

Conservative Management of Massive Intraperitoneal Hemorrhage from Corpus Luteum Cyst in a Patient on Anticoagulant Therapy Post Fontan Procedure

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Abstract. The management of massive hemorrhage from corpus luteum cysts in patients on anticoagulant therapy was practically always surgical with significant morbidity. A case of a massive intraperitoneal hemorrhage from a ruptured corpus luteum cyst that was satisfactorily managed conservatively is reported. A 15-year old girl was diagnosed as having intraperitoneal hemorrhage. She was on anticoagulant therapy for atrial fibrillation that developed after having a Fontan procedure. The decision was made for surgical intervention. During the preoperative assessment the patient became hemodynamically stable, and thus, it was decided to continue conservative management. This is the first case report where a conservative management approach was utilized successfully in a patient who had a Fontan operation. This case illustrates that conservative management may represent a reasonable option in patients who are hemodynamically stable.

Keywords: Ruptured corpus luteum cyst, Anticoagulant therapy, Fontan procedure.

Introduction

A woman receiving an anti-coagulant is at risk of developing hemorrhage at the time of ovulation, and may develop intraperitoneal hemorrhage since a bleeding vessel will be much less likely to clot with adequate

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Accepted for publication: 18 June 2011. Received: 18 March 2011.

anticoagulation. A slight but significant increase in the risk of hemorrhage was recorded for INR of 3.0-4.4, and the risk of bleeding becomes much higher for values over 4.5^[1]. In large prospective series, long term oral anticoagulation has been associated with an increased risk of hemorrhage with an annual risk of major bleeding in the range of 1.1-7.5% and fatalities in the range of 0.25-1.1%^[2].

The rupture of a corpus luteum cyst in a patient on an anti-coagulant therapy is a catastrophic event, which usually ends up by surgical intervention. Hence, with relatively major impact on the ovarian function as a result of the surgery, *e.g.*, oophorectomy, over sewing of the ovarian stroma which might compromise the organ blood supply, bilateral salpingo-oophorectomy, and in some cases, an added hysterectomy^[3]. This rare, yet serious event is well-described in the literature with aggressive surgical management. The complication of a ruptured corpus luteum cyst ranges from a simple corpus luteum hematoma to a life threatening massive intraperitoneal hemorrhage^[3]. In women on anticoagulation therapy, the likelihood of a more catastrophic event is significantly higher^[4].

The Fontan procedure, or Fontan/Kreutzer procedure, is a palliative surgical procedure used in children with complex congenital heart defects. It involves diverting the venous blood from the right atrium to the pulmonary arteries without passing through the morphologic pulmonary ventricle. It was initially described in 1971 by Drs. Fontan and Kreutzer separately as a surgical treatment for tricuspid atresia^[5].

Case Report

A fifteen-year-old single female presented to the Emergency Room with a history of sudden onset of lower abdominal pain associated with dizziness. Her last menstrual period was 15 days prior to presentation. Her menstrual cycles were regular and had been associated with normal bleeding from the time of her menarche that occurred at the age of 12 years. At the age of 14, she had a Fontan operation for a complex congenital heart anomaly. After the surgery, she developed atrial fibrillation and had been maintained on warfarin for the last 6 months.

On examination, at the time of presentation, the patient was in pain and afebrile. Blood pressure was 120/70 mmHg, pulse: 100/min. of normal volume, temperature: 36.8°C. The abdomen was soft with

marked tenderness in the hypogastric area. There was no abdominal distension at that time. Rectal examination revealed tenderness in the Pouch of Douglas (PD). Vaginal examination was deferred, as she was a virgin. Initially, the patient was seen by the cardiology service and admitted to the hospital for observation. Urgent investigations showed Hb: 9.2 g/dl, total white count: 13.3×10^3 /dl, platelets: 290×10^3 /dl, International Normalized Ratio (INR): 6.7. Urea and electrolytes were normal. A pregnancy test was negative.

An ultrasound scan (USS) disclosed a minimal amount of fluid in the pouch of Douglas and a small cystic mass measuring 3 x 3 cm. in diameter behind the uterus, thought to be most probably related to the left ovary. The radiologist suggested more evaluation by computerized axial tomography (CAT) scan. Due to the high INR, the warfarin was discontinued. On the second day of admission, the patient remained hemodynamically stable, although, the abdominal pain increased in intensity with an increasing abdominal distension. Thus, she required opiate analgesia to relieve the pain. Her hemoglobin dropped to 7.4 g/dl. The CAT scan showed a large amount of intraperitoneal fluid. There was a large complex area of different density posterior to the uterus that was interpreted as involving either, a fluid collection or hematoma. At that stage, the patient was referred to the gynecology service. Examination at that time showed a pulse of 106, BP: 120/70 mmHg and her abdomen were tender and distended. Investigation showed Hb: 5.9 g/dl, INR: 4.5.

A decision was made to give the patient fresh frozen plasma (FFP), Vitamin K and blood to correct the high INR and the anemia, respectively, before conducting a laparotomy. She received 2 units of FFP, 4 units of blood and 10 ml of Vitamin K. Over the following few hours, the INR continue to be more than 4.5 despite all the measures taken to reverse its effect. After 24 hr (48 hr from the admission time), the INR was normalized and the patient was prepared for laparotomy, as it was felt that laparoscopy was not the best option due to the abdominal distention. Prior to the laparotomy, the patient's condition improved. The abdominal pain became less severe and she needed less analgesia. Her hemoglobin was stabilized at 9.9 g/dl over the course of few hours prior to the planned surgery. The INR was 1.5. The ultrasound showed a reduction in the amount of free fluid in the peritoneal cavity. After discussion with the patient's family, the cardiologist and the

hematologist, it was decided to postpone the laparotomy, in favor of conservative management, with very close observation of the patient.

The patient's condition started to improve and by the 5th day of admission, she had no abdominal pain. Her hemoglobin had risen to 11.2 g/dl and a repeat ultrasound examination showed minimal fluid in the PD.

On the 7th day of admission, the patient was discharged home after she had received 150 mg of Depo-Medroxy Progesterone Acetate. She had no complaints on the follow up 2 weeks later, at which point the repeat Hb was 15 g/dl.

Discussion

Corpus luteum is formed during the luteal phase of ovarian cycle and 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.

Severe bleeding into the peritoneal cavity from a ruptured corpus luteum cyst is a rare complication in women receiving anticoagulation therapy. A number of surgical methods have been utilized to treat this problem. This report describes a feasible therapeutic approach in a 15-year old patient, who was presented with an acute hemoperitoneum under anticoagulation therapy for an atrial fibrillation following Fontan cardiac surgery for a complex congenital heart disease. The hemoperitoneum was successfully managed conservatively with supportive measures and normalization of the INR. In this way, both ovaries and the uterus were maintained without removal or compromising their important function in this young girl.

The outcome of patient on anticoagulant presented with intraperitoneal hemorrhage is appalling, and it may be devastating to the patients and the treating physician.

Semchyshyn and Zuspan^[3] reported the outcome of such events in 6 cases of ovarian hemorrhage in patients on anticoagulant therapy, and added 29 cases from the literature to their series for a total of 35 cases. Of the 35 cases of ruptured corpus luteum cysts in patients on anticoagulant therapy, 14 (40%) patients had one ovary removed, 8 (23%) had hysterectomy combined with bilateral salpingo-

oophorectomy, 4 (11%) had their ovaries over-sewn, 4 (11%) had ovarian cystectomies, one patient had the hemorrhagic mass removed and in one case the outcome was not reported. Three patients died as a result of the bleeding. The three fatal cases were diagnosed as having bled from a hemorrhagic cyst at autopsy, their deaths being attributed to poor anticoagulant control and the lack of early diagnoses. They concluded in this report that a woman on anticoagulant therapy should be considered at high risk of potentially life threatening ovarian hemorrhage if she continues to ovulate.

In our case the patient was managed conservatively, partly because of the unsuccessful timely reversal of the warfarin effect despite the use of Vitamin K, as well as fresh frozen plasma and even blood transfusion, which were given to the patient when her hemoglobin dropped to 5.2 g/dl. All these measures did not manage to correct the INR to an acceptable level for safe surgical intervention until 48 hr after admission. After which, the decision was made to proceed to surgery. However, at that point, the patient was hemodynamically stable with no further drop in her hemoglobin. There was no further increase in the abdominal girth. The urine output was satisfactory and the INR was corrected. Based on the foregoing, it was decided to observe the patient, who did extremely well, recovering from her bleeding episodes. She was discharged home and she was started on medroxyprogesterone-acetate. The cardiologist on discharge discontinued the warfarin, as it was felt that the patient might not need any further anticoagulation from the cardiac point of view. The same management was adapted by Lurie *et al.*^[6] where they managed to treat the patient who had a repeated massive hemoperitoneum conservatively on two occasions. Payne *et al.*^[7] reported three patients presenting with hemoperitoneum in association with factor VII deficiency, factor X deficiency and sitosterolemia. Conservative management with blood product and factor concentrate support was successful in avoiding surgery in three of the five episodes of bleeding. They concluded that preservation of ovarian function is possible with a conservative approach.

Thus, the current case supports the previous reports on the conservative management for massive bleeding from corpus luteum cyst in patient on anticoagulant therapy, albeit primarily because of the delay related to a high INR that was temporarily refractory to the treatment. Once the patient became hemodynamically stable, the decision to treat

her conservatively was much less difficult. It's believed that the massive hemoperitoneum worked as a tamponade on the bleeding source, and thus created the hemodynamic stability. Interestingly, when the patient came to the clinic two weeks after discharge, her hemoglobin was 15 g/dl.

This is the first case to be reported in the literature utilizing the conservative approach in the management of massive intraperitoneal hemorrhage from corpus luteum cyst in a patient on anticoagulant therapy post Fontan procedure.

It was concluded, that a conservative approach can be utilized in some highly selected patients with massive hemoperitoneum secondary to anticoagulation therapy. Perhaps it's recognized by increasing stability of vital signs and the adequacy of urine output, thus avoiding the risk of surgery. In such cases it is predicted that there would be a reduction in patient morbidity, or even mortality. However, more observations are required to support this prediction.

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العلاج التحفظي لنزيف شديد داخل التجويف البريتوني ناتج عن مضاعفات في كيس الجسم الأصفر في المبيض ، لمريضة قد أجريت لها عملية فونتان في القلب

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المستخلص. الطرق الطبية المستخدمة حالياً لإيقاف النزيف الشديد من كيس ليوتيم في المرضى الذين يتلقون العلاج للتخثر، دائماً تكون جراحية، مما يؤدي إلى مضاعفات قد تكون شديدة للمريضة، وتؤدي إلى فقد عضو حيوي كالمبيض. ونحن نقدم تقرير حالة نزيف داخل الغشاء البريتوني من كيس الجسم الأصفر، عولجت بطريقة متحفظة. وهذا التقرير لفتاة عمرها ١٥ سنة، وشخصت بأنها نزيف داخل الغشاء البريتوني. وكانت على علاج مانع للتخثر، نتيجة تذبذب أذيني للقلب، والتي وضعت عليه بعد إجراء عملية فونتان. وقد اتخذ القرار بالتدخل الجراحي. أثناء تقييم حالة المريضة وجد أنها مستقرة، وهكذا تقرر مواصلة الطريقة المتحفظة في العلاج. هذا هو أول تقرير عن حالة لمريضة تعالج تحفظياً، والتي كان قد أجريت لها عملية فونتان. توضح هذه الحالة الطريقة المتحفظة في العلاج، قد تمثل خياراً معقولاً في المرضى الذين تكون حالة الدورة الدموية مستقرة بعد حصول النزيف الشديد.