CORPUS LUTEAL CYST HAEMORRHAGE UNVEILING RARE PATHOLOGY AS CML

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Abstract

Rupture of an ovarian cyst is a common occurrence in women of reproductive age group. Bleeding from ruptured corpus luteum may vary from mild haemorrhage to massive haemoperitoneum. Here, a rare case of corpus luteal haemorrhage that unveiled the rare underlying pathology as Chronic myelogenous leukemia (CML) is presented.

Keywords: Myeloproliferative neoplasm, Haemoperitoneum, Haematoma, Corpus luteum

Introduction

Rupture of an ovarian cyst is a common occurrence in women of reproductive age group. Bleeding from ruptured corpus luteum may vary from mild haemorrhage to massive haemoperitoneum necessitating urgent surgical intervention. It mimics ruptured ectopic pregnancy but a negative hCG tests helps in ruling out ectopic pregnancy. Cases are reported in literature where ruptured corpus luteal cyst with significant intraperitoneal haemorrhage have been associated with chronic myelogenous leukemia (CML) or other cause of deranged coagulation.

Case Report

A-30-year-old female presented to emergency with complaints of severe pain abdomen for four days and abdominal distension for one day. For last two days patient was admitted at some private hospital where she was being managed conservatively with antibiotics and was given two blood transfusions but from there she was referred to Sri Guru Ram Das Institute of Medical Science and Research as her condition deteriorated. She was P L with all normal vaginal deliveries. Her last menstrual period (LMP) was 20 days back, normal in flow and duration. There was no history of vomiting, diarrhoea, constipation, trauma or bleeding per vaginum. On examination her general condition was poor and she had a pale blanched look and respiratory discomfort. Pulse rate (PR) was 140/min and BP was 90/60 mm of Hg, respiratory rate was 24/min. On auscultation both lungs were clear. On per abdominal (P/A) examination abdomen was distended with tenderness in all quadrants. Bowel sounds were sluggish. No mass could be palpated. Pelvic examination was not very informative except for fullness and tenderness in fornices. Laboratory investigation revealed discrete anaemia with hemoglobin of 6.5 g/dL, Total Leucocyte Count (TLC) was 21800/mm³, DLC-neutrophil 80, lymphocyte 20, Platelet count was 4 lac/mm³. Renal function test and liver function test were normal. Prothrombin Time Index (PTI) was 87%. Ultrasound showed well defined large heterogeneous hyper echoic organised haematoma measuring 11.6x11x8 cms seen in pouch of Douglas (POD) extending to left adnexa. On colour flow imaging no vascularity was seen within the haematoma. A cystic structure measuring 5.9x2.8 cm was seen adjacent to it. Left ovary was not seen separately. Right ovary was normal. Moderate amount of free fluid was seen in pelvis, perihepatic and subhepatic space. Impression was ruptured ectopic pregnancy or ruptured ovarian tumour with mild hepatosplenomegaly. Since, urine pregnancy test was negative, the provisional diagnosis of ruptured ovarian cyst with shock with septicemia was made (Figure 1& 2).

Figure 1&2. Histopathology of ruptured ovarian cyst

Blood was arranged and emergency laparotomy was done. Peroperatively about two litres of collected blood was drained from the peritoneal cavity. Ruptured left ovarian haemorrhagic cyst was seen. Left salpingo-oophrectomy was done. Abdomen was closed in layers. Patient required ventilatory support in ICU for 12 hours. She was transfused two units of packed RBCs before shifting to ward and was

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apparently on her way to recovery. About 36 hrs after surgery she complained of sudden appearance of a swelling in and around the stitch line with oozing of blood after a bout of coughing. On examination non compressible swelling was seen in middle of infraumbilical vertical midline incision with oozing of blood mixed fluid. After removing few skin stitches in ward she was shifted to OT for exploration. Per operatively rectus muscle haematoma was seen which was extending to peritoneal cavity. No active bleeding point was identified in peritoneal cavity (Figure 3).

Stumps of salpingo-oophorectomy were rechecked and were found to be intact. Abdomen was closed after putting a drain. Impaired coagulation was suspected. Haemoglobin was 7.5gm%, TLC 28000/mm³, peripheral smear examination revealed a dimorphic smear with leukocytosis and marked thrombocytosis (15, 00,000/mm³). Shift to left was noted with presence of myelocyte and metamyelocyte(both constituting 10% of all neutrophilic cells) 5%basophils 3% eosinophils. No blast cells were seen. Platelets were seen in increased number with numerous aggregate formations, many showed abnormal ‘giant platelet’ morphology, suggesting a possibility of myeloproliferative disorder. PTI was 65%, fibrinogen degradation products (FDP’s) were more than 20ug/ml. Total leucocyte count (TLC) was 22000/mm³. Subsequent haemograms showed raised TLC with platelet count more than 20 lac/mm³. Bone marrow aspiration and biopsy were done which suggested a diagnosis of essential thrombocytopenia. After second surgery patient’s clinical condition improved and she was stable but fluid in abdominal drain persisted in significant amounts and continued to be haemorrhagic for next many days. USG was again done which showed hepatosplenomegaly with organised haematoma of 7.1x2.3cm size in pelvis. It was attributed to impaired coagulation. Patient was referred to higher centres where genetic studies were done. The RT-PCR for BCR-ABL was positive and JAK2V617F mutation was negative. Ph positive (Philadelphia chromosome-positive) metaphases were 16/20. Her diagnosis was revised to CML. She was put on hydroxy urea and abdominal drain was removed approximately 25 days after second surgery. Patient remained stable afterwards.

Discussion

Chronic myelogenous leukemia, BCR-ABL1+ (CML) is a myeloproliferative disorders that originates from a pluripotent bone marrow stem cell and is associated with the BCR-ABL1 fusion gene. This genetic abnormality results from translocation of ABL1 on chromosome 9 to the region of chromosome 22. The resulting fusion gene encodes an abnormal protein with constitutively activated tyrosine kinase activity leading to abnormal bone marrow proliferation and to the clinical and morphologic manifestations of leukaemia. The natural history of untreated CML is triphasic; an initial indolent chronic phase (CP) is followed by an accelerated phase (AP), a blast phase (BP), or both.1 Thrombotic and haemorrhagic complications such as atherothrombosis, venous thrombosis and rarely gastrointestinal haemorrhage and splenic haemorrhage do occur in CML.2

Many cases of haemoperitoneum in CML because of splenic haemorrhage have been reported in literature. Ovarian cyst haemorrhage in CML had been first described by Valentik in 1951.3 A thorough review of literature reveals only one case where spontaneous haemoperitoneum in CML was due to ruptured corpus luteal cyst.4

Rupture of an ovarian cyst is a common occurrence in women of reproductive age group. Bleeding from ruptured corpus luteum may vary from mild haemorrhage to massive haemoperitoneum leading to shock necessitating urgent surgical intervention. Etiology is unknown, although abdominal trauma and congenital bleeding disorders may increase the risk. Diagnosis is by pelvic ultrasound. Treatment can be either conservative or surgical.5

Postoperative rectus sheath haematoma in our case was also due to impaired coagulation and cough. Berna et al have given CT classification of Rectus Sheath Haematoma (RSH) into three types. Type 1 are mild unilateral haematomas within muscles, Type2 are unilateral or bilateral moderate intramuscular haematomas. Type3 haematomas extend between muscle and transversalis fascia extending to peritoneum and prevesical space.6 Important risk factors that lead to RSH include female sex, older age, anticoagulation therapy, cough or other abdominal trauma.7

Conclusion

Apparently simple and a common diagnosis of ruptured corpus luteal cyst in this case has led to unveiling of a rare pathology as CML. Postoperative rectus sheath haematoma was another complication which developed during the course of treatment. Cases of ruptured corpus luteal cyst presenting with massive haemoperitoneum should be thoroughly evaluated for impaired coagulation and other systemic disorders.
Editor’s comment

Corpus luteal haemorrhage could be a possible etiology of spontaneous haemoperitoneum in patients of CML belonging to reproductive age group. However, in this case the diagnosis of CML was unveiled after surgery.

References