

Submyomatous Cornual Pregnancy Managed surgically after failed medical management

Lamya Al-Kharusi,¹ *Vaidyanathan Gowri,² Rashid Al-Sukaiti,² Wadha Al-Ghafri,¹ Kuntal Rao¹

حَمْلُ قَرْنِي فِي تَلْيَفِ رَحْمِي تَحْتَ مَخَاطِي عِلَاجٍ جِرَاحِيٍّ بَعْدَ فَشَلِّ الْعِلَاجِ الطَّبِيِّ

لمياء الخروصي، فيدياناثان جوري، راشد السكيتي، وضى الغافري، كونتال راو

الملخص: يُعد الحمل القرني من حالات الطوارئ، ويشكل تشخيصه وعلاجه تحدياً كبيراً. كما أن وجود تشوهات تشريحية في الرحم مثل الورم الليفي في أحد قرني الرحم يجعل علاج الحمل القرني أكثر صعوبة. إحدى المريضات الحوامل التي لم يسبق لها الحمل والتي كانت تعاني من ورم ليفي في قرن الرحم الأيمن تم تشخيصها كحالة حمل خارج الرحم، وكان حمل قرني تحت ورم ليفي ضخم. على الرغم من الجرعات المتعددة من الميتوتريكسيت لعلاج الحمل خارج الرحم (الحمل القرني) فإن مستوى بيتا موجة الغدد التناسلية المشيمية البشرية في الدم تضاعف في اليوم الخامس وأثبت التصوير وجود جنين حي. تم إجراء التدخل الجراحي بدايةً بعملية منظار ثم أجريت عملية فتح البطن لاستئصال الحمل والورم العضلي الليفي الكبير على قرن الرحم. بعد العملية بثلاثة أسابيع كان مستوى بيتا موجة الغدد التناسلية المشيمية البشرية سلبياً. لقد كان التدخل الجراحي في شكل استئصال الورم العضلي ضرورياً للحفاظ على الخصوبة في مثل هذه الحالات من الحمل غير العادي خارج الرحم.

مفتاح الكلمات: تشخيص، حمل خارج الرحم، ورم ليفي، ورم عضلي، أمليس، ميتوتريكسيت، جراحة، تقرير حالة، عُمان.

ABSTRACT: Cornual pregnancy constitutes an emergency while its diagnosis and management remain a challenge. Anatomical abnormalities in the uterus, such as fibroids in the cornual region, make the management even more difficult. A nulliparous patient presented with an ectopic pregnancy at the right cornua under a huge fibroid. Despite multiple doses of methotrexate for a cornual ectopic gestation, the serum beta human chorionic gonadotropin (β -hCG) levels doubled on the fifth day and a viable fetus was demonstrated on imaging. Thus surgical intervention in the form of laparoscopy followed by laparotomy, myomectomy of a large cornual fibroid and cornuostomy was performed. The serum beta human chorionic gonadotropin result was negative three weeks later. Surgical intervention in the form of myomectomy and cornuostomy was necessary to preserve fertility in this unusual presentation of cornual ectopic pregnancy.

Keywords: Diagnosis; Ectopic pregnancy; Fibroid; Leiomyoma; Methotrexate; Surgery; Case report; Oman

CORNUAL PREGNANCY ACCOUNTS FOR 2–4% of ectopic pregnancies and can be associated with a mortality rate in the range of 2.0–2.5%.¹ Cornual pregnancy constitutes a medical emergency and its diagnosis and management is challenging. We present a rare case of an ectopic pregnancy located just under a cornual fibroid, at the site of right fallopian tube insertion, which did not respond to intensive medical management thus necessitating surgical intervention.

Case Report

A 25 year-old primigravida presented with a history of mild vaginal bleeding, abdominal pain

and 7 weeks of amenorrhoea. She gave history of bleeding per vagina at home during the last four days. There were no identifiable risk factors for an ectopic pregnancy. On physical assessment, the patient was haemodynamically stable and not in distress. Abdominal palpation revealed a 16-week size mobile non-tender mass in the midline. Bimanual examination confirmed an enlarged uterus corresponding to 16 weeks with a single closed cervix and minimal cervical motion tenderness. Abdominal ultrasound showed an 8 cm right cornual fibroid and transvaginal ultrasound (US) confirmed an empty endometrial cavity (endometrial thickness of 19 mm) despite high beta human chorionic gonadotropin (β -hCG) levels of 6,600 units, normal adnexa and no free fluid

Departments of ¹Obstetrics & Gynaecology and ²Radiology & Molecular Imaging, Sultan Qaboos University Hospital, Muscat, Oman; ³Department of Obstetrics & Gynaecology, College of Medicine & Health Sciences, Sultan Qaboos University, Muscat, Oman.

*Corresponding Author email: gowrie61@hotmail.com

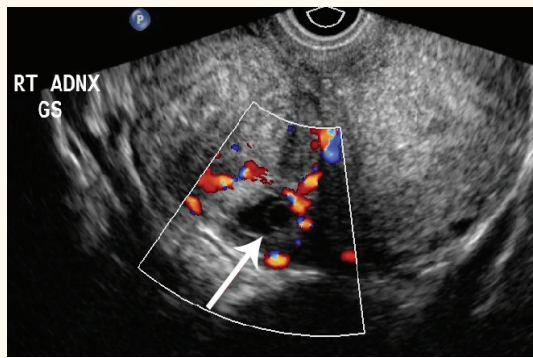


Figure 1: Transvaginal ultrasound scan of right adnexal region demonstrating a gestational sac and yolk sac (arrow).

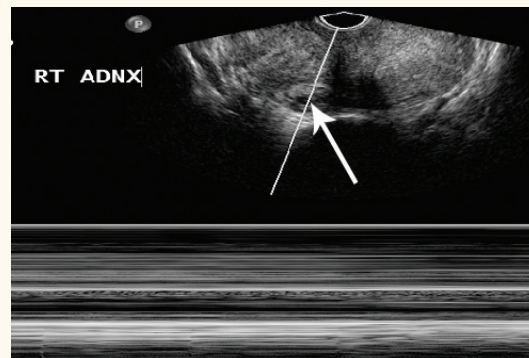


Figure 2: Transvaginal ultrasound scan of right adnexal region demonstrating a positive fetal heart activity in M-mode (arrow).

in the pelvis. A complete blood count revealed haemoglobin of 10.7 g/dL, total white cell count of $4.2 \times 10^9/L$ and platelets of $402 \times 10^9/L$. Liver function tests revealed alanine aminotransferase (ALT) 11 iu/L; aspartate aminotransferase (AST) 18 u/L; bilirubin $4 \mu\text{mol/L}$, and alkaline phosphatase 51 u/L (all within normal limits).

A right cornual ectopic was suspected in view of the location of a sac-like structure and increased Doppler flow [Figure 1]. After counselling, she opted for conservative management with a multidose methotrexate (EBEWE Pharma, Austria) regimen as an inpatient. This was given on days 1, 3 and 5 intramuscularly (50 mg/m^2 body surface area) with rescue doses of folinic acid (EBEWE Pharma, Austria) to reduce bone marrow toxicity on day 2, 4 and 6. On day 3 (after receiving a second dose of methotrexate) she passed some tissue vaginally and it was sent for histopathological examination. On day 5, after the third dose of methotrexate, the repeated $\beta\text{-hcG}$ test result was 12,220 units. She started having increasing lower abdominal pain. A transvaginal US was repeated and revealed a 1.5cm gestational sac located very close to a cornual fibroid which was now demonstrating fetal heart beat activity [Figure 2] and some free fluid in the peritoneum. At this time, histopathology was reported as a decidual reaction with no chorionic villi present in the tissue that was passed a few days before. In view of increasing abdominal pain, a viable fetus and elevated $\beta\text{-hcG}$, the concern for rupture was high. She consented to surgical intervention.

Laparoscopy revealed an 8 cm subserous fibroid at the right cornua and visible pulsations just under it (on the posterior aspect) suggesting

the location of the ectopic pregnancy. The adnexa were normal. It was not technically feasible to complete her management laparoscopically. Hence a 7cm Pfannenstiel incision was made and the laparoscopy findings were confirmed [Figure 3]. Needle aspiration of the cornual end was attempted blindly (as US guided aspiration was not possible in the operating room) to locate the ectopic gestation. Since even that did not show any evidence of the exact location of the pregnancy, a myomectomy was performed. The fibroid appeared to have undergone some degeneration as it was soft. The ectopic pregnancy was identified as a circular mass with a tiny black dot, likely representing the embryo, at the base of the myoma bed and this was evacuated. During the procedure, the endometrial cavity was not entered. A classical wedge resection was not carried out. The myoma bed was closed in layers with interrupted vicryl no. 1 sutures (Polyglactin 910, Ethicon, Johnson & Johnson) and 2/0 vicryl to the serosa. The uterus was reconstituted and looked normal at the end of the procedure. The intraoperative blood loss was 300 ml.

The serum $\beta\text{-hcG}$ test results were as follows: on admission 2,035 mIU; 48 hours later 6,684 mIU; on day 5 (after the third dose of methotrexate) 12,647 mIU; one day after cornuostomy 2200 mIU.

The postoperative period remained uneventful and the $\beta\text{-hcG}$ was monitored weekly. It became negative after three weeks. She was advised to have early evaluation at the next pregnancy to rule out a recurrent ectopic. In addition, she was advised to have an elective cesarean section during her next pregnancy. She resumed normal periods in about 4 weeks and remains well at the time of writing.

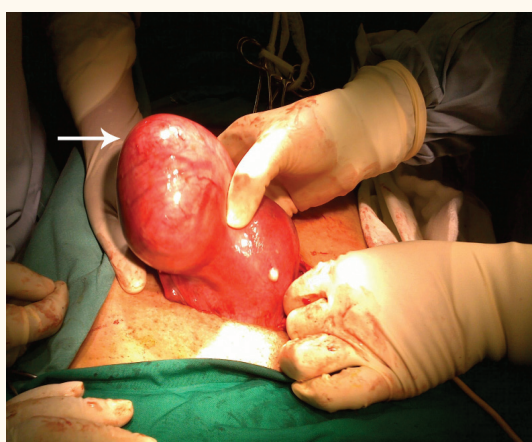


Figure 3: Intraoperative photograph of the uterus with the myoma (arrow) in the right cornual region.

Discussion

Despite recent advances, cornual pregnancy remains a diagnostic and therapeutic challenge. The three criteria defined by Timor *et al.* have increased the rate of diagnosis.² Fortunately, diagnosis was made early in our nulliparous patient and conservative measures were taken initially. Systemic treatment with multiple doses of methotrexate was given in this patient as she was haemodynamically stable, willing for follow-up and there were no absolute contraindications. In a prospective observational study at St George's Hospital Medical School, London, 17 out of 20 women with cornual pregnancy were treated with single-dose intramuscular methotrexate, which was administered on day 0. A second dose of methotrexate was given if the β -hcG levels had not fallen by 15%.³ All women with cornual pregnancy presenting with initial hcG values of <5000 mIU were treated successfully with single-dose methotrexate, but almost all women with an initial hcG of >5000 mIU required two doses according to the Royal College of Obstetricians and Gynecologists, UK.⁴ Medical treatment is not free of complications; it can be associated with uterine rupture and catastrophic haemorrhage.

A large ectopic pregnancy and the presence of a heartbeat are relative contraindications to medical treatment.¹ Multiple dose methotrexate is recommended by many authors for interstitial/cornual pregnancy despite the lack of strong evidence for or against a multidose methotrexate regimen.⁵

A US guided local injection of methotrexate/potassium chloride is described by Monteagudo *et al.* with immediate cessation of fetal heart activity.⁶ There was no time difference in the resolution of the ectopic whether they used potassium chloride or methotrexate and they reported a 100% success rate. Many studies have reported the use of laparoscopy for local methotrexate injection into a cornual pregnancy.⁷ In this case, local injection of methotrexate was not feasible either by US or laparoscopy due to the position of the ectopic under the myoma. Cornual excision, salpingostomy and repair are other approaches reported by many authors for cornual gestation.⁸ Successful cornuostomy has been reported by some other authors;^{9,10} however, a laparoscopic cornual excision and salpingostomy was not possible as an 8 cm myoma was sitting on the ectopic pregnancy; even visualising the pregnancy completely was impossible in this case. Hence a laparotomy and myomectomy, followed by a cornuostomy, were performed.

Conclusion

This is a rare case of cornual pregnancy situated under a cornual subserous fibroid. It was successfully managed by myomectomy and cornuostomy thus preserving the future fertility of the patient.

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