CASE REPORT

A painless pelvic wall ectopic pregnancy

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SUMMARY

A 38-year-old woman presented with light vaginal bleeding at approximately 8 weeks gestation. Although she initially presented to accident and emergency with vaginal bleeding and menstrual-like pain, this rapidly settled and both symptoms resolved over the following days. An initial ultrasound scan revealed an empty uterus, with no adnexal masses or free fluid. Miscarriage was presumed. However, following suboptimal β -human chorionic gonadotropin decline, further ultrasonography noted a mass next to the right ovary. The patient underwent laparoscopy, revealing an ectopic pregnancy closely adherent to the peritoneum of the right pelvic wall. The patient was treated with systemic methotrexate and responded well. This rare form of ectopic pregnancy has a high maternal morbidity and mortality, and its optimal management is not definitively decided, due to the scarcity of the condition and subsequent lack of high-powered data. We present a case of successful medical management.

BACKGROUND

Ectopic pregnancy refers to implantation of a pregnancy outside of the endometrial cavity; most commonly into the fallopian tubes.¹ However, more unusual forms of ectopic pregnancy exist including ovarian and abdominal pregnancy.² Abdominal pregnancy refers to implantation outside of the uterus, fallopian tubes, ovaries, or their ligamentous attachments.³ It is a rare occurrence, although reported incidence varies widely particularly between developed and developing settings. An influential review of 5221 abdominal pregnancies in the USA placed its incidence at 10.9/100 000 live births,⁴ while smaller studies in Africa place it as high as 36.2/100 000⁵ or even 152.9/100 000.⁶ Alternately, its incidence has been reported from as low as 0.9% of ectopic pregnancies in the US⁴ up to 4.3% in Nigeria.⁶ A potential explanation for this discrepancy is the increased prevalence of risk factors such as lower socioeconomic status and pelvic inflammatory disease (PID) in developing settings.^{7 8}

Owing to the rarity of abdominal ectopic pregnancies, the majority of the relevant literature relies on case reports and relatively small series. As a result, there is no definitive consensus regarding management.⁹ With the increasing adoption of early pregnancy assessment units (EPAUs), and advances in the technique and availability of ultrasound, there is an increase in the early recognition of these pregnancies.¹⁰ Consequently, trends in management are shifting from an open surgical approach (which was often the time of incidental diagnosis and which in several cases has resulted in

live birth^{11 12}) to laparoscopic or medical management or a combination thereof.^{10 13}

There are no strong predictors for the success of medical management,⁹ though high β -human chorionic gonadotropin (HCG; >5000 mIU/ml), moderate to large amounts of peritoneal free fluid and fetal heartbeat have been suggested as poor prognostic factors.^{1 13} Medical management is most often by systemic methotrexate. There have also been cases of successful management with intraleisional methotrexate, with some authors advocating intralesional administration after failed systemic therapy prior to resorting to surgical management.¹⁴ Interventional radiology may also have a role through embolisation of feeding vessels, either as a primary treatment modality, in preoperative optimisation for surgical removal, or postoperatively, as haemorrhage control.^{14 15} In addition, intracardiac potassium chloride administered to the embryo as an agent for feticide has also been used to augment methotrexate therapy.¹³

We present a patient with a pelvic ectopic pregnancy. Her presentation to the EPAU will be discussed, as well as the investigations she underwent and her subsequent treatment and outcome.

CASE PRESENTATION

A 38-year-old nulliparous Caucasian woman presented to the EPAU following a private ultrasound at 7 weeks+6 days gestation by her last menstrual period. Prior to this attendance, she had been assessed in the emergency department, for menstrual-like pain and vaginal bleeding, and had been reassured and discharged with instructions to return if symptoms did not settle. The pain resolved quickly, though a reduced volume of bleeding continued and the patient opted for the private scan to alleviate her anxiety. The scan revealed an empty uterus, with no evidence of adnexal masses or free fluid in the pelvis.

At presentation to the EPAU, the patient reported a 2-day history of light vaginal bleeding. She felt otherwise well with no associated abdominal or pelvic pain, and on examination her abdomen was soft and non-tender. Speculum examination revealed a small amount of bleeding only, with no other abnormality identified.

The patient had undergone two terminations in the past and had been treated for PID following one of these. She had no other risk factors for ectopic pregnancy. There was no relevant medical history, she was not taking any regular medication, did not smoke regularly and had no family history of note.

Initial blood tests performed revealed a serum β -HCG of 2208. The patient was asymptomatic

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apart from some mild bleeding, which was settling. She remained haemodynamically stable. She was therefore discharged with a plan to return 48 h later to have a repeat serum β -HCG. This showed only slight decline to 2095. The patient was asked to re-attend the unit for a repeat ultrasound scan and assessment. Again, she was well and the bleeding had settled at this point. She continued to report no abdominal pain. The repeat ultrasound scan was normal, with no adnexal masses seen, no free fluid and an empty uterus. A corpus luteum was noted on the right ovary.

The options for management, along with their respective risks and benefits, were discussed with the patient. These included expectant, medical and surgical management plans. As she felt well with no pain and only a small amount of bleeding, the patient opted for expectant management on the proviso that if her serum β -HCG continued to exhibit suboptimal decline then she would need further invasive investigation or active treatment.

Her third β -HCG rose to 2772 and she was therefore advised to re-attend for a further ultrasound scan by a specialist consultant. This third ultrasound revealed a mass near the right ovary, thought to potentially represent a gestational sac. The uterus remained empty and there was no free fluid.

The patient was booked onto the emergency list for a laparoscopy±removal of ectopic±salpingo-oophorectomy.

INVESTIGATIONS

Serial serum β -HCGs and transvaginal ultrasound scans were performed, as detailed above. Although ultrasonography remains the preferred investigation in the diagnosis of abdominal pregnancy, its sensitivity remains low even when combined with clinical acumen, demonstrating the importance of close follow-up and a high index of suspicion.¹⁵

Another potential investigation sometimes advocated in pregnancy of unknown location (such as the initial phase of this case), is dilation and curettage performed in an attempt to differentiate between a non-viable or miscarried intrauterine pregnancy and an ectopic pregnancy.¹⁶ However, in this instance, the patient had been counselled about her management options and initially elected for expectant management, a recognised option in minimally symptomatic, stable women.¹⁷ During the subsequent period of close observation, a third ultrasound scan noted the mass by the right ovary. As such, there was adequate clinical suspicion to justify diagnostic±therapeutic laparoscopy rather than further investigation.

DIFFERENTIAL DIAGNOSIS

It was evident from the trend over several serial β -HCG's that this was a case of ectopic pregnancy; the uncertainty in diagnosis related to its location, given the lack of definitive diagnosis at ultrasound. Contemporaneously, it was thought to be either a right tubal or ovarian ectopic pregnancy.

TREATMENT

Diagnostic laparoscopy was performed with a view to treat an ectopic pregnancy, if present. The right fallopian tube was noticed to be slightly thickened in the ampullary region but otherwise appeared healthy. This was thought to be atypical of a tubal ectopic. A corpus luteum was visualised on the right ovary.

An ectopic pregnancy was noted, closely adherent to the pelvic wall, inferior to the right ovary (figures 1–3). There were also endometrial deposits noted in the Pouch of Douglas and on the left pelvic wall. A second consultant was called to theatre to confirm the findings and it was agreed that systemic

Figure 1 Relation of the pregnancy to the right ovary at laparoscopy.

methotrexate would be the best management option, due to the proximity of the ureter and the high likelihood of bleeding should surgical removal be attempted.

OUTCOME AND FOLLOW-UP

The patient recovered well from surgery and the findings were explained. She was consented for methotrexate therapy and this was started the following day. Her β -HCG levels gradually declined. On day 4, her β -HCG was found to be 1815 and this fell to 1245 on day 7. Another blood test was taken 4 days later revealing it had declined further to 483. Two weeks later, her β -HCG was 34. Her liver function tests remained normal throughout.

DISCUSSION

This case demonstrates the importance of serial β -HCG follow-up in women with pregnancy of unknown location. This patient was asymptomatic and felt very well throughout the majority of her investigation, with her initial pain and vaginal bleeding settling quickly without intervention. She was keen for conservative management and, had she not been closely reviewed, she would have been at risk of significant morbidity. Likewise, given the rapid resolution of her initial symptoms, she may well have presented much later had she not arranged a private ultrasound scan to alleviate her anxiety, and this would undoubtedly have increased her risk of both morbidity and mortality.

Abdominal pregnancies have been described in a number of locations. A recent review of 225 abdominal pregnancies

Figure 2 'Close-up' perspectives of the pregnancy at laparoscopy, seen to be closely adherent to the pelvic sidewall.

Figure 3 'Close-up' perspectives of the pregnancy at laparoscopy, seen to be closely adherent to the pelvic sidewall.

reported the most common sites as (from most frequent to least): the pouches around the uterus (most commonly the Pouch of Douglas), the adnexae, omentum, bowel/appendix, liver, spleen, retroperitoneum and the abdominal wall.⁹ In addition, implantation sites at the diaphragm and renal fascia have been described.^{15 18 19}

Abdominal pregnancies are described as being primary or secondary dependent on satisfaction of Studdiford's criteria.^{9 20} A primary abdominal pregnancy refers to a pregnancy that is implanted directly into the abdominal cavity, that is, the tubes and ovaries are normal. A secondary abdominal pregnancy is more common and is likely to occur following a tubal pregnancy, which is then expelled into the abdominal cavity.^{2 3} This was likely the case for the patient described here, as her fallopian tube on the side of the pregnancy was noted to be swollen at laparoscopy. The classification, however, is of predominantly academic interest and has been suggested to have little clinical value.¹⁵

Risk factors for abdominal pregnancy are similar to those of any ectopic pregnancy and include low-socioeconomic status, PID, endometriosis, previous pelvic surgery, intrauterine contraceptive devices and history of ectopic pregnancy.^{7 8 15 21} The patient described here was not from a low socioeconomic group, although she did have a history of PID, which may have contributed to her risk of developing this rare form of ectopic pregnancy. This patient was not previously known to have endometriosis, though deposits were noted at the time of laparoscopy.

Despite its rarity, abdominal pregnancy is an important consideration due to its association with high maternal mortality: 7.7 times that of other ectopic pregnancies and 89.8 times that of uterine pregnancy.⁴ The most appropriate management of abdominal or pelvic pregnancies remains a matter of contention. Surgical management, even early in pregnancy, can lead to extensive bleeding due to the vascularity of the attachment.^{12 22} There is little evidence as to whether systemic or intralesional administration of methotrexate is more effective, and no convincing evidence for the use of markers to predict the success of medical management.⁹

As a result, each case should be considered individually. In this case, the woman was asymptomatic, her β -HCG was <3000, and surgical removal of the pregnancy was considered high risk due to its proximity to the right ureter. As such, it was decided to commence medical therapy with systemic methotrexate. Even in the event of failure of this treatment modality, methotrexate still has a role in facilitating safe surgical removal

by ameliorating the risk of haemorrhage with placental removal.³ A trend towards laparoscopically diagnosing the pregnancy before attempting medical management has been noted, with surgery reserved for advanced pregnancies and those non-responsive to medical therapy.⁹ Interventional radiology also has a role, both as a potential treatment modality and in facilitating safe surgical removal.¹⁴

The lack of definitive consensus on treatment is due to the scarcity of the condition, which makes a high-powered randomised trial practically impossible. As a result, practitioners managing these pregnancies should report their results in order to add to the collective literature and enable the optimal management to be elucidated.

Patient's perspective

- "My husband and I had been trying to conceive for 4 months, when I became pregnant. We were overjoyed and relieved that it had not taken too long. Everything seemed well in the first weeks with the pregnancy, with no sickness or other issues. Then, one Tuesday afternoon, I noticed bleeding. I began to worry as the blood was bright red and did not subside, but at this point I did not have any pain. We decided to go to A&E as I knew something was not right. When I was seen by the doctor she told me that it was quite normal to have some bleeding in early pregnancy; I tried to explain that I did not think it was normal, but as I was not in pain she dismissed my fears and I was sent home with no further investigations. I continued to bleed during the night and the following day I stayed in bed as I began to have period-like pains and was by this point very distressed. I called my GP and they said they would contact the EPAU and have them make an appointment for me. I did not hear again from anyone later in the day and by this point I was desperate to know if I had lost my baby, which I was almost sure I had. I booked an appointment that evening at a private ultrasound scan clinic. The scan confirmed that my uterus was empty. My husband and I were both very upset as we believed I had miscarried and we had lost our baby.
- The following morning, I had a call from the EPAU and was booked in to see them that afternoon. After an internal examination and a discussion about my medical history, and after blood tests, I was asked to come back in 2 days time for repeat hormone tests. I was then asked to return for a second internal ultrasound. The scan found no sign of an ectopic pregnancy and I was feeling well, the bleeding had stopped and I had no pain. The risks of a possible pregnancy of unknown location were discussed with me but I asked just to be monitored for another week as I was not convinced that I had an ectopic pregnancy as I felt physically fine and nothing had shown on the scans.
- Hormone tests during that week continued to rise and so, on the following Wednesday, I was asked to come back in to see the consultant on EPAU. She performed an ultrasound and after a lot of prodding found a mass near my ovary. To my surprise, I was asked to remain in hospital that afternoon for surgery. Up until that point I had felt physically well for a week after the initial pain and bleeding, and thought I had just had a miscarriage.

- I was told by the doctors after surgery that they had found an ectopic pregnancy but that it was in my abdomen and not in the normal tubal location, which was very rare. They could not remove it as it was close to the tube to my bladder and my ovary, and I was advised to take methotrexate. I was concerned as I knew it was a strong drug—I don't normally need to take any medication, not even paracetamol. However, I knew there was no other option. It also meant I would have to wait another 3 months to start trying to conceive again, which at the age of 39 is stressful, as every month is precious.
- Once we were home, the full measure of everything that had occurred hit me and I was inconsolable. While in hospital I had been on auto-pilot. There had not been time to fully digest what had happened and I felt I had to lose my baby twice, once after the first scan and bleeding (having believed I'd had a miscarriage) and then again after my surgery and injection in hospital.
- After being discharged, I had to return to the hospital numerous times to have my hormone levels checked. It seemed to take forever for them to return to normal, which was very frustrating as I knew I would not start my normal menstrual cycle again until that happened. Once they did I was relieved and felt I could now concentrate on recovering emotionally, and looking ahead to when we could begin trying for a baby again.
- The doctors and nurses who cared for me were very good and I cannot fault their level of care. I think the only disappointment in the whole episode was the initial visit to A&E, where I felt dismissed even though I knew something was wrong. I thought a lot afterwards about what would have happened if I had not called the GP and been referred to the EPAU, and continued to believe that I had miscarried; the situation could have been much more serious."

Learning points

- Systemic methotrexate therapy is a reasonable and safe method of management for early abdominal/pelvic pregnancies.
- Rare forms of ectopic pregnancy do exist and should be considered due to the high morbidity and mortality rates with which they are associated.
- Stringent follow-up should be in place for all women who are found to have a pregnancy in an unknown location.
- Clinicians with experience in managing these cases should report their experiences in order to help elucidate the optimum treatment for these rare pregnancies.

Contributors ZB was involved in preparation of the manuscript, liaising with and gaining the consent of the patient, and reviewing and obtaining the electronic intraoperative images. AGL contributed to the writing of the initial article as well, and editing and refining subsequent drafts. SS contributed by reviewing several drafts of the submission.

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REFERENCES

- Martínez-Varea A, Hidalgo-Mora JJ, Payá V, *et al.* Retroperitoneal ectopic pregnancy after intrauterine insemination. *Fertil Steril* 2011;95:2433.e1–3.
- Fylstra DL. Ectopic pregnancy not within the (distal) fallopian tube: etiology, diagnosis, and treatment. *Am J Obstet Gynecol* 2012;206:289–99.
- Gupta P, Sehgal A, Huria A, *et al.* Secondary abdominal pregnancy and its associated diagnostic and operative dilemma: three case reports. *J Med Case Rep* 2009;3:7382.
- Atrash HK, Friede A, Hogue CJ. Abdominal pregnancy in the United States: frequency and maternal mortality. *Obstet Gynecol* 1987;69:333–7.
- Sunday-Adeoye I, Twomey D, Ekwuatu EV, *et al.* A 30-year review of advanced abdominal pregnancy at the Mater Misericordiae Hospital, Afikpo, southeastern Nigeria (1976–2006). *Arch Gynecol Obstet* 2011;283:19–24.
- Ayinde OA, Aimakhu CO, Adeyanju OA, *et al.* Abdominal pregnancy at the University College Hospital, Ibadan: a ten-year review. *Afr J Reprod Health* 2005;9:123–7.
- Aliyu LD, Ashimi AO. A multicentre study of advanced abdominal pregnancy: a review of six cases in low resource settings. *Eur J Obstet Gynecol Reprod Biol* 2013;170:33–8.
- Yildizhan R, Kulusari A, Adali F, *et al.* Primary abdominal ectopic pregnancy: a case report. *Cases J* 2009;2:8485–90.
- Poole A, Haas D, Magann EF. Early abdominal ectopic pregnancies: a systematic review of the literature. *Gynecol Obstet Invest* 2012;74:249–60.
- Chetty M, Elson J. Treating non-tubal ectopic pregnancy. *Best Pract Res Clin Obstet Gynaecol* 2009;23:529–38.
- Muehlparzer L, Arzt W, Ebner T, *et al.* Secondary abdominal pregnancy with live birth. *Acta Obstet Gynecol Scand* 2011;90:288.
- Kwok A, Chia KK, Ford R, *et al.* Laparoscopic management of a case of abdominal ectopic pregnancy. *Aust N Z J Obstet Gynaecol* 2002;42:300–2.
- Moores KL, Keriakos RH, Anumba DO, *et al.* Management challenges of a live 12-week sub-hepatic intra-abdominal pregnancy. *BJOG* 2010;117:365–8.
- Anderson PM, Opfer EK, Busch JM, *et al.* An early abdominal wall ectopic pregnancy successfully treated with ultrasound guided intralesional methotrexate: a case report. *Obstet Gynecol Int* 2009;2009:247452.
- Agarwal N, Odejinmi F. Early abdominal ectopic pregnancy: challenges, update and review of current management. *Obstet Gynaecol* 2014;16:193–8.
- Awonuga A. Ectopic pregnancy and the dilemma of pregnancy of unknown location. *Med J Obstet Gynecol* 2013;1:1007.
- Rathod S, Samal SK, Ghose S. Pregnancy of unknown location (PUL): a case report and review of the literature. *Int J Reprod Contracept Obstet Gynecol* 2014;3:808–11.
- Fishman DA, Padilla LA, Joob A, *et al.* Ectopic pregnancy causing hemothorax managed by thoracoscopy and actinomycin D. *Obstet Gynecol* 1998;91(5 Pt 2):837–8.
- Chishima F, Kato-Suzuki E, Ichikawa G, *et al.* Rare case of primary peritoneal pregnancy infiltrated into the Gerota's fascia of the right kidney. *J Obstet Gynaecol Res* 2013;39:1073–6.
- Studdiford WE. Primary peritoneal pregnancy. *Am J Obstet Gynecol* 1942;44:94.
- Pisarska MD, Carson SA. Incidence and risk factors for ectopic pregnancy. *Clin Obstet Gynecol* 1999;42:2–8.
- Gorry A, Morelli ML, Olowu O, *et al.* Laparoscopic management of abdominal ectopic pregnancy using FLOSEAL Hemostatic Matrix. *Int J Gynaecol Obstet* 2012;117:83–4.

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