

Multiple and Bilateral Mature Cystic Teratoma: A Rare Case Report

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Abstract

Mature teratomas, also known as dermoid cysts, are the most commonly occurring benign ovarian tumors in young and middle-aged women. Bilateral occurrence of mature teratomas is observed in 10% of cases. The incidence rate of mature teratomas is equal in both the right and left ovaries, and malignant transformation is observed in 1-3% of cases. At sonographic examination, mature cystic teratomas appear as cystic, solid, or heterogeneous masses containing areas of fat and calcification. Macroscopically, areas of fat are observed in 90% of cases, while areas of calcification are observed in 50% of them. In our presented case, a 28-year-old virgin female patient complained of pain in both adnexal regions, and tumor markers were found to be normal. During imaging, a complex mass measuring 65x47 mm was observed in the adjacent right ovarian parenchyma, and a complex mass measuring 44x41 mm was observed in the adjacent left ovarian parenchyma. However, during surgery, it was discovered that there were two additional teratomas adjacent to the cyst. Given that the presence of both bilateral and multiple mature teratomas is rare, decided to present this case.

Keywords: Bilateral ovarian dermoid cyst, multiple mature cystic teratomas, bilateral ovarian mass.

Multiple ve Bilateral Matür Kistik Teratom: Nadir Bir Olgu Sunumu

Öz

Genç ve orta yaşlı kadınlarda en sık görülen iyi huylu over tümörleri, matür teratomlardır (dermoid kistler). Matür teratomların bilateralizm oranı %10'dur. Matür teratomların sağ ve sol overlerde görülme sıklığı eşittir ve malign dönüşüm olguların %1-3'ünde gözlenir. Sonografik incelemede, matür kistik teratomlar yağ ve kalsifikasyon alanlarının görüldüğü kistik, katı veya heterojen kitle olarak görülür. Makroskopik olarak, olguların %90'ında yağ alanları ve %50'sinde kalsifikasyon alanları gözlenir. 28 yaşındaki bir bekâr kadın hastamız, her iki adneksal bölgede ağrı şikayeti ile başvurdu. Tümör belirteçleri normal olarak saptandı. Yan yana sağ over parankiminde 65x47 mm boyutunda kompleks bir kitle ve yan yana sol over parankiminde 44x41 mm boyutunda kompleks bir kitle görüldü. Ancak ameliyat sırasında palpasyon yapıldığında, bu kistin yanında iki tane daha teratom olduğu görüldü. Hem bilateral hem de çoklu matür teratomlar nadir görüldüğü için olgumuzu sunmaya karar verdik.

Anahtar Sözcükler: Bilateral Over dermoid kist, bilateral matür kistik teratom, bilateral ovaryen kitle.

Olgu Sunumu (Case Report)

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Introduction

Mature teratomas, also known as dermoid cysts, are benign ovarian tumors that are most commonly observed in young and middle-aged females¹. They account for approximately 20-50% of ovarian tumors in adults and pediatric patients. The incidence of bilateralism of mature teratomas is 10%. The incidence of mature teratomas is equal in both the right and left ovaries, with a 1-3% rate of malignant transformation². Squamous cell carcinoma is the most commonly observed malignancy associated with mature ovarian teratoma (70-80%), while adenocarcinoma and malignant melanoma are rarer^{3,4}.

Most patients with mature teratomas are asymptomatic, and abdominal pain is a rare symptom. Symptoms due to hormone secretion (such as estrogen and prolactin) and paraneoplastic syndrome are extremely rare⁵.

Mature cystic teratomas appear as cystic, solid, or heterogeneous masses with areas of fat and calcification seen on sonographic examination. Macroscopically, areas of fat are present in 90% of cases, and areas of calcification are present in 50% of cases. Rokitansky nodules can be observed in radiological examinations⁶.

Bilateral and multiple mature teratomas are very rare. In our case, a total of 4 teratomas were present, with 3 in the right ovary and 1 in the left ovary. Due to the rarity of observing both bilateral and multiple mature teratomas, have decided to prepare a case report.

Case Report

In 2019, a 28-year-old female virgin patient presented to our clinic with a complaint of bilateral adnexal pain. She had no known medical conditions, had regular menstrual cycles, and was not using any medications. The adnexal region was evaluated using rectal examination, transabdominal and transrectal ultrasound examinations, revealing a complex mass measuring 65x47 mm in the adjacent right ovarian parenchyma and a complex mass measuring 44x41 mm in the adjacent left ovarian parenchyma. No free intra-abdominal fluid was observed and the uterus appeared normal in size and appearance.

Tumor marker tests were conducted, and the results were normal as follows: AFP (1.39 IU/mL with a reference range of 0-5.8), CEA (1.31 ng/mL with a reference range of 0-5), CA 125 (19.4 U/mL with a reference range of 0-35), CA 15-3 (5 U/mL with a reference range of 0-32.4), and CA 19-9 (29.44 U/mL with a reference range of 0-37). The patient underwent a radiological examination in the form of a contrast-enhanced MRI examination of the lower abdomen. This revealed the presence of multiloculated, thick, septated, and heterogeneous cystic lesions with loss of signal in both the right and left ovaries. The masses in the right and left ovaries were reported to be heterogeneous and measured 69x42 mm and 62x41 mm, respectively (as shown in Figure 1-2). Based on the recommendation of the radiology specialist, a computed tomography scan of the lower abdomen was performed to better highlight the fatty components. The results

showed the presence of complex cystic lesions with a lobular contour containing foci of fat and calcification, which were consistent with a mature cystic teratoma. The right ovarian mass measured 61x60 mm, while the left ovarian mass measured 64x45 mm.

Figure 1. MRI examination. The Right Ovary

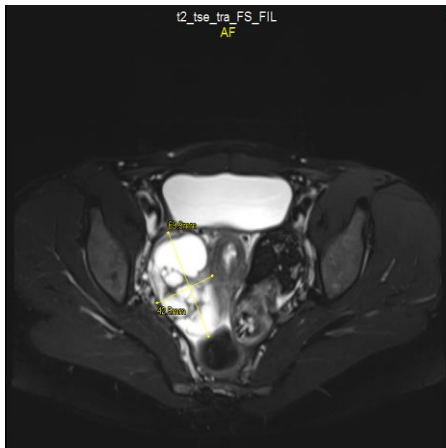
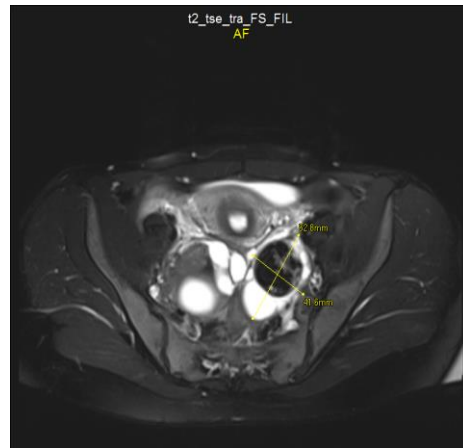


Figure 2. MRI examination. The Left Ovary



A mini-laparotomy was performed on the patient after a preliminary diagnosis of mature cystic teratoma was made. During abdominal exploration, it was observed that the uterus was of normal size and structure, and the intraabdominal organs appeared normal. No free fluid was detected within the abdomen. However, it was noted that both ovaries were larger than their normal size and appeared firm to the touch (as shown in Figure 3).

Figure 3. Intraoperative appearance of the ovaries



Initially, a scalpel incision was made in the left ovary, and the teratoma capsule was dissected from the ovarian parenchyma using sharp and blunt dissection techniques. The cyst region and intact ovarian tissue were then sutured closed. Subsequently, an incision was made in the right ovary, and the teratoma capsule was dissected off using a combination of sharp and blunt dissection techniques. During palpation, it was discovered that two additional teratomas were present adjacent to this cyst. These teratomas were also removed using the same dissection

technique on the ovary parenchyma (as seen in Figure 4-5). Hemostasis was achieved, and the parenchyma of the right ovary cyst was closed with sutures. The abdominal wall layers were then closed in accordance with anatomical principles, and the surgery was completed. The size of the teratomas removed during the operation was consistent with the preoperative evaluation.

Figure 4. Teratomas removed

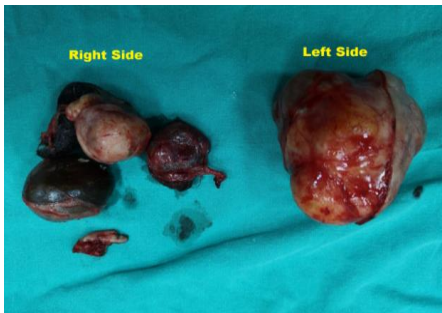


Figure 5. Contents of the teratomas removed



There were no immediate postoperative complications observed, and the patient was discharged on the second day after surgery with complete recovery. The histopathological examination confirmed the diagnosis of a mature cystic teratoma. During the two-year follow-up period, there were no signs of recurrence or any late postoperative complications. In 2022, the patient spontaneously conceived and gave birth to a healthy baby in February 2023.

Discussion

Although teratomas can occur at any age, they are more commonly seen in women of reproductive age between 20-40 years old⁷. Teratomas are germ cell tumors that generally derive from three germ layers and consist of multiple cell types. Pathological examinations of teratomas reveal features of different tissues all together⁸. Teratomas are classified as mature, immature, and monodermal. They are thought to arise from the parthenogenetic development of the haploid germ cell and are mostly of the 46, XX karyotype⁹. Mature teratomas are usually unilateral, but they may rarely be bilateral and/or multiple (10-15%)¹⁰. There are case reports in the literature of

teratomas being found as types of intraabdominal ovarian parasitic cystic teratoma¹¹, omental teratoma, teratoma of the Douglas pouch¹², and teratoma of the uterosacral ligament¹³. Pepe et al.¹⁴ reported two teratomas in the right ovary and one in the left ovary; Sinha et al.¹⁵ reported seven in the left ovary and three in the right ovary; Bournas et al.¹⁶ reported four in the right ovary and one in the left ovary; Walid et al.¹⁷ reported bilateral mature cystic teratoma in pregnant women. Elevated levels of CA 19.9 among serum tumor markers can be observed in mature cystic teratomas, but all tumor markers can be normal. In this case, tumor markers were normal¹⁸. The most important concern for these patients is future fertility, so treatment should focus on the preservation of ovarian tissue and minimizing adhesion formation. Whenever possible, laparoscopy should be preferred¹⁹. However, choose laparotomy in this case because more experienced with it and operating room conditions were not suitable for laparoscopy. Fortunately, since the capsule of the teratoma completely dissects the cyst from the ovarian tissue, enucleation does not significantly damage the ovarian tissue. Care should be taken not to rupture the cyst during enucleation. Rarely, recurrence of teratoma can occur due to incomplete enucleation or rupture of the dermoid cyst. The risk of recurrence of teratoma in the same ovary is 3-4%²⁰. The incidence of recurrence is higher in the presence of more than one of the following factors: being younger than 30 years of age, having a larger cyst diameter (diameter <8 cm), and having bilateral cysts²¹. Since patient has more than one risk factor, she should be followed up for a longer period for the recurrence of teratoma. Some cases considered as recurrence are small teratomas that are present with the main teratoma, but they cannot be detected and are left behind during surgery²². Intraoperative imaging methods can be used to visualize teratomas that cannot be detected with exploration and to avoid leaving teratoma behind, but intense acoustic shadowing can prevent the recognition of separate masses by hiding the boundaries of the teratoma and ovary²³. Therefore, it is important to palpate the ipsilateral and contralateral ovary to eliminate bilateral and multiple cases after the removal of the known cyst/mass during laparotomy. As in this case, it should be kept in mind that the teratoma can be bilateral and/or multiple, even if it is not detected on preoperative ultrasound examination.

Conclusion

Although both bilateral and multiple mature teratomas are rare, it is crucial not to overlook the possibility of their occurrence, as in this case where two teratomas were detected preoperatively, but four were found through palpation during surgery.

Consent to Participate

The patient was informed about the procedure, and her informed consent was obtained for the publication of this study.

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