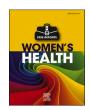
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A diagnostic dilemma of an 18-week cervical ectopic pregnancy: A case report

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ABSTRACT

Introduction: Cervical ectopic pregnancy (CEP) is characterised by the implantation of trophoblastic tissue within the cervical canal and is associated with a significant risk of maternal morbidity and mortality. This case report addresses a second-trimester CEP with unusual sonographic features suspicious of placenta accreta spectrum (PAS), which was successfully managed with an abdominal hysterectomy.

Case Presentation: A 27-year-old woman, G6P2, presented to the labour ward of a rural hospital at 18 weeks of gestation with premature rupture of membranes. The index pregnancy was complicated by an absence of any antenatal care, as well as a history of cigarette smoking and cannabis use. An ultrasound scan demonstrated a live pregnancy with the foetal head within the cervical canal. A termination of pregnancy was arranged with misoprostol 200 mg orally followed by an oxytocin induction. However, a repeat ultrasound scan, after 12 h of oxytocin infusion, which failed to terminate the pregnancy, demonstrated a still live foetus as well as increased vascularity, concerning for PAS. The patient underwent an emergency abdominal hysterectomy, with an intra-operative diagnosis of a CEP. The postoperative course was unremarkable, and the patient was discharged home on day 3 post-operatively.

Discussion: Appropriate antenatal care and early booking-in would have identified a CEP early in gestation and allowed for minimally invasive management and potential conservation of fertility. When this is not possible in such cases, meticulous pre-operative planning by a gynaecologist with experience in advanced pelvic surgery can minimise the associated morbidity and mortality.

1. Introduction

Cervical ectopic pregnancy (CEP) is characterised by the implantation of trophoblastic tissue along the lining of the endocervical canal [1]. It accounts for less than 1% of ectopic pregnancies and is associated with a significant risk of maternal morbidity and mortality due to brisk and often difficult to control haemorrhage [2,3]. Risk factors for CEP include in vitro fertilisation, cigarette smoke and previous uterine surgery, such as cervical dilation, uterine curettage or caesarean section [4,5]. An accurate diagnosis at early gestation allows appropriate planning and management of CEP using a combination of systemic or intra-sac methotrexate, uterine artery embolization and ultrasound guided endocervical curettage, with the aim to preserve fertility [6,7].

CEP typically presents as painless vaginal bleeding in early pregnancy. On a speculum examination the cervix is hyperaemic and may appear bulging with foetal membranes or products of conception protruding into the vagina and, consequently, is misdiagnosed as an

incomplete or impending miscarriage. Ultrasound features are similarly non-specific, especially with more advanced gestation. An hourglassshaped uterus with a ballooned endocervical canal, visualisation of an endometrial stripe, the absence of movement of the intracervical sac when the vaginal transducer applies pressure on the cervix (sliding sign) and a closed external cervical os are all characteristic of CEP and can help distinguish it from an incomplete or inevitable miscarriage [8,9]. Ultrasound, however, may challenge the diagnosis of CEP, especially at more advanced gestation, which may be misdiagnosed as placenta accreta spectrum (PAS), characterised by the invasion of trophoblastic villi into the myometrium and an absence of an intervening decidua basalis. The highly distorted vascular signal on colour ultrasound which can be seen in cases of CEP at more advanced gestation is also pathognomonic of PAS, which is usually managed with caesareanhysterectomy, in contrast to CEP which can be managed with fertilitysparing methotrexate therapy.

This is a case of a singleton pregnancy at 18 weeks of gestation with

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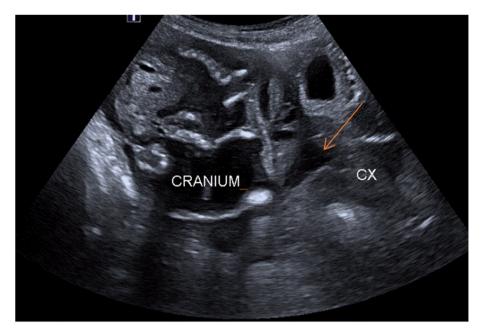


Fig. 1. Foetal head within the cervix, which appears to be funnelling (arrow), suggesting imminent miscarriage.

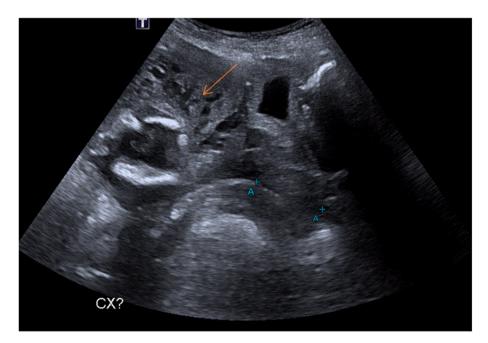


Fig. 2. Ultrasound evidence of finger-like placental projections (arrow) invading beyond decidua basalis, suspicious of placenta accreta spectrum.

sonographic features of both imminent miscarriage and PAS, and an intraoperative diagnosis of CEP. This case highlights the importance of appropriate preoperative planning and various fertility-preserving and non-fertility-preserving management options.

2. Case Presentation

A 27-year-old woman, gravida 6 para 4, presented to a rural hospital birth unit at approximately 20 weeks of gestation, having attended no antenatal care in the index pregnancy. Her presentation was on a background of four previous vaginal deliveries, all of which were at term except for the last pregnancy, which was delivered at 24 weeks gestation and resulted in an early neonatal death. A speculum examination confirmed rupture of membranes, with the cervix appearing closed.

Sonographic assessment dated the pregnancy at 18 weeks and 2 days with a foetal head seen within the cervical canal, indicating an imminent miscarriage (see Fig. 1). After no cervical change was observed for more than 24 h, the patient elected to terminate the pregnancy in view of the high likelihood of either miscarriage, delivery before viability or extreme preterm delivery, aware of the associated morbidity and mortality. She however failed a 12-h trial of an oxytocin infusion, followed by mifepristone 200 mg orally once and 5 doses of misoprostol 200 mg given buccally every 4 h. A repeat ultrasound found an open cervix with the head of a live foetus located within the internal cervical os. There were also radiological features that were concerning for PAS, including a highly distorted vascular signal and evidence of a placental bulge and finger-like trophoblastic invasion beyond the decidua basalis (see Fig. 2).



Fig. 3. A normal-sized, non-gravid uterus on top of a ballooned cervix indicative of a cervical ectopic pregnancy.



 $\textbf{Fig. 4.} \ \ \textbf{Histopathology specimen showing cervical implantation site and cervicotomy scar.}$

Following consultation with the reporting radiologist and gynecology-oncologist at the nearest tertiary center, the patient was transferred to theatre for management of a suspected PAS in the context of imminent miscarriage. A repeat vaginal exam found a 3 cm dilated cervix, with fetal tissue palpable within the endocervical canal. Based on these findings, the treating team, which included a senior gynecologist, anesthetist, a general surgeon and an interventional radiologist on

stand-by, took the decision to proceed with a total abdominal hysterectomy and bilateral salpingectomy with a provisional diagnosis of placenta accreta. Intraoperatively, the uterus appeared normal while the cervix was ballooned and found to be highly vascularized (see Fig. 3). Soft angled vascular clamps were used to isolate the uterine vessels bilaterally by perforating the broad ligament. The fetus was delivered through a transverse cervicotomy and the cord tied and cut close to the placenta, before separating from the uterus with gentle traction. The cervicotomy was repaired and proceeded to a total abdominal hysterectomy and bilateral salpingectomy. The total estimated blood loss was 500 mL. The patient was transferred to the intensive care unit (ICU) for postoperative management before being stepped down to the general surgical ward on day 1 postoperatively. She was discharged home day 3 $\,$ postoperatively. The histopathology subsequently confirmed the placenta had implanted within the endocervical canal but did not demonstrate any evidence of PAS (see Fig. 4).

3. Discussion

CEP is rare complication of pregnancy associated with a significant risk of maternal morbidity and mortality. These risks are made worse by the potential for an incorrect diagnosis, which has a significant impact on management decisions, made even more challenging in rural or resource-limited settings.

Ultrasound of CEP at more advanced gestation is prone to misinterpretation since the size of the foetus and its positioning within the endocervical canal make visualisation of the cervix and its relationship to the foetal head extremely challenging. In the case described above, the late presentation in the second trimester and the absence of previous ultrasounds for comparison resulted in an initial working diagnosis of an impending miscarriage rather than CEP. The consequence of this was a failed trial of medical termination of pregnancy with oxytocin infusion followed by mifepristone and misoprostol before proceeding to emergent surgical intervention. The abnormal implantation of trophoblastic tissue within the fibrous endocervical canal, rather than the endometrial cavity, was likely why medical management failed to result in separation of the placenta or expulsion of the foetus through the vagina.

The accurate diagnosis of CEP in early gestation allows initiation of appropriate management with a view to fertility preservation. Kim (2004) compared the outcomes of thirty-one cases of CEP that were managed with either methotrexate or uterine and endocervical curettage [10]. In that series, of twenty-two women who received methotrexate, none subsequently required hysterectomy due to haemorrhage or treatment failure. Seven women who received methotrexate therapy required blood transfusion and eight required subsequent surgical intervention for haemorrhagic control. There was also a clear correlation between more advanced gestational age and need for further intervention.

Correct diagnosis of CEP at early gestation minimises the risk of any subsequent morbidity and mortality. It is, however, increasingly challenging at more advanced gestation, especially in rural and resource-limited settings. If planning surgery at more advanced gestation, it is important that it occurs in a centre with a 24-h blood bank, high-dependency unit/ICU availability, and the support of an interventional radiologist for uterine artery embolization.

Contributors

All authors were involved in the clinical care of this patient, contributed to the drafting of the case report and have approved the final version of the article.

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Provenance and peer review

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Conflict of interest statement

All authors declare that they have no conflict of interest regarding the publication of this case report.

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