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CASE REPORT

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Uterine lipoleiomyoma misdiagnosed as uterine fibroid in a pregnant woman: a case report and literature review

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Abstract

Uterine lipoleiomyoma is a rare variant of leiomyomas, especially in pregnant women. Currently, there is only one case reported in literature, suggesting that without proper knowledge about this disease, there is a high risk that uterine lipoleiomyoma could be misdiagnosed as other gynecological diseases, particularly during pregnancy. Here, we report the case of a 38-year-old female admitted to our department due to intermittent lower abdominal pain and a small amount of vaginal bleeding after menopause. Based on her clinical manifestations, laboratory tests and imaging examinations, she was preoperatively diagnosed as pregnant with uterine fibroid fatty degeneration. Following an induced abortion, she underwent a laparoscopic myomectomy and was diagnosed with uterine lipoleiomyoma on postoperative immunohistochemistry. During a 6-month follow-up, no abnormalities were detected. The obstetric outcomes, diagnosis and treatments of pregnancy with uterine lipoleiomyoma may resemble those of pregnancy with uterine fibroid. Although comprehensive imaging examinations are helpful for preoperative diagnosis, the final diagnosis still relies on postoperative pathological examination.

Keywords

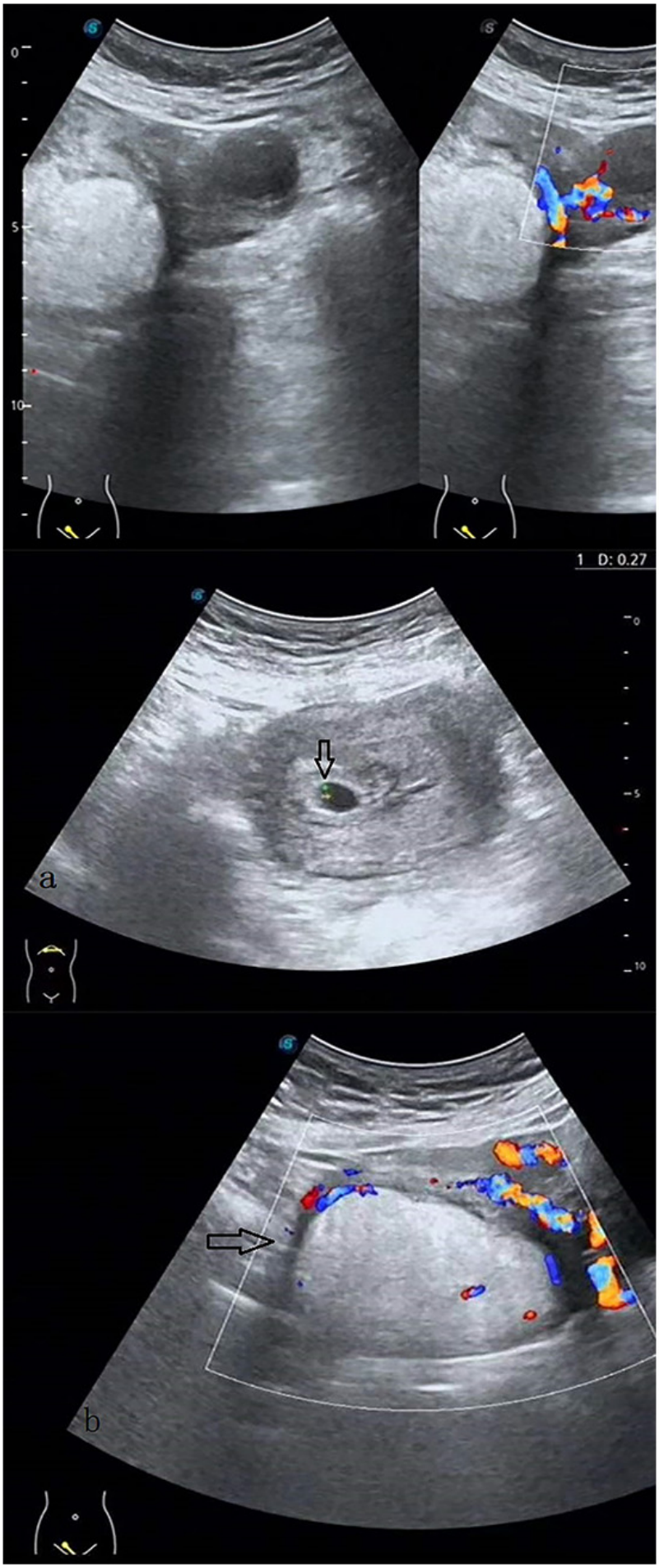
Misdiagnosis; Pregnancy; Uterine fibroid; Uterine lipoleiomyoma

1. Introduction

Uterine lipoleiomyoma is a rare and benign tumor composed of mature adipocytes and smooth muscle cells, with an incidence rate of 0.03%–0.20% [1, 2]. It commonly occurs in menopausal and postmenopausal women and is often accompanied by obesity, hypertension, gallbladder diseases, diabetes and thyroid disorders [3]. Currently, there is only one reported case of uterine lipoleiomyoma in pregnancy in literature [4]. Herein, we report the case of a 38-year-old female who was initially misdiagnosed as pregnant with uterine fibroid fatty degeneration, which we believe could be due to little knowledge on uterine lipoleiomyoma in pregnancy. Thus, we also reviewed corresponding literature to advance our understanding on this disease.

2. Case presentation

A 38-year-old woman of child-bearing age with regular menstruation and normal last menstrual period (LMP) with the same menstrual amount and blood color was observed. She was first admitted to a local county hospital and then referred to our hospital. The patient presented to our hospital with intermittent lower abdominal pain discomfort for 10 days and a small amount of vaginal bleeding for 1 day after a month of menopause. She denied other symptoms such as fever, abdominal distention, nausea, vomiting and weight loss. The patient had no remarkable past medical history and no special personal or family history. Gynecologic examinations revealed uterine enlargements with a soft texture and no tenderness and that she was approximately over three months pregnant. No abnormal sounds were detected during auscultation, and there was no tenderness, rebound pain and muscle tension over the abdominal swelling. Her urinary human chorionic gonadotropin (HCG) at the local hospital was positive for pregnancy. Additional laboratory tests at our hospital showed an increase in serum concentration of HCG to 8751.00 mIU/mL (normal range: 0.00–30.00 mIU/mL) and serum carbohydrate antigen 125 (CA-125) to 823.00 U/mL (normal range: 0.00–35.00 U/mL), while the level of other tumor markers was within normal range. Electrocardiogram, chest X-ray and arterial blood gas were also normal. The gynecological ultrasound at our hospital identified two abnormalities (Fig. 1): 1 a gestational sac in the uterine cavity with a yolk sac but without embryo tissue and a liquid dark area around the gestational sac (Fig. 1a); 1 a hyperechoic regular-shaped lesion of size 7.7cm × 5.8cm with clear boundaries, but the internal echo of the lesion was heterogenic, and the blood flow signal was found in the peripheral locations (Fig. 1b). Based on these findings, the patient was initially diagnosed with early in-



F I G U R E 1. Gynecological ultrasound: (a) A gestational sac in the uterine cavity; (b) Hyperechoic lesion at the posterior wall of the uterus.

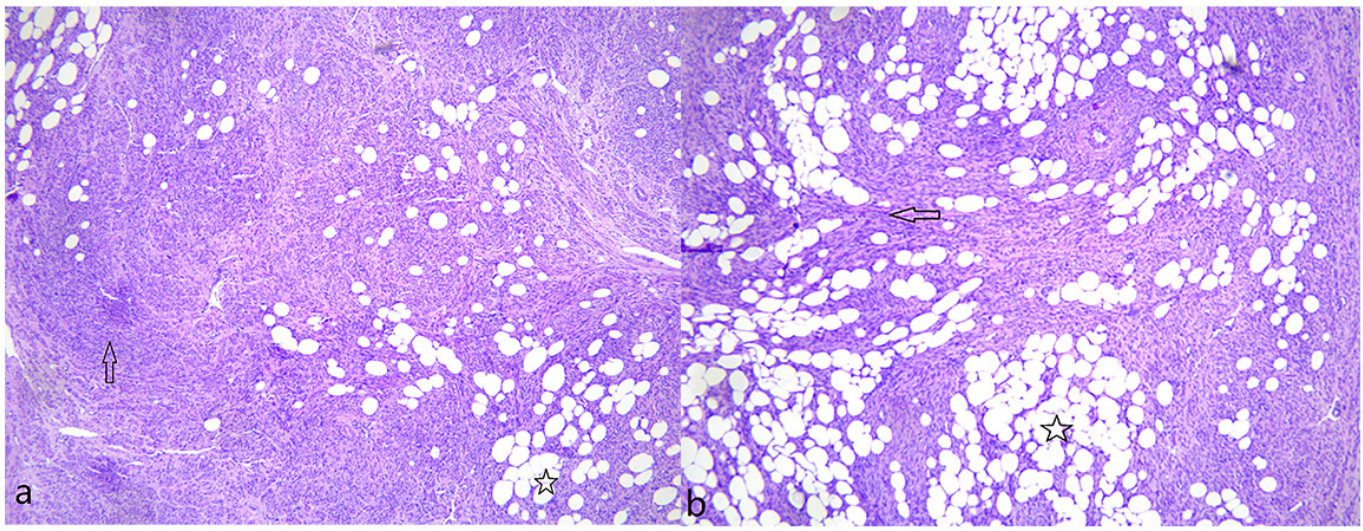


FIGURE 2. Histopathological examination of the uterine lipoleiomyoma: mature adipocytes (stars) together with smooth muscle cells (arrows). (a: hematoxylin and eosin, 40×; b: hematoxylin and eosin, 100×).

trauterine pregnancy and uterine fibroid with fatty degeneration. Since she already had three children, she requested an induced abortion, which was performed on the third day of admission. The patient was discharged from the hospital after five days of hospitalization and was recommended to return to the hospital within 2 weeks for further treatments. However, she returned after 45 days since discharge and underwent laparoscopic surgery under general anesthesia on the third day of the second admission. During laparoscopy, the lesion was of size 7cm × 6cm, originated from the back wall of the uterus and went deep into the myometrium without penetrating the endometrium. The lesion was completely removed during the surgery, and the uterus was completely repaired. The intrauterine samples were placental villi, decidua and secretory endometrial membrane, consistent with intrauterine pregnancy changes. Histopathological characteristics of the myometrium samples (Fig. 2) were compatible with lipoleiomyoma. Her postoperative immunohistochemical staining was as follows: Estrogen Receptor (ER) (+), Progesterone Receptor (PR) (+), desmin (+), h-caldesmon (+), S100 (−), β-catenin (+), Mouse Double Minute 2 (MDM2) (−), Cyclin Dependent Kinase 4 (CDK4) (−), Human Melanoma Antibody 45 (HMB45) (−), MelanA (−), Cluster of Differentiation (CD) 117 (−), CD34 (−), Dog-1 (−) and Ki67 positive cells 1%, consistent with the diagnosis of lipoleiomyoma. Thus, her final diagnosis was early intrauterine pregnancy associated with uterine lipoleiomyoma. Overall, the patient's postoperative course in the six months after surgery was uncomplicated, she was asymptomatic, ultrasound examination showed no evidence of any abnormality in her pelvic cavity, and her CA-125 returned to normal range after three months following the surgery.

3. Discussion

Uterine lipoleiomyoma is a rare and easily misdiagnosed benign uterine tumor usually found incidentally during surgery. It occurs in perimenopausal and postmenopausal women [2]. Patients with uterine lipoleiomyoma either have no obvious clinical symptoms or present with symptoms similar to uterine fibroids, such as menstrual disorders, abdominal mass, abdominal pain, urinary frequency and urinary incontinence [3, 5]. Diagnosing abdominal-pelvic masses in women is challenging because the patients' clinical features and findings from physical examination are usually nonspecific. Tumor markers, such as CA-125, are useful tools that can help distinguish between benign and malignant masses. However, according to current literature, although the combination of a pelvic mass with a high level of CA-125 arouses suspicion of gynecological malignancy, there is limited evidence to support an association between elevated CA-125 levels and uterine lipoleiomyoma [6]. Ultrasound is preferred for gynecological diseases, and most of the manifestations of uterine lipoleiomyoma in ultrasound are clearly defined as hyperechoic lesions with hypoechoic edges [7]. Computed tomography (CT) may show localized or diffused fat density with solid interlaced components, while enhanced CT scans may show slight enhancement of the solid components of the lesions but no enhancement of fat density [8]. Magnetic Resonance Imaging (MRI) is also an important imaging examination for diagnosing uterine lipoleiomyoma [9], in which fat tissue shows high signal T1/T2 as well as a signal loss on diffusion-weighted imaging (DWI). Although CT and MRI are also useful for distinguishing between lipoleiomyoma and simple lipoma, histopathology and immunohistochemistry are still required to confirm the diagnosis. On histopathology, the lesions are characterized by different proportions of mature adipocytes and smooth muscle cells, where the mature adipocytes portray a focal or diffuse distribution in the tumor without heterogeneity [2]. Immunohistochemical features of lipoleiomyoma include positive desmin protein and smooth muscle actin (SMA) in the smooth muscle cells, positive S100 protein in adipocytes, positive Vimentin, positive ER, and PR in some cells, with Ki67 cell number positivity $<1\%$ [8, 10].

Additionally, uterine lipoleiomyoma can be easily misdiagnosed as other diseases, such as uterine fibroids fat degener-

ation, a rare type of uterine fibroids degeneration that mostly occurs in the late of transparent degeneration or after necrosis and tends to occur in perimenopausal and postmenopausal women. Due to very similar clinical characteristics and features with uterine lipoleiomyoma, the two can be easily confused, especially via preoperative ultrasound. On MRI, when uterine fibroids fat degeneration occurs, mixed signals can be detected inside the lesions, often in conjunction with other degenerations such as cystic degeneration and calcification. However, lipoleiomyoma has no degeneration manifestations [10]. Uterine fibroids fat degeneration in the histopathological section manifests as smooth muscle cells containing lipid droplets instead of fat cells, while the fatty component in lipoleiomyoma is real fat cells.

The treatments of uterine lipoleiomyoma and uterine fibroids are similar and are selected based on age, fertility requirements and severity of symptoms [3]. In perimenopausal and postmenopausal asymptomatic patients, the lesion can only be suspected by follow-up observation. Those with symptoms or without follow-up are recommended to undergo surgical treatment. The gold standard surgical treatment of uterine lipoleiomyoma is total hysterectomy. Its prognosis depends on the presence of tumor necrotic foci, atypia and mitoses per high-power field (HPF) under the microscope. Although uterine lipoleiomyoma are benign lesions, they should be viewed as low-grade malignant lesions and necessitate careful follow-up when histopathological characteristics such as abnormal mitoses, cellular and nuclear atypia, local infiltrating tendencies and vascular invasion are detected [11].

In current literature, only one case of uterine lipoleiomyoma during pregnancy has been reported. The patient was diagnosed at 37 weeks of gestation, and her lesion was principally located in the anterior lip of the cervix. In addition, these two women (past literature and current case) were illiterate, belonged to the low socioeconomic group, did not receive antenatal care throughout the pregnancy, and had no specific clinical symptoms. Thus, limitations in economic conditions and lack of adequate health examinations can make it very challenging to fully understand the incidence of uterine lipoleiomyoma in pregnancy, the relationship between uterine lipoleiomyoma and pregnancy and whether pregnancy may cause uterine lipoleiomyoma changes.

Since patients with uterine lipoleiomyoma commonly present similar symptoms as uterine fibroids [3] and considering that the patient reported in this study demonstrated clinical characteristics similar to pregnancy with uterine fibroids such as abdominal pain followed by vaginal bleeding after menopause, it is speculated that the clinical symptoms of pregnancy with uterine lipoleiomyoma is similar to those of pregnancy with uterine fibroids. Although uterine fibroids during pregnancy may be asymptomatic, they could lead to obstetric complications such as early abortion, abdominal pain due to red degeneration, premature delivery, premature rupture of membranes, fetal abnormalities, intrapartum and postpartum hemorrhage and uterine torsion [10]. Based on these, we speculate that uterine lipoleiomyoma may also lead to similar adverse obstetric outcomes. In this study, the patient presented with symptoms of threatened abortion.

The general treatment principle of pregnancy with uterine fibroids should be based on the pregnancy month, fibroid size, clinical symptoms, growth site and the patient's needs [12]. Generally, conservative treatment is the first choice of treatment. Asymptomatic and small uterine fibroids in early pregnancy do not require treatment. The patient can have regular examinations to observe any changes in the size of the uterine fibroids. Those with symptoms of threatened abortion need bed rest, strengthened nutrition and abstinence from sexual intercourse. They can also be given appropriate drugs to protect the fetus. If the symptoms improve in the short term, the pregnancy can be continued; otherwise, repeated symptoms of inevitable abortion or recurrent abortion may increase the risks of obstetric complications. Further, if the fibroid is very large, may seriously influence the pregnancy, or the patient does not want to continue the pregnancy and requests for an abortion, then abortion should be considered, followed by myomectomy [13, 14]. In this study, the patient's condition was strictly distinguished from normal menstrual dysmenorrhea, abortion and ectopic pregnancy. Her gynecological ultrasound confirmed intrauterine pregnancy, indicating threatened abortion. She had symptoms of threatened abortion during the early weeks of pregnancy, the lesion was large and located between the muscular wall, and she requested an abortion. Thus, a laparoscopic myomectomy was performed after an induced abortion, aligning well with the recommended treatment principles.

Due to existing deficiencies in the diagnosis and treatment of uterine lipoleiomyoma, the patient admitted to our hospital was misdiagnosed with uterine fibroid fatty degeneration. The reasons were as follows: First, the patient did not undergo regular routine physical examinations before admission, and we could not determine the lesion's occurrence time and growth rate. Thus, based on her clinical symptoms of vaginal bleeding and lower abdominal pain after menopause, she was easily misdiagnosed with a more common disease—uterine fibroid, and it was unlikely to determine the relationship between the development of abortion and uterine lipoleiomyoma at that stage. Second, the preoperative examination of the patient was not sufficient. We only performed gynecological ultrasound without further CT and MRI examination, which may have contributed to the misdiagnosis. Third, the patient lost the possibility of continuing the pregnancy, and therefore it was not possible to observe the developments and changes of the uterine lipoleiomyoma during the whole process of pregnancy and its impact on the pregnancy outcomes. Thus, it was difficult to differentiate uterine lipoleiomyoma from uterine fibroids in this pregnant woman before surgery.

From this case, it should be noted that the misdiagnosis was due to its rarity, lack of knowledge on the disease and preoperative examinations were not comprehensive. Therefore, we propose the following suggestions to avoid similar misdiagnosis situations in the future: First, ultrasonography is a must and should be the first choice of examination. For patients with suspected benign or malignant lesions on ultrasonography, serum tumor markers should be tested. For those with positive tumor markers, CT or MRI examination should be further considered. Second, a rapid perioperative pathological examination should be considered as it could guide the determination of surgical methods and surgical scope. Lastly, it is our responsibility

and obligation to educate the population about the importance of health examinations, and it is advisable that women should have a comprehensive gynecological examination before when planning pregnancy and should also undergo regular routine prenatal check-ups as per obstetrist recommendation to timely identify and treat any potential anomalies.

4. Conclusion

In conclusion, uterine lipoleiomyoma is a rare benign tumor, and its diagnosis depends on a combination of clinical data, pathological features and immunohistochemistry. Although the clinical manifestations and treatments of pregnancy with uterine lipoleiomyoma may be similar to pregnancy with uterine fibroids, more investigations are needed to determine its optimal treatment. Here, we report this rare case to create more awareness on this disease and emphasize that all aspects of pregnancy-related complications should be considered to properly diagnose any underlying condition and formulate an appropriate individualized treatment. Further, due to the lack of literature on uterine lipoleiomyoma during pregnancy, doctors must remain highly alert to this condition and accumulate more experience in its diagnosis and treatments to better treat these patients.

AUTHOR CONTRIBUTIONS

YYW—collected and analyzed the data, drafted the manuscript; LPL and WF—were the patient's gynecologists, performed the operation and managemented the patient; RFZ—collected and analyzed the imaging findings; XFF—reviewed the literature and contributed to manuscript drafting; YZG—was responsible for the revision of the manuscript for important intellectual content. All authors issued final approval for the version to be submitted.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

The patient gave her written informed consent to publish her case. All clinical information and images used in this paper were approved by the ethical committee of the Second Hospital of Lanzhou University.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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