Case Report

An Advanced Intact Tubal Ectopic Pregnancy with an Alive Foetus of 19 Weeks Gestation; A Rare Case Report

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Abstract

Ectopic pregnancy is the leading cause of direct maternal morbidity and mortality in the first trimester of pregnancy worldwide. It can undergo spontaneous resolution or a rupture; leading to serious health consequences, making its early diagnosis imperative. Almost 98% ectopic pregnancies implant in ampullary portion of the fallopian tube. Due to the weak musculature, tubal ectopic pregnancy usually becomes symptomatic around 6-7 weeks of gestation. Rarely it can progress undiagnosed even beyond first trimester, endangering the woman's life. Here we are presenting such an unusual case of advanced tubal ectopic pregnancy of 19+2 week's gestation in a young primigravida, referred to our hospital with diagnosis of an extrauterine abdominal pregnancy, on an obstetrical ultrasound done a day earlier in a peripheral health care center. Owing to uneventful progress of pregnancy, she never had any antenatal checkup till she experienced dull lower abdominal pain for the last few days and had this ever first ultrasound. She was haemdynamically stable with mildly tender abdomen and symphysio-fundal height of 22cm. Our further workup including a detailed obstetric and Doppler ultrasound confirmed the diagnosis of abdominal pregnancy harbouring an alive but anomalous fetus. MRI was unavailable for precise localization of ectopic pregnancy. An exploratory laparotomy was proceeded, revealing an intact, mobile left tubal pregnancy mass 15×15 cm in size, which was excised intact via partial salpingectomy. This case emphasizes the importance of timely and accurate diagnosis of ectopic pregnancy to avoid life-threatening haemorrhagic consequences.

Keywords: Advanced ectopic pregnancy, Hemoperitoneum, Tubal pregnancy, Tubal rupture, Ultrasonography. .

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Introduction

A fertilized ovum implanted outside the normal uterine cavity is termed as an ectopic pregnancy (EP) and corresponds to almost 2% of all pregnancies, fluctuating between 1.3% to 2.4%.[1] It is a true gynaecological emergency as it typically ruptures within first few weeks of pregnancy resulting in intraperitoneal haemorrhage and shock, accounting for severe maternal morbidity and nearly 6% of maternal deaths in first trimester.[2] Almost 96% - 98% of all the EP occur in fallopian tube; the ampulla being the commonest site, while other parts of tube are less frequently involved. Other less common sites for EP are; ovary, cervix, broad ligament, pouch of Douglas, abdominal cavity or least likely within the myometrium of uterus or uterine scar.[3] A tubal ectopic pregnancy (tEP) is typically diagnosed at an average gestational age of 6 (±1.5) weeks. The size of tubal pregnancy before rupture is usually 1.5-3.5 cm,[2] after this a lack of submucosal layer within tube wall encourages the trophoblastic invasion rapidly through the thin tubular muscular wall which strains to keep up, often rupturing around 7.2 ± 2.2 weeks.[4,5] An EP presents with a typical symptom's triad of gestational amenorrhoea, lower abdominal pain and vaginal bleeding.[6] However presentation may be quite atypical or asymptomatic having an incidental diagnosis.

Clinical confirmation of EP requires quantitative assessment of β-HCG levels and a transvaginal ultrasonography showing the characteristic findings of 'blob sign' (indistinguishable adnexal mass), 'tubal ring' (ectopic gestational sac devoid of fetus), and an empty uterine cavity or an ambiguously located pregnancy. These findings have a positive predictive value of >94% for tEP.[7] The widely recognised risk factors are; advanced maternal age, history of previous EP or pelvic surgery, intrauterine contraceptive devices, tubal sterilisation/reconstruction, pelvic inflammatory disease, subfertility, low BMI and smoking during pregnancy.[8,9] Treatment modalities can be expectant,

medical or Surgical one, dictated by the particular clinical scenario.[1]

Owing to a thin and less compliant musculature, it is very unusual for a tEP to advance intact beyond first trimester, however literature reveals a few intact tubal EP spreading over a span of 13 to 34 weeks of gestation, [6,10-17] at term (42 weeks)[18] and even beyond term (50 weeks).[19]

Here we present another such rare case of advanced intact tubal pregnancy that went undetected until 19th week of gestation harbouring an alive anomalous foetus and then misdiagnosed as an abdominal pregnancy, putting the life of woman in grave danger.

Case Report

A 25 years old Primigravida married for 1.5 years, reported to emergency department of Aziz Bhatti Shaheed Teaching Hospital, Gujrat with the diagnosis of extrauterine abdominal pregnancy of 19+2 weeks' gestation, confirmed on an obstetrical ultrasound done one day earlier in a peripheral health care centre.

Clinical History; This was her spontaneous conception, confirmed by an early urine dip-stick pregnancy test at home. As progress of pregnancy remained uneventful, she did not go for any antenatal checkup till the time when she experienced dull lower abdominal pain for the last few days and had this ever first obstetric ultrasound on her own by a local sonologist, who referred her to our tertiary care hospital. There was no associated nausea, vomiting, fever or vaginal bleeding. Her past medical/surgical history was insignificant. No any risk factor for an EP could be detected. On examination she was haemdynamically stable. Abdomen was soft and mildly tender; symphysio-fundal height was almost 22cm and feeble foetal movements were appreciable (Figure.1). She was admitted for further workup.

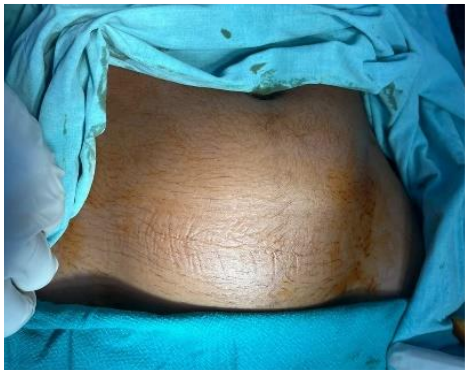

Figure 1. Abdominal Mass of Ectopic Pregnancy

Investigations & Diagnostic Assessment; Her detailed obstetric and Doppler ultrasounds were sought on urgent basis by senior sonologist. Obstetric ultrasound revealed an alive abdominal pregnancy of 19 weeks outside the uterus enclosed in a thin-walled gestational sac in lower mid abdomen just above the uterus, having no communication with uterus or cervix. Uterus was visualized normally measuring 83×49 mm with markedly thickened endometrium of 18 mm (Figure.2).

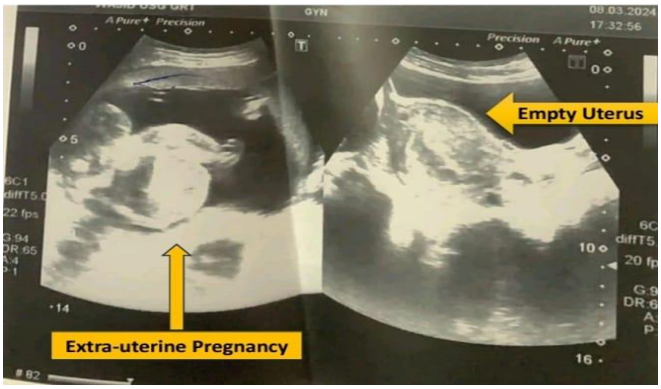

Figure 2. Ultrasound Images mimicking Ectopic (abdominal) Pregnancy with Empty Uterus.

The foetus had acrania, bilateral club feet, kyphoscoliosis and spinal dysraphism. Amniotic fluid was in excess. Placenta was located anterior just deep to anterior abdominal wall. On doppler ultrasound, there were no lacunae or increased retroplacental vascularity. Impression was of single alive extrauterine abdominal pregnancy of 19 weeks with an anomalous foetus. To further comment upon the precise location of ectopic pregnancy, MRI was planned but was unavailable. Laboratory investigations revealed a haemoglobin level of 10.8 g/dl; HCT of 33.1% while Liver Function Test, Renal Function Tests and serum electrolytes were normal.

The woman and relatives were counselled and an Exploratory laparotomy was scheduled with high-risk consent and adequate blood back up. General surgeons and haematologist were also taken on board. One unit of blood was transfused pre-operatively.

Per-operative Findings; Under general anaesthesia, exploratory laparotomy was proceeded with a subumbilical mid-line incision. Per-operative findings revealed an intact, well circumscribed (15× 15 cm) and mobile left tubal ectopic pregnancy mass, without any sign of impending rupture. The uterus, contralateral tube and both ovaries were healthy and normal. There were no peri-tubal adhesions or free fluid (Figure 3). To minimize the blood loss, the tubal pregnancy mass was excised intact with partial left salpingectomy via broad

tubal pedicles and haemostasis was secured (Figure 4). Surgery was concluded successfully with no need of further blood transfusion. In vitro, pregnancy mass was incised through its muscular wall and an anomalous foetus weighing 250gm, was delivered having acrania, limb deformity, spinal defect and ambiguous genitalia (Figure 5). Placenta was delivered completely from its anterior location in uterus, and was normal looking with a circumference of 27cm. (Figure 6)

Figure 3. Per-operative Findings; Left Tubal (ampullary) Pregnancy.

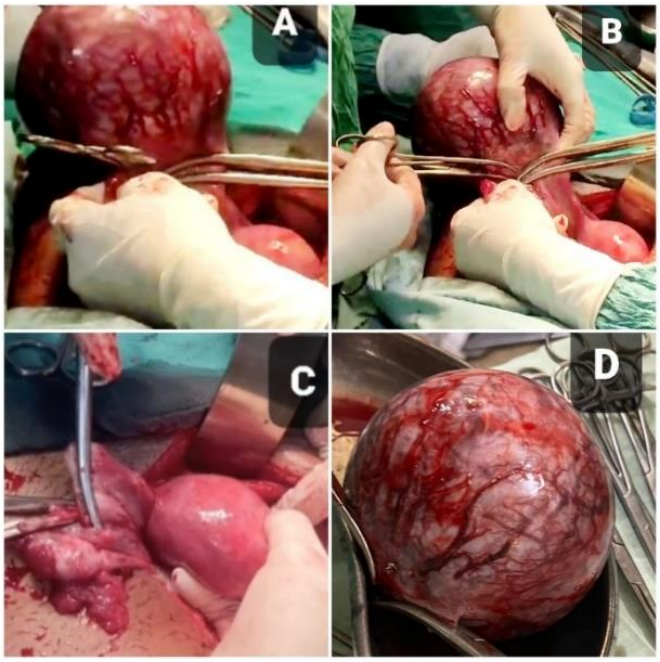

Figure 4. Excision of Intact Tubal Pregnancy Mass (A, B) via Partial Salpingectomy (C). Tubal Pregnancy Mass (D).

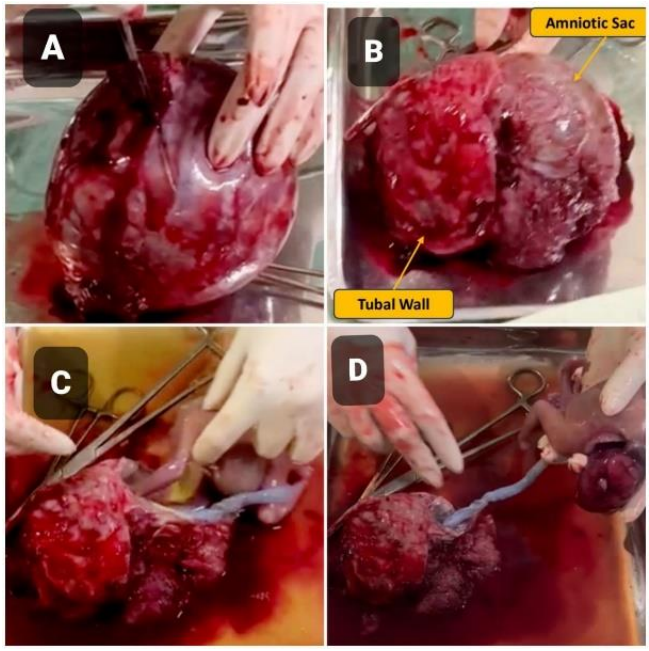

Figure 5. Tubal Pregnancy Mass incised (In-Vitro) to deliver Products of Conception.

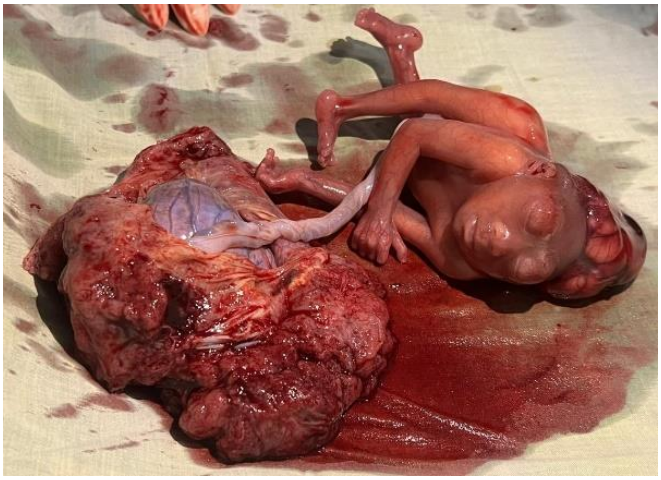

Figure 6. A 19+ weeks' gestation Anomalous Fetus, Placenta & Membranes delivered.

Follow-up; The patient had uneventful recovery and was discharged on third post-operative day.

Discussion

Ectopic pregnancy is the leading cause of direct maternal morbidity and mortality in the first trimester of pregnancy worldwide. It can undergo spontaneous resolution or a rupture; leading not only to immediate serious health consequences, but also a 10-25% risk of a subsequent ectopic pregnancy[1,6] and subfertility.[20]

Hence, the early diagnosis is imperative in reducing the brunt of morbidity, improving the success rate of subsequent management and minimizing the long term complications.[3] Unfortunately even in this modern era

an EP, can go undiagnosed or worse misdiagnosed, endangering a woman's life,[1,11] as was evident in our presented case of an alive tubal pregnancy progressing until mid second trimester, mainly due to poor antenatal awareness and care with subsequent misdiagnosis. Most of the times a tEP becomes clinically evident during early pregnancy due to its thin muscular wall, rarely it can progress undiagnosed beyond first trimester until quite an advanced gestation. Literature search reveals such rare case reports in second trimester (at the gestation of 13,[6,10] 14,[11] 15,[12] 16,[13]19,[2,14] 26,[15] and 29 weeks[16]); in third trimester (34 weeks);[17] until term (42 weeks);[18] and even beyond term; (50 weeks).[19] Though in last case an intrauterine foetal demise was diagnosed at 39 weeks but confirmation of tEP was made at 50 weeks of gestation.[19] The possible explanation for such rare situations can be; a tubal structural anomaly leading to an enhanced elasticity of the fallopian tube; and an abnormal trophoblastic invasion which does not penetrate the entire tubal wall, thereby preventing a tubal rupture.[1] In our case tEP progressed until 19 weeks, much akin to two previously reported cases.[2,14]

Although multiple risk factors have been associated with EP, but in almost 50% of the cases no risk factor is identifiable, so was in our case. Similar to few previously reported cases, our patient was a young primigravida,[1,19] who exhibited no any early clinical manifestation that could have suggested an ectopic pregnancy. Same clinical picture was seen in few other reported cases.[2,4,14,19]

The risk of tubal rupture increases with the period of time the problem remains undiagnosed and untreated.[14] Diagnosis involves a combination of clinical symptoms, beta-hCG levels, and ultrasound imaging.[6] The clinical spectrum may vary from no symptoms at all through non-specific symptoms of lower abdominal pain and vaginal bleeding (in case of intact tubal pregnancy), to hypovolemic shock (in case of a rupture). To avoid diagnostic delays and serious consequences, all women of reproductive age with such non-specific symptoms (mimicking appendicitis, ovarian cysts, or urolithiasis), should be evaluated as potential case of EP, with subsequent beta hCG levels and pelvic ultrasound.[21]

HCG level has a pivotal role in diagnosis and monitoring of early pregnancy. HCG levels >1500 - 2000 mIU/mL, in absence of intrauterine pregnancy on ultrasound, strongly favour for an ectopic pregnancy. Serial monitoring every 48 hours, with an increase by < 35% of hCG level is typical of EP.[6]

A transvaginal ultrasound, is the diagnostic imaging of choice for EP due to its availability, efficiency, non-invasiveness and is associated with high sensitivity (92.3%), specificity (75%), positive predictive value (96%), and accuracy (90%) in diagnosing suspected EP. [6] In developed world, high-resolution ultrasound has enabled detection of >80% of EP before rupture, and >50% are diagnosed while asymptomatic. However, the situation is not the same in developing countries having constrained health logistics or standard antenatal care.[6]

Notably, in an EP greater than 12 weeks of gestation; an ultrasound study alone may not be diagnostic, and requires an advanced imaging study like Magnetic Resonance Imaging (MRI) for precise localization of ectopic mass.[2] However, MRI is not readily available at all health care centres especially in developing countries, so our case was misdiagnosed as abdominal pregnancy with ultrasound assessment alone. Multiple cases of advanced tEP reported in past were also initially misdiagnosed as abdominal pregnancy and later on were confirmed as tEP, per-operatively.[1,6,13,22]

The management of tEP is scenario specific and depends largely upon status of pregnancy and maternal condition. A conservative management is better reserved for cases of EP undergoing early spontaneous resolution or a tubal abortion, provided woman remains asymptomatic, stable, hCG <1000 IU/L, and is followed up subsequently with periodic hCG levels and pelvic ultrasound.[1] A medical management with single or multiple doses of intramuscular methotrexate (a folate antagonist-DNA synthesis suppressor), is offered to asymptomatic, haemodynamically stable woman with intact EP mass ≤ 5 cm in size, no free intraperitoneal fluid on ultrasound scan, and β-hCG level ≤10,000 mIU/mL.[1,22-24]

However, in case of a ruptured EP, surgery becomes mandatory. A laparoscopic approach is best opted for hemodynamically stable patient; is associated with lesser intra-operative blood loss and analgesia requirements, with consequent shorter operative time and hospital stay. Whereas, a laparotomy should be planned in case of acute abdomen or shock due to rupture.[1] The surgical procedure could either be; A salpingectomy, involving the removal of tubal segment harbouring the EP, or; A salpingostomy, involving the

removal of ectopic pregnancy mass by incising the fallopian tube, in situ, to preserve future fertility. Current guidelines support for laparoscopic salpingectomy as the method of choice when the contralateral tube is healthy.[25]

We planned an exploratory laparotomy for our case with the primary diagnosis of an extrauterine abdominal pregnancy. However, per-operatively it turned out to be a large tubal ectopic pregnancy mass and surgery was concluded with partial salpingectomy at the expense of a healthy contralateral fallopian tube.

The importance of presenting this case is twofold. One is the rarity of such a case of an advanced tEP progressing into second trimester and then with an alive fetus, which is exceptionally rare. To the best of our knowledge, no such an advanced case of tEP has yet been reported in Pakistan. Secondly, to endorse the importance of early and correct diagnosis of this obstetric catastrophe to minimize the consequent health hazards. In developing countries like ours, multiple factors like; poor antenatal awareness and booking; the constrained health resources with inadequate diagnostic facilities; in addition to, often an atypical presentation of ectopic pregnancy; can contribute towards failure of the timely diagnosis resulting in life threatening haemorrhagic consequences.

The persistent reports of advanced tEP from all around the world, ring the alert alarm aloud that, even in this modern era, ectopic pregnancies are allowed to progress undiagnosed or, worse, misdiagnosed, endangering a woman's life.

Recommendations; In view of the grave morbidity associated with advanced tEP, there is a dire need for aggressive and effective antenatal booking awareness programmes, emphasis on improved antenatal care, an urge for medical vigilance and knowledge dissemination, as well as continuous professional development training courses of healthcare professionals to enhance their diagnostic abilities and managing the ectopic pregnancies effectively.

Conclusion

Irrespective of location, ectopic pregnancy is a life-threatening condition. The progression of an ectopic pregnancy to more advanced stage is rare and has serious consequences of subfertility, recurrence of ectopic pregnancy and even maternal death. Sonographic imaging can often be deceiving, mimicking an abdominal pregnancy. Therefore, in addition to an enhanced antenatal booking awareness, the key step in early pregnancy management is to determine the exact location and to confirm an extra-uterine pregnancy. Once diagnosed, surgical intervention must be undertaken immediately.

References

1. Ilea C, Ilie OD, Marcu OA, Stoian I, Doroftei B. The Very First Romanian Unruptured 13-Weeks Gestation Tubal Ectopic Pregnancy. Medicina (Kaunas). 2022 Aug 25;58(9):1160. https://doi.org/10.3390/medicina58091160
2. Huerta AL, Navarro JJA, Pompa AR, Uscanga OAM, Chavira MEP, Vilchis JAB. Advanced tubal ectopic pregnancy of 19 weeks gestation: case report and review of the literature. Int J Reprod Contracept Obstet Gynecol 2018;7:4794-7. https://doi.org/10.18203/2320-1770.ijrcog20184551
3. Flanagan HC, Duncan WC, Lin CJ, Spears N, Horne AW. Recent advances in the understanding of tubal ectopic pregnancy. Fac Rev. 2023 Nov 1;12:26. https://doi.org/10.12703/r/12-26
4. Fan YY, Liu YN, Mao XT, Fu Y. The Prevalence of Ectopic Gestation: A Five-Year Study of 1273 Cases. Int J Gen Med. 2021 Dec 14;14:9657-61. https://doi.org/10.2147/IJGM.S344648
5. Kim M. Unusually large, unruptured tubal ectopic pregnancy mass in a woman with bilateral tubal obstruction treated by laparoscopy: a case report. J Med Cases. 2016; 7: 396-98. https://doi.org/10.14740/jmc2613w
6. Ademi Ibishi V, Rafuna N, Kolgeci K. An Uncommon Instance of Tubal Pregnancy with a Vital Fetus at the Thirteenth Week of Gestation. J Med Cases. 2024 Jan;15(1):15-19. https://doi.org/10.14740/jmc4173
7. Restaino S. Degano M. Padovani D. Biasioli A. Capodicasa V. Vizzielli G. Driul L. A Case of Advanced Tubal Ectopic Pregnancy after Emergency Contraception. Healthcare 2022, 10; 1590. https://doi.org/10.3390/ healthcare10081590
8. Huang CC, Huang CC, Lin SY, Chang CY, Lin WC, Chung CH, Lin FH, Tsao CH, Lo CM, Chien WC. Association of pelvic inflammatory disease (PID) with ectopic pregnancy and preterm labor in Taiwan: A nationwide population-based retrospective cohort study. PLoS One. 2019 Aug 13;14(8):e0219351. https://doi.org/10.1371/journal.pone.0219351
9. Bellver J. Female underweight and risk of ectopic pregnancy. BJOG. 2021 Feb;128(3):551. https://doi.org/10.1111/1471-0528.16418
10. Gari R, Abdulgader R, Abdulqader O. A Live 13 Weeks Ruptured Ectopic Pregnancy: A Case Report. Cureus. 2020 Oct 16;12(10):e10993. https://doi.org/10.7759/cureus.10993
11. Elmoheen A, Salem W, Eltawagny M, Elmoheen R, Bashir K. The Largest Tubal Pregnancy: 14th Week. Case Rep Obstet Gynecol. 2020 May 20;2020:4728730. https://doi.org/10.1155/2020/4728730
12. Stremick JK, Couperus K, Ashworth SW. Ruptured Tubal Ectopic Pregnancy at Fifteen Weeks Gestational Age. Clin Pract Cases Emerg Med. 2019 Jan 29;3(1):62-64. https://doi.org/10.5811/cpcem.2019.1.40860
13. Ngene NC, Lunda O. Ectopic pregnancy in the ampulla of the fallopian tube at 16 gestational weeks: lessons from a case

report. Afr Health Sci. 2020 Dec;20(4):1895-1897. https://doi.org/10.4314/ahs.v20i4.47
14. Bursać D, Zadro M, Haramina S, Bošnjak DC, Tučkar N, Kuna K. Unusual presentation of late-stage ectopic tubal pregnancy (19 weeks' gestation): Case report and literature review. Eur J Obstet Gynecol Reprod Biol. 2024 Apr;295:150-152. https://doi.org/10.1016/j.ejogrb.2024.02.015
15. Nkwabong E, Tincho EF. A case of a 26-week ampullary pregnancy mimicking intrauterine fetal death. Anatolian J Obstet Gynecol. 2012;1(1). https://doi.org/10.18203/2320-1770.ijrcog20184551
16. 16. Hong LJ, Chen SS, Oshiro BT. Third trimester tubal pregnancy. A case report. J Reprod Med. 2015 Jan-Feb;60(1-2):83-6. PMID: 25745758.
17. Liu, Y., Xu, X., Liu, Q. et al. Advanced tubal pregnancy at 34 weeks with eclampsia and HELLP syndrome: a case report and literature re.BMC Pregnancy Childbirth 23, 142 (2023). https://doi.org/10.1186/s12884-023-05469-w
18. FRACHTMAN KG. Unruptured tubal term pregnancy. Am J Surg. 1953 Aug;86(2):161-8. https://doi.org/10.1016/0002-9610(53)90425-5
19. Van der Bijl P, Mabesa TP. Tubal pregnancy beyond term. S Afr Med J. 2007 Mar;97(3):184-6. PMID: 17440663.
20. Baggio S, Garzon S, Russo A, et al.: Fertility and reproductive outcome after tubal ectopic pregnancy:comparison among methotrexate, surgery and expectant management. Arch Gynecol Obstet. 2021, 303:259-268. https://doi.org/10.1007/s00404-020-05749-2
21. Hendriks E, Rosenberg R, Prine L. Ectopic Pregnancy: Diagnosis and Management. Am Fam Physician. 2020 May 15;101(10):599-606. PMID: 32412215.
22. Khalil A, Saber A, Aljohani K, Khan M. The Efficacy and Success Rate of Methotrexate in the Management of Ectopic Pregnancy. Cureus. 2022 Jul 11;14(7):e26737. https://doi.org/10.7759/cureus.26737
23. Naveed AK, Anjum MU, Hassan A, Mahmood SN. Methotrexate versus expectant management in ectopic pregnancy: a meta-analysis. Arch Gynecol Obstet. 2022 Mar;305(3):547-553. https://doi.org/10.1007/s00404-021-06236-y
24. Zhang J, Zhang Y, Gan L, Liu XY, Du SP. Predictors and clinical features of methotrexate (MTX) therapy for ectopic pregnancy. BMC Pregnancy Childbirth. 2020 Oct 29;20(1):654. https://doi.org/10.1186/s12884-020-03350-8