

DOI: <https://dx.doi.org/10.18203/2320-1770.ijrcog20252354>

Case Report

Non-tubal ectopic pregnancy – case reports on diagnosis and management of ruptured primary abdominal ectopic and caesarean scar ectopic pregnancy in low resource setting

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Received: 13 June 2025

Revised: 07 July 2025

Accepted: 08 July 2025

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ABSTRACT

Non-tubal ectopic pregnancy is a rare form of ectopic pregnancy with high morbidity and mortality. The diagnosis of non-tubal ectopic pregnancy requires a high index of suspicion. Clinical history, physical examination, and laboratory and ultrasonographic features may all be non-specific. Fortunately, a low threshold for diagnosis, urine and serum beta-human chorionic gonadotrophin (β -hCG) assays and transvaginal sonography allow earlier diagnosis. Consequently, both maternal survival rates and conservation of reproductive capacity are improved. We present 2 cases of non-tubal ectopic pregnancy. The first is a case of a 17-year-old female para 0+0 with a period of amenorrhea, who presented with lower abdominal pain and moderate bleeding per vaginam. A suspicion of a ruptured ectopic gestation was made and an emergency exploratory laparotomy done which revealed a ruptured ectopic gestation implanted on the lower edge of the omentum with intact tubes and ovaries bilaterally was seen intra op. The second case is a 32-year-old female gravida 4 para 2 + 1 ectopic (2 previous caesarean section) who presented with recurrent bleeding per vaginam and lower abdominal pain. Transvaginal ultrasound (USG) report showed a cervical ectopic gestation and patient was counselled accordingly and prepared for curettage or total abdominal hysterectomy. However, about 18weeks gestation uterus, a gestation implanted in an old caesarean section scar extending into the cervix was seen intra op. No obvious intrauterine gestation, tubes and ovaries were all normal and hence a total abdominal hysterectomy was done with conservation of both ovaries. Non-tubal ectopic pregnancy is a rare but potentially life-threatening and often misdiagnosed condition. Our case presents an opportunity to discuss 2 different cases of rare forms of ectopic pregnancy and the importance of having a high index of suspicion to make a diagnosis and prompt management to reduce maternal morbidity and mortality.

Keywords: Ectopic pregnancy, β -human chorionic gonadotrophin hormone, Exploratory laparotomy, Caesarean section, Gestation

INTRODUCTION

In normal pregnancy, after fertilisation of the egg by a sperm in the fallopian tube, the fertilized ovum moves from the tube and goes to implant in the endometrial cavity of the uterus. Ectopic pregnancy is when a fertilized ovum implants anywhere outside the endometrial cavity of the

uterus.¹ Ectopic pregnancy may be classified as tubal and non-tubal of which the most common is tubal (95%).² Non-tubal ectopic pregnancy, as the name suggests, are seen in sites other than the fallopian tubes including cervix (<1%), previous caesarean scar (<1%) and the peritoneal surface or abdomen (1%) which are altogether termed non-tubal ectopic pregnancy.^{3,4}

Ectopic pregnancies occurs in 1-2% of pregnancies worldwide.⁴ Prevalence of ectopic pregnancy is 1 in 90 pregnancies in the UK, 1 in 50 pregnancies in the USA while that in Ghana is 1 in 100 pregnancies.⁵

Though causes of ectopic pregnancy are not well understood, studies suggests that ectopic pregnancy may be from both abnormal transport of the fertilised ovum and changes in the tubal environment hence leading to abnormal implantation.⁶

However, though several risk factors have been implicated, some patients may not have any of these risk factors. The commonest risk factor of ectopic pregnancy is partial tubal blockage. Other risk factors include tubal damage from abdominopelvic surgeries and pelvic inflammatory disease, failed tubal ligation, previous ectopic pregnancy (increases risk by 10-20%), previous tubal surgery, uterine fibroids, congenital anomalies of the fallopian tube (congenital tubal diverticula, abnormally long tube, accessory ostia, tubal stenosis).⁷

In about 50% of the time, a woman with an ectopic pregnancy presents with the classic triad of a period of amenorrhoea, abdominal pain and bleeding per vaginam (74%).⁸ Others may present with symptoms of early pregnancy like nausea, vomiting and breast fullness. The presence of severe abdominal pain (96%), dizziness, difficulty in breathing, fast breathing, shoulder tip pain, syncope, pallor, fast heart rate, low blood pressures, and signs of peritonism (abdominal rigidity, guarding and severe abdominal tenderness) suggest a haemoperitoneum from a ruptured ectopic pregnancy and this is a surgical emergency.⁹

In the unruptured or subacute ectopic presentation, a serial β -hCG, pelvic or transvaginal ultrasound is requested to confirm diagnosis. Laparoscopy is the gold standard for diagnosis; however, diagnosis may be missed in up to 4% of early ectopic pregnancies.¹⁰ In low resource settings ultrasonography, be it transabdominal or transvaginal can be used to make a diagnosis. Ectopic pregnancy is usually seen as an empty uterus but slightly enlarged uterus due to hormonal stimulation and an extrauterine gestational sac.¹⁰ When there is a ruptured ectopic gestation, free floating bowel, fluid filled pouch of Douglas may be seen on ultrasound. A standard quantitative test that complements the diagnosis is the serum β -hCG.¹¹ This biochemical assay correlates with growth of early intrauterine gestation by doubling every 48-72 hours but in ectopic pregnancies there is a lower rise in serum β -hCG (i.e. fails to double after 48 hours).¹¹

Management options in ectopic pregnancy include expectant management, medical and surgical management depending on the type of ectopic, patient's presentation, expertise of the clinical team, and patient choice.¹²

With an acute ruptured ectopic presentation, tubal or non-tubal, where patient may or may not be haemodynamically stable, emergency advanced resuscitation is instituted followed by emergency surgical intervention laparotomy or laparoscopy is the treatment of choice.¹³ For unruptured ectopic, the patient and clinician have the option of either expectant management where patient is observed for resorption of the pregnancy or some pharmacological agents (like methotrexate, potassium chloride, hyperosmolar glucose) are given if patient the criteria for medical management to end the pregnancy.¹⁴ In special cases like caesarean scar ectopic expectant management is highly discouraged due to risk of haemorrhage, uterine rupture, morbidly adhered placenta and death.¹⁵ The most reliable and quickest option being surgery by laparoscopy or laparotomy may also be employed to terminate the pregnancy.

However, in advanced abdominal pregnancy, unlike the other types of ectopic, the placenta is left in-situ for either spontaneous resolution or hasten with the help of methotrexate but this has been reported to cause accumulation of necrotic tissue and infection with abscess formation.¹⁶

Besides the most feared complication of ectopic which is rupture, haemorrhage and subsequently shock leading to death, damage to the fallopian tube also predisposes patients to recurrence and even subfertility in the future.¹⁷ Treatment of ectopic pregnancy also come with its own complications like surgical site infection, iatrogenic damage to tube and thus subfertility.

CASE REPORTS

Case 1

We report a case of a 17-year-old female para 0+0, who was referred to the emergency unit of the Eastern Regional Hospital as a case of lower abdominal pain in cyesis and malaria. She presented with a 6-week period of amenorrhea prior to a 10-day history of lower abdominal pain. She described the lower abdominal pain as gradual in onset, constant, non-radiating which had progressively worsened over the period and associated with a day's duration of unprovoked bleeding per vaginam for which she reported to the referral facility where she was then sent to our emergency for further management. At presentation, she was generally stable, asymptomatic of anaemia though vulva pad was soaked with bright red blood.

On examination we saw a young female not pale, afebrile, anicteric, hydration satisfactory, and positive urine pregnancy test. Vital signs recorded temperature 36.4 blood pressure 122/67 mmHg with a pulse rate of 102 bpm regular and of good volume, breathing at 20cpm and with oxygen saturation on room air being 98%. Her abdominal examination findings were flat soft abdomen that moved with respiration, moderate tenderness in right iliac, suprapubic and left iliac regions. There was no guarding,

no rebound tenderness, no organomegaly and bowel sounds were present and normal. All other systems were normal. A speculum examination done showed healthy vulvovaginal wall, os closed, blood clots in the posterior fornix. A bedside scan showed an empty uterus and both adnexa were not visualized.

An impression of bleeding in early cyesis to rule out ruptured ectopic gestation was made, samples were taken for full blood count, grouping and cross matching, serum β -hCG, and a formal pelvic scan done to confirm the diagnosis. Ultrasound scan report showed complex mass with a centrally located gestational sac abutting the right adnexa estimated gestational age of 6 weeks 2 days, Uterus empty with 88 ml of fluid in Pouch of Douglas (Figure 1).

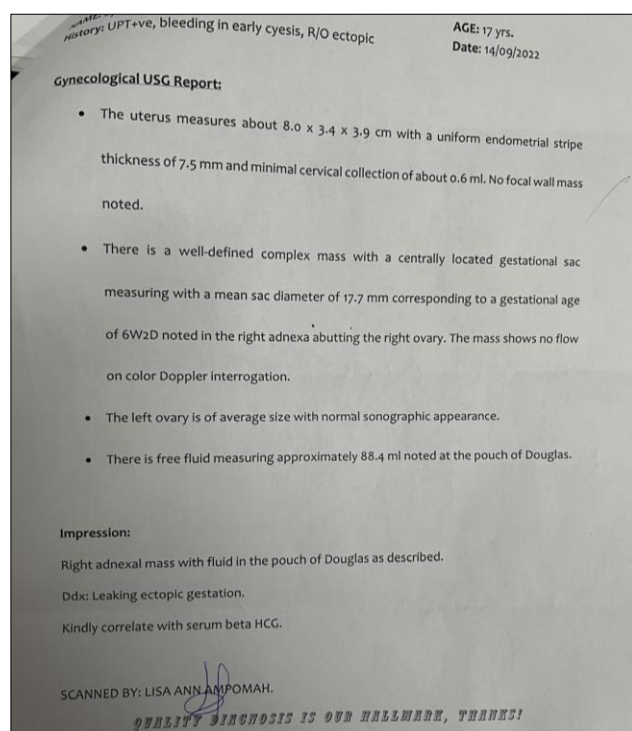


Figure 1: Transabdominal ultrasound report of patient discussed in case 1.

Patient was counselled on the findings, an informed consent sought for and sent for emergency laparotomy o/a ruptured ectopic gestation. Under asepsis and subarachnoid block, the abdominopelvic cavity was accessed via a lower transverse abdominal incision. Hemoperitoneum of about 100 ml was noted, gestational sac (ruptured with clots) attached to the omentum was seen intraoperatively (Figure 2). Both tubes and ovaries were normal. Ruptured gestational sac was resected (Figure 3), haemostasis secured and wound closed.

Patient remained well during the post-operative period. Patient was informed of the intra-op findings and the need to report to any health facility when she misses her menses. Patient was counselled on the need for serial β -hCG checks, family planning and discharged home post op day

3 on oral antibiotics and analgesics to be reviewed at on outpatient basis. She never did the β -hCG because of financial constraints and has since been lost to follow.

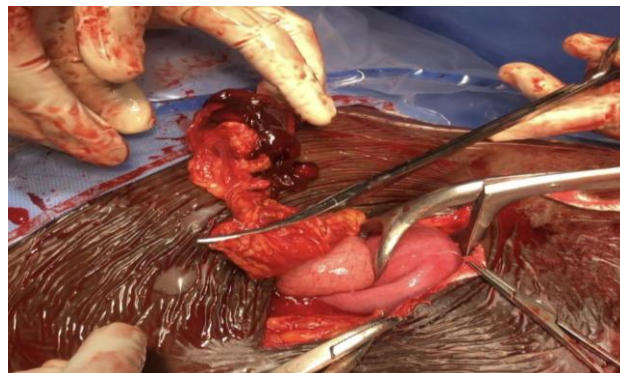


Figure 2: Ruptured ectopic gestation which was implanted on the omentum.



Figure 3: Ruptured ectopic gestation with some part of the omentum on which it was attached (resected).

Case 2

We also present a 32-year-old gravida 4 para 2(all caesarean deliveries) + 1 ectopic female who presented with recurrent spotting and lower abdominal pain. She was admitted and investigations done. Serum β -hCG came out as 59933 mIU/ml (Figure 4) and all other blood work (full blood count, liver panel, urea, creatinine and electrolyte) came out normal. The transvaginal ultrasound done showed single foetal pole inside a gestational sac with foetal heart rate of 160 bpm, crown rump length of 17.4 mm, and estimated gestational age of 8 weeks 1 day. Gestation with foetal pole was seen at the cervical region (Figure 5). A repeat serum β -hCG done 7 days after the first saw it increased to 114065 mIU/ml (Figure 6). Patient was counselled on medical and surgical management, and she consented to the latter.

She was prepared for curettage or total abdominal hysterectomy on account of cervical ectopic gestation. About 8weeks gestation uterus, a gestation implanted in an old caesarean section scar extending into the cervix was seen intra op (Figure 7). No obvious intrauterine gestation, tubes and ovaries were all normal but with a bulky uterus.

Total abdominal hysterectomy with conservation of both ovaries was done. Post op condition was satisfactory, and she was discharged on post op day 3 on antibiotics, and analgesics. Recovery was uneventful with no complications.

MEDICAL LABORATORY REPORT			
HOSPITAL LAB. KOPORINDA DEPOT			
CHAMA			
Patient :	NOT AVAILABLE	Quarantee :	CASE N
Doctor Ref :	32 / 8 / 1300708	Medial :	0245 878 183
Age/Date/DOB :	1108/02/2010	Tel :	(N) NOT AVAILABLE
Lab Ref :	70112529	Collection Date :	08/11/22 1453
Ref No. :	000182712	Received Date :	08/11/22 1544
Spec N :	1108/02/2010	FINAL Report Date :	09/11/22 1523
Requested : .. QHCG			
ENDOCRINOLOGY			
Test	Result	Reference	

h-HCG	13408.0 mIU/ml		
Healthy non pregnant perimenopausal women	2 - 5.3 mIU/ml		
Healthy post menopausal women	2 - 2.6 mIU/ml		
Male	2 - 2.6 mIU/ml		
Weeks of gestation	hCG mIU/ml		
3	5-10		
4	5-10		
5	5-10		
6	5-10		
7	5-10		
8	5-10		
9	5-10		
10	5-10		
11	5-10		
12	5-10		
13	5-10		
14	5-10		
15	5-10		
16	5-10		
17	5-10		
18	5-10		

**The gestational weeks 14 to 18 are the relevant weeks **			
***** for the trisomy 21 risk assessment *****			
** Continued on Next Page **			

Figure 4: First β -HCG results of patient discussed in case 2.

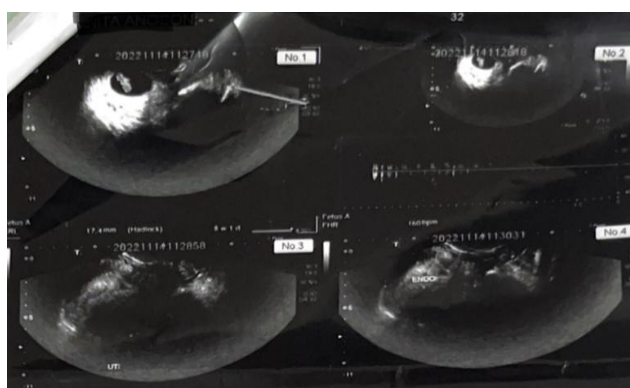


Figure 5: Sonogram of patient discussed in case 2 showing fetal pole in a gestational sac located close to the cervix.

MEDICAL LABORATORY REPORT			
HOSPITAL LAB. KOPORINDA DEPOT			
CHAMA			
Patient :	NOT AVAILABLE	Quarantee :	CASE N
Doctor Ref :	32 / 8 / 1300708	Medial :	0245 878 183
Age/Date/DOB :	1108/02/2010	Tel :	(N) NOT AVAILABLE
Lab Ref :	70112529	Collection Date :	18/11/22 1300
Ref No. :	000182712	Received Date :	18/11/22 1338
Spec N :	1108/02/2010	FINAL Report Date :	18/11/22 1338
Requested : .. QHCG			
ENDOCRINOLOGY			
Test	Result	Reference	

h-HCG	13408.0 mIU/ml		
Healthy non pregnant perimenopausal women	2 - 5.3 mIU/ml		
Healthy post menopausal women	2 - 2.6 mIU/ml		
Male	2 - 2.6 mIU/ml		
Weeks of gestation	hCG mIU/ml		
3	5-10		
4	5-10		
5	5-10		
6	5-10		
7	5-10		
8	5-10		
9	5-10		
10	5-10		
11	5-10		
12	5-10		
13	5-10		
14	5-10		
15	5-10		
16	5-10		
17	5-10		
18	5-10		

**The gestational weeks 14 to 18 are the relevant weeks **			
***** for the trisomy 21 risk assessment *****			
LIMITATION OF PROCEDURE			
As with all diagnostic tests, a definite clinical diagnosis should not be based on the results of a single test, but should be made by physician after all clinical and laboratory findings have been evaluated. Please notify the Lab to repeat the test if clinical suspicion and/or clinical symptoms are in discordance with our results for consultation by referring doctor only.			
Dr David Rumbau +2733 358 0800		Dr H M Van Deventer +2733 358 0800	
Dr Peter P. Tshameka +2733 358 0800		Dr Jacques De Groot +2733 358 0800	

Figure 6: Repeat β -HCG results of patient discussed in case 2.



Figure 7: Intra-op pictures of gestational sac implanted on the old CS scar extending to the cervix anteriorly.

DISCUSSION

Abdominal pregnancy is a rare type of ectopic pregnancy that occurs when there is implantation of the embryo in structures in the peritoneal cavity. It may be primary, where fertilization and implantation occur in peritoneal cavity and abdominal organs or secondary, where there is detachment into the abdominal cavity through tubal abortion, rupture of tube or uterus.¹⁸ The latter being commoner.

Diagnosis is usually difficult. An abdominal pregnancy can go undetected until an advanced gestational age, at which most abdominal pregnancies are discovered, complicating further management.¹⁹

Clinical history, physical examination, and laboratory and ultrasonographic features are all non-specific but it must be suspected if patient complains of recurrent or persistent abdominal pain throughout pregnancy, easily palpable fetal parts, persistent and fixed abnormal lie of the foetus and also the uterus can be palpated separately from the foetus on bimanual examination.²⁰ Confirmation is by ultrasound and very occasionally by plain abdominal x-ray showing the presence of fetal parts superimposed on the maternal spine. Ultrasonographic features of abdominal pregnancy include no uterine wall visualised between the maternal urinary bladder and the foetus, the placenta location outside the uterus, foetal parts are close to the maternal abdominal wall, the foetus lies abnormally, and no amniotic fluid is present between the placenta and foetus.²¹ Magnetic resonance imaging (MRI) is an excellent modality to assess the definitive area of placental implantation. However, it may not be readily available in many centres and is rarely required.²²

To diagnose a primary abdominal pregnancy, the Studdiford criteria should be met: normal tubes and ovaries, no evidence of uteroperitoneal fistula, and pregnancy related solely to the peritoneal surface and no evidence of secondary implantation following initial primary tubal nidation.²³

With this in hindsight our first case report is most certainly a primary abdominal pregnancy.²⁴

Although the diagnostic tools to identify an abdominal pregnancy are well established, optimal treatment guidelines is less certain. Current treatment options consist of conservative management, surgery with termination of the pregnancy (removal of the fetus) via minimally invasive laparoscopic surgery or medical management with use of methotrexate, embolization, and combinations of these.^{25,26} Because abdominal pregnancies typically implant on highly vascular surfaces such as the liver, spleen, omentum, large blood vessels, or abdominal serosa, the most minimally invasive but most effective means of treatment must be used.^{12,25} Many different agents have been used to treat ectopic pregnancies including systemic and local methotrexate, local potassium chloride and hyperosmolar glucose, prostaglandins, danazol, etoposide, and mifepristone. Most investigators have reported varying success rates in the medical treatment of abdominal pregnancies with local potassium chloride and/or local methotrexate, sometimes with the addition of systemic methotrexate.²⁶

Sapuri and Klufio indicate that conservative treatment is also possible though there is little published information on the duration of conservative treatment that can be safely employed beyond this stage to gain further fetal maturity.²⁷ Ideal management of abdominal pregnancy is multidisciplinary. The choice, however, is largely dependent on gestational age of the foetus, patient condition, and understanding and compliance to her choice of management. In our case emergency exploratory laparotomy was the only option and that was done for her.

The management of abdominal pregnancy clearly depends on the stage at which it is diagnosed. Our patient was a case of ruptured early abdominal pregnancy hence emergency laparotomy. If diagnosed before 28 weeks' gestation then conservative management is feasible provided there is absence of a major congenital malformation, a live fetus, continuous hospitalization in a well-equipped and well-staffed maternity unit with immediate blood transfusion facilities available, careful monitoring of maternal and fetal wellbeing and placental implantation in the lower abdomen away from the liver and spleen. In such a case, once sufficient viability is reached (after 28 weeks' gestation) immediate laparotomy and delivery is recommended.²⁸

There is continuing controversy with regard to the management of the placenta.^{29,30} Whether the abdominal pregnancy is early or advanced, it is recommended that the umbilical cord be ligated as close as possible to the placenta. The removal of the placenta is likely to be associated with torrential and uncontrollable intraabdominal haemorrhage.²⁸ Since this case was early cyesis with placenta barely formed, all of the gestation

together with its attachment to the omentum was resected and hemostasis secured.

Where the placenta is left in situ, it may continue functioning for a number of weeks, and even as long as five and a half years. Both serial β -hCG levels and MRI can be used to follow up placental involution postoperatively. Methotrexate may be added to hasten placental involution, as we done by Ombelet et al.²⁹

In keeping with best practice, we would have loved to do serial β -hCG, but client was not forthcoming and was lost to follow up.

Caesarean scar ectopic pregnancy (CSP) occurs when a pregnancy implants on a caesarean scar. It is rarest of all ectopic pregnancies. Its incidence approximates 1 in 2000 normal pregnancies and has increased along with the cesarean delivery rate.^{31,32} Increasing caesarean delivery rates over the past two decades and advances in prenatal imaging have led to an increase in the number of patients with CSP diagnosis. Despite the high burden of maternal morbidity associated with this condition, CSP is commonly misdiagnosed.³³

As the name suggests, the single most important risk factor is previous caesarean section which our patient had had on 2 occasions. She also had history of an exploratory laparotomy for ectopic gestation and this further increases her chance of recurrence.

Clinical manifestation of the pregnancy implanted in the scar after caesarean section may vary from symptomless to sharp abdominal pain with vaginal bleeding like seen in our patient. It is life threatening condition, causes excessive haemorrhage and risk of uterine rupture. Women with CSP usually present early, and pain and bleeding are common. Still, up to 40 percent of women are asymptomatic, and the diagnosis is made during routine sonographic examination.³²

The ultrasound criteria for CSP have been redefined recently, proposing transvaginal ultrasound as the imaging technique of choice at early gestation and MRI to be of no significant help. CSP is diagnosed in the presence of an early gestational sac and/or placenta in close proximity to the previous hysterotomy scar/niche in a patient with previous CS and a positive pregnancy test.¹⁵ Our radiologist could not tell whether gestational sac was in close proximity to the patient's previous scar on ultrasound but with history 2 previous caesarean delivery and a positive pregnancy a diagnosis of CSP was probable.

Definitive algorithm of CSP treatment is still not established. Pharmacological and operative methods are approved while expectant observation is considered unsafe due to possible risk of complications for the patient, including hemorrhage, placenta accreta, uterine rupture and death. Nonetheless in a review by Maheux-Lacroix et

al, live birth rates of 57 percent were recorded with expectant management.³⁴ For patient desiring sterilization, hysterectomy is an acceptable initial choice. Pharmacological treatment options include systemic or local administration of methotrexate, chloride potassium, hyperosmolar glucose solution, prostaglandin or combination of the medications in laparoscopic assist or its direct injection to the gestational sack under transabdominal or transvaginal ultrasound control.

Surgical management include visually guided suction curettage, hysteroscopic removal, or isthmic excision done abdominally or vaginally. These may be done alone or with adjunctive methotrexate.³⁵

Fertility-preserving options include systemic or locally injected methotrexate either alone or combined with conservative surgery is available and was discussed with our patient but she opted for surgical termination by either curettage or hysterectomy.³⁶

Treatment of CSP is challenging. Because of definitive consensus of CSP treatment is still not established — the type of treatment method depends on many factors such as size of pregnancy, presence or absence of uterine continuity, level, the possibility of further fertility and patient's hemodynamic state and should be based on the experience of the treatment centre.

CONCLUSION

Non-tubal ectopic pregnancy is a rare but potentially life-threatening and often misdiagnosed condition. Successful diagnosis and management depend on a high index of suspicion. This is particularly pertinent for caesarean scar ectopic pregnancies, the prevalence of which is increasing due to the rising proportion of women having Caesarean sections. While ultrasound and serial serum β -hCG may help in the diagnosis, there is no single diagnostic tool available. Even in the era of increased access to advanced diagnostic imaging modalities, the diagnosis and management is still a challenge to obstetricians. Albeit the relative rarity of these advanced healthcare system in sub-Saharan Africa, this case reports highlights the importance of thorough early clinical assessment and comprehensive ultrasound assessment of patients with presumptive symptoms of ectopic pregnancy.

Practitioners and radiologists alike ought to have a high index of suspicion, improve their skills, understanding and interpretation of clinical and imaging findings is imperative in making a diagnosis, optimize management and increase patient safety.

ACKNOWLEDGEMENTS

The authors are grateful to the patients who gave consent for their cases to be shared and all staff of Gynaecology Department of the Eastern Regional Hospital, Koforidua.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

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Cite this article as: Ahorklo IMK, Nimoh-Brema NA, Attuah SK, Wuobar F. Non-tubal ectopic pregnancy – case reports on diagnosis and management of ruptured primary abdominal ectopic and caesarean scar ectopic pregnancy in low resource setting. *Int J Reprod Contracept Obstet Gynecol* 2025;14:2747-53.