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Intramural pregnancy: A case report

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ABSTRACT

A 24-year-old woman, who had undergone neither fertility treatment nor uterine surgery other than a cesarean section, presented with an intramural ectopic pregnancy. A laparotomy with uterine wedge resection including the embryonic tissue was performed. The postoperative course was uneventful, with falling βHCG levels. Two months after surgery she presented again with an intrauterine pregnancy.

1. Introduction

The vast majority of ectopic pregnancies (EP) are found in the fallopian tubes [1]. The probability of EP increases after fertility treatment, insertion of intrauterine devices, pelvic inflammatory disease as well as after uterine surgery [2]. Very rarely, intramural pregnancies are localized in the uterus without communication with the endometrial cavity. Here we present the case of a young woman with an intramural pregnancy and a previous history of cesarean section.

2. Case

The 24-year-old patient's medical history included one miscarriage during early pregnancy without any need for surgical intervention, bronchial asthma, an allergy to animal hair, foot surgery, and mild obesity. She used a beclometasone dipropionate/ formoterol fumarate inhaler on demand to control her asthma. The patient was in the 9th week of pregnancy (8 + 4 weeks) and had been delivered by cesarean section 6 months previously due to obstructed labor. All pregnancies were conceived without fertility treatment and she had never used an intrauterine contraceptive device and had no history of pelvic inflammatory disease. She had used condoms for contraception.

2.1. Clinical Findings

One month before presentation, the patient had a positive home pregnancy test. Two weeks later she visited her primary care gynecologist; although no pregnancy could be visualized by ultrasound, the pregnancy test remained positive. Further ultrasound assessment after two weeks led to the diagnosis of ectopic pregnancy with an intramural pregnancy being suspected. She was immediately admitted to hospital. During the pregnancy, the patient had not experienced pain or vaginal bleeding. Clinical examination was unremarkable.

2.2. Diagnostic Assessment

Ultrasound examination (Figs. 1, 2) showed a moderately developed endometrium, inconspicuous adnexa on both sides, and no signs of free intraabdominal fluid. On the left side of the uterus, a 43x35mm mass containing a gestational sac with a viable embryo (crown-rump length 21 mm) was seen. The mass did not appear to communicate with either the fallopian tube or uterine cavity. Serum testing revealed a βHCG of 53,000 miU/ml, which was consistent with the calculated gestational age; all other laboratory parameters were unremarkable. An intramural ectopic pregnancy (iEP) was therefore suspected and treatment options discussed with the patient.

2.3. Therapeutic Interventions

There are several ways of treating an intramural pregnancy. In a review in 2013, Kirk et al. summarized the following options: surgical

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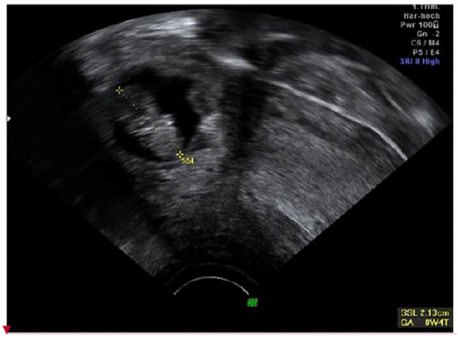

Fig. 1. Transvaginal scan showing the iEP without communication with the uterine cavity.

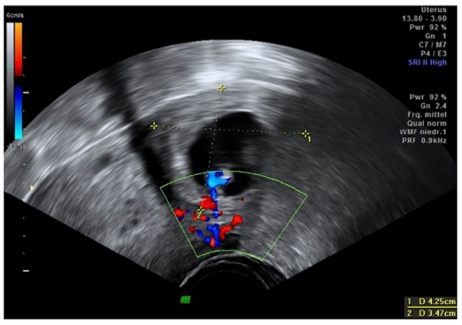

Fig. 2. Transvaginal scan showing the size of the iEP and color Doppler.

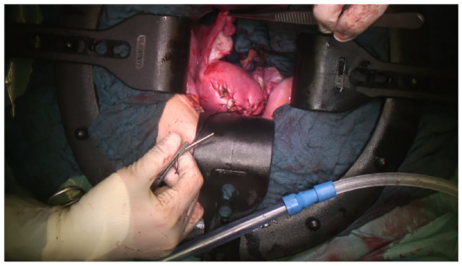

Fig. 4. Intraoperative finding: after wedge resection again unremarkable ovary and fallopian tube.

Fig. 5. iEP as a whole structure.

therapy by laparoscopy or laparotomy, if necessary, after an injection with vasopressin or methotrexate; methotrexate administration intramuscularly (50 mg/m^2) or as an infusion; methotrexate injection controlled by ultrasound; or a wait-and-see approach. Due to the size and location of the structure, the alternatives of surgery or administration of methotrexate were discussed with the patient. She explicitly requested primary surgery. Due to her cesarean section 6 months previously, the extent of the findings, and the patient's obesity, the operation was planned as primarily laparotomy, possibly with additional hysteroscopy and curettage. The patient was informed in advance of the possibility of partial uterine and fallopian tube resection as well as the possibility of a supracervical hysterectomy as the last resort in the event that bleeding could not be stopped using any other method. She agreed to the procedure and gave her written consent. During the surgery, the sonographic findings of a pregnancy were confirmed intramurally near the left tubal os (Figs. 3, 4). After dissection (Figs. 5, 6) and preservation of the tube, the affected area was removed using the LigaSure System for uterine wedge resection. Subsequently, the myometrial edges were approximated, a drain inserted into the rectouterine pouch, and the wound was closed.

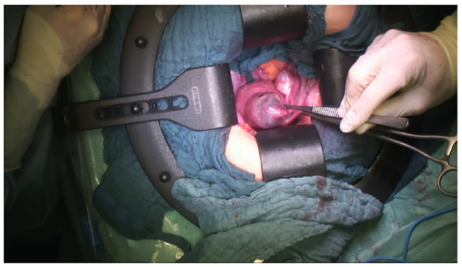

Fig. 3. Intraoperative finding: iEP and unremarkable ovary and fallopian tube.

2.4. Follow-up and Outcome

The patient was monitored postoperatively for one night in the intermediate care unit. She was transferred to the normal care unit on the first day after surgery. The drain was removed on the second postoperative day. The βHCG levels dropped to 5800 mIE/ml, and the patient was discharged on the third postoperative day. She was advised to have βHCG levels checked by her gynecologist until they were normal, to wait at least one year before becoming pregnant again, and then, in the event of a further pregnancy, to deliver the baby by elective cesarean section. Histopathological examination showed partial resection of the uterus (left) with an intact pregnancy at the isthmus. The

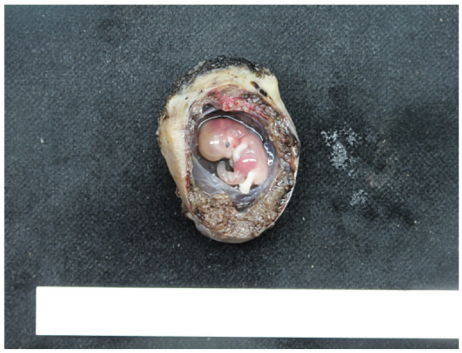

Fig. 6. open iEP with embryonic structure.

placental tissue was unremarkable and age-appropriate. No evidence of malignancy was found in the material analyzed.

The postoperative course was uneventful. Two and a half months after surgery, the patient presented at the clinic again with an intrauterine pregnancy. We discussed the two possible approaches with the patient: termination or continuation of the pregnancy with close monitoring and premature delivery after 34 weeks of pregnancy at the latest. At the time of writing this case report, the patient has not decided whether to continue with the pregnancy or not.

3. Discussion

This case offers a good example for diagnosing and treating an intramural pregnancy. Because the patient was always asymptomatic, there was enough time for diagnosis and we were able to evaluate therapies in accordance with her wishes and needs.

The rarest location of an ectopic pregnancy is inside the myometrium, at a rate of less than 1% [3], and even today the etiology and pathogenesis of intramural pregnancies are not known. Possible causes include adenomyosis, IVF, uterine curettage, cesarean section, myomectomy, or pelvic infection [2]. In this case, the patient had undergone a cesarean section 7 months previously; however, she did not have any other risk factors in her medical history. Symptoms of an intramural pregnancy can be abdominal pain, vaginal bleeding, or amenorrhea, and also nausea and vomiting. In the case of a rupture, hypovolemic shock can develop [4]; therefore, early diagnosis is important. The diagnostic algorithm always includes ultrasound, measurement of βHCG levels, and a clinical examination; however, early diagnosis is sometimes difficult [5]. Indeed, ultrasound may not differentiate between an intramural or a normally located pregnancy, as documented in a patient in whom the intramural pregnancy was only detected by surgery in the 26th week of pregnancy, after unremarkable ultrasound findings at the 7th and 13th week [6]. Sonographic characteristics of intramural pregnancies include: completely surrounded by the myometrium, no contact with either the endometrial cavity or the fallopian tube, and, usually, identification of an embryonic structure [4]. Color Doppler may help to distinguish the structure from fibroids. Gestational trophoblast disease represents another important differential diagnosis; here, the border between the structure and the endometrium is often not distinct, whereas an intramural pregnancy is completely and clearly surrounded by myometrium. It is not unusual for an intramural pregnancy to be diagnosed at surgery or even later, histologically. The therapeutic options for intramural pregnancies include surgery (excision or hysterectomy), medication (methotrexate ± vasopressin), or taking a wait-and-see approach, depending on the clinical findings.

Intramural pregnancies represent an important differential diagnosis in patients with a positive pregnancy test without evidence of an intrauterine pregnancy. Due to the possible complications, rapid diagnosis and management are crucial.

Contributors

Juliane Nees drafted the manuscript and contributed substantially to revision of the manuscript.

Gesine Faigle-Krehl edited the manuscript and contributed substantially to revision of the manuscript.

Janina Brucker contributed substantially to revision of the manuscript.

Dagmar Leucht contributed substantially to revision of the manuscript.

Lisa Katharina Platzer edited the manuscript and contributed substantially to revision of the manuscript.

Christa Flechtenmacher edited the manuscript and contributed substantially to revision of the manuscript.

Christoph Sohn contributed substantially to revision of the manuscript.

Markus Wallwiener drafted the manuscript and contributed substantially to revision of the manuscript.

Conflict of Interest

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

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Patient Consent

Obtained.

Provenance and Peer Review

This case report was peer reviewed.

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