Massive hemoperitoneum secondary to ruptured corpus luteum cyst of pregnancy in 17-year old female with Hemoglobin SC disease

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Citation

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Abstract

We report a case of a 17 year old female with Hemoglobin SC (Hgb SC) disease and an early intrauterine pregnancy who sustained a significant hemoperitoneum secondary to a hemorrhagic corpus luteum cyst. A ruptured corpus luteum cyst presenting with significant hemoperitoneum is a rare but life threatening condition requiring immediate surgical intervention. It may occur both in the pregnant and non-pregnant state. If it occurs at an early gestational age the differential diagnosis between ectopic pregnancy and ruptured corpus luteum cyst of pregnancy can be extremely difficult.

CASE PRESENTATION

A 17 year old G1P0 female LMP beginning of March with a known positive pregnancy test and known Hgb SC disease presented to the emergency department (ED) on 9th of April complaining of severe abdominal pain which started 6 hours prior to admission. The pain was described as sharp, radiating to the back, chest and shoulders, measured 10/10 in severity and started in the umbilical area. The patient denied fever, cough, chest tightness, headache, dizziness, nausea and vomiting. The patient denied vaginal bleeding or discharge. The patient denied any such episodes in the past. Despite this, the emergency physician considered the pain to be consistent with the pain of a vaso-occlusive crisis requiring pain management with analgesics and IV hydration as per standard protocols used in the ED.

Her past medical history is significant for Hgb SC disease. The patient was regularly followed at a hematology clinic and denied any history of blood transfusions, acute chest syndrome or stroke. Her hemoglobin counts ranged between 9.6 and 11.2 g/dl. The patient was admitted to a hospital 4 years prior with abdominal pain and was diagnosed with cholelitiasis and had been asymptomatic since then. Her past medical history was thus remarkable for the lack of serious symptomatology caused by her Hgb SC disease. She had no past surgical history nor known drug allergies. Her gynecologic history was significant for regular menses since age of 13, lasting 3-5 days, occurring every 28-30 days. She

was sexually active with one partner and used condoms as a means of contraception but not regularly. She was treated for Chlamydia one year ago and denied prior obstetrical history. Patient took folic acid and multivitamins for Hgb SC disease.

On examination the patient appeared writhing on the stretcher in severe distress due to her pain. Her vital signs on examination were: blood pressure 106/55 mm Hg, pulse rate 106 beats/min, respiratory rate 18 breaths/min, and oral temperature 37.8 C. Head, eyes, ears, nose, throat, neck appeared to be normal. Skin exam revealed no lesions, masses, or erythema. On heart exam tachycardia was noted up to 106 beats/min, S1 and S2 – normal, no murmurs. Lungs were clear to auscultation bilaterally. The patient's abdomen was non distended, but significant tenderness and rebound were noted in all quadrants. On speculum exam there were no products of conception or blood in the vaginal vault. The cervical os was closed. On bimanual exam she was found to have a normal size anteverved uterus and no cervical motion tenderness. The adnexae were tender to palpation bilaterally. The rest of the physical exam was within normal limits.

Her initial laboratory results were: Complete blood count: WBC 13.5 (N: 4 - 10.5 K/uL), Hgb 8.7 (N: 12 – 15 g/dL), Hct 24.4 (N: 35-45%), Plt 267 (N: 130 – 400 K/uL). Reticulocyte count: 4.06 (N: 0.5 - 2.9%). \square -HCG – 2171

mIU/ml. Basic metabolic profile – within normal limits.

After gynecologic assessment a pelvic sonogram was performed by a radiologist. It revealed an anteverted uterus measuring 6.8x5.5x4.1 cm. No uterine masses were noted. The endometrial echo was 1.35 cm thick and no intrauterine pregnancy was identified. The right adnexa was unremarkable measuring 2.7x2.9x1.5 cm. The left adnexa measured 6.5x4.5x3 cm and included a 2.5 cm complex cystic structure with septations and a mural nodular component with blood flow within it. There was a small amount of pelvic fluid. The radiologist felt that the findings were suggestive of ectopic pregnancy in the left adnexa.

The patient was taken to the operating room with a presumptive diagnosis of a ruptured ectopic The patient underwent exploratory laparotomy via Pfannensteil skin incision. A hemoperitoneum of approximately one liter of fresh blood along with dark blood clots was appreciated. Both fallopian tubes, the right ovary and the uterus appeared normal. A left ovarian cyst resembled a corpus luteum with a small point which was bleeding. No other bleeding site was appreciated. A left ovarian cystectomy was performed and the bleeder was sutured. Excellent hemostasis was noted.

Patient was not interested in continuing pregnancy and a dilatation and curettage (D&C) was performed. The estimated blood loss was 1200 cc. The patient tolerated the procedure well. Immediate postoperative Hgb was 7.0 g/dL and Hct 20.4% and she received 2 units of packed red blood cells immediately after surgery for symptomatic anemia. Overall the postoperative course was unremarkable. She was discharged home in stable condition. The final pathology report showed corpus luteum cyst with hemorrhage. D&C tissue contained products of conception with chorionic villi.

DISCUSSION

The differential diagnosis for women with acute lower abdominal pain and intraabdominal hemorrhage includes: ectopic pregnancy, ruptured cyst, adnexal torsion. In patients with Hgb SC disease this differential may be obscured or pursued late because of the trigger reaction to consider and treat a vaso-occlusive crisis as it is so common in this group. I-HCG helps to differentiate ectopic pregnancy from other condition associated with lower abdominal pain and intraabdominal hemorrhage. Besides IIHCG serum progesterone has been studied in the evaluation of early and ectopic pregnancy. Low serum progesterone (less than 5 ng/ml) indicates either a nonviable intrauterine or ectopic

pregnancy in almost all cases. High serum progesterone level (greater than 25 ng/ml) predict a viable intrauterine pregnancy and the chance of ectopic pregnancy is reduced to 2% [1]. However 52% of women with ectopic pregnancies have progesterone levels between 10 and 20 ng/ml [1,2]. Moreover the test is not available in every institution and the sample processing may take 2-3 days. Thus, measurement of serum progesterone may be of limited utility in the evaluation of ectopic pregnancy. As with any test, it is not to be used in isolation and must be used in strong conjunction with pelvic sonography. Usually the level of I-HCG required for intrauterine gestational sac detection (the discriminatory zone) varies between 1500 mIU/ml and 2000 mIU/ml using a transvaginal transducer with 5.0 -7.0 MHz scanning frequency [3]. The sac may be seen in women with I-HCG levels as low as 800 mIU/mL, but should be identified by experienced ultrasonographer when the level is above 2000 mIU/mL. [4,5]. In one representative study 185 of 188 (98%) intrauterine pregnancies were visualized in women with I-HCG above 1500 mIU/L [5]. An early gestational sac may be difficult to distinguish from either a small collection of fluid or blood or the pseudogestational sac of an ectopic pregnancy. The detection of intrauterine gestational sac depends on experience of the sonographer/radiologist, the equipment used and concomitant intrauterine pathology such as fibroids or polyps, which may obscure the view. In the case of a hemorrhagic corpus luteum cyst, an ultrasound examination shows a heterogenic mass beside the uterus, sometimes accompanied by various amounts of intraabdominal fluid [6,7]. However due to its relative small size, low incidence and a need of experienced sonographer, hemorrhagic corpus luteun cysts is often missed on ultrasound studies [7].

The corpus luteum is formed during the luteal phase of ovarian cycle and supports the developing pregnancy. Spontaneous but limited bleeding fills the central cavity of the maturing corpus luteum with blood. Blood is absorbed and sequence of the small cystic space is formed. When hemorrhage is excessive, the corpus luteum enlarges and sometimes forms a large cystic structure. Most of the time, a ruptured corpus luteum doesn't result in significant bleeding. However blood loss may be catastrophic, necessitating surgical intervention and blood transfusion. Rupture and subsequent bleeding is often triggered by exercise, coitus, trauma or a pelvic examination [8]. According to Tang L, Cho H, Chan S. most ruptures occur between 20th and 26th day of the menstrual cycle [9].

In our case report, the <code>I-HCG</code> level on presentation was 2171 mIU/ml which is slightly above the discriminatory zone. However, the sonogram performed by radiologist did not reveal an intrauterine gestational sac and the presence of culde-sac fluid was noted. Moreover, it was highly suggestive of ectopic pregnancy in the left adnexa.

Based on the clinical presentation including severe abdominal pain, physical examination, laboratory results of **I-HCG** of 2171mIU/ml in the absence of intrauterine gestational sac on pelvic sonogram and the presence of free fluid, the diagnosis of ruptured ectopic pregnancy was made. The exploratomy laparotomy was performed versus laparoscopy due to intraabdominal hemorrhage, the presence of severe abdominal pain and emergency setting involving teenage female. The presence of hemoperitoneum is not a contraindication to laparoscopic intervention if patient is hemodynamically stable, the equipment is available and surgeon is trained to perform laparoscopy in emergent clinical settings. Three prospective randomized trials have shown definitively that laparoscopic surgery is superior to laparotomy in hemodynamically stable patients [10]. They demonstrated lower cost, less operative time, less blood loss, less analgesia required, shorter hospital stay, and shorter recovery period in the laparoscopic group. The rates of subsequent intrauterine pregnancy were 61% after laparotomy and 53% after laparoscopy. However the rates of ectopic pregnancy were higher in laparoscopic group - 14% versus 7% after laparotomy [10].

After we could not identify an ectopic pregnancy despite finding significant intraabdominal hemorrhage of 1200 cc we started to search for another source of bleeding. Retrospectively, laparotomy was the best approach in that clinical settings. We were able to thoroughly examine abdominal cavity including epigastrium to rule out other bleeding source, search for potential gestational sac in case of tubal abortion, abdominal, and ovarian pregnancy. However no other source of bleeding was identified besides

hemorrhagic ovarian cyst. The intraoperative diagnosis changed from ruptured ectopic pregnancy to possible intrauterine pregnancy with significant hemorrhage from hemorrhagic ovarian cyst. However we couldn't exclude 100% the possibility of tubal abortion with ceased bleeding. The definitive diagnostic tool available in the setting was uterine curettage. The use of curettage as diagnostic tool is limited by the potential for disruption of a viable pregnancy [11]. In our atypical and difficult clinical setting and non desirable pregnancy the team performed dilation and curettage to confirm the presence of intrauterine pregnancy. Knowing the results of curettage avoids unnecessary methotrexate treatment. Moreover methotrexate treatment was absolutely contraindicated for this patient secondary to presence of Hgb SC disease and significant anemia.

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