Pseudo-Meigs' Syndrome Secondary to Subserous Myoma Uteri : A Case Report

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Abstract

A 45 year-old Thai woman, gravida 5, para 5 presented with a huge pelvic mass as well as ascites and right pleural effusion. Right thoracocentesis was performed pre-operatively and malignant cells could not be detected on the cytological examination of the pleural fluid. Surgical exploration revealed a large pedunculated subserous leiomyoma of the uterus without malignant transformation. Total abdominal hysterectomy with bilateral salpingooophorectomy and appendectomy were performed. Both ascites and pleural effusion resolved post-operatively and did not recur during the 12-month follow-up. This case demonstrated the extremely rare case of pseudo-Meigs' syndrome caused by a subserous uterine leiomyoma.

Key word : Meigs' Syndrome, Pseudo-Meigs' Syndrome, Myoma Uteri

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It was in 1937 that Meigs' syndrome was first described as the combination of unilateral ovarian fibroma, ascites, and right-sided pleural effusion(1). Atypical Meigs' syndrome, also known as pseudo-Meigs' syndrome, occurs with the clinical triad of ascites, pleural effusion and pelvic mass other than ovarian fibroma(2,3). These syndromes are largely the results of ovarian pathologies, not that of uterine origin, and normally resolve when the pelvic pathologies are removed. The authors report here the extremely rare case of pseudo-Meigs' syndrome arising from a large subserous uterine leiomyoma. The presentations of pelvic mass together with ascites and pleural effusion may easily mislead physicians to

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believe that the condition was due to a malignant process. Although rare, this case is another example of the curable cause of such combinations.

CASE REPORT

A 45-year-old, Thai woman, gravida 5, para 5 had a ten-month history of increasing abdominal girth and a three-month history of palpable pelvic mass. The patient also experienced progressive dyspnea for the last two weeks prior to admission to our hospital. She had regular menstruation and experienced no history of abnormal uterine bleeding. Upon examination, the authors found that there was decreased breath sound and increased vocal resonance at the right lower lung field. The patient's abdomen was distended with ascites while the liver and spleen were impalpable. A gynecologic examination revealed a normal size uterus and a large firm mass approximately 15 x 10 x 10 cm in size located on the right side of the uterus. This mass was easily mobilized during bimanual examination. Laboratory findings included hemoglobin of 8.4 g/dl, hematocrit of 26 vol%, leucocytes of 8,800 cells/mm^3, platelets adequate, fasting blood sugar of 82 mg/dl, BUN of 5.9 mg/dl, serum creatinine of 0.9 mg/dl, sodium of 139 mEq/L, potassium of 3.9 mEq/L, bicarbonate of 24.0 mEq/L, chloride of 107 mEq/L, cholesterol of 188 mg/dl, total protein of 7.2 g/dl, albumin of 4.0 g/dl, globulin of 3.2 g/dl, total bilirubin of 1.0 mg/dl, direct bilirubin of 0.2 mg/dl, SGPT of 16 U/L, SGOT of 22 U/L, alkaline phosphatase of 52 U/L and LDH of 172 U/L. The results of urine analysis and Pap smear were within normal limits. Chest X-ray revealed moderate right pleural effusion with otherwise normal appearance of lung parenchyma. Pelvic ultrasonographic study demonstrated a moderate amount of ascitic fluid with a huge suprapubic solid mass showing internal hypoechogenic spaces and normal size of the uterus. Right thoracocentesis was performed pre-operatively for diagnostic purpose and pleural fluid was sent for further evaluation. Laboratory findings of the pleural fluid included erythrocyte count of 1,200 cells/mm^3, leucocyte count of 1,800 cells/mm^3, LDH of 86 U/L, sugar of 130 mg/dl, protein of 2.1 g/dl. The cytologic evaluation of pleural fluid turned out negative for malignant cells. Gram stain of pleural fluid revealed a small amount of neutrophils without bacteria detected. AFB staining also demonstrated negative finding for tuberculous bacteria. From these findings, the most likely diagnosis was malignant ovarian tumor, thus, the patient was counseled and prepared for exploratory laparotomy. Before the surgery, lung function test and a three-day bowel preparation were conducted. The patient underwent laparotomy under general anesthesia. The findings at surgery included 300 ml of yellow and clear ascites, and a firm pedunculated mass, 19 x 15 x 10 cm in size arising from the right uterine fundus. This mass was freely mobile and showed no adhesion to the surrounding structures. Its peduncle was approximately 7 cm in length. The ovaries and both fallopian tubes appeared grossly normal. Other intra-abdominal organs such as pelvic peritoneum, omentum, appendix, liver and spleen were grossly normal. Both pelvic and para-aortic lymph nodes were impalpable. A total abdominal hysterectomy with bilateral salpingooophorectomy with omental biopsy and appendectomy were performed. After being taken from the patient's pelvis, this uterine mass was carefully evaluated by the surgical team. Cut surface of the mass revealed whorl-like appearance with multiple small cystic areas containing yellowish fluid. Immediate postoperative diagnosis of subserous myoma with pseudo-Meigs' syndrome was thus made but the definite diagnosis could not yet be made without histological confirmation. The surgery concluded with estimated blood loss of 200 ml and no intra-operative blood transfusion was required. The patient began to regain her appetite 2 days after the operation. The patient's nutritional and physical status improved rapidly with no dyspnea presented. A follow-up chest roentgenography, performed 4 days after the surgery, showed clearing of right pleural effusion. The patient's postoperative course was uncomplicated, and she was ultimately discharged from the hospital in stable condition 7 days after the surgery. The pathological results, obtained two weeks later, demonstrated a minute intramural leiomyoma and huge subserous leiomyoma with degenerative changes. The cervix, adnexa, omentum and appendix were normal. Upon one-year follow-up, there was no evidence of recurrent ascites or pleural effusion. The final diagnosis of pseudo-Meigs' syndrome caused by a large pedunculated subserous myoma was, thus, confirmed.

DISCUSSION

Meigs' syndrome is presented by a benign tumor of the ovary (such as fibroma, thecoma, or Brenner tumor) associated with ascites and pleural effusion that respond to removal of the tumor[4,5]. Pseudo-Meigs' syndrome includes all other pelvic tumors (both benign and malignant) associated with

ascites and pleural effusion, in which the fluid does not reaccumulate after removal of the tumor. Andinolfi first used the term pseudo-Meigs' syndrome in 1966 to describe a uterine myoma causing ascites and pleural effusion[6]. A pseudo-Meigs' syndrome rarely occurs in patients with a uterine leiomyoma and, to the best of the authors' knowledge, only 20 cases of pseudo-Meigs' syndrome arising from myoma uteri have been reported in both English and Japanese literature[7-13]. In the previous review by Migishima et al, the ages of the 20 patients presenting with pseudo-Meigs' syndrome due to leiomyoma were from 30 to 73 years, with a mean age of 45.4 years[13]. Dyspnea and abdominal distention were found in a large proportion of these patients (55 and 70% of the cases, respectively)[13]. The case reported here also presented with both manifestations together with evidence of right pleural effusion demonstrated by chest X-ray. The study by Leuallen and Carr demonstrated that among 436 patients presenting with hydrothorax, only 1 case (0.2%) was due to Meigs' syndrome. Based on this report, it is thus reasonable to postulate that hydrothorax secondary to pseudo-Meigs' syndrome should be far less common. Concerning the laterality of pleural effusion, it was claimed that, in Meigs' or pseudo-Meigs' syndrome, collection of fluid in the pleural space is more common on the right side. The review by Migishima et al also confirmed this notion since among the 20 cases reviewed by his group hydrothorax was present largely on the right pleural space (45%) followed by the left (25%) and both sides (20%)[13].

This report emphasize the fact that the findings of pelvic mass, ascites and pleural effusion, which usually results from a malignant process, does not necessarily mean that the patients have an untreatable surgical condition. If there is no firm evidence of distant metastasis, such as no malignant cells detected from the pleural fluid, surgical treatment shall remain one of the alternative treatment option. Thorough discussion with the patient and their relatives should also be made regarding the possible treatment choices in such cases since, although rare, benign conditions of Meigs' or pseudo-Meigs' syndromes are still possible.

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กลุ่มอาการ pseudo–Meigs ซึ่งเกิดจากเนื้องอกกล้ามเนื้อมดลูก : รายงานผู้ป่วย 1 ราย

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รายงานผู้ป่วยหนึ่งรายซึ่งเป็นหญิงไทยคู่ อายุ 45 ปี มีบุตรมาแล้ว 5 คน มาพบแพทย์ที่โรงพยาบาลศรีนครินทร์ คณะแพทยศาสตร์ มหาวิทยาลัยขอนแก่น เนื่องจากคลำพบก้อนในท้องน้อยมาประมาณ 3 เดือน และมีอาการหายใจลำบากมาประมาณ 2 สัปดาห์ ตรวจร่างกายพบว่าเสียงหายใจบริเวณชายปอดข้างขวาลดลง ตรวจพบน้ำในช่องท้อง และก้อนในอุ้งเชิงกรานขนาดประมาณ 15 x 10 x 10 เซนติเมตรที่ด้านขวาของมดลูก ผลการถ่ายภาพรังสีทรวงอก พบน้ำในเยื่อหุ้มปอดข้างขวา การตรวจคลื่นเสียงความถี่สูงทางนรีเวชวิทยาพบว่ามดลูกขนาดปกติ และมีก้อนขนาดใหญ่ในอุ้งเชิงกราน ซึ่งมี hypo-echogenic spaces จำนวนมากอยู่ภายในก้อนนี้ ได้ทำการเจาะน้ำจากเยื่อหุ้มปอดข้างขวาเพื่อส่งตรวจซึ่งไม่พบเซลล์มะเร็ง ได้ทำการผ่าตัดเปิดหน้าท้องพบ pedunculated subserous myoma ขนาดใหญ่มีก้านยาวประมาณ 7 เซนติเมตร และมีน้ำในช่องท้องสีเหลืองใสประมาณ 300 มิลลิลิตร ได้ผ่าตัดเอามดลูก ท่อนำไข่ และรังไข่ทั้งสองข้างออก พร้อมทั้งตัดไส้ติ่งและทำการตัดชิ้นเนื้อของโอเมนตัม (omental biopsy) ผลทางพยาธิวิทยาพบว่าเป็น subserous myoma with degenerative changes และไม่พบบริเวณที่มีการเปลี่ยนแปลงไปเป็นมะเร็ง ผู้ป่วยมีอาการดีขึ้นอย่างรวดเร็วภายหลังการผ่าตัด การตรวจภาพถ่ายรังสีทรวงอกติดตามหลังผ่าตัด 4 วัน พบว่าน้ำในเยื่อหุ้มปอดข้างขวาหายไป ได้ติดตามผู้ป่วยต่อมาเป็นเวลา 1 ปี ก็ไม่พบว่ามีการกลับเป็นซ้ำของน้ำในช่องท้อง หรือ น้ำในเยื่อหุ้มปอดอีก จึงให้การวินิจฉัยว่าเป็น pseudo–Meigs' syndrome ที่เกิดขึ้นจาก pedunculated subserous myoma ซึ่งเป็นกรณีที่พบได้น้อย แต่ก็ควรจะอยู่ในการวินิจฉัยแยกโรคของแพทย์ด้วย เมื่อผู้ป่วยมาพบด้วยกลุ่มอาการลักษณะเช่นนี้

คำสำคัญ : กลุ่มอาการ Meigs, กลุ่มอาการ pseudo–Meigs, เนื้องอกในมดลูก

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