



Large Twisted Ovarian Fibroma Complicated with Meigs Syndrome and Acute Abdomen in A Pregnant Woman During the Second Trimester of Gestation Treated by Excisional Laboratory, Case Report and Review of the Literature

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Received Date: April 23, 2022

Published Date: May 01, 2022

DOI: [10.1027/margy.2022.0138](https://doi.org/10.1027/margy.2022.0138)

Abstract

Introduction: Ovarian fibromas belong to the group of sex cord-stromal cell tumors and are the most common benign solid tumors of the ovary, accounting for 1–4% of all benign ovarian tumors. The most frequent symptoms are abdominal discomfort and pain, but many patients do not experience any specific symptoms. These solid tumors are often difficult to diagnose based on preoperative ultrasonography findings and are commonly misdiagnosed as uterine myomas. They are also sometimes misdiagnosed as malignant ovarian tumors because of accompanying ascites and an increased serum CA-125 level.

Ovarian fibromas account for the majority of benign tumors causing Meigs' syndrome, which is a rare but well-known syndrome defined as the triad of benign solid ovarian tumor, ascites and pleural effusion. Almost all cases of ovarian fibroma can be cured by surgical excision. The Meigs' syndrome is a rare but well-known syndrome defined as the triad of benign solid ovarian tumor, ascites, and pleural effusion. It always requires surgical treatment. However, the optimal approach for its management has not been sufficiently investigated. Adnexal torsion is a rare cause of acute abdominal pain during pregnancy. The clinical, laboratory and imaging findings are non-specific. It requires differential diagnosis from other diseases presenting with abdominal pain & necessitates a prompt surgical intervention, as any delay leads to irreversible ovarian necrosis, so that adnexectomy is ultimately required. Despite the technological advances in ultrasonography, the diagnosis of the disease is difficult, especially during pregnancy and occasionally remains a diagnostic dilemma (1_3)

Case presentation: We report a pregnant patient at 16 weeks of gestation with a large twisted ovarian fibroma associated with Meigs' syndrome, abdominal pain and anemia that was treated by lapratomy excision of the twisted tumor. This case highlights the difficulties that may be encountered in the management of patients with Meigs' syndrome, including potential misdiagnosis of the tumor as a malignant ovarian neoplasm that may influence the medical and surgical approach and the adverse impact that Meigs' syndrome can have on the patient's condition, especially if it is associated with acute pain and severe anemia.

Considering the patient's serious clinical condition and assuming that she had Meigs' syndrome with a twisted large ovarian mass and possible hemolytic anemia, we first concentrated on effective medical management of our patient and chose the most appropriate surgical treatment. The main aim of our initial approach was preoperative management of the anemia and improve the general condition of the patient. Blood transfusions, glucocorticoid, antibiotics & analgesic therapy resulted in stabilization of the hemoglobin level & minimize the pain of the patient, which confirmed the appropriateness of this approach.

Conclusions:

This case highlights the difficulties that may be encountered in the management of patients with Meigs' syndrome, including potential misdiagnosis of the tumor as a malignant ovarian neoplasm that may influence the medical and surgical approach, and the adverse impact that Meigs' syndrome can have on the patient's condition, especially if it is associated with acute pain and severe anemia and this deteriorate the pregnancy condition. Adnexal torsion is a rare cause of acute abdominal pain during pregnancy. The clinical, laboratory and imaging findings are non-specific. It requires differential diagnosis from other diseases presenting with abdominal pain during pregnancy. It necessitates a prompt surgical intervention, because any delay leads to irreversible ovarian necrosis, so that adnexectomy is ultimately required. In spite the technological advances in ultrasonography, the diagnosis of the disease is difficult, especially during pregnancy and occasionally remains a diagnostic dilemma.

Case Presentation

A 25-year-old woman, gravida 3, para 2 in the 16th week of pregnancy was admitted to our clinic in the Department of obstetrics & Gynecology at Tanta University Hospital, Tanta, Egypt because of an acute abdomen, vomiting, difficulty of breathing & fainting. She was febrile and physical examination showed a point of maximal tenderness in the right lower abdominal quadrant, with a 4-days history of abdominal pain that started in the right lower quadrant and subsequently spread to the whole abdomen. Acute appendicitis and renal colic were included in the differential diagnosis.

As regards her vitals: She was pale and in obvious discomfort. Her temperature was 38°C, blood pressure was 100/60 mmHg and heart rate was 120 beats/min. Her past medical history was unremarkable

except for intermittent episodes of abdominal pain and a sensation of abdominal heaviness during the preceding months.

Physical examination revealed a mass that occupied almost the entire abdomen, extending from the lower abdomen to above the umbilicus and restricting mobility. The fundal level of the gravid uterus was about mid-way between the symphysis pubis and the umbilicus. The fetal heart rate was heard and it was about 160 beat/min, also the fetal movements were felt by both physician & the mother. Meanwhile a huge regular tender right adnexal mass can be assessed on bimanual pelvic examination, it was reaching to the right hypochondria region.

Pelvic ultrasonography showed mild ascites throughout the abdomen and an a gravid, enlarged uterus with a single , viable ,living fetus with a changeable presentation, and the fetal biometry was about 16+5 weeks of gestation . The kidneys, pancreas, gallbladder and the placenta had no abnormalities. The adnexal structures were not recognizable. A non-homogeneous mass (185 × 120 × 155 mm) adjacent to the uterus occupied almost the right side of abdomen, extending from the pelvis to the right hypochondria region. No flow was detected in the mass on color Doppler or power Doppler ultrasonography. In addition, a large amount of fluid was revealed in the pouch of Douglas. MRI showed a large solid right adnexal mass, ascites and bilateral pleural effusions; a thickened and twisted Fallopian tube with a whirlpool sign was also observed, suggesting torsion of huge adnexal mass. MRI did not show enhancement of the adnexal structures, confirming the hypothesis of torsion and necrosis.

As regards the patient's hematological parameters at admission, she had severe anemia, hemoglobin was 8 g/dl, with leukocytosis & the renal and liver functions were normal. Meanwhile, there was elevated CA-125 level and elevated levels of inflammatory markers including the total white blood cell count, percentage of neutrophils, C-reactive protein (CRP) and fibrinogen. Considering the patient's serious clinical condition and assuming that she had Meigs' syndrome with a twisted large ovarian mass and possible hemolytic anemia, we implemented intensive medical therapy to prepare her for surgery. She received 2 units of packed red blood cells to correct the anemia and one unit of fresh frozen plasma, she received intravenous antibiotics, full fluids & methylprednisolone 500 mg/day. We consider that a long waiting time before surgery should be avoided, as cure depends on surgical excision of the tumor, and the pathogenic mechanisms that trigger autoimmune responses in patients with benign ovarian tumors are unknown.

It seems appropriate to administer medical therapy, including transfusion, to ensure that patient can undergo early surgery. In our patient, this approach was necessary because of the pregnancy, fetal life & well-being, and the size of the ovarian mass, the ovarian torsion and the resulting severe pain. Furthermore, it is known that large ovarian masses in Meigs' syndrome are often associated with intra-abdominal hypertension up to abdominal compartment especially in pregnant females.

After improvement of the patient's condition, we performed urgent laparotomy for her. A large amount of ascites was aspirated and a large solid mass was observed. The mass was huge bluish whitish and occupied the whole right side of the abdominal cavity, with twisted fallopian tube & the ovarian ligament (Figure 1, 2).

The mass was carefully freed from the surrounding adherent structures and was found to arise from the twisted right adnexa. The utero-ovarian ligament, Fallopian tube and infundibulo- pelvic ligament, which were twisted together, undue of the twisted pedicle and unilateral salpingoophorectomy was done, and intraperitoneal drain was omitted and removed 12 hours after the surgery. There was no significant blood loss and no anesthesia-related complications were observed.

Postoperative pathological examination of the surgical specimen showed complete hemorrhagic necrosis of an ovarian fibroma with evidence of stromal edema (weight 1,800 g) and a 10-cm long necrotic Fallopian tube. Peritoneal cytology showed inflammation but no malignant cells. The patient was discharged 7 days after surgery with a small pleural effusion which resolved approximately 2 weeks after discharge. Fortunately, the pregnancy continued and the fetal wellbeing was confirmed by repeated ultrasound scans. Seven days after discharge she reported a satisfactory return to her normal social but with instructions to minimize her working activities. One month later she had recovered well and was asymptomatic.

Post-operative, she also received subcutaneous low-molecular-weight heparin, antibiotic therapy, albumin infusion and minimal diuretics to avoid progression of her ascites and pleural effusions. Her hematological parameters improved significantly after 3 days. The temporal association between the cessation of hemolysis and glucocorticoid therapy supported our hypothesis of an association between the ovarian tumor and the hemolytic anemia.

Background & Discussion

Ovarian fibromas belong to the group of sex cord-stromal cell tumors and are the most common benign solid tumors of the ovary, accounting for 1–4% of all benign ovarian tumors. The most frequent symptoms are abdominal discomfort and pain, but many patients do not experience any specific symptoms. These solid tumors are often difficult to diagnose based on preoperative ultrasonography findings and are commonly misdiagnosed as uterine myomas. They are also sometimes misdiagnosed as malignant ovarian tumors because of accompanying ascites and an increased serum CA-125 level. The high level of CA-125 associated with anemia and high levels of inflammatory markers (CRP and fibrinogen) & these associations may indicate advanced ovarian cancer and our patient could potentially have been misdiagnosed with a malignant ovarian tumor.

However, a high CA-125 level does not necessarily indicate ovarian cancer and can also be associated with ovarian fibroma and the accompanying ascites. In addition, torsion of an ovarian fibroma with subsequent necrosis and inflammation can result in increased serum levels of CA-125 and inflammatory markers. (3,4, 5)

Ovarian fibromas account for the majority of benign tumors causing Meigs' syndrome, which is a rare but well-known syndrome defined as the triad of benign solid ovarian tumor, ascites and pleural effusion. Almost all cases of ovarian fibroma can be cured by surgical excision. However, the optimal approach for the management of ovarian fibromas has not been sufficiently investigated. Surgeons may be reluctant to remove the tumor laparoscopically as it can be difficult to safely remove the excised tumor from the abdominal cavity. However, recent advancements in operative instruments and techniques have resulted in laparoscopic surgery becoming increasingly popular among gynecological surgeons. (1, 2)

The incidence of adnexal masses during pregnancy before 14 weeks of gestation varies from 6% to 25% in longitudinal studies. Most of these adnexal masses are functional ovarian cysts and generally resolve during pregnancy, leaving between 0.7% and 1.7% of women with persistent masses. Although ovarian cyst torsion, hemorrhage, or rupture leading to abdominal pain are uncommon in pregnancy, some women may require emergency surgery for these complications. Women with persistent masses may also opt for surgery in the second trimester of pregnancy if malignancy is suspected on ultrasonography images or to prevent cyst complications, which may necessitate emergency surgery in the third trimester, which adds an increased risk of complications. (5,6)

Although several studies have reported the safety of adnexal mass removal during the first trimester of pregnancy, it is generally avoided to allow time for spontaneous resolution and to reduce the risk of spontaneous miscarriage. However, delaying surgery until the second trimester of pregnancy poses a technical challenge, especially if laparoscopic surgery is performed. In a study by Whitecar et al, women who underwent laparotomy after 23 weeks' gestation had a 50% risk of adverse perinatal outcome. Traditionally, laparotomy has been used to remove adnexal masses during pregnancy. However, increasing evidence suggests that laparoscopic treatment of adnexal masses in pregnancy is safe and effective. (6)

One of the main concerns with an adnexal mass is the risk of malignancy. In most of the published series, the reported incidence of ovarian cancer in pregnancy ranges from 1 in 5000 to 1 in 47 000 live births, with 2% to 6% of persistent adnexal masses found to be malignant. In the present study, the rate of malignancy was 1 in 10 000 live births, or 8.6% of persistent masses, which is comparable with the reported rates.

However, in a retrospective study of 60 adnexal masses resected during pregnancies over a 12-year period, Sherard et al, 13 reported a malignancy rate of 13%, which was more than twice the previously reported incidence. (7)

Adnexal torsion is a rare cause of acute abdominal pain during pregnancy. It is frequently associated with ovarian stimulation for in vitro fertilization (IVF) or with ovarian masses, mainly of functional origin. The clinical, laboratory and imaging findings are non-specific. The diagnosis of adnexal torsion is difficult, especially during pregnancy, and occasionally remains a diagnostic dilemma. It necessitates a prompt surgical intervention, because any delay leads to irreversible ovarian necrosis, so that adnexectomy is ultimately required. Adnexal torsion is rarely observed during pregnancy. Its incidence is approximately 1 in 5000 pregnancies. (6,7)

The clinical symptoms are non-specific and could be confused with other acute abdominal conditions, such as acute appendicitis, renal colic, and cholecystitis. Traditionally, abdominal complications during pregnancy have been treated by means of laparotomy. Nowadays, laparoscopy is considered the preferable surgical option until approximately the 16th week of gestation. A prompt diagnosis is essential for a conservative, organ-preserving management, because after 36–48 h of torsion irreversible lesions of the ovary are likely. (8)

The majority of lesions surgically excised at the second trimester are persistent corpus luteum cysts (20%), cystadenomas (24%), dermoids (37%), paraovarian cysts (5%), endometriomas (5%) and leiomyomas (5%). Malignant tumors are found in 5.9% of the cases. Most of the described torsions have been in the right adnexa, probably because the sigmoid limits the mobility of the left ovary. Adnexal torsion usually presents in the first trimester, when the uterus is moving out of the pelvis, although some cases have been described in the second and, rarely, in the third trimester.

The preoperative diagnosis is difficult, especially in pregnant women. Torsion of the ovarian pedicle results in circulatory stasis that is initially venous, but becomes arterial as the torsion and the resultant edema progress. When complete torsion with hemorrhagic necrosis is suspected, immediate surgery is necessary. If there is a delay in the diagnosis and the torsion persists for more than 36–48 h, the lesions of the ovary are irreversible and a conservative, organ-preserving approach is not possible. However, it has been described that viable ovarian tissue can be detected even after 48 h of torsion. Conclusively, ovarian torsion often remains a diagnostic dilemma, largely because of the non-specific clinical, laboratory and imaging findings. In our case, there was a critical delay in the establishment of the diagnosis, which had, as a result, adnexal removal. Except for the difficulty in diagnosis, many obstetricians hesitate to proceed on time to an explorative laparotomy's, fearing the potential risks that the operation and general anesthesia may cause to the fetus. (7, 9)

In the case of oophorectomy in the first 12 weeks of gestation, substitution of progesterone is recommended in order to support an early pregnancy. After this period, progesterone is produced by the placenta. Ultrasound findings, including solid, cystic and complex pelvic tumors, with or without fluid collection in the pouch of Douglas, are non-specific for the diagnosis of adnexal torsion.

Color Doppler sonography has been proved useful and the main sign is the absence of intraparenchymal ovarian blood flow. The disappearance of ovarian flow depends on the stage of torsion, therefore close monitoring is necessary in order to achieve timely management with a conservative approach. Doppler sonography, although highly specific, has low sensitivity, as it may miss the diagnosis in approximately 60% of cases. (10,11)

MRI can be also applied in pregnant women without the hazards associated with ionizing radiation. MRI findings in adnexal torsion include a thick edematous pedicle and ovary, lack of enhancement and signal intensities indicative of hemorrhage. Adnexal torsion is a surgical emergency during pregnancy.(11) In the past, the traditional treatment was adnexal removal, in order to prevent the increased risk of pulmonary embolism. Nevertheless, a case report describes a patient who developed pulmonary embolism after adnexal resection. Nowadays, de-torsion of the twisted adnexa should be considered a safe procedure and does not increase the risk of pulmonary embolism, compared with excision of the adnexa without untwisting. (10)

Cases of adnexal torsion occurring during the first trimester of pregnancy should preferably undergo laparoscopy, which is suitable for diagnosis, evaluation and treatment. However, laparoscopy may also be carried out during advanced pregnancy. In order to avoid penetrative injuries with the insufflation needle, an open laparoscopy is a possible option. The procedure is better tolerated by the patient compared to laparotomy, because of less postoperative pain and the absence of an abdominal scar. (10,11)

Nevertheless, under certain circumstances, laparotomy remains a traditional approach. The surgery for an adnexal mass any time during pregnancy increases the risk of pregnancy loss and the likelihood of intrauterine growth restriction (IUGR) and preterm delivery. Considerations during laparotomy when performing a laparotomy or cesarean section for an adnexal mass, the surgeon must take into account a number of variables when selecting the type of incision (i.e., vertical vs transverse). Surgery is generally not recommended during the first trimester. Among the reasons are the high likelihood of a corpus luteum cyst, the low likelihood of an invasive malignancy, the low risk of adnexal complications associated with observation, and the potential for pregnancy loss or teratogenicity. However, as pregnancy progresses beyond the first trimester, surgery poses other problems. In general, if malignancy is suspected, or if uterine manipulation is to be minimized, a vertical incision is best. Other considerations include a prior scar, body habitus, obstetric issues, and the patient's wishes. (12, 13)

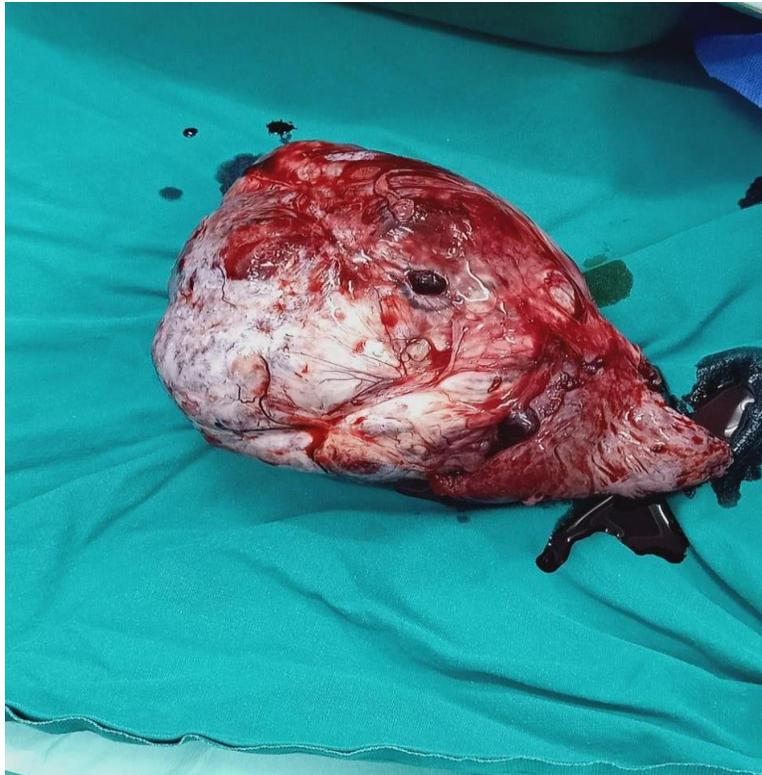


Fig. 1,2; Showing Twisted Pedicle of The Right Adnexal Mass & The Mass After Removal

Citation: Yasmine I.M. El-Masry "Large Twisted Ovarian Fibroma Complicated with Meigs Syndrome and Acute Abdomen in A Pregnant Woman During the Second Trimester of Gestation Treated by Excisional Laboratory, Case Report and Review of the Literature." MAR Gynecology 3.1 www.medicalandresearch.com (pg. 9)

Conclusion

This report presents a rare case of a large twisted ovarian fibroma associated with pain, Meigs' syndrome and hemolytic anemia in a pregnant female in the 2nd trimester of gestation. This case highlights the complexities associated with the diagnosis and treatment of patients with severe complications of benign ovarian disease during pregnancy. In rare cases, ovarian fibromas are associated with ascites and pleural effusions, which is known as Meigs' syndrome. This syndrome is usually associated with large fibromas and high CA-125 levels (1). Surgical management of adnexal masses during pregnancy appears to have favorable outcomes for the mother and the fetus.

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