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Abdominal pregnancy implanted on surface of pedunculated subserosal uterine leiomyoma: A case report

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ABSTRACT

Abdominal pregnancy is a rare form of ectopic pregnancy. Various sites of implantation in abdominal pregnancy have been reported. Uterine serosa is an extremely rare implantation site, with only a few cases reported to date. No case of abdominal pregnancy implanted on the surface of a subserosal uterine leiomyoma has been reported. We herein report the case of a 40-year-old primigravida woman who was diagnosed with abdominal pregnancy implanted on the surface of a pedunculated subserosal uterine leiomyoma. The uterine leiomyoma with gestational tissue was resected laparoscopically and the postoperative course was uneventful. It is necessary to remember the possibility of unexpected implantation sites and that laparoscopic surgery may be more difficult in such cases than that for fallopian tube pregnancy.

1. Introduction

Abdominal pregnancy is a rare form of ectopic pregnancy. Ectopic pregnancy accounts for 1% of all pregnancies; abdominal pregnancy accounts for 1%–4% of all ectopic pregnancies [1]. Early diagnosis of abdominal pregnancy is difficult; the diagnosis is not generally made until ectopic mass rupture with bleeding in the abdominal cavity. The maternal mortality rate of abdominal pregnancy is about 8 times greater than that of tubal pregnancy.

In abdominal ectopic pregnancy, products of conception commonly implant on the peritoneum of the vesicouterine pouch or on the peritoneum of the posterior cul-de-sac. Uterine serosal pregnancy is extremely rare. In uterine serosal pregnancy, the fetus is implanted within the uterine serosa, without a connection to the endometrial cavity, fallopian tubes, or round ligament. Only a few cases of uterine serosal pregnancy have been reported [2]. No case of abdominal ectopic pregnancy implanted on the surface of a subserosal uterine leiomyoma has been reported. We herein report the case of a 40-year-old primigravida woman who was diagnosed via laparoscopic intervention with abdominal ectopic pregnancy on the surface of a pedunculated subserosal uterine leiomyoma.

2. Case report

A 40-year-old primigravida woman was referred for further examination and treatment after suspected ectopic pregnancy was diagnosed at a fertility clinic. A subserosal uterine leiomyoma measuring 6 cm in diameter had been diagnosed about 2 years before the pregnancy.

The pregnancy had been achieved by in vitro fertilization and a 2-step embryo transfer procedure, with cleaved-embryo transfer on day 2 and blastocyst transfer on day 5 in the clinic. The patient's serum β-human chorionic gonadotropin (hCG) was 0.9 IU/L at 4 weeks, 1 day of gestation, estimated according to the embryo transfer day. About 1 week later, a urinary hCG test was positive. Transvaginal ultrasound revealed no gestational sac (GS) in the uterus and serum β-hCG was 385 IU/L at 5 weeks, 5 days of gestation. At 6 weeks, 4 days of gestation, the patient was asymptomatic and no GS was detected in the pelvic organs, including the uterus, although serum β-hCG had risen to 1174 IU/L. An ectopic pregnancy was suspected, and the patient was referred from the fertility clinic on the next day.

At presentation, the patient was asymptomatic and her clinical findings had not changed. Transvaginal ultrasound revealed a subserosal uterine leiomyoma measuring 6.1 cm in diameter. No GS

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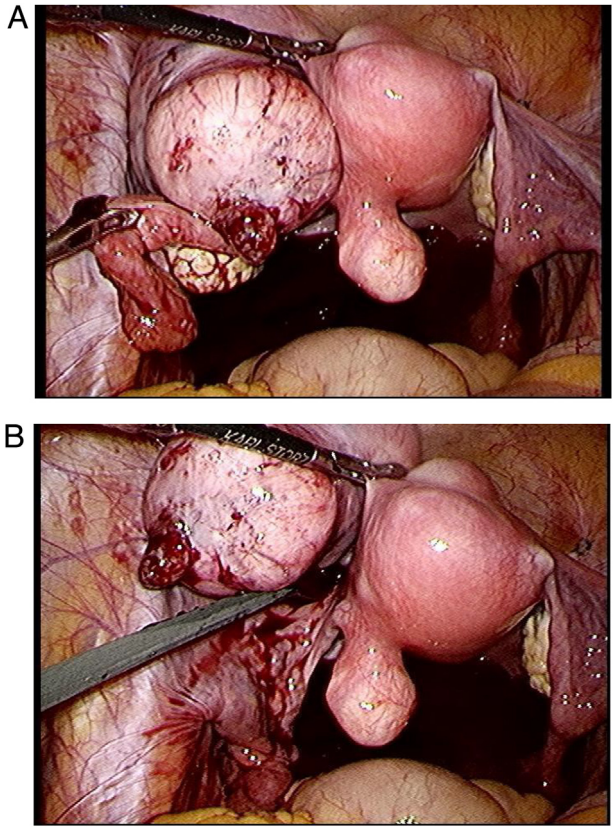

Fig. 1. (A↘B)Laparoscopic findings. A ruptured ectopic mass is observed on the surface of the pedunculated subserosal leiomyoma. Both adnexae were grossly normal. Another small pedunculated subserosal leiomyoma was found on the left uterine corner.

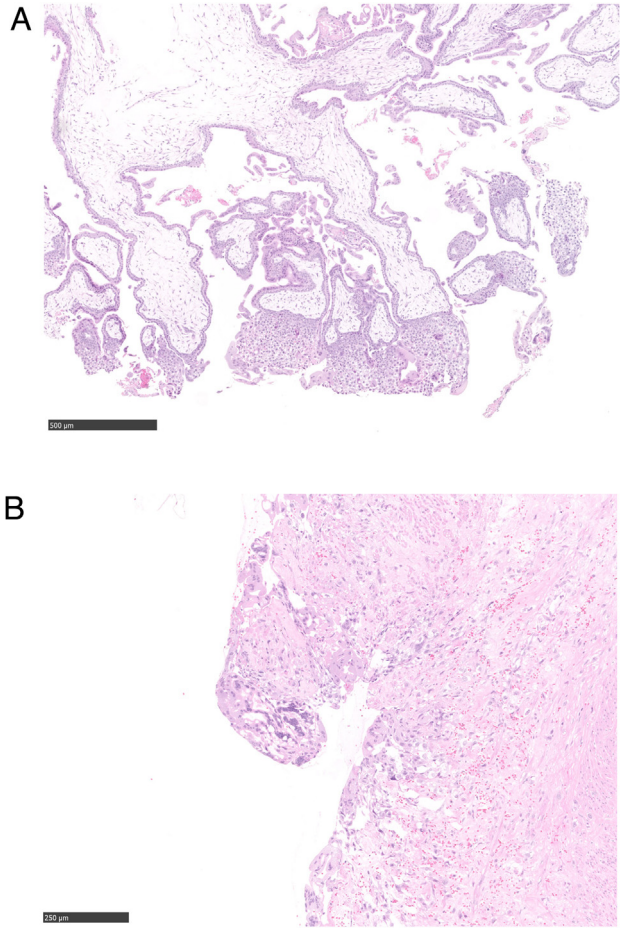

Fig. 2. (A) Microscopic photograph of trophoblastic villi removed from surface of pedunculated subserosal leiomyoma. (Hematoxylin & eosin stain, ×100).
(B) Trophoblast cells are present on surface of removed uterine leiomyoma. (Hematoxylin & eosin stain, ×200).

was detected; the endometrial thickness was 3.4 mm. The patient's serum β-hCG was 1396.3 IU/L, which was relatively low, although the presumed diagnosis was ectopic pregnancy. Intrauterine pregnancy, such as miscarriage, was possible, although it was unlikely. If the patient's serum β-hCG had decreased at the next visit, we could have managed the case expectantly. At that time, we concluded that it was premature to perform medical and/or surgical treatment of an ectopic pregnancy. Therefore, close observation was planned. The patient reported lower abdominal pain 5 days after her initial visit. Her vital signs were stable. Transvaginal ultrasound revealed fluid in the cul-de-sac, suggesting the presence of intraperitoneal bleeding resulting from rupture of an ectopic pregnancy.

Laparoscopic surgery was performed. A hemoperitoneum of approximately 700 mL was present. A pedunculated subserosal uterine leiomyoma was found on the left side of the uterine fundus. Gestational tissue was observed on the surface of the leiomyoma. Both uterine adnexae were normal (Fig. 1A, B). Another small pedunculated subserosal uterine leiomyoma was found on the left uterine corner. Two small non-pedunculated subserosal uterine leiomyomas were found at the anterior wall of the uterus and at the fundus of the uterus. No findings of congenital uterine malformation were seen. There were no other findings suggesting ectopic pregnancy anywhere in the pelvis. The laparoscopic diagnosis was ruptured abdominal ectopic pregnancy on the surface of a pedunculated subserosal uterine leiomyoma.

The stalk of the pedunculated uterine leiomyoma was coagulated and transected during laparoscopic surgery. The resected uterine leiomyoma with gestational tissue was placed in a polyurethane bag in the abdominal cavity and removed through minilaparotomy without electric morcellation. Allogenic transfusion was not needed.

The patient's postoperative course was uneventful and her serum hCG level declined rapidly. Serum β-hCG was 1.9 IU/L 20 days after surgery. Histopathologic examination confirmed trophoblastic villi and trophoblast cells on the surface of the removed uterine leiomyoma (Fig. 2A, B). Written, informed consent was given by the patient for investigation and publication.

3. Discussion

Abdominal ectopic pregnancies comprise less than 1% to 4% of all ectopic pregnancies. [1] However, abdominal ectopic pregnancy has a maternal mortality rate about 8 times greater than tubal ectopic pregnancies. Uterine serosal pregnancy is extremely rare. In uterine serosal pregnancy, the fetus is implanted within the uterine serosa, without a connection to the endometrial cavity, fallopian tubes, or round ligament [2]. Various sites of implantation have been reported in abdominal pregnancy. Our case may be broadly classified as uterine serosal pregnancy. To our knowledge, this is the first case of an ectopic pregnancy implanted on the surface of a pedunculated subserosal leiomyoma.

Primary abdominal pregnancies are exceedingly rare. The diagnosis is based on the following criteria: (i) normal fallopian tubes and ovaries; (ii) absence of uteroperitoneal fistula; and (iii) pregnancy that is attached only to the peritoneal surface and is diagnosed early, excluding the possibility of secondary abdominal pregnancy [3]. Our case met these criteria because the ectopic preg-

nancy was implanted on the surface of a pedunculated subserosal leiomyoma.

Yoder et al. reported a systematic review of 29 cases of abdominal pregnancy after IVF [4]. Several trends were identified. Tubal factor infertility is a known risk factor for ectopic pregnancy following IVF. A history of tubal pregnancy or prior tubal surgery was particularly common among abdominal ectopic cases in the systematic review. The cause of infertility in our patient was unknown. She had no history of previous abdominal or pelvic surgery. Her fallopian tubes were grossly normal. However, she had two pedunculated subserosal uterine leiomyomas. They were at the left side of uterus and near the left uterine corner. The left fallopian tube was easily compressed, with movable pedunculated subserosal uterine leiomyomas. They may have prevented normal function of the left fallopian tube and have led the abdominal pregnancy. Pedunculated subserosal leiomyomas increase the surface area of the uterine serosa more than other types of uterine leiomyoma. This increased surface area may have contributed to the abdominal pregnancy in the present case.

Several studies have investigated the value of β-hCG in the prediction of viable pregnancy after IVF and have found correlations between β-hCG levels and pregnancy outcome [5]. Therefore, serumβ-hCG levels are often evaluated after IVF. In our patient, the serum β-hCG level was very low at 4 weeks, 1 day after embryo transfer. Irani et al. reported a case of abdominal pregnancy in which serum β-hCG was undetectable 9 days after blastocyst transfer [6]. A delayed rise in serum β-hCG titers may raise the suspicion of abdominal pregnancy. Therefore, close monitoring of serum β-hCG titers is necessary.

Pedunculated subserosal leiomyoma can be have complications such as acute torsion and acute pain caused by red degeneration during pregnancy [7]. Pedunculated subserosal leiomyoma was diagnosed before pregnancy in our patient. Although the question of whether myomectomy should precede pregnancy is controversial, resection of a pedunculated subserosal leiomyoma before pregnancy may be recommended.

In this patient, resection of the pedunculated subserosal myoma was not difficult. Control of bleeding was easy. However, the operation would have been more difficult and required more time if the subserosal leiomyoma had not been pedunculated. Leiomyomas need to be morcellated electrically or manually to be retrieved from the abdominal cavity. Morcellation without bag may allow some morcellated fragments to drop into the abdominal cavity. Electrical morcellation may mix gestational tissue with the morcellated leiomyoma. In the present case, the resected leiomyoma with gestational tissue was placed in a bag and morcellated manually with a surgical scalpel to allow histological evaluation of gestational tissue and to prevent secondary ectopic pregnancy caused by spread of trophoblastic cells.

As the number of IVF procedures performed continues to rise, the incidence of ectopic and abdominal ectopic pregnancy will likely also rise. In hemodynamically stable patients with ectopic pregnancy, laparoscopic surgery is usually selected. The fallopian tube is the implantation site in more than 90% of cases. Fallopian tube pregnancy is treatable with salpingectomy or salpingotomy. However, the mode of operation in abdominal pregnancy depends on the implantation site. In uterine serosa pregnancy, the myometrium at the implantation site should usually be resected. After resection, suturing of the uterine myometrium is often necessary. In resection of other implantation sites, surgeons should avoid injury to adjacent organs, such as the bladder, ureter, and rectum. In ectopic pregnancy, it is necessary to remember the possibility of unexpected implantation sites and that laparoscopic surgery may be more difficult in such cases than that for fallopian tube pregnancy.

Contributors

Hiroshi Sato designed the study, and wrote the initial draft of the manuscript.

Masaya Hirose contributed to analysis and interpretation of data, and assisted in the preparation of the manuscript.

All other authors contributed to data collection and interpretation, and critically reviewed the manuscript. All authors saw and approved the final version of the manuscript.

Declaration of competing interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

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Patient consent

Written, informed consent was given by the patient for investigation and publication.

Provenance and peer review

This case report was peer reviewed.

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