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Case Report

Torsion Haematosalphinx During the Second Trimester of Pregnancy - A Rare Cause of Acute Abdomen

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Abstract

A 26 year old young primigravid woman who was known to have an ovarian cyst of 12 x 8 cm during her first trimester presented to our emergency obstetric services at 26+5 weeks period of gestation as acute abdomen. She was treated with analgesics and intravenous fluids for three days and discharged as symptoms reduced. But she was readmitted again after three days of discharge with pain localised to the right iliac fossa and two episodes of vomiting. She was evaluated by USG and MRI, diagnosed provisionally as a torsion ovarian cyst and emergency laparotomy was undertaken. A 10 x 8 cm right-sided haematosalphinx has undergone torsion twice in the clockwise direction and was excised. She was treated with tocolytics, progesterones and antibiotics peri-operatively and had regular antenatal care. She was diagnosed to have mild features of IHCP and underwent induction of labour 38+5 weeks gestation and was delivered by vacuum extraction because of fetal distress.

Keywords: Pregnancy; Acute Recurrent Pain Abdomen; Haematosalphinx; Torsion

Abbreviations

POG: Period of Gestation; USG: Ultrasound; HDU: High-Dependency Unit; MRI: Magnetic Resonance Imaging; IHCP: Intrahepatic Cholestasis of Pregnancy

Introduction

Acute abdomen during pregnancy is rare and occurs 1 in 500 to 1 in 635 [1]. A delay in diagnosis and misdiagnosis can increase maternal and fetal morbidity. Difficulties are encountered in arriving at an accurate diagnosis. The most common cause of pain in the right iliac fossa during pregnancy is acute appendicitis. Torsion haematosalphinx during pregnancy is very rare and not usually diagnosed pre-operatively. If not managed timely, it can lead to rupture and haemoperitoneum and may result in mortality. The aetiology of the development of haematosalphinx during pregnancy is unclear in the literature. The reports available in the literature revealed that haematosalphinx can develop or be associated with ectopic pregnancy, endometriosis and also after torsion of the normal fallopian tube. We could find only one case report of haematosalphinx during normal pregnancy, which was managed by laparoscopic surgery at 13weeks of pregnancy [2]. This case is reported because of its rarity and also due to the difficulties experienced in diagnosis.

Case

A 26 years old young primigravid woman, at 26+5 weeks period of gestation (POG), presented with acute onset abdominal pain and vomiting with an ultrasound (USG) report showing a right adnexal cyst of 12 x 8 cm. She was diagnosed to be pregnant at 8 weeks by a transvaginal USG at which time the adnexa were reported to be normal. But at NT scan a ovarian cyst of 12x8cms was reported and she was asymptomatic since then. At 26+5 weeks, she presented with a history of sudden onset pain in the lower abdomen pain, in the right shoulder and back, followed by vomiting, pain and heaviness in the lower abdomen. She sought immediate consultation in a nearby hospital and was referred to us with USG report. On examination, her pulse rate was 95 per minute, and her blood pressure (BP) was 105/75 mmHg. Her respiratory and cardiovascular system examinations were normal. Per abdomen, the uterus was 24 weeks in size. There was tenderness in the right iliac fossa (RIF) and no guarding or rigidity. USG at admission showed a right adnexal anechoic cyst 10 x 6cm with preserved vascularity on Doppler. She was hospitalised and was treated with intravenous fluids and analgesics. Her urinary ketones were positive, and she was treated with intravenous fluids (crystalloids) with restriction of dextrose. Her pain was reduced after analgesics, and she was discharged the following day.

She was re-admitted at 27+4 weeks (within 8 days) with pain on the right side of the abdomen and two episodes of vomiting. On examination, her pulse was 86 per minute, and her blood pressure was 110/72 mmHg. Abdominal examination revealed tenderness in right iliac fossa with no guarding or rigidity. USG examination showed the uterus deviated to the left side and a right-sided multiloculated cyst of 8.4 x 7.6cm with a cyst wall thickness of 8mm. The cyst wall had poor vascularity on Doppler flow, and partial torsion was suspected. She was kept on conservative management with intravenous fluids and nil per oral, and analgesics were withheld.

The differential diagnosis was as follows: 1. Torsion/Haemorrhagic Ovarian cyst: A torsion or a bleed inside the ovarian cyst presents as acute abdomen. The most common type of ovarian cyst during pregnancy is a corpus luteal cyst. But rarely, a corpus luteal cyst persists beyond 20 weeks. Others include luteoma of pregnancy and theca lutein cyst. The possibility of a malignant ovarian cyst was ruled out as the tumour markers were normal, and USG criteria for a malignant ovarian cyst were not fulfilled. 2. Acute appendicitis and appendicular abscess: It is also a common surgical emergency presenting as an acute abdomen in the second trimester of pregnancy. The pain also recurred after the initial treatment with analgesics and antibiotics. However, she had no fever, obstipation, migratory pain, or rebound tenderness, and imaging showed no features suggestive of appendicitis or abscess. 3. Torsion sub-serosal pedunculated fibroid/red degeneration: Torsion of a subserosal pedunculated fibroid is more common in the first trimester and post-partum periods, whereas red degeneration is common in the second trimester. Both form a differential diagnosis of adnexal masses. On USG the typical whorl pattern of appearance is lacking and it was mainly anechoic. On MRI, peripheral hyperintensity signals were demonstrated in T1-weighted images and variable-intensity signals in T2-weighted images. Her first-trimester sonography did not reveal the presence of sub-serosal fibroid, so the possibility of this differential was ruled out. 4. Rudimentary horn ectopic pregnancy: An ectopic of the rudimentary horn is an extremely rare form of ectopic which ruptures late at the end of the first trimester or second trimester, depending on the capacity and the musculature of the uterine cavity. Chronic pain abdomen or acute on chronic pain abdomen can occur when rupture results. Though the availability of 3-D sonography can detect early gestation, missed cases in routine ultrasound can result in catastrophic intraperitoneal haemorrhage mimicking acute abdomen.

Her investigations are shown in Table 1.

Table 1

Parameters	Patient value	Reference range
Tumor markers		
CA-125 (U/ml)	15.6	<35
CEA (ng/ml)	0.9	<5
CA 19-9 (U/ml)	17.3	<35
Ultrasonogram (USG)		
12 weeks	Right adnexal clear cyst measured 12*8cm	
20 weeks	Right adnexal clear cyst measured 10.5*7cm	
27+4 weeks	Right-side multiloculated cyst of 8.4 x 7.6cm Cyst wall thickness of 8mm Poor cyst wall vascularity, ? partial torsion No free fluid in the pelvis	
Magnetic resonance Imaging (MRI) - pelvis		
Right adnexal 7.2*7.9cm cystic lesion with internal septations with no solid components was attached to the uterus by a pedicle. Mild hyperintensity in the T1 weighted image and proper hyperintensity was demonstrated in the T2 weighted image, which is highly suggestive of haemorrhage inside the cyst.		
Liver function tests (LFT) at 32 weeks		
Total bilirubin (mg/dL)	0.4	0.3 - 1.2
Direct bilirubin (mg/dL)	0.1	0 - 0.2
Aspartate transaminase (IU/L)	35	0 - 35
Alanine transaminase (IU/L)	52	0 - 35
Bile acid (µmol/L)	11.8	0.5 - 10

Monitoring was done in a high-dependency unit (HDU), and she was investigated further. The tumour markers were normal. Magnetic Resonance Imaging (MRI) was performed to rule out torsion and to know the nature of the cyst in detail. MRI showed a 7.2 x 7.9cm cystic lesion with internal septations in the right adnexa with no solid components, and the lesion was attached to the uterus by a pedicle. The cyst showed mild hyperintensity in the T1 image, and proper hyperintensity was demonstrated in T2 weighted images, highly suggestive of haemorrhage inside the cyst and the origin of the cyst was not made out (Figures 1 and 2). The provisional diagnosis was Ovarian cyst with internal haemorrhage on MRI.

As the pain in the abdomen increased over 48 hours of conservative management, an emergency laparotomy was undertaken with a provisional diagnosis of torsion ovarian cyst. The abdomen was opened by a vertical right paramedian incision of 8 cm given 4 cm below the fundus. A 10 x 8 cm right-sided bluish-coloured cyst was found to be torted twice in the clockwise direction (after

exteriorization). De-torsion and excision of haematosalphinx was performed (Figure 3). There was no haemoperitoneum. Bilateral ovaries and left fallopian tube were normal, with no evidence of endometriosis. The gross specimen which was cut open had 150g of blood clots. A final diagnosis of torsion haematosalphinx was made. Perioperative tocolytics and progesterones were continued for seven days. She was discharged on the 7th postoperative day with the advice to come for regular antenatal checkups.

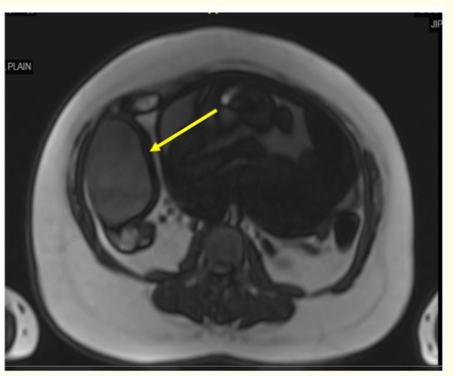

Figure 1: Shows T1-weighted MRI image of abdomen. Arrow points to the right adnexal cyst.

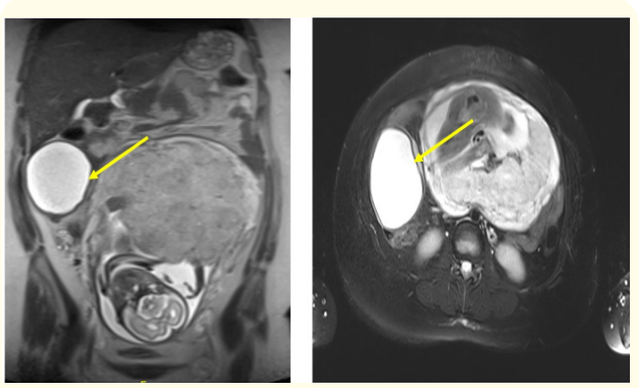

Figure 2: Shows T2-weighted MRI images of the abdomen. Arrow points to the right adnexal cyst.

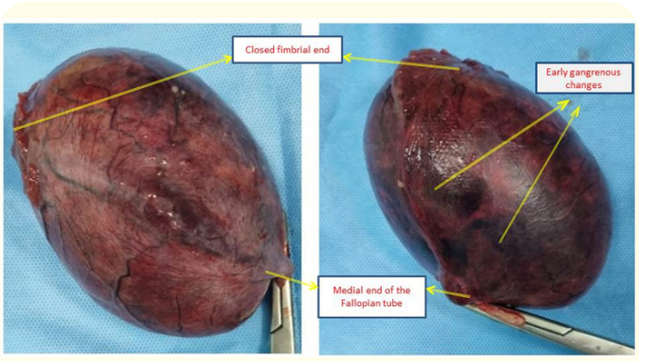

Figure 3: Shows the gross specimen of the haematosalphinx.

Outcome and follow-up

The histopathological features of the specimen were consistent with haematosalphinx with no evidence of dysplastic cells, endometriosis or malignancy. On a regular antenatal visit at 32 weeks, she was started on Ursodeoxycholic acid (UDCA) 300mg TDS as she had generalized pruritis with a borderline elevation of liver enzymes, bilirubin, and serum bile acid. There was clinical and biochemical improvement following UDCA administration. She was admitted at 38+5 weeks for safe confinement, and the labour was induced with 25µg vaginal misoprostol due to suspected intrahepatic cholestasis of pregnancy (IHCP). The labour was augmented with syntocinon, and epidural analgesia was provided for pain relief. The first stage lasted 18 hours, and the second stage lasted 1 hour. She was delivered by vacuum extraction due to a second-stage pathological cardiotocograph (CTG). The baby weighed 2.6kgand was healthy. Her postpartum period was uneventful, and she was discharged on post-natal day 3 with contraceptive counselling for copper-T insertion after six weeks.

Discussion

In any case of an acute abdomen during pregnancy, obstetric, gynaecological and non-gynaecological causes should be considered, and for accurate diagnosis and imaging plays an important role. The obstetric causes in early pregnancy include ectopic pregnancy and abortion, and gynaecological causes are torsion or haemorrhage into the ovarian cyst, red degeneration and torsion of the subserous fibroid uterus. The most common non-gynaecological and non-obstetric cause is acute appendicitis, which accounts for 25% of surgeries performed during pregnancy [3]. Zacharia and colleagues listed the causes of acute abdomen in pregnancy in four primary categories, viz., Pregnancy-related causes, non-pregnancy-related causes, conditions exacerbated by pregnancy and extra-abdominal causes. They have also illustrated how normal pregnancy can predispose to torsion of adnexa [4]. However, isolated fallopian tubal aetiology was not included in this. Haematosalphinx during pregnancy mimics ectopic pregnancy, misleading to an incorrect diagnosis in early gestation. Another possibility is that an unruptured tubal ectopic pregnancy may lead to haematosalphinx when the fimbrial ends are blocked. A case of bilateral hematosalpinx associated with an ectopic pregnancy at eight weeks of gestation was reported by Sindos M and colleagues [5]. The aetiology of haematosalphinx is ambiguous and can be attributed to long or coiled tubes, infective or endometriotic foci, ciliary motility disorders, peritubular adhesions and sometimes, rarely tubal carcinoma [6]. Other causes include fallopian tube torsion and retrograde menstruation with cervical stenosis. The most common cause of haematosalpinx during pregnancy is reported to be ectopic gestation [7]. Torsion can rarely occur in normal tubes, leading to hematosalphinx or vice versa, as internal bleeding occurs due to compromised arterial blood flow and venous congestion. Our patient had a USG showing normal adnexa at eight weeks, followed by the development of hydrosalpinx possibly (which was

misdiagnosed as an ovarian cyst) within four weeks. Haematosalphinx may sometimes be asymptomatic and can be an incidental finding in imaging. The symptoms are proportional to the volume and rate of bleeding inside the fallopian tubes. If the tension inside the tube increases due to ongoing bleeding, it can lead to tubal rupture, haemoperitoneum and haemorrhagic shock [8]. Torsion is uncommon in the late second and third trimester, especially when the size of the adnexal cyst is large. Still, in the present case, torsion of haematosalphinx occurred at around 26-27 weeks of gestation. The classical retort shape of the dilated tube in the adnexa is challenging to appreciate during the USG examination in an ongoing pregnancy, and it is difficult to differentiate it from an ovarian cyst. A dilated tube with multiple mucosal folds can be confused with a multiloculated ovarian mass. Similarly, bowel loops mimic dilated tubes and could be differentiated by gas inside the lumen and bowel peristalsis. Visualising the ovaries separately from the dilated tubes is difficult with a USG, and a higher imaging modality is required. Hydrosalpinx appears anechoic, fresh haematosalphinx are hypoechoic, and an old organised haematosalphinx appears hyperechoic with varying amplitude. The use of colour dopplers guides clinicians regarding its vascularity. Retrospectively, on assessing our case, we picked up torsion early on USG and did an MRI before haemodynamic deterioration set in. A normal fallopian tube is not seen in MRI unless surrounded by fluid. T1hyperintensity signal of the adnexal cyst in an MRI image denotes a haemorrhage inside the cyst, which is best demonstrated by fat suppression [7]. The MRI images of our patient had a T1 and T2 hyperintensity signal, which was highly suggestive of a haemorrhage inside the cyst. Hence on MRI it was interpreted as ovarian cyst with haemorrhage. USG is the initial modality of imaging. The features of Ovarian cyst torsion on USG include peripherally displaced follicles with central echogenicity, which is described as a "follicular ring sign" but not pathognomonic. A long-standing infracted ovarian cyst appears complex and echogenic due to chronic haemorrhage. "Whorl-pool sign" on Doppler and probe tendernesspoint towards torsion [9]. Oedema of the cyst wall may reflect an increased thickness of the cyst wall. In the present case, we could not demonstrate these two signs. Still, the thickness of the cyst wall was 8 mm. USG appearance tubal mass is described as a "Cogwheel sign" due to the presence of tubal folds, and the cystic appearance is usually elongated and 'c" shaped or 's' shaped, but in chronic cases it appears as septations either complete or incomplete. It is difficult to differentiate from the ovarian mass [10]. The decision for surgery during pregnancy is a difficult task, especially when the woman is not in shock, and the imaging features are equivocal, as in this case. Persistent symptoms of pain in the abdomen and vomiting and tenderness of the abdomen usually guide us in contemplating surgery. Laparoscopic management is preferred in early gestation, though it is also reported in a few cases during the second trimester. De-torsion and salphingotomy reveal the presence or absence of the ectopic gestational sac. Then, one may proceed with salpingostomy or salpingectomy based on the clinical circumstances and external morphology of the contralateral tube. An early diagnosis and prompt treatment must be made to salvage the tubes. In the caseof bilateral haematosalphinx, an aspiration rather than salpingectomy is preferred, especially when the woman is nulliparous. However, there is a higher chance of tubal block or recurrence of hematosalphinx due to the destruction of the ciliated columnar epithelium, and it requires assisted reproductive techniques for future fertility. In this case, the hydrosalpinx, which was misdiagnosed as an ovarian cyst during the first trimester, could have undergone torsion, resulting in hematosalphinx. This event could have started at her first admission at 26 weeks due to anatomical alteration of position and size of the uterus, and full-blown clinical features were present during her second admission, in which the torsion led to almost gangrenous features. We did have a dilemma about whether to aspirate under USG guidance or do a laparotomy. We chose laparotomy because of the multilocularity and suspicion of torsion and haemorrhage in the cyst.

Conclusion

Torsion haematosalphinx is one of the differential diagnoses for acute abdomen during pregnancy. Pregnant women with an adnexal mass in the first trimester should be warned about the possibility of torsion and should be appraised of the acute sypmtoms so that they can report early.

The learning points from this case are: 1. Acute abdomen during pregnancy must be evaluated immediately to arrive at an accurate diagnosis, and imaging is essential for this. 2. Timely intervention reduces the morbidity, and mortality associated with torsion and subsequent rupture 3. Good clinical acumen and vigilant monitoring is necessary to decide when to stop conservative management and undertake surgical management. It is challenging to differentiate Ovarian cyst torsion from that of torsion of haematosalphinx. A pre-operative diagnosis of haemato-salphinx may not be possible in late pregnancy. 4. Whether aspiration of adenaxal cysts of a benign nature should be undertaken to prevent torsion should be investigated in the future.

Conflict of Interest

No financial interest or any conflict of interest exists.

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