identified separately in preoperative MRI and struma ovarii was found in dermoid tumor accompanying endometriosis. In our second case, unlike the first, the diagnosis of dermoid and endometrioma was considered in the initial diagnosis. However, cystectomy was performed with laparoscopy, since no definitive diagnosis could be made.

Previously reported cases underwent operations because of the suspicion of malignancy, rather than expectant management, as in our cases. In our first case, laparoscopic ovarian cystectomy was performed because the patient was young and infertile. The second case underwent a right oophorectomy due to torsion of the dermoid tumor 8 years ago. Therefore the cystectomy was performed carefully and meticulously to protect the ovarian reserve during the left ovarian endometrioma and dermoid tumor excision.

Six cystectomies, 3 oophorectomies, and 1 cystectomy with contralateral oophorectomy were performed in the previously reported cases. Laparoscopy was performed in 6 patients, laparotomy in 3 patients, and oophorectomy during cesarean section in 1 patient (**Table 1**). Laparoscopy was performed in the cases reported in recent years. When the indications of laparotomy were examined, in the first patient, since a high incidence of suspected malignancy was noted due to increased vascularization caused by decidualization of the endometrioma, oophorectomy was performed instead of cystectomy during cesarean section.[5] In the second case, laparotomy was performed due to the increased mass size (40 × 23 × 20 cm) and in the third patient emergency laparotomy was performed due to acute abdominal pain.[8,21]

MRI can differentiate between a dermoid tumor and endometrioma in the same ovary. In atypical endometrioma and ovarian cancer, papillary projections can be observed in the cyst. Although vascular flow is observed in the papillary projections in cases of ovarian cancer, there is no vascular flow in atypical endometriomas. In patients with atypical endometrioma, vascular flow after decidualization can be observed and is accompanied with a high-risk of malignancy, which increases the incidence of midline incision and oophorectomy. As in the case presented by Taylor et al. conservative followup is important during pregnancy. However, endometriomas with papillary endometriomas determined during pregnancy in patients before the delivery should be followed up after pregnancy and should be re-evaluated after the effect of decidualization disappears.

In conclusion, the ultrasonographic findings of our patients showed more complicated and rare presentations than those expected in teratoma or endometrioma cases. This was due to the rare coexistence of a teratoma and an endometrioma. Despite the progress in sonographic imaging approaches, the coexistence of these two pathologic tumors in the same ovary leads to a challenging diagnostic process. Albeit rare, the sonographer should consider the possibility of the coexistence of an endometrioma and a dermoid tumor and should not hesitate to recommend an MRI, which may easily resolve this complexity. A meticulous clinical and radiologic approach is essential in making the correct diagnosis, planning the patient's treatment, and identifying the surgical means of treatment for the patient.

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