Infected Ovarian Cyst in a Homozygous Beta-Thalassemic Patient

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Abstract

An infected ovarian cyst in a thalassemic patient is rarely reported. We describe the case of a 22-year old woman with splenectomized homozygous beta-thalassemia who developed high fever and was diagnosed as having an infected ovarian cyst. The mechanisms which ß-thalassemia might predispose to infection and considered to be immunocompromized are discussed. She was given an intravenous antibiotic regimen and the infected ovarian cyst was removed. The difficulties in the diagnosis of an infected ovarian cyst is because of its rarity and the paucity of information on it in the literature. Therefore, the triad of ovarian cyst, immunocompromized host, and signs of infection with failure to identify any other source of infection should raise the suspicion of an infected ovarian cyst.

Key word : Infected Ovarian Cyst, Beta-Thalassemia

Infected ovarian cysts have become a rare complication over the last 3 decades for many reasons, including the changing of the treatment of ovarian cysts by removal rather than aspiration and the extensive use of antibiotics. A case of infected ovarian cyst in homozygous beta-thalassemic patient is described.

CASE REPORT

A 22-year-old female patient was admitted to the hospital because of high fever with chills for 20 days. She noted the abrupt onset of fever associated with left lower abdominal pain. She denied ever having had sexual contact. She had a history of homozygous ß-thalassemia. Since childhood, she had been dependent upon blood transfusion every 3-4 weeks to maintain her hemoglobin level. As anticipated, a severe hypersplenism had developed, and splenectomy was performed at the age of 9 years.

Physical examination revealed a thin, chronically ill, febrile woman with marked pallor and thalassemic facies. Her blood pressure was 100/70 mmHg, regular pulse 100 beats per minute, respira-

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tion 16 breaths per minute, and body temperature of 39°C. The liver edge extended 4 cm below the right costal margin. There was a large, smooth cyst of 24 cm diameter occupying the pelvis and extending to the whole left lower quadrant, which on bimanual examination was slightly mobile, and tender.

The hemoglobin was 4.7 g/dl, hematocrit 13 per cent, mean corpuscular volume (MCV) 50.9 fl, mean corpuscular hemoglobin (MCH) 18.3 pg, mean corpuscular hemoglobin concentration (MCHC) 36 per cent. Examination of the peripheral blood smear showed marked anisopoikilocytosis with hypochromia, microcytosis and target cells formation. The white cell count was 10,000/mm^3 with 95 per cent neutrophils and 5 per cent lymphocytes. Platelets were 1,200,000/mm^3. Urinalysis, renal and liver function tests were all within normal limits. A chest X-ray was unremarkable. Thick smears for malarial parasites were negative as were titers for salmonella antigens. Blood and urine culture was negative. Pelvic ultrasound examination showed a large uniloculated cyst in the left pelvic region with an internal interface between upper hypoechoic part and lower hyperechoic part which was gravity dependent as shown in Fig. 1. The uterus and kidneys were normal. The liver was generally enlarged. A presumptive diagnosis of infected ovarian cyst was made. Antibiotic therapy was promptly initiated with ampicillin 1 g IV every 8 hours and gentamicin 80 mg IV every 8 hours.

After improvement of her general status, she was explored through a lower midline incision on the third day of admission. Four units of packed red blood cells were given before and during operation. A left 24-cm ovarian cyst occupied the pelvic cavity without adhesion to the adjacent organs. The uterus, right ovary and both fallopian tubes were normal. Left salpingo-oophorectomy was performed. Pathological evaluation of the left ovary showed a uniloculated cyst with smooth surface. Its content was purulent discharge with debris sediment in the lower part of the cyst. On microscopic examination of hematoxylin and eosin section, the cyst was lined by single layer mucinous epithelium. Infiltration of poly-morphonuclear cells was noted in the cavity and cyst wall as shown in Fig. 2. The left fallopian tube was slightly inflamed. On culture of the content from the ovarian cyst, there was no growth of any bacteria. Intravenous antibiotics were continued. She became afebrile the fourth day after operation. The postoperative course was uneventful,

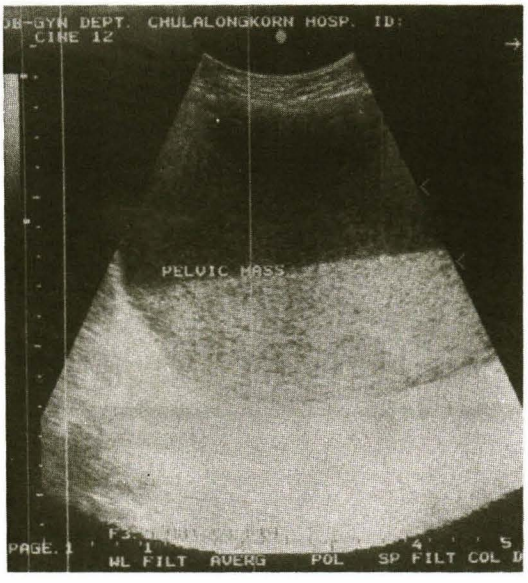

Fig. 1. Sonogram showing a large uniloculated cyst in the left pelvic region with an internal interface between upper hypoechoic part and lower hyperechoic part which is gravity dependent.

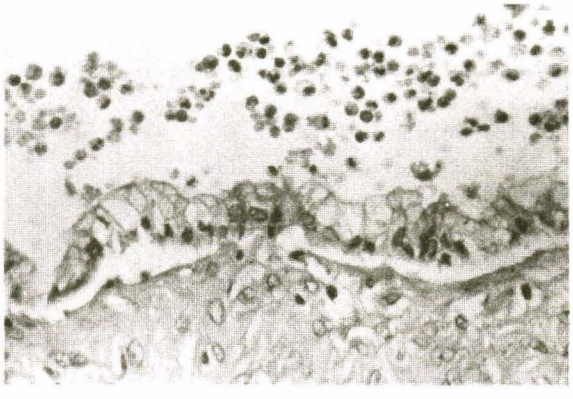

Fig. 2. Microscopic examination showing the cyst is lined by single layer mucinous epithelium. Infiltration of polymorphonuclear cells is noted in the cavity and cyst wall.

with a gradual return to normal for all clinical and laboratory findings. She was discharged 7 days after the operation and has remained well since.

DISCUSSION

Nowadays, an infected ovarian cyst is a rare but sometimes fatal complication. In the previous

review, many factors associated with the development of an infected ovarian cyst were listed. These included introduction of infection at the time of abdominal aspiration or by needle aspiration through the posterior vaginal fornix, hematogenous spreading during the transient bacteremia, lymphatic spreading from postpartum or postabortal infection, tubal infection, vaginal surgery, torsion of the ovarian cyst and adhesion to the adjacent organs including the bowel and appendix[1]. Two important factors contributing to the dramatic decrease of this complication are changing of the treatment of ovarian cysts by removal rather than aspiration and the extensive use of antibiotics. We hypothesized that an infected ovarian cyst developed from seeding of bacteria into the left mucinous cystadenoma during transient bacteremia which occurs occasionally in the case of an immunocompromised host such as a thalassemic patient. In this case, an ascending lower genital tract infection or spreading from the adjacent organs is less likely. The clinical presentations of infected ovarian cyst in this case were characterized by high fever, tachycardia, tender at the ovarian cyst and general malaise. Because of the difficulty in diagnosis and the rarity of this complication, they often go undetected and death occurs from sepsis. In this case, a sonographic picture of ovarian cyst with an internal interface between upper hypoechoic part and lower hyperechoic part which was gravity dependent helped us to confirm the diagnosis. The sonographic findings were confirmed by the pathological findings that the lower hyperechoic part was compatible with the dense debris sediment of the purulent material. The sonographic findings of internal interface with gravity dependence can also be found in the case of resolved pyosalpinx. The treatment of an infected ovarian cyst involves the use of an intravenous antibiotic regimen administered in a hospital with surgical exploration and removal of the infected cyst.

Infected ovarian cyst should be considered in the differential diagnosis of complications of ovarian cysts, especially in an immunocompromized patient, such as thalassemic patients. Thalassemia is a major hereditary hemoglobinopathy characterized by absent or deficient synthesis of globin chain. ß-Thalassemia is characterized by deficient synthesis of the beta globin component of the hemoglobin molecule, which leads to severe ineffective erythropoiesis. Individuals afflicted with homozygous ß-thalassemia suffer from severe anemia and are dependent on blood transfusion and susceptible to infection more often than the normal population[2]. However, the defects that predispose thalassemic patients to infection are not well understood. Previous studies have suggested splenectomy[3], abnormal immunoglobulin production[4], impaired T-cell mitogen response and decreased T-cell numbers[5], deficient activity of the complement system[6], defective phagocytic power of white blood cells[7], presence of immune complexes[8], and iron overload[9] as possible causes. Infections represent a common cause of death in thalassemia, especially in splenectomized cases[3]. Splenectomy has been considered to affect the immune system. Patients splenectomized for thalassemia have an 800-fold increase in sepsis. The causative organisms can be either Gram-positive or Gram-negative bacteria, and most patients die in a few days after they become infected. Therefore, it is recommended that when a thalassemic patient, especially if splenectomized, has fever, a complete evaluation is mandatory and empirical antibiotic therapy should not await confirmation by culture[10].

Thus, our report is the first in the literature possibly associating infected ovarian cyst to immunocompromized hosts, such as thalassemic patients. An infected ovarian cyst is usually not included in the initial differential diagnosis because of its rarity and the paucity of information on it in the literature. In conclusion, it is important to keep the diagnosis of infected ovarian cyst in mind, especially in a patient with ovarian cyst, immunocompromized host, and signs of infection with failure to identify any other source of infection should raise the suspicion of an infected ovarian cyst.

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การติดเชื้อของถุงน้ำรังไข่ในผู้ป่วยฮีโมซัยกัส บีตา–ธาลัสซีเมีย

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ภาวะติดเชื้อของถุงน้ำรังไข่พบได้น้อยมาก ได้รายงานผู้ป่วยหญิงอายุ 22 ปี มีประวัติเป็น โฮโมซัยกัส บีตา–ธาลัสซีเมีย ซึ่งได้รับการตัดม้ามแล้ว มาพบแพทย์ด้วยปัญหามีไข้สูงและได้รับการวินิจฉัยว่ามีภาวะติดเชื้อของถุงน้ำรังไข่ บีตา–ธาลัสซีเมีย เป็นสาเหตุให้มีความบกพร่องของระบบภูมิคุ้มกันของร่างกายและอาจส่งเสริมให้เกิดภาวะติดเชื้อได้ ได้รักษาผู้ป่วยโดยการให้ยาปฏิชีวนะและผ่าตัดเอาถุงน้ำที่รังไข่ออก การวินิจฉัยการติดเชื้อที่ถุงน้ำของรังไข่ทำได้ยากเนื่องจากเป็นภาวะที่พบได้น้อยและมีข้อมูลจากรายงานต่างๆน้อยมาก ดังนั้นในผู้ป่วยที่มีถุงน้ำที่รังไข่, ภาวะที่ผู้ป่วยมีความบกพร่องของระบบภูมิคุ้มกันของร่างกาย, มีอาการของการติดเชื้อโดยที่ไม่สามารถหาแหล่งของการติดเชื้อได้ควรสงสัยภาวะติดเชื้อของถุงน้ำที่รังไข่ด้วย

คำสำคัญ : ถุงน้ำรังไข่ติดเชื้อ, บีตา–ธาลัสซีเมีย

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