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Case Series

Corpus luteal cyst rupture presenting as massive spontaneous hemoperitoneum in women with dengue infection: A case series

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ABSTRACT

Corpus luteal cyst rupture presents with acute pain abdomen, hemoperitoneum and features of shock. The condition clinically closely mimics acute rupture ectopic pregnancy but has negative urine pregnancy test. Massive hemoperitoneum requires urgent surgical exploration and achievement of hemostasis. Here is a case series describing 3 cases of corpus luteal cyst rupture in patients with diagnosed dengue fever presenting with features of acute abdomen with or without shock at emergency. All were managed surgically, did well in postoperative period, had transfusions of blood and blood products and finally discharged in satisfactory condition. The objective of this series is to highlight the importance of this condition as a differential diagnosis in patients with acute abdomen which is not much described in medical literature.

1. Introduction

There are many causes of acute abdominal pain. Out of which, the common clinical encounter with gynaecologists is rupture ectopic pregnancy which is surgically managed. An uncommon cause of acute abdominal pain is spontaneous hemoperitoneum.[1] It is rare but life threatening, usually caused by atraumatic rupture of abdomen viscera like liver, spleen or other diseased abdominal vasculature.[2]

In luteal phase of ovarian cycle, there is formation a cystic structure called corpus luteum. It is a thin -walled structure, that synthesizes progesterone. As it is richly vascular, it is prone to haemorrhage, however bleeding is usually limited to itself. Sometimes it ruptures and cause minimal hemorrhage inside the abdominal cavity that causes acute pain but it is self limiting and is managed conservatively.[3]

Massive hemoperitoneum resulting from ruptured corpus luteal cyst may happen in cases of abdominal trauma, bleeding disorders, patients on dialysis, patients on anticoagulants and recent sexual intercourse.[4]

Dengue is a viral infectious disease that is mosquito-borne, and is primarily transmitted to humans by the female Aedes mosquito. It is caused by any one out of the four dengue virus serotypes: DENVs 1–4. Infection with dengue virus cause a wide spectrum of medical illness ranging from mild asymptomatic fever (dengue fever) to severe hemorrhagic disorder (dengue hemorrhagic fever, DHF) and stage of circulatory collapse and shock (dengue shock syndrome, DSS). DHF and DSS may turn fatal.[5]

Hemorrhagic manifestations in dengue illness are multifactorial. Predisposing factors for the same are (a) hemoconcentration, (b) increased prothrombin time, (c) elevated alanine transaminase (ALT) and (d) platelet count of less than 50,000 cells/mm^3.[6]

In literature, There are few case reports of dengue illness with hemoperitoneum secondary to spontaneous rupture of

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the spleen.[6] corpus luteal cyst rupture has been discussed commonly as radiological differential diagnosis. Only one case report is available in literature suggesting similar findings in a female with dengue infection thus leading us to a conclusion that corpus luteal cyst rupture associated with dengue infection is a rare entity.

Here is a case series of three patients of spontaneous massive hemoperitoneum with ruptured corpus luteal cyst in patients with diagnosed dengue fever. Though hemoperitoneum due to corpus luteal cyst rupture is common clinical entity, case reports or protocols for its management are still lacking.

2. Case Presentations

2.1. Case 1

A 21year old unmarried female was admitted in emergency with history of fever, vomiting and body ache for past four days. There was no history of bleeding diasthesis, no history of blunt trauma and injury over abdomen. Her last menstrual period was 2 weeks back. On examination she was pale with Pulse – 104/ min, BP – 110/70 mm Hg, Temperature – 100 F,. Abdominal examination revealed mild tenderness in left iliac fossa. Her investigations were Haemoglobin- 8.62 gm%, Total leukocyte count –15, 000 / dl, Platelet count 1,36,000/ l, BUN 28.6 mg/dl, Serum Creatinine 1.4 mg/dl Serum Bilirubin 0.11 mg/dl, SGOT – 314 U/ litre, SGPT- 208 U/ litre. True nat for COVID 19 was negative, Dengue NS1, Ig G and Ig M for Dengue was positive. Her ultrasound whole abdomen showed gross peritoneal fluid with internal echoes suggestive of hemoperitoneum. Left tube fallopian tube was edematous raising suspicion of ruptured ectopic pregnancy. However, her urine pregnancy test was negative and serum beta HCG 0.12 mIU/ml. After 24 hours, her abdominal pain worsened, she had perspiration, her vitals were PR- 124/min, BP- 90/60 mm Hg. USG guided abdominal tapping revealed dark coloured blood suggestive of hemoperitoneum. Urgent investigations at same time, hemoglobin was 4.8 mg%, platelet count was 1.6lacs, TLC 7,400/ dl, PT -47 seconds, INR 1.21.

A decision for exploratory laparotomy was taken in view of falling haemoglobin levels, unstable haemodynamic status and acute abdomen. Intraoperatively, 1.5 litre of haemoperioneum was found. Uterus was normal sized, right tube and ovary were normal, left tube was edematous and on left side about 4 X 4 cm ruptured ovarian cyst with organised clot were present. Cystectomy was done followed by securing of haemostasis. A thorough peritoneal lavage was done and pelvic drain was placed. Patient received 1 unit of PRBC

and 4 bags of random donor platelets preoperatively. 2unit PRBC and 4unit of fresh frozen plasma (FFP) were given intraoperatively.1unit PRBC was transfused postoperatively. In postoperative period patient was kept on antibiotics and analgesics. Postoperative period was uneventful and patient was discharged on day 9 after stitch removal with Haemoglobin of 10.5 gm%, Platelet count of 2.6 Lac/ mm3.

2.2. Case 2

A 35year old married female P3L3 with history of tubal ligation 3 years back was referred from a private hospital to our emergency as a case of ruptured ectopic pregnancy with severe anaemia with Dengue NS1 Positive report. She had already received 2 unit packed red blood cells and 4unit platelets (RDP).There was no history of blunt trauma, bleeding disorder or recent sexual activity. On admission her Pulse Rate was 114/ minutes, BP 90/60 mm Hg, chest on auscultation had bilateral fine crepitation. P/A gross distension with tenderness in lower abdomen. On USG guided paracentesis haemorrhagic fluid was aspirated. Urine pregnancy test was negative. Her investigations showed Haemoglobin of 3.4 gm%, TLC 2415/ dl, Platelet count 115000/microlitre BUN 30.6 mg/dl, Serum Creatinine 1.7 mg/dl, Serum Bilirubin 0.88 mg/dl, SGOT 370 U/ litre, SGPT 387 U/ litre. PT -45 seconds, INR 1.18. Her Dengue virus Ig G and Ig M antibodies were also positive. Her ultrasound showed a 6 cm anechoic left ovarian cyst with massive free fluid in peritoneal cavity. Patient was taken up for emergency laparotomy. Per operatively around 2 litres of hemoperitoneum was present. Uterus and right ovary were normal. On left side ovarian rupture was present with oozing from its margins. Haemostatic sutures were taken and ovarian tissue repaired. Peritoneal lavage done and abdomen closure was done after insertion of drain. 2 bags of PRBC and 4 bags of FFP were transfused per operatively. In post op period of patient was 2 units of PRBC was transfused and patient was kept on antibiotics and analgesics. She was discharged uneventfully on postoperative day 10 with haemoglobin of 10.3 gm% and platelet count 2.8lac/mm3.

2.3. Case 3

A 33year old married female P2L2 admitted to the emergency with complain of fever since 7 days along with new onset pain abdomen since two days. Her last menstrual period was 21 days back. There was no history of blunt trauma, bleeding disorder or recent sexual activity. Her pulse rate was 110 beats per minute, BP 90/60 mm of Hg, and temperature of 100.8 F. There was mild tenderness in left side of lower abdomen during examination. Her haemoglobin was 7.8 gm%. Dengue fever was confirmed with positive NS1 Antigen and Dengue virus Ig G and Ig M antibodies. Her urine pregnancy test was negative, serum beta HCG was 0.10 mIU/ml. Her investigations were Haemoglobin- 8.5 gm%, Total leukocyte count – 18, 000 / dl, Platelet count 1,06,000/ microlitre, BUN 25.6 mg/dl, Serum Creatinine 1.2 mg/dl Serum Bilirubin 0.16

mg/dl, SGOT – 304 U/ litre, SGPT- 220 U/ litre. True nat for COVID 19 was negative, PT -45 seconds, INR 1.18. Ultrasound revealed an anechoic 4X5 cm left ovarian cyst along with massive haemoperitoneum. On exploratory laparotomy, a 5 cm ruptured left corpus luteal cyst was found along with one litre of haemoperitonum. Cystectomy with residual ovarian preservation was done followed by a peritoneal lavage and abdominal drain was put. One bag of packed RBC was transfused intraoperatively. Her post-operative period was uneventful. She was kept on antibiotics and analgesics. She was discharged uneventfully on postoperative day 10 with haemoglobin of 10.2 gm% and platelet count 2.5lac/mm3.

Figure 1:

3. Discussion

After ovulation occurs, the remains of mature ovarian follicle get deposition of pigment lutein, get a yellowish appearance and thus named as corpus luteum. Function of corpus luteum is synthesis of hormone progesterone. It regresses to corpus albicans if pregnancy does not happen, progesterone secretion declines, support to endometrium is withdrawn and menstruation begins. When pregnancy occurs, it does not degenerate and continue to secrete progesterone to maintain pregnancy till placenta is developed by 9 to 11 weeks (luteo-placental shift).[7]

Corpus luteum is a thin walled richly vascular structure. The rate of blood flow to it exceeds any other adult organ which is needed to deliver substrates for hormone production.[7] This results in formation of haemorrhagic cyst.[8] Ongoing bleeding into the cyst especially associated with coagulopathy, patients on anticoagulants and other predisposing factors, results in its rapid growth in size and subsequent rupture leading into hemoperitoneum.

One of the common condition of admission in emergency room in female of child bearing age is acute abdomen and it needs urgent evaluation. Its common differential diagnoses are gastrointestinal & urinary tract emergencies. In case of acute lower abdomen pain, with history of amenorrhoea, vaginal bleeding and fainting attack in reproductive age group female, the first provisional diagnosis is of rupture ectopic pregnancy.[9] Other common causes of haemoperitoneum causing acute abdomen are endometriosis, ruptured hydropyosalpinx, uterine rupture, rupture of corpus luteal cyst etc.[10]

The diagnosis of ruptured corpus luteal cyst begins with clinical suspicion from history, clinical features and lab investigations. Patients usually present in luteal phase of menstrual cycle or may have delayed cycles. Rupture corpus luteal cyst in reproductive age group women leads to acute pain in abdomen and slight haemoperitoneum.[11]

Diagnosis of hemoperitoneum due to corpus luteal cyst rupture should be considered especially in patients with bleeding disorder like congenital hypofibrinogenemia, von-willebrand disease, factor V deficiency, aplastic anemia, factor XIII deficiency, sickle cell anemia. Thrombocytopenia in patients with dengue may also be a cause for the same.

In our case series, all three women presented with acute abdomen and massive haemoperitoneum with features of shock. All women were diagnosed to have Dengue infection. None of the women had any haemorrhagic manifestation like petechiae, purpura and rash. There coagulation profiles were unremarkable. However, they had thrombocytopenia which could be explained by dengue spectrum. Coagulopathies bleeding disorders and vigourous sexual activity are known causes of rupture of corpus luteal cyst. However, in our patients no such factors were present.

Patient may have no apparent clinical signs resulting in haemodynamic instability due to acute blood loss. Peritoneal irritation due to haemoperitoneum may mimick acute appendicitis. Complete blood count estimation is a must and urine pregnancy test is necessary to differentiate ruptured corpus luteal cyst from ruptured ectopic pregnancy both of which have similar clinical presentations.[12] Other features include anaemia, increasd CRP and mild leucocytosis.[13]

For any case of acute abdomen with suspected hemoperitoneum, Ultrasound is the first imaging modality due to its easy availability & high sensitivity. Ultrasound findings may show with free hypoechoic moving echoes suggestive of fluid in peritoneal cavity with focal collection of higher echogenicity (eg: clotted blood) in pelvic cavity, with a complex adnexal cyst with increased peripheral echogenicity surrounding cystic component.

Once intrauterine pregnancy is ruled out, CT can be done. Though CT is more sensitive than ultrasound, it is less specific in detecting ovarian cyst. CT scan is considered best option for imaging hemorrhagic ovarian cyst or hemoperitoneum due to cyst rupture and can easily differentiate other intrabdominal causes of acute haemorrhage.[14] On CT, corpus luteal cyst appears like a well circumscribed unilocular adnexal lesion. The cyst show a characteristic inhomogeneous contrast medium due to increased vascularity.[15]

Table 1: Comparison of details of all cases of corpus luteal cyst rupture

Features	Case 1	Case 2	Case 3
Age, marital status, GPLA	21year, unmarried, P0L0	A 35 years married P3L3	33 years, married P2L2
Presenting complaints	Pain Abdomen +shock(later)	acute pain abdomen +abdomen distension.	Fever + acute pain abdomen + abdomen distension.
Vitals (PR, BP) Features of shock	PR- 124/min, BP- 90/60 mm Hg, present	PR 114/ minutes, BP 90/60 mm Hg, present	PR -110 bpm, BP 90/60 mm of Hg, and temp of 100.8 F
Investigations; HB, TLC, Platelet count	Hb- 4.8 mg% TLC 7,400/ dl, pl/c-1,60,000-,/microlitre	Hb-3.4 gm%, TLC 2,415/dl, P/c1,15,000/microlitre	Hb- 8.5 gm%, TLc – 18, 000 / dl, P/c 1,06,000/ microlitre,
S. LFT, S.RFT, PT/ INR	BUN 28.6 mg/dl, S.Cr 1.4 mg/dl S.Bil 0.11 mg/dl, SGOT – 314 U/ litre, SGPT-208 U/ litre. PT -47 sec, INR 1.21.	BUN 30.6 mg/dl, S.Cr 1.7 mg/dl, S. Bil 0.88 mg/dl, SGOT 370 U/ litre, SGPT 387 U/ litre. PT -45 sec, INR 1.18.	BUN 25.6 mg/dl, S. Cr 1.2 mg/dl S. Bil 0.16 mg/dl, SGOT – 304 U/ litre, SGPT-220 U/ litre. PT -45 sec, INR 1.3
UPT , Beta hcg,	Negative, 0.12 mIU/ml	Negative, 0.14 mIU/ml	Negative, 0.10 mIU/ml.
Dengue IgG&IgM	Positive	Positive	Positive
Management and Intraop finding (hemoperitoneum, cyst size)	Exploratory laparotomy, 4 cm corpus luteal cyst left side, Left tube edmatous Uterus normal, Right adnexa normal 1.5 lt HP	Exploratory laparotomy, 6 cm corpus luteal cyst left side, Uterus normal, Right adnexa normal 2 lt HP	Exploratory laparotomy, a 5 cm ruptured left corpus luteal cyst, 1 ltHP Uterus normal, Right adnexa normal
Blood and blood products transfused. (total)	4 unit of PRBC + 4 units FFP+ 4units RDP.	4 unitsPRBC + 4 units FFP+ 4unit RDP.	One bag of packed RBC
Postoperative period discharge day	Uneventful, Postoperative day 9	Uneventful Postoperative Day 10	Uneventful Postoperative Day 8

Though MRI is the most accurate technique for pelvic evaluation, it is usually not used in emergency settings due to its considerably limited availability and high cost.

For corpus luteal cyst rupture, if patient is hemodynamically stable, clear cut diagnosis on ultrasound is made and haemoglobin levels over 4-6 hrs of monitoring is not much declining, conservative approach can be followed, using intravenous fluid, tranexamic acid and blood and blood products transfusion.[16] In patients presenting with haemodynamic instability, emergency laparoscopy/ laparotomy with cystectomy should be done. In our set up laparoscopy is not available in emergency hours. So, we were only left with choice of exploratory laparotomy. Out of 3 cases two had features of shock at the time of admission and the other case of unmarried girl was initially hemodynamically stable but she showed features of shock after 24 hours of conservative management. In patients with recurrent corpus luteal cyst, oral contraceptive pills are used for management.[17]

4. Conclusion

Corpus luteal cyst rupture is a rare and life threatening complication.[2] In women with Dengue fever, associated thrombocytopenia further increases the chances of spontaneous hemoperitoneum in luteal phase of menstrual cycle.[6] Diagnosis may be difficult due to multiple conditions mimicking its presentation like acute appendicitis, rupture ectopic pregnancy, acute urinary tract infection etc. Prompt diagnosis (specially using ultrasound), negative urine pregnancy test, other blood biochemical investigations and eventually surgical management are mainstay of treatment.

Literature suggests that conservative approach has also been tried to manage haemorrhagic complications but hemodynamic instability in our cases required immediate surgical management along with blood and blood products transfusion. All cases had uneventful post operative course and follow up periods.

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None.

6. Conflicts of Interest

None.

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