International Journal of Reproduction, Contraception, Obstetrics and Gynecology
Bhatia R et al. Int J Reprod Contracept Obstet Gynecol. 2024 Oct;13(10):2947-2951
www.ijrcog.org
pISSN 2320-1770 | eISSN 2320-1789

DOI: https://dx.doi.org/10.18203/2320-1770.ijrcog20242839

Case Report

A rare case of giant endometrioma-with ultrasound features mimicking malignancy

Ruby Bhatia, Pooja Shah*, Rishu Goel, Vidushi Tewari, Sanjum Yasmin

Department of Obstetrics and Gynecology, MMIMSR, Mullana, Ambala, Haryana, India

Received: 27 July 2024
Revised: 07 September 2024
Accepted: 09 September 2024

***Correspondence:**
Dr. Pooja Shah,
E-mail: drpoojapshah@gmail.com

ABSTRACT

Ovarian endometriomas are common in women of reproductive age but rarely exceed 6 cm in diameter. Ovarian endometrioma exceeding 10 cm in dimension are referred as giant endometrioma. They are rare and pose a major diagnostic dilemma to the clinicians. We present a rare case of ovarian endometrioma of 18.7 cm in largest diameter, with internal septations, with a solid component, with low grade neovascularization on ultrasonography having RMI I score of 294 suggesting risk of malignancy, and IOTA simple rules suggesting the mass as inconclusive in nature, and the diagnosis of a benign mass was made on frozen section and confirmed as endometrioma on postoperative histopathologic examination. The challenges in making diagnosis of a huge ovarian endometrioma are highlighted and various literature on giant ovarian endometrioma are reviewed.

Keywords: Diagnostic dilemma, Giant endometrioma, Risk of malignancy index, IOTA simple rules

INTRODUCTION

Endometriosis is a benign estrogen-dependent, chronic inflammatory disorder characterized by the presence of endometrial-like tissue, including endometrial glands and stroma, outside the confinement of uterine cavity. It affects 10% of reproductive age women and 50% of women with infertility.[1] Pelvic endometriosis usually involves the ovaries, bilateral involvement occurs in one third to one half of the cases.[2,3] Ovarian endometriomas are also known as chocolate cysts, which refer to the presence of endometrial tissue within the ovary. Ovarian endometriomas rarely exceed 10-15 cm in diameter. 0.7% to 1.0% of patients with endometriosis have lesions that undergo malignant transformation.[4] When the diameter of the ovarian cyst exceeds 10 cm, malignancy is suspected. It is important to distinguish a benign adnexal mass from a malignant one as a benign ovarian mass requires a more conservative approach like a close follow up, and rarely laparoscopic surgery, while a malignant tumor requires extensive workup and usually urgent laparotomy with systemic consultations at a tertiary care centre. Various combined methods have been developed to evaluate the risk of ovarian cancer like RMI, ADNEX model, IOTA simple rules. Usually endometriomas are diagnosed by ultrasonography, but sometimes it is difficult to make a precise diagnosis preoperatively. We present a case of giant ovarian endometrioma of 18 cm in largest diameter with raised RMI index and IOTA simple rules suggesting the mass to be of inconclusive nature, hence raising a suspicion of malignancy, diagnosis of endometrioma was determined by postoperative histopathological examination.

CASE REPORT

A 24-year-old, unmarried female presented with abdominal distention since, 4 months, which was insidious in onset and progressed slowly, with recurrent abdominal pain and dysmenorrhea. She had no complaints of

menstrual irregularity. She had no history of heavy menstrual bleeding. She denied any history of weight loss, urinary and gastrointestinal symptoms. History of dyspareunia could not be ascertained as patient was not sexually active.

Her general physical examination was normal with a pale conjunctiva. No lymphadenopathy was noted. The abdomen was distended and revealed a tense, palpable mass of 20-week size with regular margins, filling the entire abdomen in diameter extending till xiphoid.

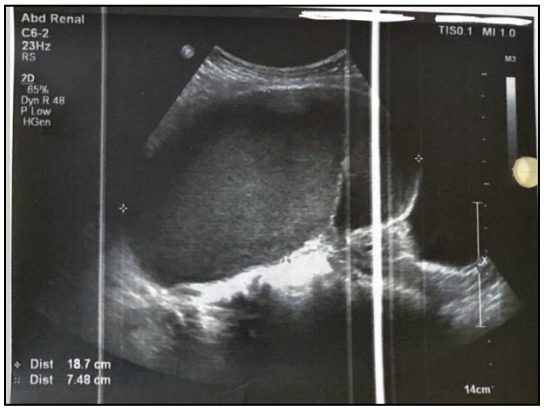

Figure 1: Ultrasound image of the mass.

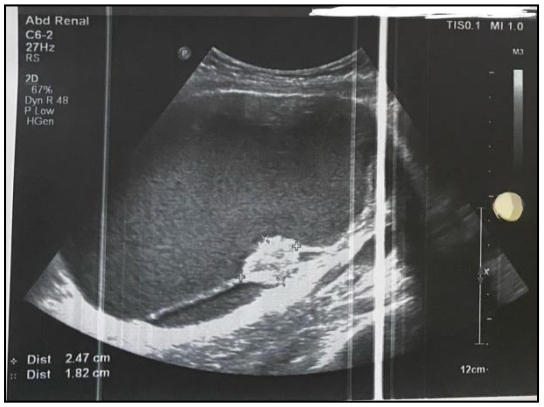

Figure 2: Ultrasound image of the mass showing the solid component within it.

Full blood count and serum biochemistry tests were normal. Serum cancer antigen 125 was slightly elevated to 98 U/ml and other tumor markers were CA19-9-31 U/ml, βHCG-0.100 mIU/ml, AFP, 1.66 ng/ml, CEA, 3.2 ng/ml.

The USG examination (Figure 1,2) showed a large multiloculated cystic mass of 18.7×12.4×7.4 cm in the left adnexal region. Heterogeneous in nature, with low-level internal echoes, a well-defined wall, and a solid component of 2.4×1.8 cm with low level internal vascularisation on doppler. The right ovary was normal, but the left ovary was not visualized. RMI, I index was calculated and it scored 294.

Table 1: RMI 1 score calculation.

Parameter		RMI 1 score	Patient RMI 1 score
USG score (U)	No feature	0	
	1 feature	1	
	>/=2 feature	3	3 (solid areas+multilocular cyst+)
Menopausal state (M)	Premenopausal state	1	1
Postmenopausal state		3	
CA-125 (IU/ml)			98

RMI 1=U×M×CA125, 3×1×98=294.

Table 2: IOTA simple rules assessment.

(M rules)	Patients score	(B rules)	Patient score
M1 irregular solid tumor	X	B1 unilocular	X
M2 presence of ascites	X	B2 presence of solid components with largest solid diameter <7 mm	X
M3 at least 4 papillary structures	X	B3 presence of acoustic shadows	X
M4 irregular muitilocular solid tumor with largest diameter >100 mm	X	B4 smooth multilocular tumor with largest diameter <10 cm	X
M5 very strong blood flow (color score 4)	X	B5 no blood flow (color score 1)	X

The adnexal mass is inconclusive in nature according to IOTA simple rules as neither malignant nor benign features apply. MRI (Figure 3-5) was performed, which showed a large multilocular complex ovarian mass.

A multidisciplinary team involving the surgeons and pathologists was made and decision for surgery was made primarily. Exploratory laparotomy was performed after informed consent, a midline incision extending from the supra-pubic region to the xiphisternum was given; which revealed a huge cystic mass originating from left adnexal region and filling the entire abdomen (Figure 6), the mass was adhered to the sigmoid colon (Figure 7).

Figure 3: MRI Image of the mass.

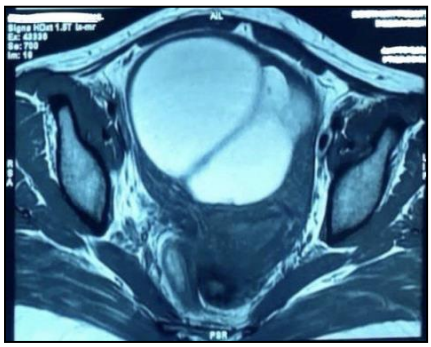

Figure 4: MRI Image of the mass showing septations.

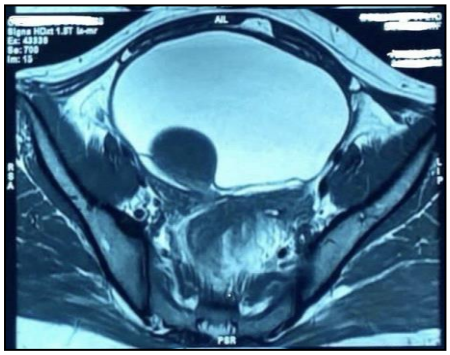

Figure 5: MRI Image of the mass.

The uterus, tubes, and right ovary were grossly normal except for adhesions. No ascites was seen. Adhesiolysis was done; however, the cyst content spilled during adhesiolysis due to dense adhesions between the cyst and colon, it released fluid with a chocolate-like appearance (Figure 8). For confirmation, the cut open cyst was sent for frozen section intraoperatively-which revealed it as a benign mass.

On gross examination, a large number of locules with thick septations were drained with solid areas in between. A total of 1200 ml of dark brown fluid was drained. The left ovarian tissue was identified after the cyst. Content was emptied and ovarian reconstruction was done. Other pelvic and abdominal organs were grossly normal except for the adhesions. Patient was discharged on the 4th postoperative day on Injection Leuprolide. She came to hospital for follow-up for suture removal with the histopathology report stating that the cyst wall was lined by inflamed granulation tissue with siderophages, fibrosis, endometrial glands, and chronic inflammatory infiltrates in the cyst wall, suggesting ovarian endometrioma. She had no new complaints, and the suture line had healed. The cytology of the fluid drained from the cyst content revealed a mixture of RBCs, neutrophils, lymphocytes, and a few degenerate epithelial cells.

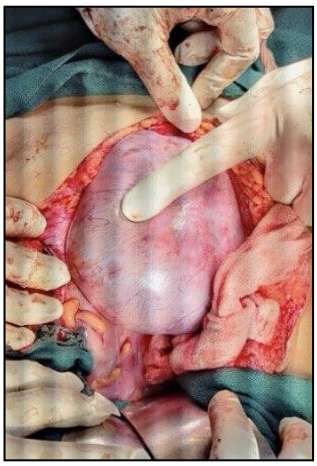

Figure 6: Intra operative picture of the mass.

Figure 7: Mass adhered to sigmoid colon.

Figure 8: Brown chocolate like fluid from the mass.

DISCUSSION

Giant ovarian endometriomas are uncommon and usually cause diagnostic dilemma to the clinicians due to their atypical and rare presentation. Very few cases have been reported in the literature, and in most of them, diagnosis was made post-operatively.

Mishra et al reported a 30×12×10 cm ovarian endometrioma in a 38-year-old multipara which mimicked ovarian malignancy pre-operatively.[5] Yaşar et al also reported ovarian endometrioma of 26×18×17 cm which contained 3250 ml of chocolate fluid in a 33-year multipara from Turkey where the diagnosis was also missed pre-operatively.[6] In our case, the size of the endometrioma was 18.7×12.4×7.4 cm in size and it contained approximately 1200 ml of chocolate brown fluid, and the diagnosis was made postoperatively. Endometriosis, including ovarian endometrioma, typically presents with symptoms of chronic pelvic pain, dysmenorrhea, dyspareunia, and infertility.[7] The presenting complaints in our patient were chronic pelvic pain with dysmenorrhoea and abdominal swelling. History of dyspareunia and infertility were not elicited as the patient was sexually inactive.

Transvaginal ultrasound (TVS) scan play an important role in the initial evaluation of women with suspected ovarian endometriosis, which was not performed in our patient as she was not sexually active. Ovarian endometriomas have a typical appearance of homogenous low-level internal echoes and thick walls on ultrasound scan. Also, the ground glass echogenicity of cyst fluid within the ovarian endometrioma is said to have a sensitivity of 73% and a specificity of 94% in detecting ovarian endometrioma and is single best ultrasound variable to differentiate between endometriomas and other adnexal masses in premenopausal women which was absent in our patient.[8]

In our case, Trans-abdominal ultrasonographic examination showed a large multiloculated cystic mass of 18.7×12.4×7.4 cm in the left adnexal region. Heterogeneous in nature, with low level internal echoes, a well-defined wall, and a solid component of 2.4 cm×1.8 cm with low level internal vascularisation on doppler. A RMI score (sensitivity 85%, specificity 97%) of more than 200, has 42 times higher risk of cancer compared to 0.15 times the risk in those with a lower score.[9] The RMI1 score of our patient was 294, which raised a suspicious of malignancy. As per IOTA simple rules the mass was inconclusive in nature.

A prospective internal validation from the IOTA group showed that these rules can be applied to 77% of adnexal masses and that, when applied, the diagnostic performance is high.[10] Magnetic resonance imaging (MRI) is considered the best diagnostic imaging technique for ovarian endometriosis. The state-of-the-art MRI protocol for the diagnosis of endometriosis includes T2-and fat-suppressed T1-weighted sequences.[11] The "shading sign" seen on T2-weighted images is pathognomonic of ovarian endometrioma, which was absent in our index case. The treatment of ovarian endometriomas is still a controversial issue. This is mostly because, prior to creating a treatment plan, various factors are taken into account, like patient's age, cyst laterality, current ovarian reserve, and fertility goals. The literature that is currently available shows that large ovarian endometriomas have been managed surgically. In all cases exploratory laparotomy was the surgical approach with salpingo-oophorectomy, cystectomy, or total abdominal hysterectomy combined with salpingo-oophorectomy.[5,6]

The primary surgical intervention in the index case was explorative laparotomy followed by cystectomy because of the presence a huge adnexal mass reaching the xiphoid and multiple thick internal septations inside it, with RMI 1 of 294 and IOTA simple rules classifying it as inconclusive mass. Also, the patient did not accept the possibility of malignant tumor spillage that will lead to chemotherapy during laparoscopic surgery. It is clear that there is no method making definitive differential diagnosis of adnexal masses. Therefore, laparotomy was the modality chosen for the therapy in our case. Malignant transformation risk (0.7%) in ovarian endometrioma is present.[12] The most prevalent histological types are endometrioid and clear cell carcinoma.[13]

None of the cases of large ovarian endometriomas in the literature have mentioned this. Prognosis after treatment depends on the type of treatment offered, however, recurrence is quite common irrespective of the type of treatment and is one of the most important problems in management of ovarian endometriosis. A persistence rate of 9.87% and a recurrence rate of 20.27% after surgical treatment have been reported.[14] Hence patients with endometriomas should have a close follow up even after surgical removal.

CONCLUSION

We presented this case because of its rarity as well as due to high suspicion of the mass being malignant or borderline as RMI I score was 294 and IOTA simple rules classified the mass as borderline/inconclusive in nature. Giant endometriomas are rare and pose a diagnostic dilemma to clinicians. High index of suspicion is needed for early diagnosis and prompt management of endometrioma as there can be a risk of malignancy.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Bhatia R, Shah P, Goel R, Tewari V, Yasmin S. A rare case of giant endometrioma-with ultrasound features mimicking malignancy. Int J Reprod Contracept Obstet Gynecol 2024;13:2947-51.