

Intramural pregnancy following transcervical resection of adhesion: a case and review literatures

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Summary

An intramural pregnancy is an unusual type of ectopic pregnancy in which the gestational sac is situated in the myometrium, separate from the endometrial cavity and fallopian tubes. There are only 22 cases in the published literature via search of PubMed from 2010 to 2018. The authors report an additional rare case of ectopic pregnancy which underwent previous surgery for intrauterine adhesions (IUA) treated by laparoscopy and they summarize the current data regarding etiology, diagnosis, and management from literature.

Key words: Intramural pregnancy; Ectopic pregnancy; Intrauterine adhesions.

Introduction

Intramural pregnancy is an infrequent form of ectopic pregnancy. Although it accounts for less than 1% of all the ectopic pregnancy [1], and little is known about its etiology, it can seriously threaten maternal life. However, its diagnosis and therapy remain a clinical challenge. If early diagnosis is missed, the woman's future fertility and even her life are at risk.

The authors reported a case of intramural pregnancy in a woman with previous surgery for intrauterine adhesions (IUA). This study was approved by the ethics committee of this hospital.

Case Report

A 32-year-old female, G4P1A3, had a normal pregnancy in 2015. She then underwent transcervical resection of adhesion that was performed in 2016, because her postpartum menstruation did not return. Postoperative recovery was achieved. On March 5, 2018, she presented to the hospital for a missed abortion. In another hospital, one week prior, she was initially misdiagnosed at approximately eight weeks of gestation and then medical abortion and suction curettage were performed. However, the pregnancy tissue was not completely removed after that and ultrasound image confirmed intrauterine fetal residual. Therefore, a ten-week scan showed a normal fetus which was described to be highly localized in the uterus (Figure A). The obstetric examination showed no vaginal hemorrhage, no uterine dilatation, neither uterine contractions nor abdominal pain. The patient vital signs were stable. These findings aroused suspicion of either an angular pregnancy or an intramural pregnancy. This is what prompted the gynecologists to perform an ultrasound-guided revision of the uterine cavity. Unfortunately, during this procedure, it was not possible to reach the gestational sac, so it was decided to perform hysteroscopy which revealed an empty uterine cavity with endometrial thinness and visible bilateral ostia,

which negated the possibility of cornual pregnancy. Then it was deemed necessary to introduce diagnostic laparoscopy, which revealed that the uterine size was larger with an unruptured mass which protruded from the fundal myometrium and was distinct from both fallopian tubes (Figure B). the ovaries and the fallopian tubes appeared normal. After the diagnosis of intramural preg-

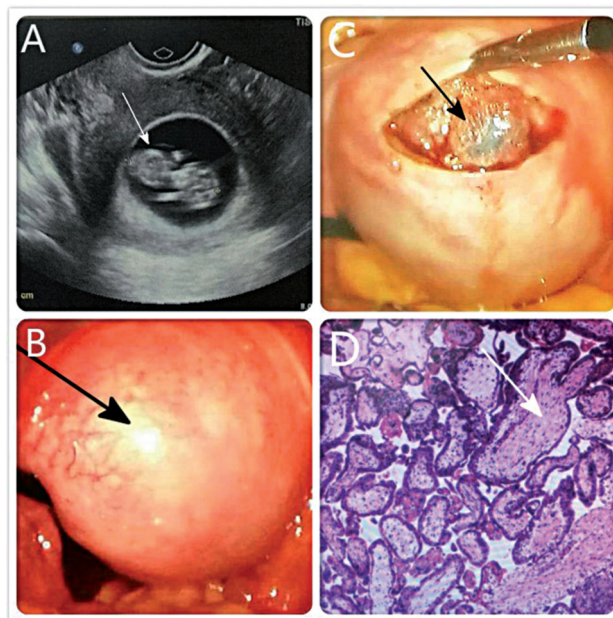


Figure 1. — (A) Preoperative transvaginal ultrasound scan reveals a 10-week normal fetus which was described to be highly localized in the uterus. (B) Laparoscopic view shows that the uterine size is larger with an unruptured mass which protrudes from the fundal myometrium and is distinct from both fallopian tubes. (C) After incision of the serous layer, the fetus and clear amniotic fluid outflow are observed. (D) Pathological diagnosis showed the presence of chorionic villi in the intramural pregnancy.

nancy, terlipressin 6U was injected into the myometrium around the base of the mass. Surgery was performed and an incision was made in the uterine serosa. After incising the serous layer, the fetus and clear amniotic fluid outflow were observed (Figure C), and the tissue and fetal body were absorbed. Then the authors found that the tissue was implanted in the myometrium without a connection to the endometrial cavity or fallopian tubes. Then the wound was sutured with number 1 monocryl sutures. The patient's postoperative condition was stable five days after surgery, and the pathological diagnosis confirmed the presence of chorionic villi in the myometrium (Figure D).

Discussion

Intramural pregnancy refers to a gestational sac located within myometrium, separated from the endometrial cavity and the fallopian tubes [2]. There have been only a handful of cases reported in the world literature to date. It was reported in 1913 by Doederlein *et al.* [3] for the first time that the incidence rate was about 1/30,000 of the pregnancies, which represents less than 1% of all ectopic pregnancies [1]. The authors performed a systemic search in PubMed from 2010 to 2018. The database search with the query term "intramural pregnancy", resulted in 22 reports which were selected, and of these only six reports describe laparoscopic resection of intramural pregnancy, while the other reports discussed surgical management which were often performed via laparotomy and included hysterectomy. Due to the lack of specificity in the early diagnosis of intramural pregnancy, most of the clinical cases are diagnosed when emergency surgery is performed on an acute abdomen, hence optimal treatment time is missed. According to statistics, the uterine rupture of intramural pregnancy in China is up to 28% [4], and the uterine rupture occurs during the period from 10 to 30 weeks of pregnancy. The report of delivery of live births is extremely rare except for two of them that survived with delivered by cesarean hysterectomy, one at 30 weeks, the other at 37 weeks [5]. However, both the uteri were ruptured.

The cause of intramural pregnancy is unclear. There are many theories for the etiological factors of this infrequent ectopic pregnancy. The most commonly cited etiological factor is previous uterine trauma (such as dilatation and curettage, cesarean section, manual removal of the placenta, and myomectomy), resulting in a sinus tract within the endometrium, in which the zygote is implanted in the myometrium and continues to grow and develop from the defective endometrium. In the present case, the patient had previously undergone hysteroscopic surgery for the IUA, consistent with endometrial damage theory. Other etiological factors are increased trophoblastic activity and defective decidualization which allow the conceptus to implant into the myometrium. It is also believed to be associated with in vitro fertilization and embryo transfer, or even difficult embryo transfer, are implicated in some cases [6].

Pelvic pain and uterine hemorrhage are the hallmark pre-

sensation of ectopic pregnancy. However, early diagnosis of intramural pregnancy is extremely difficult and always made intraoperatively. Only few cases of intramural pregnancy have been correctly diagnosed preoperatively by ultrasound and magnetic resonance imaging [7].

It is simple to misdiagnose intramural pregnancy because it is extremely rare, and ultrasonic appearance is not specificity. In the present report, the patient's pregnancy appeared to be from six to ten weeks, and multiple color ultrasonography showed intrauterine pregnancy. It eventually led to the failure of the abortion. Lack of knowledge regarding the condition and its rarity are possible reasons why the diagnosis may not be made on ultrasound. Therefore sonographers should improve the understandings of the disease, and closely combine the clinical features to determine the location of gestational sac to reduce the misdiagnosis. In the cases where clinical diagnosis is unclear, hysteroscopy may be a mini-invasive surgical treatment to exclude angular pregnancy and other ectopic pregnancy. In the present case, although the diagnosis was delayed, the authors eventually successfully resolved the intramural pregnancy via laparoscopy and hysteroscopy. The mini-invasive surgical treatment can preserve the uterus and fertility, as well as improve the quality of the patients' life. Literature review showed that there are other conservative managements, such as, uterine artery embolization, surgical enucleation, or local or systematic injection of methotrexate [2, 8].

An early diagnosis permits an expectant management to intramural pregnancy, which can be either medical or surgical. The main surgical procedure is local excision by laparoscopy or laparotomy [9]. There are few cases of conservative management with surgical excision, and of these, only six cases exist describe the laparoscopic resection of intramural pregnancy. The present authors report an additional rare case of ectopic pregnancy which underwent previous surgery for intrauterine adhesions treated by laparoscopy. In this report, the uterus was as large as ten weeks of gestation, which is an intramural ectopic pregnancy near the second trimester. The uterus was not ruptured which was different from previous cases.

Conclusion

Although intramural pregnancy is rare, it is important to reinforce the knowledge of this disease. Early diagnosis prevents potential life-threatening bleeding and allows fertility preservation. Both surgical and medical treatments of intramural pregnancy have been described in a small number of cases. The present authors' experience with hysteroscopic and laparoscopic management of intramural pregnancy reveals that this minimally invasive procedure is effective and safe.

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