Abstract

Live unilateral twin ectopic gestations are a rare ultrasound finding with only 12 previous cases documented, none in Africa. This 13th case highlights a 30 year old primigravida who presented with abdominal pain. Transabdominal ultrasound found live right adnexal monochorionic diamniotic twin ectopic gestations and 200 mls of free fluid in the pouch of Douglas. Laparotomy showed two gestational sacs within the right fimbria and a right Salpingectomy was performed with good maternal outcome.

This case is different from most previous cases in having previous appendectomy as a risk factor and also for its use of transabdominal ultrasound. During transabdominal ultrasound, Sonologists must carefully evaluate any surgical scars and other risk factors, as these may raise the index of suspicion and lead to a prompt diagnosis of live unilateral monochorionic diamniotic twin ectopic gestations.

Key Words: Live, Monochorionic Diamniotic, Transabdominal Ultrasound, Twin Ectopic, Ghana.

Background

Ectopic pregnancy (EP) is a type of pregnancy in which the gestational sac is implanted anywhere outside the endometrial cavity. Such locations include the ampulla, isthmus and fimbria of the fallopian tubes and in a minority of patients the abdominal cavity, cervix and ovaries. Patients with a history of pelvic surgery including appendectomy show an increased risk of developing EP because of post-inflammatory scarring of the fallopian tubes. A unilateral twin ectopic pregnancy is a rare condition and occurs in about 1 in 200 ectopic gestations. Twin ectopic pregnancy occurs at a frequency of 1/125 000 and only 12 cases have been reported with documented fetal cardiac activity. When the diagnosis of EP is missed, there is a huge likelihood of a bad maternal outcome due to haemorrhage.

Case Report

A 30-year-old primigravida of Nigerian origin presented with a history of 2 month’s amenorrhea and 1 month’s lower abdominal pain. Although initially relieved by analgesics, pain became aggravated a day prior to presentation and was more severe on the right iliac fossa. There was no associated bleeding per vaginam or symptoms of anaemia. She had previously been investigated for infertility. She neither smoked nor drank alcohol. On examination, patient looked acutely ill, was in pain and showed mild pallor. Her cardio-respiratory systems were stable. The pelvis was full and tender on palpation with rebound tenderness and guarding, more within the right adnexa. Vaginal examination showed a normal introitus. There was fullness at the pouch of Douglas and cervical motion tenderness. The cervix was closed and there was no blood in the vagina. No pelvic masses were noted on bimanual examination.

Ultrasound Findings

Transabdominal ultrasound showed an empty uterus measuring 8.2 x 5.8 x 4.4 cm. The endometrium was mildly thickened and echogenic. No pseudogestational sac seen. There was a right adnexal gestational sac with twin gestations, both with active foetal heart activities (Fig 1-3).
Crown-Rump length measurements were 1.33cm and 1.27cm, corresponding to estimated gestational ages of 7 weeks 4 days each. A solitary chorion was noted and the “Lambda sign” was absent. A thin intervening membrane was noted between the two gestations, conforming to the “T – sign” seen in monochorionic diamniotic gestations. In addition, there was free intraperitoneal fluid estimated at 200mls.

Fig 1. Longitudinal and Transverse transabdominal ultrasound of the pelvis showing an empty uterus. There is a Folley’s catheter in the urinary bladder.

Fig 2. Pelvic Ultrasound images demonstrating a gestational sac with two embryos within the right adnexa.

Fig 3. Colour Doppler depicts 2 gestations with active foetal heart beats

The left adnexa was unremarkable. A diagnosis of haemoperitoneum secondary to leaking right adnexal live monochorionic diamniotic twin ectopic gestations was made. Patient and her partner were counselled for an emergency laparotomy.

Intraoperative findings

Exploratory laparotomy showed an unruptured right ectopic fimbrial gestation containing 2 sacs with attachment to the omentum, suggesting pelvic adhesions (Fig 4). There were additional moderate adhesions within the right adnexa and right iliac fossa, as well as 500 mls of haemoperitoneum. The right ovary was normal. The left fallopian tube was normal. There was a 2.0 x 2.0 cm left adnexal simple cyst.

Fig 4. Intraoperative photograph showing 2 embryos obtained from the right adnexa

After a right salpingectomy was performed, patient was monitored and was discharged home on the third post-operative day.

Discussion

Ectopic pregnancies are potentially life-threatening gynaecologic emergencies and require early diagnosis and prompt management for good maternal outcomes. Some studies suggest that there is a delay in ovum transport and consequently implantation, which increases the risk of occurrence of monozygotic twin pregnancies⁶.

The first report of a unilateral twin ectopic pregnancy was made in 1891 by De Ott⁷. In 1990, Sherer⁸ described unilateral twin ectopic gestations with the beating of the foetal heart by transabdominal ultrasound. Then in 1994, Gualandi⁹ documented live unilateral twin tubal pregnancy detected by transvaginal ultrasonography. Since then, an average of one case of live unilateral twin ectopic gestation has been reported per year by transvaginal ultrasound. Most recently, in 2018 Chang-Ihll et al⁵ reported the 12th case of a live unilateral twin tubal pregnancy making this current report the 13th case. Of the 12 previous cases of unilateral twin tubal pregnancies presenting with cardiac activity reported between 1994 and 2018, 4 cases had no risk factors and all were managed by surgical intervention except one⁵.

In the current case, the patient had a scar at McBurney’s point, from a previous appendectomy. This observation is consistent with a previous finding by Fernandez⁷ that appendectomy is associated with an
increased risk of EP in general. The presence of any previous appendectomy scars on a patient presenting for early pregnancy ultrasound should therefore raise the index of suspicion for EP. Other risk factors\textsuperscript{11} for EP include a prior ectopic pregnancy, prior genital surgery, psychiatric disorders, pelvic inflammatory disease, infertility, use of intrauterine contraceptive devices, tubal surgery, tubal ligation, smoking and previous exposure to Diethylstilboestrol, and Sonologists must be aware of the presence of any of these risk factors at the time of the ultrasound examination.

The patient in this case was of Nigerian origin and had a positive family history of twin gestations. This is consistent with the findings in literature that Nigeria is the country with the highest rate of twin pregnancy worldwide\textsuperscript{2}. The positive family history of twinning also corroborates previous research findings that twin pregnancy rates are higher in patients with a positive family history\textsuperscript{11} and also that twinning may be attributable to genetic and environmental factors\textsuperscript{12}. Indeed, globally, the highest burden of multiple pregnancy has been found not just in Nigeria, but in the entire sub-Saharan Africa, with an average twinning rate of 20 per 1,000 deliveries\textsuperscript{13}, double the figures from Europe\textsuperscript{14} and quadruple the rates in Asia\textsuperscript{13,14}. The evidence further shows that twins and multiple pregnancies worldwide are associated with a higher risk of maternal and perinatal morbidity and mortality as compared to singletons. The higher morbidity risk is manifested in prematurity, low birthweight, and intrauterine growth restriction\textsuperscript{15,16,17}. Interestingly, in the current case the diagnosis of live unilateral EP was made on transabdominal sonography. Several studies have compared the effectiveness of transabdominal and transvaginal sonography in the diagnosis of EP and confirmed the transvaginal approach to be vastly superior\textsuperscript{18-21}. One of the prerequisites for using the transabdominal approach is a full maternal bladder. Waiting for bladder filling may lead to unnecessary delays in diagnosis or possibly a suboptimal ultrasound procedure if a full bladder is not attained. In addition, there may be an undue need for a speedy procedure when the patient’s urinary bladder is overfilled and is no longer able to hold urine, potentially compromising the results. Transvaginal sonography, however helps identify subtle adnexal lesions which may be missed with other methods. Besides, patients do not require bladder filling for transvaginal ultrasound, enabling a quicker diagnosis. It must be noted however, that many ultrasound units in Ghana only have a convex probe and are not equipped for transvaginal ultrasound, as occurred in this case report. It therefore remains the responsibility of Sonologists to educate management in hospitals and clinics as to the value of investing in such valuable transvaginal transducers to avoid missing EP. Difficult cases may be referred for repeat ultrasound at centers with endovaginal transducers. In the interim, sonographers who only work with transabdominal ultrasound may still employ optimum bladder filling and a careful and systematic approach to aid the prompt diagnosis and management of EP.

This current case is distinguished from other cases of live unilateral twin ectopic gestations reported. Firstly, it is the first live unilateral twin ectopic pregnancy reported with previous appendectomy as a risk factor. Secondly, to the best of our knowledge this is the first report of a live unilateral monochorionic diamniotic twin ectopic gestation to be reported in Ghana and Africa. Thirdly, it is only the second report after Sherer\textsuperscript{a} et al in 1990 to diagnose a live unilateral twin ectopic gestation by transabdominal ultrasound. Sonologists will be encouraged by such diagnoses to continue to scan the adnexa even after one EP has been found, and even when they only have transabdominal ultrasound probes.

Conclusion
Careful and systematic ultrasound procedures are required for the early diagnosis and prompt management of live unilateral twin ectopic gestations. During sonography, it is still vital to review other areas of the pelvis even when an ectopic gestation has been found, because of the possibility of a second gestation. Radiologists must take into account any scars on the patient’s abdomen and other risk factors, as these may raise the index of suspicion and lead to prompt diagnosis of live unilateral twin ectopic gestations. Finally, though surpassed by transvaginal ultrasound, clinicians can still diagnose cases of live unilateral monochorionic diamniotic twin ectopic pregnancy on transabdominal ultrasound through careful ultrasound technique.

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JNO was responsible for the design of the manuscript for intellectual presentation. He also contributed to the collection, management, analysis and interpretation of the ultrasound images. EEA assisted in the operative management and subsequent follow up of the patient. Both authors read and approved the final manuscript.

EKO assisted in the preparation and reviewing of the manuscript.

RA assisted in the ultrasound image acquisition and formatting of this work.

References