Management of a spontaneously conceived live unilateral twin ectopic pregnancy in Australia: A case report

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A B S T R A C T
Introduction: Unilateral twin ectopic pregnancies are exceedingly rare, occurring in one in every 250,000 pregnancies. While clear guidelines exist regarding the management of singleton ectopic pregnancies, no such recommendations exist for multi-gestational ectopic pregnancies.

Case presentation: A 36-year-old woman, gravida 3 para 1, presented to the emergency department at 6 weeks and 4 days of gestation with a live twin ectopic pregnancy diagnosed on a dating ultrasound scan. Given the high likelihood of rupture, she underwent a laparoscopic salpingectomy and was discharged home the following day. The patient was followed-up with weekly serum β-hCG tests, to ensure there was no remaining pregnancy.

Discussion: While the morbidity and mortality associated with singleton ectopic pregnancies has gradually declined, the risk of rupture is higher in twin ectopic pregnancies and rupture is estimated to occur in 30–50% of cases. Surgical intervention remains the mainstay of treatment for these pregnancies.

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1. Introduction

Ectopic pregnancies are extrauterine gestations, accounting for 1.62% of all pregnancies in Australia [1]. Risk factors include a history of ectopic pregnancies, cigarette smoke, the use of an intrauterine contraceptive device, tubal sterilisation surgery, assisted reproductive techniques (ART) and anatomy distorted by pelvic inflammatory disease or operative trauma [2].

Guidelines recommend salpingectomy in the case of ectopic pregnancy with bHCG greater than 5000 IU/L due to increased risk of treatment failure and rupture with medical management [3]. bHCG levels in twin ectopic pregnancies are higher than in singleton ectopic pregnancies, which tend to be lower than in normal intrauterine pregnancies [2]. While the morbidity and mortality associated with singleton ectopic pregnancies is declining, the risk of hypovolemic shock as a result of rupture is higher in twin ectopic pregnancies and rupture is estimated to occur in 30–50% of cases [4]. While clear guidelines exist regarding the management of singleton ectopic pregnancies, neither the American College of Obstetricians and Gynecologists (ACOG) nor the UK National Institute for Health and Care Excellence (NICE) make recommendations on the management of multigestational ectopic pregnancies [5,6].

We present the case of a unilateral live twin ectopic pregnancy requiring a laparoscopic salpingectomy and discuss the management strategies reported in the literature as well as the implications for future fertility.

2. Case Presentation

A 36-year-old woman, gravida 3 para 1, presented to the emergency department at 6 weeks and 4 days of gestation with live twin ectopic pregnancies, diagnosed on a first-trimester dating ultrasound scan (see Fig. 1). The patient was asymptomatic, with no abdominal pain or vaginal bleeding, but was referred to the department by the radiologist. Twin A had a crown-rump length (CRL) of 7.7 mm and a foetal heart rate (fHR) of 133 bpm, while twin B had a CRL of 6.9 mm and a fHR of 125 bpm. This was a planned, spontaneous pregnancy on a background of no significant past medical or surgical history. The patient’s previous obstetric history included a first-trimester miscarriage, managed conservatively, followed by an uncomplicated pregnancy, delivered vaginally at term.

The patient was hemodynamically stable and physical examination found only mild tenderness in the right iliac fossa on deep palpation, without any evidence of peritonism. The serum β-hCG was 11,870 units/L and haemoglobin level 134 g/L. Given the high likelihood of rupture, the decision was made for definitive surgical management. A diagnostic laparoscopy and right salpingectomy were performed and an unruptured ectopic pregnancy was confirmed, localised at the cornual end of the right fallopian tube (see Fig. 2).

The patient was discharged home the following day and followed up with weekly serum β-hCG tests, to ensure there was no remaining...
pregnancy. Histopathology confirmed a fallopian tube and products of conception. No other abnormalities were found.

3. Discussion

Live twin ectopic pregnancies are exceedingly rare, occurring in one in every 250,000 pregnancies and are only sporadically reported in the literature [7]. We report a case of live unilateral twin ectopic pregnancy successfully managed with salpingectomy. In this case, ectopic pregnancy was an unexpected diagnosis in an asymptomatic, haemodynamically stable patient without risk factors for ectopic pregnancy, having conceived spontaneously without ART. Additionally, a transvaginal ultrasound scan had identified two distinct gestational sacs, indicating the pregnancy was dichorionic and diamniotic, whereas most twin ectopic pregnancies are monochorionic and monoamniotic.

We conducted a literature review of unilateral live multiple ectopic pregnancies to compare management decisions and outcomes. We identified 21 cases of live twin ectopic pregnancies [2,4,8–11,13–16,18,19,21–23]. Of these, 14 were diagnosed between six and eight weeks of gestation [2,4,6,8–11,18–22], while one pregnancy was diagnosed at just four weeks of gestation after presenting with abdominal pain and vaginal bleeding [14]. Only three pregnancies progressed beyond eight weeks of gestation [13,16,23], and the gestational age of the remaining two was not reported [12,17]. The serum β-HCG at diagnosis ranged from 3,374 to 110,774 units/L. Of all 21 cases in this series 16 were symptomatic with a combination of abdominal pain (n = 15) [2,4,8–11,13–16,18,19,21–23] and vaginal bleeding (n = 8) [2,11,13–15,19–21]. In comparison, only 45% of singleton ectopic pregnancies present with the clinical triad of amenorrhea, abdominal pain and vaginal bleeding [18]). There was no discernible correlation between gestational age, patient’s symptoms or serum β-hCG and the likelihood of rupture in the 21 live twin ectopic pregnancies in this series.

All except three of the 21 live twin ectopic pregnancies underwent a salpingectomy of the affected tube. Kim et al. [22] and Atye et al. [10] both reported performing cornual wedge resections on interstitial, multi-gestational ectopic pregnancies. Atye et al. also used postoperative intramuscular methotrexate and followed the serum β-hCG level to negative. Finally, Summa et al. [5] reported successful reanastomosis of the fallopian tube following a partial salpingectomy in an attempt for fertility preservation. No cases underwent medical management with methotrexate alone.

Our patient expressed concern regarding future fertility when salpingectomy was recommended. While systematic review of randomised trials found that fertility rates are similar after treatment of ectopic pregnancies with either salpingostomy, salpingectomy, or intramuscular methotrexate [24], women with a single fallopian tube or bilateral tubal ectopic pregnancies may benefit from fertility preservation with salpingotomy or methotrexate therapy. Betti et al. [25] and Berkes et al. [26] both described managing multi-gestational ectopic pregnancies without cardiac activity with a multiple dose regimen of intramuscular methotrexate. Both resulted in rupture of the fallopian tube and significant haemoperitoneum and required surgical intervention. The serum β-hCG level at diagnosis in both cases was greater than 5,000 units/L (13,217 units/L [26]; 60,348 units/L [25]). The use of intramuscular methotrexate in women with multigestational ectopic pregnancy must therefore be recommended with caution, balancing fertility preservation with the increased likelihood of rupture and the chance of treatment failure, and patients should be thoroughly counselled regarding these risks.

Fig. 1. Transvaginal ultrasound scan of the right adnexa, demonstrating two distinct gestational sacs, situated within the right fallopian tube.

Fig. 2. Tubal ectopic pregnancy localised towards the cornual end of the fallopian tube, demonstrated on diagnostic laparoscopy.
4. Conclusion

Live tubal ectopic pregnancies are rare. In the absence of specific guidelines or recommendations, surgical intervention appears the most appropriate course of management.

Contributors

A Martin drafted the initial manuscript.
K Balachandar drafted the initial manuscript.
P Bland conceived the report and provided support and feedback.
All authors approved the final manuscript before submission.

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

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Provenance and Peer Review

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