



ORIGINAL RESEARCH PAPER

General Surgery

SPONTANEOUS HEMOPERITONEUM IN A CASE OF BLEEDING DIATHESIS. CHANGING TRENDS.

KEY WORDS: Factor X Deficiency, von willebrand disease, Spontaneous Hemoperitoneum.

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ABSTRACT

Background: In woman with Coagulation disorder, spontaneous massive hemoperitoneum is a lethal Complication. Woman with Factor X (Stuart Prower) factor deficiency and von Willebrand disease are particularly prone for massive hemoperitoneum, which most of the times will be serious and life threatening, if not managed appropriately. **Description:** We report on a 25 year unmarried woman, with Factor X deficiency who presented to us with a massive Spontaneous Hemoperitoneum. This patient in the past (2016)have undergone an Exploratory Laparotomy , for similar presentation, in our Institution by a team of Surgeons & Gynaecologist with ligation of bleeding region in the left Ovary. **Conclusion:** Multimodality approach (as in Oncology) will be the Management Protocol in the future for this life threatening condition, with the Surgical faculty incorporating the developing specialities such as Intervention Radiology and Applied Hematology.

INTRODUCTION

Most of the documented Spontaneous Intra abdominal bleeding in woman of Reproductive age group, occurs during a a normal Ovulation and so this complications can be expected any time during her reproductive period. Von Willebrand disease (VWD) is the most common inherited bleeding condition that involves extended or excessive bleeding, caused by the deficiency or defect of von Willebrand factor (VWF). Hematoperitonerm as a complication of gynecologic diseases represents an acute condition which is usually caused by the hemorrhagic corpus luteum or a rupture of either ectopic pregnancy or a hemorrhagic ovarian cyst (1). Corpora lutea are cystic with gradual resorption of a limited amount of haemorrhage. Intrafollicular bleeding does not occur during ovulation. Rather, it typically occurs 2- 4 days later during the stage of vascularisation when the thin walled capillaries invade the granulosa cells from the theca interna. Spontaneous limited bleeding fills the central cavity of the maturing corpus luteum with blood. If haemorrhage is brisk, the intracystic pressure increases, and rupture of the corpus luteum is possible (2). Corpus luteum cysts may cause intraperitoneal bleeding after coitus, trauma, exercise, or pelvic examination (3).

Hemoperitoneum is a Surgical complication and so usually present to the General Surgeons and most of the times the patient will have to undergo a Exploratory Laparotomy in the presence of a Gynaecologist and may even end up in Oophorectomy and prolonged post op care and sub-fertility.

In recent years, management has become multimodal with involvement of General Surgeon, Gynaecologist, Intervention Radiologist, Haematologist and Anaesthetist towards a more minimally invasive and Conservative line of management thereby avoiding Laparotomy, Oophorectomy and its complication.

CASE REPORT

We report on a 25 year unmarried woman, with history of Von willebrands and Factor X deficiency who presented to us with a massive Spontaneous Hemoperitoneum. This patient in the past (2016)have undergone an Exploratory Laparotomy , for similar presentation, in our Institution by a team of Surgeons & Gynaecologist with ligation of bleeding region in the left Ovary.



Fig 1 : Patient At Presentation

On Admission her white cell count was 13.1x 1000/ micro-litre, Haemoglobin 5.8 g/dl, Haematocrit of 14.4%. Her Platelets were 216 x 1000. Her pH was 7.37, pCo2 39.4, pO2 91.3 and chCO3 19.3 mol/L. Her Renal Function and Liver Function test was essentially normal. Her Sodium was 13.1 and Potassium 4.0 men/ L

CECT Abdomen was done which showed multiple soft tissue density lesions noted along the Fundus of Uterus , largest measuring 3.8 x 3.6 cms. Also evidence of free fluid noted in the Hepatorenal pouch and Paracolic Gutter and Pelvis with + 35 Hounsfield unit. Impression was given as Haemoperitoneum with multiple Uterine Fibroids.

She was immediately admitted in Surgical Intensive care unit with 24 hour monitoring with Acute Abdomen Chart with a view to undertake a Laparotomy if her condition deteriorated.

During the intensive care management, she was seen by other specialists including Haematologist, Intervention Radiologist , Medical Gastroenterologist, Gynaecologist and Physician. With the advent of expert biochemical techniques to evaluate the exact level of each clotting factors, the availability of specific blood products, in our case

Cryoprecipitate, FFP, Tranexamic acid and FEIBA (Anti-Inhibitor Coagulant Complex) and minimally invasive Interventional Radiology had contributed to the expert management.

It was decided to take her to Digital Subtraction Angiogram (DSA), which is a fluoroscopic technique with a view to Embolization or Coiling in case of any continuing Intraperitoneal bleed. Patient after Anaesthetic fitness underwent intervention procedure with a view to embolize her Uterine (R) artery. But no bleeding or oozing was detected in any of the Aortic branches.



Fig 2: Patient at the Intervention Radiology Theatres

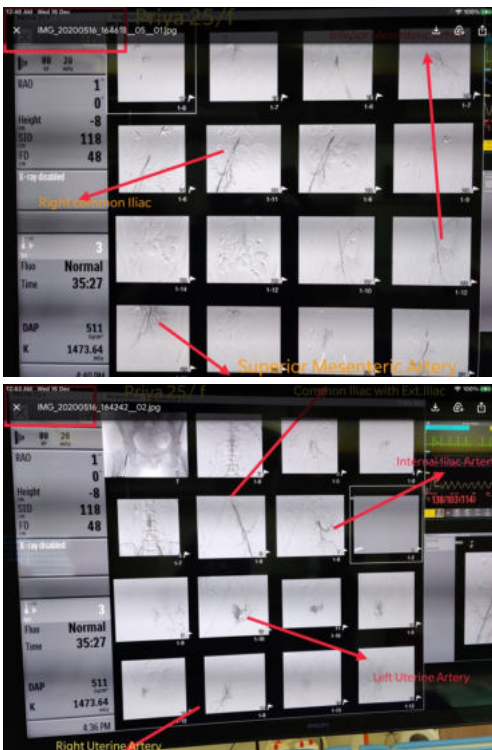


Fig 3&4 : Aortic Angiogram images (for planning of Embolization / Coiling of Bleeding vessels)

As there was no Active Bleeding in any of the Aortic Branches including the Iliac , Ovarian and Uterine Arteries , patient was managed conservatively.

She was monitored in Intensive Care Unit with 24 hours monitoring of Abdominal Girth, Pulse Rate, SpO2, Blood Pressure on hourly Basis, with Haemoglobin and ABG on 6th hourly basis.

As Per the Hematologist instructions apart from Packed Red Cells, regular Cryoprecipitate and Fresh Frozen Plasma she was given FEIBA (Anti-Inhibitor Coagulation Complex) with a dosage of 100 units/ kg every 12th hour for 3 days.

The Hemoperitoneum was allowed to resolve spontaneously with USG quantification. The patient was able to completely resolve her Hemoperitoneum from a quantity of 2000 ml to less than 200 ml in 10 days time and was discharged to be followed up with her routine Haematology Team.

DISCUSSION

Spontaneous Haemoperitoneum is a lethal but expected complication in patients with coagulation disorder. This is more so, in woman in reproductive age group. In women, heavy menstrual bleeding (HMB), recurrent ovulation bleeding with haemoperitoneum and bleeding complications in pregnancy such as retroplacental haematoma and postpartum haemorrhage have been reported (4). Since, spontaneous internal bleeding can start even during normal ovulation, more studies are needed to prevent the complication in such woman.

Most of the time, the woman presents with an Acute abdomen, with abdominal distension and severe anaemia and hypotension The follicle ruptures at the time of ovulation and fills with blood, forming a corpus hemorrhagicum. Minor bleeding from the follicle into the abdominal cavity may cause peritoneal irritation and, when it occurs in a patient with a defect of primary hemostasis, hemoperitoneum can occur. Von Willebrand disease and afibrinogenemia are two important bleeding disorders in which both primary hemostasis and coagulation are involved (5).

As a life saving measure the patient will be subjected to emergency Laparotomy. In recent years, such situations are managed conservatively with strict monitoring and multimodal management. In our particular patient, six years before she went underwent an emergency Laparotomy for a similar presentation. Needless to say, such woman if not for recent advances, will end up with multiple laparotomy, not only for repeated Hemoperitoneum, but in addition to her Obstetric surgery like Emergency Caesarean sections.

Regarding prevention, recurrent bleeding episodes following ovulation could be prevented by suppression of ovulation using oral contraceptive pills In women of child-bearing age, we are faced with a big challenge when the patient stops oral contraceptives to become pregnant. An infusion of factor VIII concentrate in severe von Willebrand's disease has been suggested in the past [3] as a prophylactic approach but there are no specific guidelines yet for mild von Willebrand's disease. An ultrasound follow-up with the possibility of using DDAVP in case of detecting a bleeding follicle could be an alternative (6). Desmopressin (also known as DDAVP, which stands for (1-deamino-8-D-arginine vasopressin) is a synthetic medicine that boosts levels of factor VIII (FVIII) and von Willebrand factor (VWF) to prevent or control bleeding.

By our case discussion, we will like to involve a multidisciplinary approach in tertiary centres with involvement of Physicians, Haematologist and Intervention Radiologists to try the conservative option instead of a 'Surgical only' option for this recurring Spontaneous Hemoperitoneum in patients with Bleeding Diathesis. We not only managed her conservatively and avoided an unnecessary Emergency Laparotomy, but also shown the medical fraternity that a multidisciplinary approach is best for these patients rather than a 'Surgery only' approach.

CONCLUSION:

A multimodality approach (as in Oncology) will be the Management Protocol in the future for this life threatening condition of Spontaneous Hemoperitoneum in woman with Bleeding Diathesis , with the Surgical faculty incorporating the ever developing biochemistry, applied hematology and minimally invasive procedures including intervention radiology in his/her armament .

REFERENCES

- (1) Conservative management of Hemoperitoneum in von wilebrands disease: Terzic M, Likic I, Pilic I, Bila J, Knezevic N. Conservative management of massive hematoperitoneum caused by ovulation in a patient with severe form of von Willebrand disease--a case report. *Clin Exp Obstet Gynecol.* 2012;39(4):537-40. PMID:23444764.
- (2) Droegmueller W. Benign gynecologic lesions. In: Stenchever MA, Droegmueller W, Herbst AL, Mishell DR, eds. *Comprehensive gynecology.* 4th ed. St. Louis, MO: Mosby, 2001:507-11.
- (3) Von wilebrands disease presenting as recurrent corpus haemorrhagicum. Jarvis RR, Olsen ME. Type I von Willebrand's disease presenting as recurrent corpus hemorrhagicum. *Obstet Gynecol.* 2002 May;99(5 Pt 2):887-8. doi: 10.1016/s0029-7844(01)01627-1. PMID:11975946.
- (4) Congenital Factor X Deficiency in Woman: Spiliopoulos D, Kadir RA. Congenital Factor X deficiency in women: A systematic review of the literature. *Haemophilia.* 2019 Mar;25(2):195-204. doi: 10.1111/hae.13729. PMID:30901144.
- (5) Prevention of Hemoperitoneum during Ovulation: Bottini E, Pareti FI, Mari D, Mannucci PM, Muggiasca ML, Conti M. Prevention of hemoperitoneum during ovulation by oral contraceptives in women with type III von Willebrand disease and afibrinogenemia. Case reports. *Haematologica.* 1991 Sep-Oct;76(5):431-3. PMID:1806451.
- (6) Recurrent Hemoperitoneum in Von wilebrands disease: Meschengieser SS, Alberto MF, Salviu J, Bermejo E, Lazzari MA. Recurrent haemoperitoneum in a mild von Willebrand's disease combined with a storage pool deficit. *Blood Coagul Fibrinolysis.* 2001 Apr;12(3):207-9. doi: 10.1097/00001721-200104000-00007. PMID:11414635.