

Laparoscopic management of a twisted ovarian leiomyoma in a woman with 10 weeks' gestation

Case report and literature review

Myounghwan Kim, MD*

Abstract

Background: Primary leiomyoma of the ovary is a rare benign ovarian tumor that only seldom causes acute abdomen.

Case summary: A 35-year-old gravida 1, para 0 woman presented with a history of acute lower abdominal pain, and 10 weeks of amenorrhea. The patient's physical examination revealed abdominal tenderness, defense, and rebound. On ultrasonographic examination, a solid mass measuring 9.3 × 7.8 cm was detected adjacent to the uterine fundus. The mass was preoperatively diagnosed as a twisted pedunculated subserosal uterine myoma. Upon entering the pelvic cavity, the mass in the right adnexa appeared twisted clockwise. Therefore, a laparoscopic salpingo-oophorectomy was performed. The tumor was pathologically diagnosed as ovarian leiomyoma. The patient delivered a healthy girl at 40 1/7 weeks of pregnancy.

Conclusion: Despite its low incidence, torsion of ovarian leiomyoma should be considered in the differential diagnosis of acute abdomen. Furthermore, laparoscopic exploration should be the preferred way of removing twisted ovarian leiomyoma, even during pregnancy. It seems that primary ovarian leiomyomata have a tendency to grow rapidly during early pregnancy. However, because of the low incidence of ovarian leiomyoma, the effects of estrogen and pregnancy on this condition remain unclear.

Abbreviation: CRL = crown rump length.

Keywords: acute abdomen, laparoscopy, ovarian leiomyoma, pregnancy, torsion of ovarian tumors

1. Introduction

Although the uterus is the most common origin site of leiomyomata, the disease may develop at any site where smooth muscle cells are found because leiomyoma lesions arise as proliferations of such cells. Unusual sites of origin for leiomyomata include the vulva, ovaries, urinary bladder, and urethra.^[1] Primary leiomyoma of the ovary is a rare benign tumor most often occurring in premenopausal women (up to 85% of cases), and which accounts for 0.5% to 1% of all cases of benign ovarian neoplasia.^[2] Primary leiomyomata of the ovary are usually smaller than 3 cm in diameter and rarely induce acute abdomen.^[3,4]

It is difficult to diagnose primary ovarian leiomyoma preoperatively based on ultrasonographic results, or even during surgery. Primary ovarian leiomyoma is often misdiagnosed as

pedunculated subserosal parasitic uterine leiomyoma that has lost its attachment to the uterus and is found attached to the ovary instead. It is also misdiagnosed as ovarian fibroma, or even as malignant ovarian tumor, which represents a more common condition.^[5]

A case of twisted primary ovarian leiomyoma in a 10-week pregnant woman with acute abdomen is presented here along with full details of the laparoscopic treatment solution applied. To the author's best knowledge, this is the first case where the laparoscopic approach is applied to manage twisted ovarian leiomyoma in a pregnant woman. This case report was approved by the Institutional Review Board of Sanggye Paik Hospital in Seoul, Korea. The patient provided written informed consent for the publication of details regarding this case.

2. Case presentation

A 35-year-old gravida 1 para 0 woman presented with a history of acute lower abdominal pain and 10 weeks of amenorrhea. The patient had been diagnosed with uterine fibroid during a gynecologic check-up at a private clinic 1 year before the events reported here. Later, 1 month before the events reported here, upon confirming the pregnancy by ultrasonography, she was informed that the mass size had remained stable. Upon arrival at the emergency room, the patient reported having intense and constant pain that had started 3 hours before admission to the emergency room. The following vital signs were noted upon admission: blood pressure, 108/72 mm Hg; pulse rate, 73 beats per minute; and body temperature, 36.5 °C. The patient's physical examination revealed abdominal tenderness, defense, and rebound. The following blood test results were noted: serum hemoglobin, 13.3 g/dL; hematocrit, 37.9%; and leukocyte count, 13,900/μL. Ultrasonographic examination indicated

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Department of Obstetrics and Gynecology, Sanggye Paik Hospital, Inje University, School of Medicine, Seoul, Korea.

* Correspondence: Myounghwan Kim, Department of Obstetrics and Gynecology, Sanggye Paik Hospital, Inje University, School of Medicine, Seoul, Korea (e-mail: myankim@paik.ac.kr).

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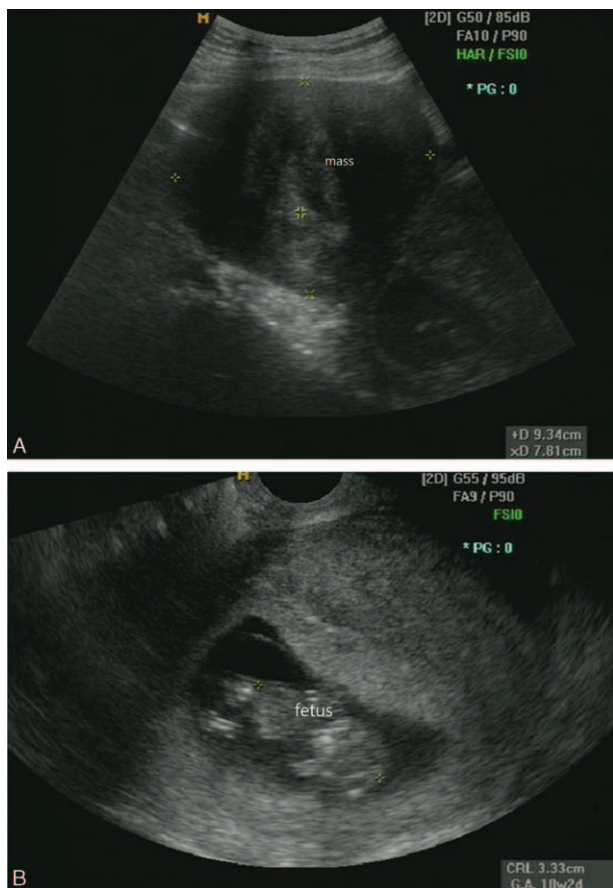


Figure 1. Ultrasound scan of a 35-year-old gravida 1, para 0 woman with a history of acute lower abdominal pain and 10 weeks of amenorrhea. The scan shows a solid mass (9.3 × 7.8 cm) adjacent to the uterine fundus (A), and a single live fetus with a crown rump length (CRL) corresponding to that of a fetus with a gestational age of 10 weeks and 2 days (B). CRL = crown rump length.

a solid mass adjacent to the uterine fundus, measuring 9.3 × 7.8 cm (Fig. 1A), and a single live fetus with a crown rump length corresponding to that of a fetus with a gestational age of 10 weeks and 2 days (Fig. 1B). The mass was preoperatively diagnosed as a twisted pedunculated subserosal uterine leiomyoma. The patient received meperidine for pain control (50 mg intramuscularly, twice, 4 hours apart), but pain was not relieved. Emergency laparoscopic exploration was performed. Upon entering the pelvic cavity, a right adnexal mass of dark bluish color to which the fimbriae were adhered was noted (Fig. 2). The mass was twisted clockwise, necrotic, and completely enclosed within the ovary itself, so it could not be separated from the ovarian cortex. Therefore, it was concluded that the mass originated from the right ovary. Laparoscopic salpingo-oophorectomy was performed. The mass was morcellated using a cold knife and extracted through an extended suprapubic trocar site. The surgery duration, defined as the time from skin incision to wound closure, was 56 minutes, and the estimated blood loss was 50 mL. The patient had no complications after the surgery, and it was confirmed by ultrasonography that the fetus was not affected by the procedure. The patient was discharged from the hospital on the third day postoperatively. The right ovarian tumor was pathologically diagnosed as ovarian leiomyoma. The patient underwent routine antenatal care after discharge. Finally, the

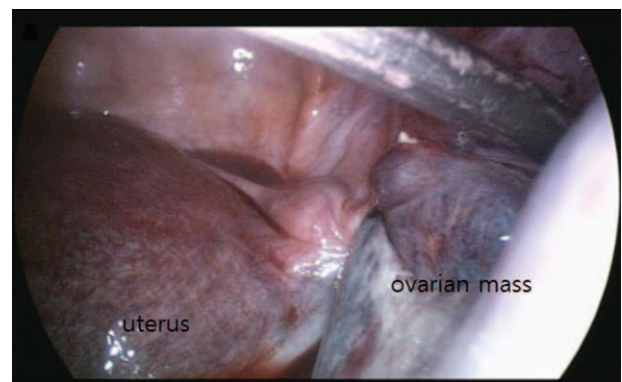


Figure 2. Laparoscopic view revealing a clockwise-twisted right ovarian mass in a 35-year-old gravida 1, para 0 woman with a history of acute lower abdominal pain and 10 weeks of amenorrhea.

patient delivered at 40 1/7 weeks of gestation, by normal birth, a healthy female infant who weighed 3160 g.

3. Discussion

Unlike the uterus, which is the most common site of leiomyomata, the ovary is an unusual site of origin for the disease.^[1] Primary leiomyoma of the ovary, a rare benign tumor, occurs most often in premenopausal women and accounts for 0.5% to 1% of all cases of benign ovarian neoplasia.^[2,6] Since Fallahzadeh et al^[4] first described ovarian leiomyoma in 1862, fewer than 100 cases of primary ovarian leiomyomata have been reported. However, the exact incidence is unknown because there seems to be a considerable number of unreported cases, as well as of cases misdiagnosed as fibroma.^[7] Most cases exhibit a small and asymptomatic tumor, typically detected incidentally during routine gynecologic examinations, during surgery, or during autopsy.^[8] In some cases, the tumor becomes large enough to cause pelvic pain and presents as a palpable mass. Associations with ascites, hydrothorax, hydronephrosis, polymyositis, Meigs' syndrome, or acute abdomen have rarely been reported.^[7] Primary ovarian leiomyomata are frequently associated with their uterine counterpart, and both tumors develop in patients of similar age. Primary ovarian leiomyoma must be differentiated from other solid ovarian tumors. Further, differential diagnoses should be made for smooth muscle tumors that secondarily involve the ovary, such as parasitic uterine leiomyoma, intravenous leiomyoma, and disseminated peritoneal leiomyoma.

The characteristic imaging findings of primary ovarian leiomyoma have seldom been described in the literature. Homogeneous density may be the most common finding indicating ovarian leiomyoma, as is the case for the classic, solid uterine leiomyoma. Because of this similarity, the radiographic imaging features of these tumors have often been misattributed. Moreover, the heterogeneous and complex nature of many of these tumors, particularly when they are large, often suggests malignancy, especially in the presence of associated ascites.^[9]

Leiomyomata are benign, monoclonal tumors involving the smooth muscle cells of the myometrium, and contain large aggregations of extracellular matrix composed of collagen, elastin, fibronectin, and proteoglycan.^[10]

Although the exact causes of leiomyoma are unknown, advances have been made towards understanding the molecular biology of these benign tumors as well as their growth, genetic,

Table 1

Primary ovarian leiomyomata during pregnancy.

Author	Patient age	Detection	Presenting symptoms	Rapid growth	Weeks of gestation on surgery	Size	Surgical procedure	Obstetric course
Olshausen ^[14]	38	Increase of abdominal girth	Rapid increase of abdominal girth; breathing difficulty;	0	12	Human's head size	Laparotomy	Natural birth, with the infant weighing 3160 g at full term.
Brachetto-Brian and Casco ^[15]	17	Incidentally on surgery	Abdominal pain	Not available	Not available	Not available	Left oophorectomy	Management of hemoperitoneum caused by ovarian pregnancy.
Moore and Forks ^[16]	34	Gynecologic exam	Rapid increase of abdominal girth	0	12	16 × 13 cm on surgery; absent during follow-up; size unknown a year before;	Right oophorectomy	Natural birth, with the infant weighing 3538 g at full term.
Zorlu and Gengiz ^[17]	32	Pregnancy test	Asymptomatic	0	6	14 × 9 cm	Left adnexectomy	Termination at request.
Daniel et al ^[16]	31	Antenatal care	Asymptomatic	0	Full term	Right 8 × 5 cm; left 10 × 7 cm;	Bilateral enucleation with residual tumor	Cesarean delivery at full term.
Kohno et al ^[19]	32	Gynecologic exam	Asymptomatic	0	40	23 × 23 × 20 cm before surgery	Left oophorectomy during cesarean section	Cesarean delivery of female infant at 40 weeks of gestation.
Hsiao et al ^[20]	42	Gynecologic exam	Asymptomatic	Not available	Not available	4.5 × 4.4 × 3.2 cm 3.5 × 2.4 × 2.5 cm a year before	Left oophorectomy during emergency cesarean section performed due to fetal distress	Emergency cesarean delivery of female infant weighing 2930 g.
Zhao et al ^[21]	28	Gynecologic exam	Mild intermittent abdominal pain	0	14	18 × 16 × 10 cm on surgery; 10.4 × 10 × 6.6 cm a week before; 4.1 × 3.2 cm 5 weeks before; 2.4 cm 3 years before;	Elective laparoscopic right ovarian tumorectomy	Natural birth, with the infant weighing 3160 g at full term.
Kim	35	Gynecologic exam	Severe right lower quadrant pain due to torsion	0	10	9.3 × 7.8 cm before surgery; 8.5 cm 1 month before; 7 cm 18 months before;	Emergency laparoscopic right adnexectomy	Natural birth of female infant weighing 3160 g at 40 1/7 weeks of gestation.

and hormonal factors.^[11] There are a number of theories regarding the origin of primary ovarian leiomyomata. The tumors most likely arise from smooth muscle cells in the ovarian hilar blood vessels, but other possible origins include cells of the ovarian ligament, smooth muscle or multipotent cells in the ovarian stroma, undifferentiated germ cells, and cortical smooth muscle metaplasia.^[4,12,13]

A number of cases of ovarian leiomyomata in pregnant women have been reported, and a summary of these cases is given in Table 1. Olshausen^[14] first reported a case of ovarian leiomyoma during pregnancy, which was managed in 1893. Brachetto-Brian and Casco^[15] reported the case of a 17-year-old girl who underwent an emergency operation for appendicitis. The girl's appendix was normal, but her left ovary was removed because it was causing hemoperitoneum. The ovary presented with impregnated ovum, endometriosis, and leiomyoma. Moore and Forks^[16] reported the case of a 34-year-old woman who underwent oophorectomy at 3 months of gestation. The mass measured 16 × 13 cm and was diagnosed as ovarian leiomyoma. No recurrence was observed at the 1-year follow-up, and the patient delivered a healthy male infant by natural birth at full term. Zorlu et al^[17] reported the case of a 32-year-old woman who requested the termination of pregnancy at 6 weeks of gestation; a solid adnexal mass was incidentally found during the examination. Daniel et al^[18] reported the case of a 31-year-old woman admitted at term for elective cesarean section. At surgery, bilateral ovarian leiomyoma was noted. With the purpose of preserving the ovaries, only the most accessible tumors were enucleated, and a few residual small tumors remained in situ. The residual tumors regressed after pregnancy. Kohno et al^[19] described the case of a 32-year-old woman with a large, rapidly growing pelvic mass noted at 16 weeks of gestation. Subsequent laparotomy at 20 weeks of gestation found a left ovarian leiomyoma weighing 11 kg, measuring 23 × 23 × 20 cm, and exhibiting extensive degeneration, with hyalinization and edema. Hsiao et al^[20] reported the case of a 42-year-old woman who underwent emergency cesarean section for fetal distress and concurrent oophorectomy for an ovarian leiomyoma detected a year before. Zhao et al^[21] reported the case of a 28-year-old woman with an ovarian leiomyoma excised laparoscopically at 14 weeks of gestation. The mass had exhibited rapid growth during early pregnancy. The surgery duration was 118 minutes, and the estimated blood loss was 800 mL. Examination of frozen sections of the tumor revealed a benign spindle cell tumor with focal ischemic infarction, which was suggestive of ovarian leiomyoma. Following surgery, prophylactic progesterone was prescribed for 4 days to prevent miscarriage. A 3.4 kg healthy baby was delivered by natural birth at full term. The authors concluded that, because of the high probability that the tumor would grow rapidly during pregnancy, surgery prior to gestation is advised in patients with ovarian solid tumors, even for tumors smaller than 3 cm in diameter. Nonetheless, their advice seems controversial because of the difficulty associated with diagnosing ovarian leiomyoma preoperatively. The present report describes the first case of laparoscopic excision of ovarian leiomyoma during pregnancy.

The case described in the present report is similar to that described by Zhao et al^[21] specifically, the patient was a 35-year-old woman who underwent emergency laparoscopic right salpingo-oophorectomy at 10 weeks of gestation due to acute abdomen caused by a twisted ovarian leiomyoma, and she delivered a healthy female infant by natural birth at 40 1/7 weeks of gestation. Nevertheless, several differences should be noted. Importantly, the patient described here had a twisted ovarian leiomyoma with intractable acute abdomen, and therefore

emergency laparoscopic surgery had to be performed. The tumor size was stable during early pregnancy. The mass was twisted clockwise, necrotic, and tightly enclosed within the ovary itself, so it could not be separated from the ovarian cortex. A salpingo-oophorectomy had to be performed, which resulted in shorter operation time and negligible blood loss. The mass was removed through the extended trocar site, after the tumor was morcellated using a cold knife to prevent intraperitoneal dissemination.

Surgeons should make an effort to perform an ovary-preserving surgery instead of oophorectomy, particularly in young patients. For cases such as this, when the mass is completely enclosed within the ovary, it is difficult to perform ovary-preserving surgery (e.g., cystectomy or ovarian wedge resection). Wei et al^[22] found that ovary-sparing surgery was performed in only 50% (4 of 8) of the cases of ovarian leiomyomata noted in pediatric and young adult patients (aged under 25 years). Lim and Jeon^[23] reported an unfortunate case of a 17-year-old patient with bilateral ovarian leiomyoma who underwent bilateral salpingo-oophorectomy, where no histologically recognizable normal ovarian tissue was present. Malignant ovarian tumors are more common than ovarian leiomyoma in all age groups, including the reproductive age group, and they are associated with a much poorer prognosis than that of ovarian leiomyomata. Therefore, the decision to perform an ovary-preserving surgery should be carefully considered after a thorough evaluation of the possibility of malignancy.^[6] Progesterone, which is typically prescribed to prevent miscarriage in early pregnancy, was not prescribed postoperatively in the case described here because the patient was admitted at 10 weeks of gestation, whereas the production of progesterone shifts from the corpus luteum to the placenta at ~8 weeks of gestation.^[24] Indeed, the patient described here delivered a healthy baby at full term, and no miscarriage occurred even in the absence of progesterone supplementation. Similarly, the patient described by Zhao et al^[21] was admitted at 14 weeks of gestation, and therefore did not require progesterone supplementation after laparoscopic excision of the ovarian leiomyoma; however, Zhao et al did prescribe progesterone for 4 days postoperatively.

A frozen section examination may be helpful to determine the necessary extent of surgery, considering the high sensitivity and specificity of the technique for ovarian tumor (87–100% and 99–100%, respectively).^[25] However, frozen section examination could not be performed in the case described here because the procedure was unavailable in the hospital during the night, which is when emergency laparoscopic exploration was performed.

In general, it is very worrisome to have surgery during pregnancy. However, patients with torsion of pedunculated myoma or adnexal mass should be indicated for emergent surgery even during pregnancy. Complete occlusion of the ovarian blood supply will cause severe pain and ultimately result in loss of ovarian function and necrosis of the torsioned tissues. Although hemorrhage and peritonitis may also occur as complications of such conditions, sepsis and hemorrhage requiring blood transfusion have rarely been reported. The necrotic tissue will involute over time, but there may be pelvic adhesion formation, which can result in pelvic pain or infertility. If the patient is pregnant, fetal loss can also occur.

Mathevet et al^[26] reviewed 47 pregnant women (17 in the first trimester, 27 in the second trimester, and 4 in the third trimester) who underwent laparoscopic management of ovarian cysts (n = 36), torsion (n = 8), or pelvic mass (n = 3). In this case series, only 1 pregnancy loss occurred, in the form of fetal death on the fourth day after an uncomplicated intra-peritoneal laparoscopic ovarian

cystectomy performed at 17 weeks of gestation. These observations suggest that the laparoscopic approach remains a safe option even in pregnancy. Regarding the case described in the present report, despite the fact that the patient was pregnant, the author decided to perform emergent laparoscopic surgery especially because of the patient's intractable abdominal pain. After being informed of the risks, the patient provided informed consent to undergo the procedure.

Anesthetic agents are not known teratogens; however, because it is currently not possible to conclusively rule out adverse effects, exposure to medications should be minimized during pregnancy. Furthermore, during surgery, the uterus should be displaced left laterally, in order to reduce the risk of hypotension in the second half of pregnancy. It is important to minimize disruption of fetal homeostasis by avoiding maternal hypotension, hypoxemia, and hyper- or hypocarbia. Postoperative care may include opioid and antiemetic drugs, as needed, to control postoperative pain and nausea. Nonsteroidal anti-inflammatory drugs should be avoided, especially after 32 weeks of gestation, because they may cause premature closure of the fetal ductus arteriosus. The patient described in the present report was administered meperidine for pain control. Additionally, the patient received cephalosporin, which is known to be safe for use during pregnancy, as is penicillin. Fetal monitoring using ultrasound is also helpful.

Previously, Wei et al^[22] also suggested that estrogens may be a significant factor for the development of ovarian leiomyoma. On the other hand, most uterine leiomyomata do not increase in size during pregnancy. Pregnancy has a variable and unpredictable effect on leiomyoma growth, which is likely related to individual differences in leiomyoma gene expression, circulating growth factors, and fibroid cell receptors.^[27,28] Out of the 9 cases of primary ovarian leiomyomata in pregnancy described to date, 5 cases^[14,16,18,19,21] exhibited a rapid increase in tumor size during early pregnancy, whereas the growth of ovarian leiomyomata in 2 cases^[9,20] was similar to that of uterine leiomyoma typically found in patients of reproductive age. It seems that primary ovarian leiomyomata have a tendency to grow rapidly during early pregnancy. If a woman has an adnexal mass suspected to be either pedunculated subserosal leiomyoma or ovarian fibroma, it is reasonable to suspect that the tumor is a primary ovarian leiomyoma provided that the mass grows rapidly during early pregnancy.

4. Conclusion

This case report is the first to describe the successful laparoscopic management of twisted ovarian leiomyoma in a pregnant woman with acute abdomen. Ovarian leiomyoma is a rare ovarian tumor and rarely causes acute abdomen. It is difficult to diagnose ovarian leiomyomata preoperatively or even during surgery, because this condition is clinically indistinguishable from subserosal leiomyoma and ovarian fibroma. Even during pregnancy, laparoscopic exploration is the preferable way to diagnose and remove twisted ovarian leiomyoma through the extended trocar site after the mass has been morcellated manually using a cold knife. Primary ovarian leiomyomata have a tendency to grow rapidly during early pregnancy, whereas uterine leiomyomata and benign ovarian tumors seldom grow during pregnancy. However, the effects of estrogen and pregnancy on ovarian leiomyoma remain unclear because of the low incidence of ovarian leiomyomata.

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