CASE REPORT

First ever case of spontaneous triplet cornual pregnancy: management of laparoscopic cornual resection with modified endoloop method

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SUMMARY

A 23-year-old woman, gravida 2 para 0, presented at 8 weeks gestation with a spontaneously conceived triplet cornual ectopic pregnancy. She was at high risk of ectopic pregnancy as she had been previously treated for pelvic inflammatory disease and had also undergone laparoscopic salpingostomy for right-sided ectopic pregnancy. She was clinically stable and her abdomen was soft and non-tender. The diagnosis was made on transvaginal ultrasound scan and this was confirmed on the three-dimensional scan. She was counselled about her treatment options and subsequently underwent laparoscopic cornual resection using the modified endoloop method. The estimated blood loss was 20 ml intraoperatively and the patient recovered well. She subsequently conceived spontaneously with an intrauterine pregnancy and underwent lower segment caesarean section at 37 weeks in view of previous laparoscopic cornual resection. Intraoperatively, the right cornua appeared normal and there was no sign of thinning.

had been isolated in the vaginal swab then and she had been adequately treated with oral antibiotics. Clinically, her vital parameters were normal. Her abdomen was soft and non-tender. Bimanual examination revealed an 8-week-sized uterus with no adnexal mass or tenderness.

INVESTIGATIONS

Transvaginal scan showed live triplet pregnancy at the cornual region, with the crown-rump length correlating to 8 weeks gestation (figure 1). It appeared as a mass of 8 cm×7.6 cm, situated 4 mm away from the endometrial lining. A high index of suspicion after the initial scan followed by availability of a three-dimensional scan confirmed the diagnosis without delay (figure 2). The serum quantitative β -human chorionic gonadotropin (β -hCG) was 97 927 IU/L. The patient was counselled regarding surgical management via the laparoscopic approach as well as the high risk of bleeding and other possible complications of the laparoscopic approach. Informed consent was obtained from the patient.

BACKGROUND

There have been several case reports of heterotopic pregnancies and ectopic pregnancies following assisted reproduction. However, to the best of our knowledge, a triplet cornual ectopic pregnancy after spontaneous conception has not been published in the literature. We therefore report the first case of spontaneous triplet cornual pregnancy. While the use of laparoscopic technique using the endoloop has generally been deemed feasible only for smaller defects, we describe the successful management of a large ectopic pregnancy using this technique with minimal morbidity. We recommend this technique for the management of all cornual pregnancies to minimise haemorrhagic complications, even if the size exceeds 5 cm.

CASE PRESENTATION

A 23-year-old woman, gravida 2 para 0, who was in a monogamous relationship, presented to the Emergency Department of the National University Hospital, Singapore, at 8 weeks of gestation. She had been referred by her general practitioner for suspected ectopic pregnancy. She was asymptomatic and had a significant history of a right-sided ectopic pregnancy 1 year prior to presentation, which was managed by laparoscopic salpingostomy. She also had a history of pelvic inflammatory disease 2 years prior to presentation. *Chlamydia*

TREATMENT

Diagnostic laparoscopy was performed via one 10 mm umbilical main port, two 5 mm ancillary ports in the iliac fossae and one 10 mm port in the suprapubic region. Intraoperatively, we found an anteverted uterus with an 8 cm mass at the right cornua. The mass had a broad base about 6 cm in diameter (figure 3A, B). The right fallopian tube was stretched over one side of this mass and the left tube and ovary were healthy. We then proceeded with laparoscopic resection of the cornual

Figure 1 A two-dimensional ultrasound scan showing the cornual ectopic pregnancy (thin arrow) separate from the endometrial lining (thick arrow).

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Figure 2 A three-dimensional ultrasound scan showing the triplet cornual ectopic pregnancy (3 thin arrows showing the 3 separate gestational sacs) separate from the endometrial lining (thick arrow).

ectopic pregnancy. A 20-gauge spinal needle, inserted directly through the skin, was used to infiltrate the base of the right cornual area with 10 mL of dilute vasopressin solution (10U in 100 mL 0.9% normal saline) to decrease the vascularity (figure 4A). This made the base prominent and created an angle to place and position the endoloop. Next we transected the right fallopian tube about 2 cm distal to the cornual mass. An endoloop (made of Vicryl 1–0 suture) was placed at the base of the mass. Gentle traction was applied to the transected end of the fallopian tube to ensure that the endoloop was below the bulge of the cornual pregnancy. The loop was then tightened carefully, sufficiently enough to strangulate the blood flow (figure 4B). A small incision was then made with diathermy over this bulge. The contents were sucked out using 5 mm suction through the suprapubic port and sent for histopathological examination (figure 4C). As the cornual mass reduced in size, the loop was tightened further to prevent bleeding from the cut cornual end. The excess wall of the cornual bulge was excised, edges were diathermised and the specimen was removed through the 10 mm port. No additional suturing was required.

OUTCOME AND FOLLOW-UP

There was minimal blood loss. The patient recovered well and was discharged 24 h postoperatively. The β -HCG 1 week after the surgery had dropped to 938 IU/L and fell further to 50.4 IU/L 3 weeks postoperatively. The patient subsequently conceived spontaneously in 2012 and it was an intrauterine pregnancy. The patient underwent lower segment caesarean section at 37 weeks in view of previous laparoscopic cornual resection. Intraoperatively, the right cornua appeared normal

and there was no sign of thinning. The placenta appeared normal, there was otherwise no abruption and no umbilical cord thrombosis noted.

DISCUSSION

Cornual pregnancies constitute 2–6% of all ectopic pregnancies with an incidence of 1 per 2500–5000 live births.¹ The incidence of cornual pregnancies after in vitro fertilisation is reported to be higher in some studies.² The Confidential Enquiry into Maternal and Child Health 2000–2002 stated that cornual pregnancy is a rare but dangerous type of ectopic pregnancy. Four in 11 deaths from ruptured ectopic pregnancies were due to ruptured cornual pregnancies and in all of them the diagnosis was made only after rupture.³ Hence, this type of pregnancy has traditionally been treated via laparotomy with cornual excision or hysterectomy.⁴ Various laparoscopic techniques have been subsequently described to be safe and effective, with more rapid postoperative recovery. Methods include laparoscopic salpingostomy and cornual resection with or without the endoloop technique. Unilateral ectopic multiple pregnancy is a very rare condition. The incidence of unilateral twin ectopic gestation is approximately 1 in 200 ectopic gestations.⁵

Risk factors associated with the higher incidence of interstitial ectopic pregnancy include previous ectopic pregnancy or salpingectomy, previous pelvic inflammatory disease, in vitro fertilisation and ovulation induction and uterine anomalies.⁶ In our index case, the patient had two risk factors due to previous ectopic pregnancy on the same side as well as previously treated pelvic inflammatory disease. There should be a higher index of suspicion particularly for patients with risk factors as cornual pregnancies are associated with a higher risk of shock and haemoperitoneum as well as higher risk of maternal mortality due to delayed diagnosis and high vascularity of the myometrium.³

In recent years, owing to advances in sonography, the diagnosis of cornual ectopic pregnancy is often made earlier and with precision regarding measurements and base dimensions. In the case of an early and asymptomatic ectopic gestation, methotrexate given either intramuscularly or intramyometrally has been shown to be an effective treatment.⁷ In cases not amenable to medical management, the feasibility of conservative surgical repair of a cornual pregnancy has also been reported during open surgery.^{8,9}

There has been a report of a unilateral triplet ectopic pregnancy reported after in vitro fertilisation and embryo transfer (two at interstitial and one at ampullary location), which failed medical management and required laparotomy, and removal of the tube and the cornual part of the uterus on the affected side.¹⁰

Figure 3 Laparoscopic picture of the uterus and cornual pregnancy depicting the 6 cm broad base (A) connecting to the 8 cm mass at the right cornua (B).

Figure 4 Steps of the laparoscopic cornual excisions depicting injecting of vasopressin into the base of the cornual mass (A), tightening of the endoloop (B) and suctioning of the contents from the cornual mass (C).

Laparoscopy has proven to have several advantages over laparotomy, such as shorter hospital stay, faster recovery and decreased health service costs.¹¹ In recent years, there have been several reports of laparoscopic management of cornual gestations describing different techniques. Hill *et al*¹² were the first to describe the endoloop technique in a patient who presented at 10 weeks gestation with an unruptured cornual pregnancy. They performed a salpingostomy by placing an endoloop (Ethicon) around the cornua and then successfully evacuated the pregnancy using unipolar current to incise over the pregnancy while tightening the endoloop. Subsequently, there have been further reports describing the technique of laparoscopic cornual excision to manage ruptured as well as unruptured interstitial pregnancies.¹³

In recent years, other methods have also been described for laparoscopic resection of the cornual pregnancy. Cucinella *et al*¹⁴ have described placing a temporary purse-string suture at the base of the cornual pregnancy, followed by resection of the cornual mass and suturing to reconstruct the myometrium over the cornua. Zou *et al* have described bipolar coagulation at the base of the cornual mass followed by resection and reconstruction.¹⁵ These methods require good laparoscopic suturing skills, which the general gynaecologist may not have. The endoloop method as described has the advantage of not requiring any additional laparoscopic suturing hence it can potentially have wider acceptability.

Less extensive laparoscopic procedures have been described for small cornual pregnancies by evacuation through a 1 cm salpingostomy, using electrocoagulation for achieving haemostasis.¹⁶ Tulandi *et al* later confirmed that conservative laparoscopic surgery could be used successfully for larger cornual pregnancies as well.¹² In their series, one patient with a cornual gestation measuring 3.5 cm underwent a laparoscopic salpingostomy. The authors noted, however, that cornual excision might be more appropriate for larger interstitial pregnancies measuring more than 4 cm.

Most reports have shown that many cases of cornual pregnancies were diagnosed after uterine rupture,³ with the median age of 11–20 weeks.¹⁷ The myometrium around the cornual ectopic

pregnancy is thick and highly vascularised and may potentially allow the gestation to expand, and protects it from rupture until the second trimester, which may lead to severe haemorrhage.¹⁸ In our article, though the diagnosis was made at 8 weeks gestation, it was a triplet pregnancy, which led to a larger than expected volume occupying the cornua, hence resulting in symptoms arising earlier than usually reported. This case is unique in that the cornual pregnancy was large since it was a triplet pregnancy with a broad base, and that would make laparoscopic excision difficult due to higher bleeding risks. Placing an endoloop is also difficult as the base is broad. Applying it more distally may result in an increased risk of residual trophoblastic tissue, while applying it more proximally may result in more uterine muscle being inevitably removed. Tightening of the endoloop was also challenging in view of the risk of cutting through friable and vascular tissues. Injection of vasopressin helps to delineate this border more clearly by causing the uterine muscle to contract, hence producing an hour glass demarcation, which helps in the exact placement of the endoloop. In addition, many articles have also reported the use of vasopressin injection into the cornua prior to incision to optimise haemostasis.⁴

Moon *et al* reported in a retrospective review the use of endoloop and encircling suture methods in 24 cases, and described the methods to be simple, safe and effective. There were no uterine ruptures reported in 11 patients with subsequent term pregnancies.¹⁹ It has also been reported that pre-operative uterine artery embolisation can be employed as an adjunctive measure to decrease intraoperative blood loss prior to laparoscopic cornual resection.²⁰

There is a concern regarding risk of uterine rupture in future pregnancies due to a defective area at the uterine cornua, especially if the endometrial cavity is involved. Hence most authors recommend elective caesarean section for delivery in subsequent pregnancies. Although it has been recommended that the uterine horn should be reconstructed and sutured in a double-layered manner to preserve its integrity, and to prevent uterine rupture in future pregnancies,¹⁵ term vaginal deliveries have been reported following laparoscopic treatment of cornual pregnancy without reinforcing sutures.²¹

While there have been several reports of laparoscopic management of cornual pregnancies, it needs to be emphasised that severe haemorrhage may occur during the surgical procedure. Hence the laparoscopic approach should only be attempted if the surgeon is well-skilled in laparoscopic technique and has the capability to convert the operation quickly to laparotomy if the situation calls for it.

Learning points

- ▶ This is the first ever case report of an extremely rare and dangerous condition of spontaneously conceived triplet cornual ectopic pregnancy.
- ▶ Risk factors associated with the higher incidence of interstitial ectopic pregnancy include previous ectopic pregnancy or salpingectomy, previous pelvic inflammatory disease, in vitro fertilisation and ovulation induction and uterine anomalies.
- ▶ In recent years, owing to advances in sonography, the diagnosis of cornual ectopic pregnancy is often made earlier and with precision about the measurements and base dimensions, allowing both conservative and surgical management.
- ▶ The combination of techniques employed during the laparoscopic cornual resection including vasopressin injection, endoloop placement and suction of contents, proved to be an easy and safe technique, with low morbidity and insignificant blood loss even in large cornual ectopic pregnancies.

Contributors AG was involved in the conception of the study, acquisition of patient data, drafting and multiple revisions of the work. LML was involved in the acquisition of patient data, drafting and multiple revisions of the work. AA was involved in the conception of the study, drafting and multiple revisions of the work. YFF was involved in the conception of the study, drafting and multiple revisions of the work as well as the final approval of the final version of the study for submission.

Competing interests None.

Patient consent Obtained.

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