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Case Report

Massive Intra-Abdominal Bleeding from A Haemorrhagic Corpus Luteal Cyst During an Immune Thrombocytopenic Purpura Relapse

Adewale Adeyemo[1*], Ryan Levi Seah[3], Deepak Kumar[2], Beena Subba[1]

[1]Department of Obstetrics and Gynecology, North Middlesex University Hospital, UK

[2]Department of Anaesthesia, North Middlesex University Hospital, UK

[3]Department of Medical Sciences, University College London, UK

[*]**Corresponding author:** Adewale Adeyemo, Department of Obstetrics and Gynecology, North Middlesex University Hospital, NHS Trust Sterling Way, London, UK. Tel: +44-02088872826; Email: a.adeyemo@nhs.net

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Abstract

Haem peritoneum after ovulation during an ITP flare is a rare occurrence, and in the setting of severe thrombocytopenia, can be a life-threatening event. Our patient, a 25-year-old nulligravida with a background of ITP presented with spontaneous gum bleeding and a widespread petechial rash preceded by a flu-like illness, followed by worsening abdominal pain and dizzy spells during admission. Urine beta-hCG was negative, haemoglobin was 54 g/L with a platelet count of 2x109 /L. Abdominopelvic ultrasound scan revealed free fluid within the peritoneal cavity. She was treated with intravenous fluids, immunoglobulins and was taken to theatre for diagnostic laparoscopy, at which an ovarian cystectomy was performed after the discovery of a haemorrhagic cyst. Histological examination of this specimen confirmed a corpus luteal cyst. This case discusses the need for surgical intervention in massive haemoperitoneum and highlights the difficulty in diagnosing an intra-abdominal bleed on a background of a bleeding diathesis and ruptured ovarian cyst.

Introduction

Immune Thrombocytopenia Purpura (ITP) is a haematological disease characterised by autoimmune-mediated platelet destruction and reduced platelet production. Typically, this disease manifests either acutely or in the chronic form. ITP usually follows a relatively benign course and remains largely asymptomatic, with symptoms occurring at very low platelet count levels. Haemorrhagic ovarian cysts are not uncommon but rupture leading to spontaneous severe hemoperitoneum is rare, and in the setting of severe thrombocytopenia, can be a life-threatening event. We describe the case of a young Asian woman known to have an acute relapse of ITP, with subsequent development of a massive haemoperitoneum as a result of a coincidental corpus luteal rupture after ovulation.

Case Report

A 25-year-old Asian female postgraduate student with a background history of ITP presented to the Accident & Emergency department of her local district UK hospital with complaints of spontaneous gum bleeding and a petechial rash over the arms and chest. She had been diagnosed with ITP in her home country 2 years ago and was treated with steroids (weaned off over the course of a year) and platelet transfusion. She gave a history of flu-like illness precipitating a lower respiratory tract infection for which she had self-medicated with a penicillin-based antibiotic a few weeks preceding her presentation. Her last menstrual period was 3 weeks before the presentation and a urine pregnancy test was negative. At presentation, she was haemodynamically stable, and her blood tests revealed a haemoglobin level of 126 g/L, platelet count of 2×10^9/L, neutrophil count of 1.95×10^9/L and a normal clotting profile. She was admitted under the haematologists and commenced on high dose oral prednisolone with proton pump inhibitor gastric protection cover.

On the second day post-admission, she started to complain of gradually worsening generalised abdominal pains, dizzy spells and had a fainting episode. She was noted to be persistently hypotensive despite adequate intravenous fluids and haemoglobin levels revealed severe anaemia with a significant drop to 54 g/L.

Urgent radiological imaging with a trans-abdominopelvic scan confirmed significant intra-abdominal fluid collection suggestive of haem peritoneum and an 11 mm adnexal cyst (Figure 1).

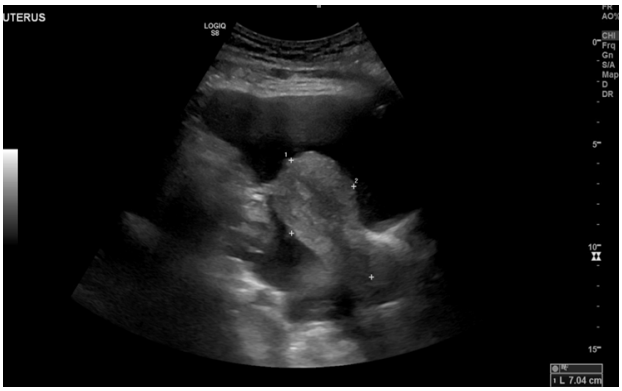

Figure 1: An ultrasound of the pelvis showed a massive collection of fluid within the pelvis. The deepest pool measured about 11.6 cm in AP diameter and multiple echogenicities was visualised within the fluid.

She was immediately commenced on Intravenous Immunoglobulin (IVIg) along with platelet transfusion and other blood products whilst preparation for surgery was initiated. Platelet count improved to 110 x 10^9/L and the fibrinogen level was 2.64. After successful resuscitation and hemodynamic stability attained, a diagnostic laparoscopy was undertaken by the gynaecologists with general surgeons in attendance. The intra-operative finding was 5 litres of intra-abdominal blood and clots which was evacuated, and a bleeding 2 cm left ovarian cyst. An ovarian cystectomy was performed, and histological examination of this specimen confirmed a corpus luteal cyst (Figure 2). A source of upper abdominal bleed was excluded by systematic examination undertaken by the surgeons. She made a good post-operative recovery on the intensive care unit and was discharged home on day 7 post-admission with 40mg prednisolone and outpatient haematology follow-up.

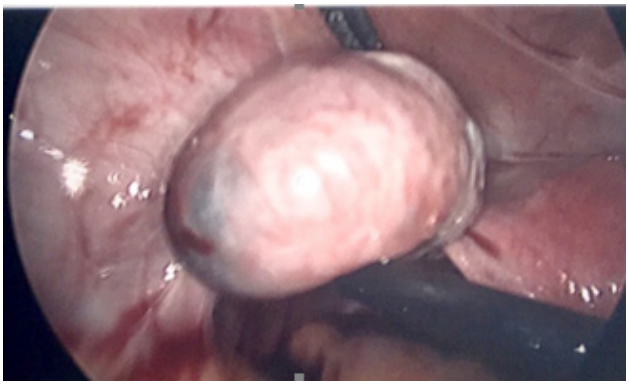

Figure 2: A small area of bleed over a haemorrhagic corpus luteal cyst was discovered on the left ovary. A left ovarian cystectomy was performed.

Outpatient immunomodulation consisted of mycophenolate mofetil, azathioprine and prednisolone which was weaned off successfully. Azathioprine as a stand-alone medication was successful in maintaining remission long-term. She was made aware that azathioprine could be continued in a future pregnancy with rescue steroids (prednisolone) used in case of any ITP flare-up.

Discussion

The aetiology of ITP is well characterised in the medical literature and can be classified according to primary and secondary causes. Primary ITP is a diagnosis of exclusion. While the pathogenesis in some cases remains idiopathic, it is largely accepted to be an immune-mediated process. Secondary causes include a preceding viral infection, medications (e.g. NSAIDs, penicillin, quinine) and malignant disorders such as leukaemia [1]. In ITP, an increased risk of bleeding from trauma happens when platelet levels fall below 50 × 10^9/L, with an increased risk of spontaneous bleeding at levels below 20 × 10^9/L [2]. It commonly presents with mucocutaneous manifestations, such as bleeding from mucosal membranes and a petechial rash. Other symptoms may include epistaxis and menorrhagia, with more severe features involving gastrointestinal bleeding or intracerebral haemorrhage. In a woman of childbearing age with progressing hypotension and syncopal episodes, important differentials to rule out would include rupture of an ectopic pregnancy or a haemorrhagic ovarian cyst. In our case, a urine pregnancy test was negative. Therefore, a high index of suspicion was the likelihood of bleeding from a ruptured ovarian cyst, which could only be confirmed with diagnostic laparoscopy.

Case reports of spontaneous haemoperitoneum secondary to ruptured ovarian cysts have been well documented [3]. During an ovulation cycle, the ovum develops inside the follicle and is released from the ovarian surface and into the peritoneal cavity. In the presence of a bleeding diathesis such as ITP, this normal physiological process of ovulation could be complicated by bleeding from the ruptured follicle and increase the risk of spontaneous haemoperitoneum. This period corresponds to the luteal phase of the menstrual cycle, the corpus luteum develops as a functional cyst and recedes if pregnancy does not occur. The thin-walled structure and highly vascular nature of the corpus luteum renders it more prone to haemorrhage [4]. While bleeding may be contained within the cyst, there is a possibility that it may spread into the peritoneal cavity. This occurrence was described by Hallatt et al, who elaborated the first significant series of patients with corpus luteum haemorrhage and haemoperitoneum. They recognised the possibility of this event at any stage of the reproductive life and noted varied volumes of haemoperitoneum at the time of exploration [5].

Corpus luteum haemorrhage may be a cause of spontaneous hemoperitoneum in patients with bleeding disorders, and has

been described in women with aplastic anaemia, Immune Thrombocytopenic Purpura (ITP), hemophilia or hemophilia carrier status, afibrinogenemia, von Willebrand disease, and factor X, VII, V, II, and XIII deficiencies [6-8]. However, instances of women experiencing massive haemorrhage due to ovulation on a background of ITP is extremely rare. There has been a case report of a newly diagnosed woman with systemic lupus erythematosus and autoimmune thrombocytopaenia who experienced intra-abdominal bleeding. In contrast to our case, she was successfully managed medically and did not require surgical intervention [9]. Typically, a conservative approach is the most appropriate strategy to adopt in patients with a ruptured ovarian cyst and haemoperitoneum. This is the case in most women who do not have any bleeding diathesis. Frequent monitoring of vital signs, haematocrit levels and reassessing the patient for signs of active bleeding by repeat imaging are suggested as the mainstay of management. Indications for surgical intervention include unstable observations, decrease in haemoglobin levels or increasing levels of haemoperitoneum detected on repeat imaging, or persistent abdominal pain that cannot be managed with analgesia [10].

In the case of our patient, she was symptomatic, hypotensive with a low haemoglobin level and radiology confirmed the presence of a large amount of haemoperitoneum, which prompted surgical intervention after correction of the platelet count and clotting factors. The realisation that our patient was having an intra-abdominal bleed was slightly delayed as this event occurred during the admission whilst under a non-surgical team. Additionally, the rare occurrence of ITP in combination with a ruptured corpus luteum may have led to diagnostic bias against the possibility of an intra-peritoneal bleed. Haemoglobin and haemodynamic status were within normal limits at presentation with dizziness and sudden syncopal episodes occurring much later on. While sonographic findings can be useful in identifying the presence of an intra-abdominal bleed, there may be limitations in identifying the underlying cause. Hence, there is a need to consider the exploration of the upper abdomen for other potential sources, such as the splenic area. In conclusion, we have described a rare case of spontaneous haemoperitoneum secondary to ovulation on a background of acute relapse. A complex case such as this highlights the need for a multidisciplinary approach to management involving the local GPs and hospital specialists.

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