Case Report

An Unusual Presentation of a Corpus Luteum Rupture

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Abstract

Rheumatic heart disease (RHD) is one of the most common cardiac conditions seen in India with mitral stenosis as the most prevalent cause affecting females more than males. With the increasing number of patients undergoing mitral valve replacement (MVR) and mandatory use of anticoagulants post-MVR, the patients presenting with drug-induced coagulopathy have increased. One of the rare complications of coagulopathy-related hemorrhage may be associated with a gynecological cause with maximum risk in women of reproductive age group. This chance of hemorrhage has increased due to various events that occur in reproductive organs, namely, ovulation, menstruation, trauma due to sexual intercourse, or pregnancy-related bleeding. Such bleeding is evident as external vaginal bleeding or hemoperitoneum. Hereby, we present a rare case of a 30-year-old woman, on anticoagulant therapy for MVR who presented with congestive cardiac failure associated with massive hemoperitoneum. On ultrasound-guided paracentesis, the cause of mild-to-moderate ascites was normal ovulatory bleed evident by the bleeding from the corpus luteal cyst.

Keywords: Ascites, chronic anticoagulants, corpus luteum hemorrhage, hemoperitoneum, mitral valve replacement

INTRODUCTION

Acute pain in the pelvic region in young females of the reproductive age group is a common presentation to the emergency room. The presence of distension of the abdomen along with pain needs urgent evaluation with special investigations. [1] Wide range of differential diagnoses should be considered for assessing such conditions as a ruptured appendix, endometriosis, ruptured hemorrhagic corpus luteum, or ruptured ectopic pregnancy. Early diagnosis is necessary to preserve the reproductive systems and the life of the patient in severe cases.

Mitral stenosis (MS) is the most common rheumatic heart disease (RHD) in India affecting females more than males. A diseased heart valve is surgically removed and replaced by a prosthetic valve to correct the defect. This, along with it, brings prosthesis-related complications. With the increasing number of patients undergoing mitral valve replacement (MVR)

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and mandatory use of anticoagulants post-MVR, the patients presenting with drug-induced coagulopathy have increased. Congestive cardiac failure (CCF) is a known and common complication in these patients. Commonly faced associated complications in clinical practice include bleeding, embolic events, valve obstruction, paravalvular regurgitation, infective endocarditis, and hemolytic anemia. Anticoagulant-associated problems and embolic events are the most common complications of mechanical valves.^[2]

Anticoagulants as a cause of ruptured corpus luteum with hemoperitoneum are a rare entity. Patient on anticoagulants is at increased risk of developing bleed during ovulation and may rarely develop intraperitoneal hemorrhage. The main reason behind a bleeding vessel is that it is less likely to clot due to adequate anticoagulation. Literature suggests that the

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risk of bleeding increases when international normalized ratio (INR) ranges from 3.0 to 4.4 and the risk increases by several folds when INR values are over 4.5.^[3] It can present in varied ways, and hence its management also ranges from a conservative approach to surgical intervention and blood transfusions. Therefore, a prompt and early diagnosis based on clinical features and investigation is essential for proper management.^[4]

CASE REPORT

A 30-year-old unmarried female presented to the emergency department complaining of pain in the lower abdomen with distension associated with palpitations and breathlessness on exertion (New York Heart Association Class III) in the past 5 days. She had undergone a MVR surgery for severe MS 12 years back and was currently on tablet warfarin 2 mg once daily. The patient had a history of excessive bleeding during her menstrual cycles past 2 months with the last menstrual period 20 days back. There was no history of blood in vomiting, malena, hematochezia, rash over the body, or hematuria. The patient also had a history of cerebrovascular attack 8 years back. On examination, she was conscious and well oriented, but her general condition was not satisfactory. She looked pale, and her initial pulse rate was 110 bpm, irregularly irregular, blood pressure – 100/70 mmHg with raised JVP measuring 12 cm H₂O, not associated with pedal edema. Bilateral equal air entry was present in the lungs with no adventitious sounds heard. A metallic click with a loud P2 was heard on the cardiovascular examination. On her per abdomen examination, she had a distended abdomen with shifting dullness and tenderness in the paraumbilical region. Gynecological examination excluded any pathology in the external genital region. Her urine pregnancy test (UPT) was negative.

After combined evaluation based on history and clinical examination, a probable diagnosis of RHD with MS with Status post MVR with atrial fibrillation with severe anemia and CCF was made and treatment was started accordingly, while the laboratory reports were awaited. Laboratory workup revealed hemoglobin: 5.3 g%, mean corpuscular volume (MCV) 80 fL, total blood count: 9900/cumm, total platelet count: 1.86 lakh/cumm, prothrombin time: 55.8 s, INR: 4.46, activated partial thromboplastin time (aPTT): 57 s, total bilirubin 2.0 mg/dL, unconjugated bilirubin 1.2 mg/dL, total proteins 6.4 g/dL, albumin 3.2 g/dL, creatinine 1.9 mg/dL, urea 56 mg/dL, sodium 139 mEq/L, and potassium 5.4 mmol/L. Tumor markers such as AFP 4.14 ng/mL and CEA 0.44 ng/mL were within normal range, but CA 125 was raised to 364 u/mL. The diagnostic ascitic tap was apparent of hemorrhagic fluid which was sent for biochemical and cytological analysis. It was of transudative type with ADA: 7.222 (negative). Two-dimensional echo was suggestive of ejection fraction 60%, hugely dilated LA/RA, severe pulmonary hypertension, PASP = 65 mmHg, severe TR, and mitral valve (MV) gradient 16/8 mmHg.

Based on the radiological investigation, transabdominal ultrasound revealed a mild-to-moderate collection of fluid in the peritoneum which was highly echogenic in nature. It was suggestive of hemoperitoneum in the abdomen and the pouch of Douglas along with a complex left adnexal mass with multiple septations as shown in Figure 1. Congestive hepatomegaly showing early changes of cirrhosis and minimal right-sided pleural effusion was also visible as shown in Figure 2. All the differentials were excluded by sonography, and a ruptured corpus luteum leading to hemoperitoneum because of deranged coagulation profile was well thought out as a provisional diagnosis.

Tablet warfarin was stopped on admission and along with the restricted use of intravenous fluids to avoid volume overload, 8 units of fresh frozen plasma and 3 units of packed red cells was transfused under diuretic cover and intravenous antibiotics such as piperacillin-tazobactam 4.5 g IV TDS and levofloxacin 500 mg OD were started for the prevention of infective endocarditis for a total of 14 days during the hospital stay. The patient was monitored in a serial fashion for improvement in complete blood count and coagulation profile. Vital parameters were monitored regularly, and transabdominal ultrasound was repeated after 48 h which did not show any further increase in hemoperitoneum levels. The patient became hemodynamically stable, and there was no fall in hemoglobin levels observed during her hospital stay, and therefore, we did not opt for any surgical intervention. The patient's condition showed improvement by the 5th day of admission with a considerable reduction in pain and distension of the abdomen. Five days later, tablet warfarin 1 mg once daily was reintroduced, and the



Figure 1: Ruptured complex left-sided adnexal mass with hemoperitoneum



Figure 2: Congestive hepatomegaly (1) and right kidney (2) visible along with mild ascitic fluid with raised echogenicity (3)

dose was titrated until optimum INR levels were achieved. She was observed for the next 7 days in the hospital, and on discharge, her hemoglobin was 11.6 g%, MCV: 85.6 fL, total blood count: 5200/cumm, total platelet counts: 1.76 lakh/cumm, prothrombin time: 16.9 s, INR: 1.35, aPTT: 30 s, total bilirubin: 1.2 mg/dL, unconjugated bilirubin: 0.7 mg/dL, total proteins: 5.3 g/dL, albumin: 2.5 g/dL, creatinine: 0.8 mg/dL, urea: 34 mg/dL, sodium: 139 mEq/L, and potassium: 4.3 mmol/L. On discharge, injection Depot medroxyprogesterone acetate (DMPA) of 150 mg was given as a stat dose. Follow-up in gynecology OPD was advised on the 3rd day of her next menstrual cycle for the consumption of GnRH analogs to avoid such catastrophic events in future. In case if she plans for parenthood after her marriage, spontaneous conception with regular hormonal levels can be achieved. She was explained that if she plans pregnancy in future, then she would be required to stop tablet warfarin and would be switched over to low-molecular weight heparin from the beginning of pregnancy till the end of 12 weeks.

Discussion

Patients with a clotting factor deficiency or on long-term oral anticoagulants can have episodes of minor or major bleeding. The most frequent manifestation is bleeding in the urinary tract. Other manifestations include gastrointestinal bleeding, gum bleeding, epistaxis, ecchymosis, menorrhagia, and postmenopausal bleeding, intracranial hemorrhage, and ovarian hemorrhage. Uncommon sites of bleeding are retroperitoneal bleeding, rectus sheath hematoma, bilateral adnexal hemorrhages, and intramural bowel hematoma. Most of the cases of bleeding from a ruptured corpus luteum are self-limiting and goes unnoticed with a spontaneous cure. [5] Only a few cases present as massive hemoperitoneum presenting as an acute abdomen, or even in shock. In 1957,

Weseley was the first one to report a case of hemoperitoneum secondary to anticoagulants.

Follicular cysts and corpus luteum cysts are the two types of functional ovarian cysts. The hemorrhage characteristically occurs in the second half of the menstrual cycle (day 16–29), subsequent to the vascularization phase that follows ovulation. Corpus luteum is highly vascular in nature as it helps to deliver substrates, for hormone production and helps to nurture the rapidly proliferating luteal cells. This nature of corpus luteum may increase the blood collection within, resulting in distension and hematoma formation. These hematomas undergo partial or complete resorption, resulting in the formation of a cyst. There occurs a rise in intraluminal pressure which leads to spontaneous rupture and intra-abdominal hemorrhage if bleeding continues. [6]

Corpus luteum rupture presenting as hemoperitoneum is a common entity among women of reproductive age group. Such patients present in the emergency room with an acute abdomen along with lower abdomen tenderness, nausea, vomiting, bleeding per vagina, and uncommonly with syncope. It mimics acute salpingitis, acute appendicitis, and ruptured ectopic pregnancy. A UPT is compulsory in such cases and is usually negative for corpus luteal hemorrhage. Such life-threatening conditions should be promptly recognized and treated to rescue the fertility of women, especially of the reproductive age group. Furthermore, among the various imaging modalities, ultrasonography is a primarily done evaluation to confirm the presence or absence of intraperitoneal fluid. However, sometimes, it is not possible to identify the mystical bleeding source and may require a computed tomography scan to diagnose the same. Surgical intervention is indicated only when the patient is hemodynamically unstable or the hemoperitoneum is very large.^[7]

In 1937, Israel was the first one who had recommended a conservative approach in the treatment of corpus luteum rupture. Later, Lurie *et al.*^[8] also reported a case of massive hemoperitoneum of ovarian origin managed conservatively. This patient was on anticoagulant therapy with a prosthetic MV *in situ*. Payne *et al.*^[9] reported three patients presenting with hemoperitoneum in association with factor VII deficiency, factor X deficiency, and sitosterolemia. Conservative management with blood product and factor concentrates support was successful in avoiding surgery in three of the five episodes of bleeding. These studies concluded that a conservative approach can not only prevent high-risk surgical interventions but also can preserve ovarian function.

Suppression of ovulation is recommended to prevent recurrent hemorrhagic corpus luteum, especially when life-long anticoagulants are mandatory. According to the WHO, long-term anovulatory therapy with the use of progestational agents, low-dose combined oral pills, or gonadotropin-releasing hormone analogs can be recommended to patients with congenital and iatrogenic clotting disorders to avoid potential ovarian hemorrhage as they are safe and do not increase the risk of thromboembolism. Injection DMPA is an effective contraceptive agent to prevent recurrent corpus luteum hemorrhage as it has a minimal effect on most of the coagulation parameters. Combined hormonal pills can also be used to prevent ovulation but are not recommended with patients on anticoagulants.^[10]

Since the patient was not desirous of childbearing in near future, she was counseled for ovulation suppression therapy to avoid such catastrophic complications in future. Levonorgestrel intrauterine devices and injectable contraceptives are safe and effective options with noncontraceptive benefits. She was given injection DMPA of 150 mg on discharge and was put on GnRH analogs for the next few cycles for ovulation suppression. Meticulous surveillance should be provided during follow-up including close monitoring of anticoagulation intensity, lipid profile, measurement of systolic and diastolic blood pressures, and weight along with general physical and gynecological examination.

CONCLUSION

The complication of a ruptured corpus luteum cyst ranges from a simple corpus luteum hematoma to a life-threatening massive intraperitoneal hemorrhage. Patients who are on anticoagulant therapy in the reproductive age group are at risk of having bleeding related to reproductive tract organs mainly ovarian and menstrual bleeding because of their dynamic functions. The ovulatory bleed may be asymptomatic as it is self-limiting and painless in healthy women and hence may go unnoticed with a spontaneous cure. Ovarian function suppression should be discussed and prescribed to women in the reproductive age group who are not desirous of pregnancy and are on anticoagulant therapy along with uncontrolled bleeding. Strict monitoring of coagulation profile and ovarian suppression of these women can avoid such near-catastrophic events in these cases. Low diastolic blood pressure and a massive hemoperitoneum suggest the need for immediate surgical intervention. In patients on anticoagulant therapy presenting with spontaneous hemoperitoneum, there is always a dilemma of the management to be given to such patients, and so it is important to manage conservatively rather give surgical stress to the body. Such patients can be managed with the support of higher antibiotics along with multiple transfusions of blood and blood products and surgery may be avoided as surgical intervention in such cases can itself pose a risk of death due to bleeding tendencies.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that name and initial will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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