Case report

Pregnancy of unknown location which was discovered to be an intra mural pregnancy at the previous uterine perforation site

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Abstract

A 35-year-old obese (BMI 35) woman presented to the antenatal clinic with a positive pregnancy test at a period of amenorrhea of six weeks. She was initially managed according to the Pregnancy of Unknown Location (PUL) care pathway. However, she presented with throbbing abdominal pain and required an emergency laparoscopy which revealed an intramural pregnancy at the site of a previous uterine perforation. The ectopic site was successfully excised laparoscopically. A detailed history, good knowledge of various rare presentations and the skills to make tailor-made decisions when dealing with PUL are crucial to avoid complications. Caregivers evaluating patients presenting with PUL should be well equipped to handle challenging cases such as these.

Key words: intramural pregnancy, pregnancy of unknown location, ectopic pregnancy, early pregnancy complication, minimal access surgery

Introduction

Pregnancy of Unknown Location (PUL) is a pregnancy with a positive pregnancy test but an undetectable intrauterine or extrauterine pregnancy on transvaginal ultra-sonography[1]. Availability of high-resolution scans, use of serum beta HCG and diagnostic laparoscopic facilities have improved the management of PUL. However, the location of the pregnancy in a minority of cases of PUL is never confirmed, due to the fact that both miscarriages and ectopic pregnancies may resolve spontaneously without intervention. Delay in locating a pregnancy may lead to life-threatening complications such as a ruptured or a leaking ectopic pregnancy leading to shock.

Protocols and diagnostic algorithms are in place to evaluate, diagnose and resolve women who present as PUL. However, there can be rare and unusual presentations that require careful evaluation. Intramural pregnancy is a rare presentation that can be the cause of diagnostic and management dilemmas[2]. Failing to diagnose and initiate optimal treatment can lead to life-threatening uterine rupture[3]. We describe the successful management of a rare presentation of PUL, that later

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turned out to be an intramural pregnancy in a previous perforation site of the uterus.

Case presentation

A 35-year-old obese (BMI 35) woman presented to the antenatal clinic with a positive pregnancy test. She was on medication for Systemic Lupus Erythematosus (SLE). She had three previous first trimester miscarriages and an ectopic pregnancy that was conservatively managed. Ten months before her current pregnancy, she underwent a laparoscopy to investigate secondary subfertility and pelvic pain during which she had a uterine perforation. Clinical records revealed a small fundal perforation that was not initially sutured, and it being a minimal bleeder was ablated using bipolar diathermy.

At the time of presentation, she was at a period of amenorrhea of 6 weeks. Transvaginal ultrasound scan showed no intrauterine pregnancy or adnexal masses suggestive of an ectopic pregnancy. Her initial serum beta HCG was 176 miu/ ml which dropped to 169 miu/ml after 48 hours. Since she did not have any symptoms or evidence of an ectopic site, she was advised follow up with repeat beta HCG levels and ultrasound scan.

However, she presented to the emergency unit with throbbing lower abdominal pain, and it was decided to perform a diagnostic laparoscopy. Laparoscopy revealed a sub serosal blackish bulge on the uterine fundus. This was not ruptured and measured 2 cm × 2 cm. Rest of the pelvis was normal and no evidence of any other sites of pregnancy were noted. This site was consistent with the previous site of perforation (Figure 1).

We injected diluted Vasopressin (20 units of Vasopressin diluted in 200 ml of normal saline (1 unit/ml)) to the myometrium at the site of the possible ectopic implantation to make it avascular (Figure 2). Then, using monopolar diathermy, a small incision at the top of the ectopic site was placed and the contents were sucked out from the sac. The base of the site was ablated to arrest bleeding and a saline irrigation was performed to confirm hemostasis (Figure 3, 4).

A follow up Serum HCG next day was 25 miu/ml. Patient was asymptomatic. Histology confirmed trophoblastic tissues from the site.

Key stages of the surgical procedure have been described in the figures below.

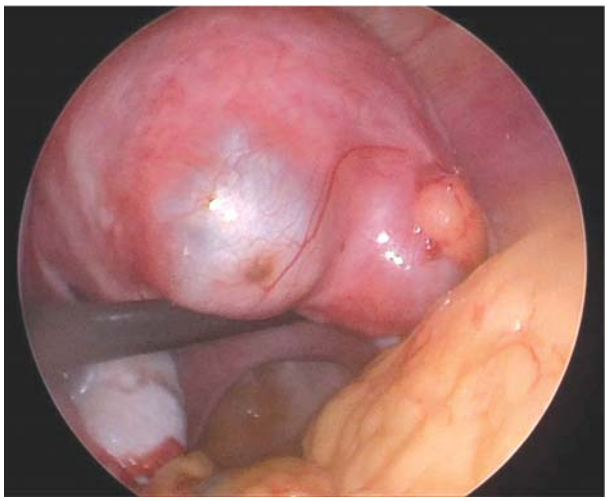

Figure 1. **The bulge suggestive of an ectopic pregnancy.**

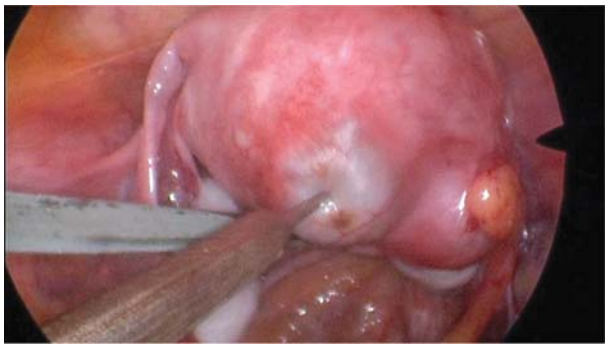

Figure 2. **Injecting diluted Vasopressin to the myometrial area at the site of possible ectopic to make the site avascular.**

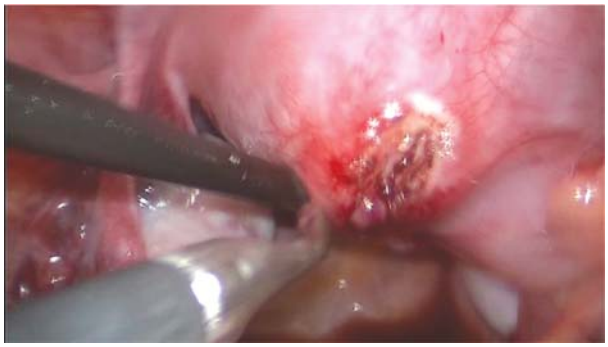

Figure 3. **Ectopic tissues were removed using grasping forceps.**

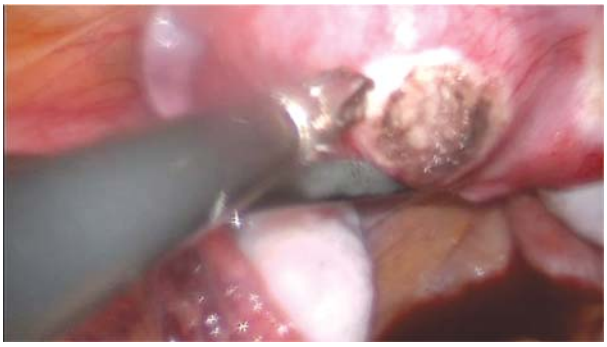

Figure 4. **Base of the site that was ablated to arrest bleeding.**

Discussion

Intramural pregnancy represents an abnormal implantation where the gestational sac is located within the myometrium without any connection to the endometrial cavity, fallopian tubes, or round ligament[2]. This condition is difficult to detect in early stages even though early diagnosis and management are crucial to prevent life threatening consequences. Intramural pregnancy is extremely rare, accounting for less than 1% of ectopic pregnancies[3]. The patient may present with lower abdominal pain, vaginal bleeding and a positive pregnancy test or may remain asymptomatic[2].

Reported risk factors are uterine surgery including dilation and curettage, caesarean section, or myomectomy. Invasion of the myometrium via microscopic tracts created by surgical instruments or following difficult in vitro fertilization (IVF) transfers resulting in migration and implantation in an unusual site have been proposed as a possible mechanism[4,5].

In our case, previous trauma in the uterus during the laparoscopic procedure could have formed a track in the uterus and implantation would have occurred along that path. Intramural pregnancy following uterine perforation has never been reported in the literature before.

This case report emphasizes the importance of a high index of suspicion been needed when evaluating patients with PUL especially if they have risk factors and should include intramural pregnancy in the differential diagnosis. Surgeons who perform procedures and manage uterine perforation should debrief and inform the patients regarding future implications and clearly document it in the patients' records. In the pre ultrasound era, intramural pregnancies were diagnosed only when they presented with uterine rupture. However, the advent of high frequency vaginal ultrasound allows a diagnosis when the gestational sac in the uterine wall is surrounded by myometrium, without visualization of a gestational sac in the endometrium or fallopian tubes.

These ectopic trophoblastic tissues are found beyond endometrial-myocetrial junction and can be covered endometrium partly or completely[5]. Demonstration of peri trophoblastic flow using a colour Doppler, three dimensional US and MRI can aid the diagnosis[4]. However, in our patient, body habitus and early presentation were limiting factors for early ultrasonic detection.

Management of intramural pregnancy can be expectant, medical, or surgical depending on presenting complaint, time of presentation, viability, size, location of the pregnancy and wishes for future fertility[6]. In our case, the diagnosis of intramural pregnancy and a decision to excise it laparoscopically were made at the time of laparoscopy. Although one similar case was managed expectantly, we decided to proceed with a surgical approach as the patient had presented with pain.

Although systemic or local administration of methotrexate is a successful intervention for management of non-tubal ectopic pregnancies in patients who wish to preserve fertility[7],we considered surgical management to be a reasonable approach due to her symptoms.

Surgeons performing laparoscopic surgery in intramural pregnancy should be aware that this could theoretically increase the risk of uterine rupture in the future[8] in addition to other complications of laparoscopy and patients should be adequately informed prior to the procedure.

This case report emphasizes the importance of history, good knowledge of various rare presentations and the skills to make tailor-made decisions when dealing with unusual presentations of intramural pregnancies. Caregivers evaluating the patients presenting with PUL should be well equipped to handle challenging such cases to avoid serious consequences.

Conflict of interests

The authors declare no conflict of interest.

References

1. Sagili H, Mohamed K. Pregnancy of unknown location: An evidence-based approach to management. The Obstetrician and Gynaecologist, 2008; 10(4): 224-30. https://doi.org/10.1576/toag.10.4.224.27438
2. Zang L, Hu Y. Intramural Pregnancy Misdiagnosed as Gestational Trophoblastic Neoplasia: Case Report and Review of the Literature. Obstetrics and Gynaecology Cases – Reviews 2021; 8(5). https://doi.org/10.23937/2377-9004/1410213.
3. Zhang Q, Xing X, Liu S, Xie X, Liu X, Qian F, Liu Y. Intramural ectopic pregnancy following pelvic adhesion: Case report and literature review. Archives of Gynecology and Obstetrics 2019; 300(6): 1507-20. https://doi.org/10.1007/s00404-019-05379-3
4. Liu Y, Wu Y. Intramyometrial pregnancy after cryopreserved embryo transfer: A case report. BMC Pregnancy and Childbirth 2020; 20(1): 90. https://doi.org/10.1186/s12884-020-2784-7.
5. Khalifa Y, Redgment CJ, Yazdani N, Taranissi M, Craft IL. Intramural pregnancy following difficult embryo transfer. Hum Reprod. 1994; 9: 2427-8.
6. Memtsa M, Jamil A, Sebire N, Jauniaux E, Jurkovic D. Diagnosis and management of intramural ectopic pregnancy. Ultrasound Obstet Gynecol. 2013; 42: 359-62.
7. Bechev B, Konovalova M. A very rare case of ectopic intramural pregnancy after IVF-ET. Clin Exp Obstet Gynecol 2017; 44: 802-3.
8. Chen X, Gao L, Yu H, Liu M, Kong S, Li S. . Intramural Ectopic Pregnancy: Clinical Characteristics, Risk Factors for Uterine Rupture and Hysterectomy. Frontiers in Medicine 2021; 8: 769627.https://doi.org/10.3389/fmed.2021.769627.