CEOG Clinical and Experimental Obstetrics & Gynecology

Case Report

Primary borderline paraovarian serous tumor in pregnancy: case report and review of the literature

F. Sorrentino, F. Boncristiano, F. Greco, A. D'Aloia, P. Di Gioia, L. Nappi

Department of Medical and Surgical Sciences, Institute of Obstetrics and Gynecology, University of Foggia, Foggia (Italy)

Summary

There are few reports of pregnancy complicated by a primary borderline paraovarian tumor. A 37-year-old woman after four pregnancies was found to have an adnexal tumor. At 17.4 weeks of gestation, laparotomic right salpingo-oophorectomy was performed and a diagnosis of primary borderline paraovarian serous tumor was made. At 40 weeks of gestation a spontaneous vaginal delivery was performed. Currently, the patient is being followed after the initial surgical treatment and all imaging data show no evidence of recurrence. This case demonstrates the appropriate management for a primary borderline paraovarian tumor during pregnancy.

Key Words: Paraovarian cyst; Borderline ovarian tumors; Pregnancy.

Introduction

A paraovarian cyst represents 5-20% of the adnexal masses and it originates from the broad ligament in the area of the fallopian tube or ovary. In 76% of cases it originates from the remnants of paramesonephric (Müllerian) or mesonephric (Wolffian) ducts that are present during urogenital embryologic development.

A simple, asymptomatic paratubal or paraovarian cyst can be managed expectantly without further follow-up. Surgical removal is indicated in case of torsion, persistent pain or pressure symptoms, or if they appear neoplastic [1, 2].

It is reported that 0.2% to 2% of pregnancies are complicated by an adnexal mass, and approximately 1% to 6% of these masses are malignant [3, 4].

Between the adnexal masses in pregnancy, paraovarian cysts are the third most common type after benign cystic teratomas and serous cystadenomas [5]. Surgical treatment for paraovarian cysts can be performed with laparotomy or operative laparoscopy.

It is almost impossible to diagnose primary borderline paraovarian tumor prior to surgery and treatment for this tumor has not been suggested since its biological characteristics have not been addressed due to its rare occurrence.

The authors report a very rare case of primary borderline paraovarian tumor. They observed this case through a histological examination after performing a laparotomic salpingo-oophorectomy on a 37-year-old female patient who attended the present clinic with an ovarian cystic tumor in the 14th week of her pregnancy.

Case Report

A 37-year-old multiparous woman gravida 5, para 3 was referred to the present Unit at gestational age of 14.4 weeks after her first trimester ultrasound scan had revealed a right ovarian mass. The patient's history included a laparotomic appendicectomy in 2002. The patient was asymptomatic, with no weight loss and abdominal pain. She had conceived spontaneously and on examination, her vital signs were normal. Standard tumor markers (AFP, CEA, CA15-3, and CA19-9) were within normal limits except for CA125 (320.0 UI/ml). Ultrasound scan showed a singleton regular pregnancy, an intramural myoma of 52×36 mm in the anterior wall of uterus, a normal left ovary. An adnexal mass, measuring approximately 79×58×65 mm, was detected arising from the right ovary. This mass had the features of an unilocular solid cyst with ground glass echogenicity and papillary structures measuring 24×33mm (color-score 2 with ovarian crescent sign) (Figure 1).

The authors' first diagnostic hypothesis was a decidualization of an ovarian endometrioma. The surgical procedure (type of incision, risks, possible complications...) was explained by a consultant and informed consent for laparotomic monolateral sapling-oophorectomy was signed by the patient. Laparotomy was performed under epidural anesthesia at 17.4 weeks of gestation. No ascites was detected. The right ovary was intact and a right paraovarian cystic mass, with an intact capsule, was detected (Figure 2). The uterus and the left ovary were normal. A right salpingo-oophorectomy was performed (Figure 3).

The histopathologic examination was consistent with a borderline paraovarian serous tumor; homolateral ovary and tube were clear by tumor. There was any cytological evidence of malignance after peritoneal washing examination. No intraoperative or postoperative complications were reported and patient was discharged three days after surgery. After the consultant extensively explained to the patient the available treatment methods and their possible risks, the pregnancy was allowed to continue under close observation. An abdominal MRI scan was performed three months after surgery with no signs of extra-adnexal localization.

Published: 15 February 2020

Clin. Exp. Obstet. Gynecol. - ISSN: 0390-6663
XLVII, n. 1, 2020
doi: 10.31083/j.ceog.2020.01.5123

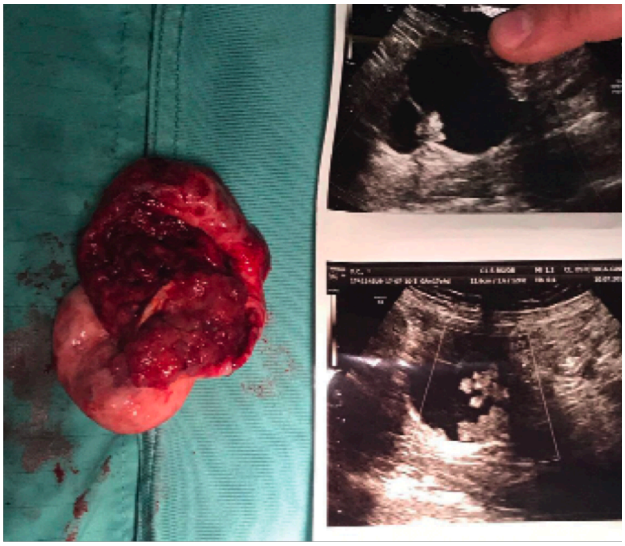

Figure 1. — Ultrasound images and cyst.

The pregnancy progressed normally until the 40th week of gestation, when spontaneous rupture of membranes and uterine contractions occurred. A healthy infant weighing 3.9 kg was delivered vaginally. Histological examination of the placenta was performed after delivery and no sign of pathology was detected.

Currently, the authors are monitoring the patient (ultrasound scan, MRI, CT, blood tests, tumor markers) for 12 months after the initial treatment and there is no evidence of recurrence.

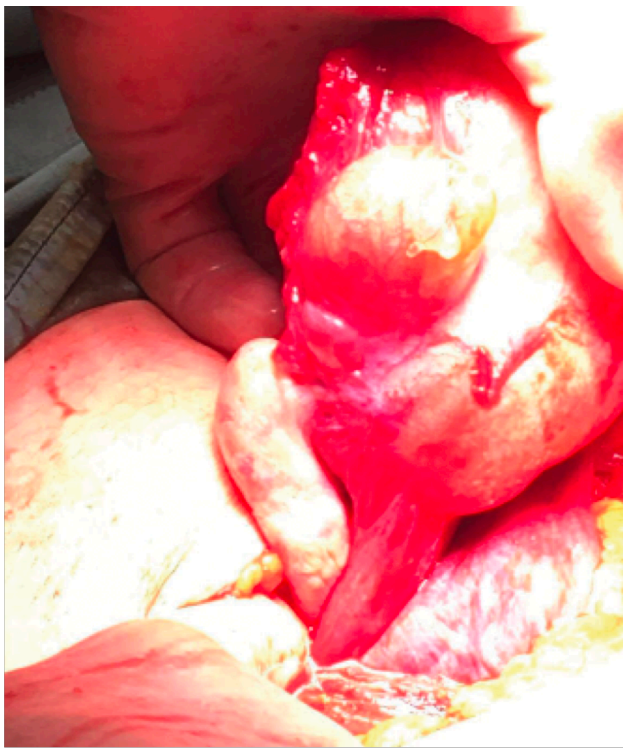

Figure 2. — Right paraovarian cystic mass.

Discussion

Incidence of adnexal mass during pregnancy ranges from 0.2% to 2% [3]. The majority of these masses are discovered incidentally during routine follow–up in the first two trimesters of pregnancy. Most of the adnexal masses are functional cysts and approximately 90% will regress spontaneously during pregnancy [6].

The majority of lesions that persist are benign, and include teratomas, serous cystadenoma, paraovarian cysts, mucinous cystadenoma, and endometriomas. There is a limited percentage (3%-6%) of malignant lesions. The most frequent malignant ovarian tumors in pregnancy are germ-cell tumors, followed by borderline ovarian tumors (BOTs) and epithelial ovarian cancer [7, 8].

The management of adnexal masses in pregnancy has been a subject of debate for years with no consensus regarding the best management plan. It is usually expectant because most lesions resolve spontaneously and the incidence of malignancy is rare [1]. However, in certain clinical situations, surgery can be lifesaving.

Ideally, it should be performed electively in the second trimester (at approximately 15 weeks of gestation) to minimize the risks of spontaneous abortion, preterm labor, and intrauterine fetal death. Paraovarian cysts are embryological remnants of the paramesonephric or mesonephric ducts. They typically are found in the mesosalpinx. Also, they are not clinically significant and are discovered incidentally during pelvic sonography or surgery [9].

Most tumors developed from paraovarian cysts are benign serous tumors [10]. Primary borderline paraovarian tumor is very rare and its complication with pregnancy is even rarer [11]. It is very difficult to diagnose borderline tumor and malignant tumor in the paraovarian before surgery. The pathological diagnostic criteria of a primary borderline tumor are the following: localization within the paraovarian, separation from the ovary, fallopian tube and uterus, characteristics of a paraovarian cystic tumor under microscopic examination, stratification of the papillary surface epithelium, dysplasia of nucleus and cells, polymorphism and active mitosis of the epithelium, and no infiltration into the cell substrate [11].

Since primary borderline paraovarian tumor is very rare, there is no consensus on effective treatment. Its treatment usually depends on its stage and on patient desire of fertility. Conservative surgery (cystectomy, oophorectomy, or salpingo-oophorectomy) is indicated when these criteria are present: lack of external vegetations, lack of adhesions, tumor excision without rupture, no ascites, no other structures involved, low-grade malignancy, and wish to maintain fertility. Nonetheless, if one of these criteria is not accomplished or fertility is not wanted, more aggressive surgery is recommended (a total hysterectomy and bilateral salpingo-oophorectomy should be performed and omentectomy and lymphadenectomy might be performed if re-

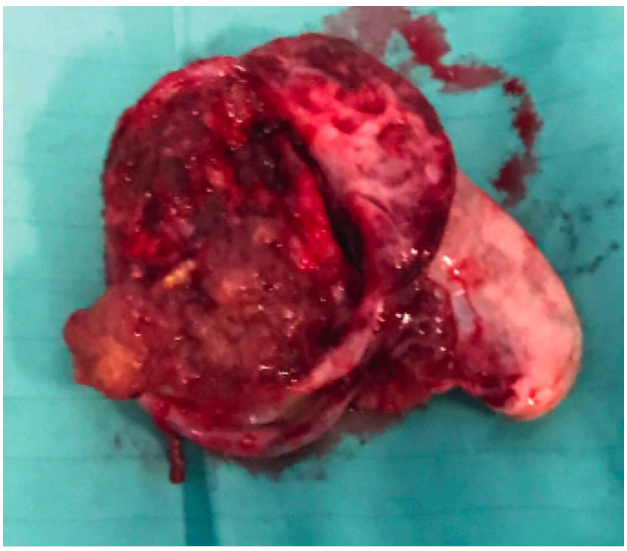

Figure 3. — Specimen for pathology.

quired). Paraovarian borderline tumors seem to have a good prognosis [12], but an accurate follow-up is required to detect late recurrences.

Conclusion

The early diagnosis and the early onset of treatment, a careful evaluation of possible malignancy in patients with adnexal masses (physical examinations and ultrasonographic examinations are required especially during the early stage of pregnancy) are important factors for good prognosis in the case of malignant ovarian tumors. During pregnancy, it is very important. The best timing for treatment and the best options should be selected to guarantee optimal prognosis and progression of a pregnancy.

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Corresponding Author:
F. SORRENTINO, M.D.
Department of Medical and Surgical Sciences
Institute of Obstetrics and Gynecology
University of Foggia
Viale L. Pinto
71100 Foggia (Italy)
e-mail: felice.sorrentino.1983@gmail.com