

Thecoma associated with pregnancy: a case report

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Summary

Ovarian thecoma, which belongs to the group of sex-cord stromal tumors, is a relatively rare neoplasm. In this report we present a pregnant woman with a solid ovarian mass diagnosed during pregnancy and operated on at the 19th gestational week. At surgical exploration, torsion of the ovarian tumor was observed together with ascites and unilateral salpingo-oophorectomy was performed. Pathological examination revealed a luteinized thecoma. After the surgery, the pregnancy continued uneventfully until term.

Key words: Luteinized thecoma; Pregnancy; Torsion; Ascites.

Introduction

Ovarian thecoma is a relatively rare tumor, most commonly encountered during the fifth to sixth decades of life [1, 2]. It belongs to the group of tumors derived from special gonadal stroma, which includes all those that contain granulosa cells, theca cells, luteinized derivatives of these cells, Sertoli cells, Leydig cells, and fibroblasts of gonadal stromal origin [3]. Excessive secretion of sex steroids is an important characteristic of ovarian thecomas as well as of the other sex-cord stromal tumors which contain cells capable of active steroid hormone production [1, 2]. Approximately 5% of all ovarian neoplasms are of the sex-cord stromal type and it is quite rare to come across them during pregnancy [3, 4]. In this report we present a case of luteinized thecoma associated with pregnancy.

Case Report

A 21-year-old primigravid woman, who had not had any previous gynecological examinations, attended for antenatal follow-up. Uterus bicornus was suspected at the first transabdominal ultrasonographic (US) examination performed at the 9th week of pregnancy. Four weeks later, the US appearance was consistent with a left-sided solid adnexal mass 63 x 44 mm in size and most probably ovarian in origin because the left ovary could not be visualized separately (Figure 1). The follow-up examinations until the 18th week of pregnancy did not reveal any clinical symptoms or any marked progressive change in the appearance of the mass but only a minimal increase in size (Figure 2). It was planned to refer the patient to a university hospital for an elective surgical exploration at the 19th gestational week. Interestingly, a few days before the planned referral time, ascitic fluid accumulated progressively in the abdomen, as observed at US examination, and the patient complained of pelvic pain but there were no signs suggesting an acute abdomen. Serum CA-125 level was 134.96 IU/ml.

After urgent preparation for the surgery at the university hospital, laparotomy was performed via a midline incision crossing the left side of the umbilicus and extending to the upper abdomen. Ascitic fluid of approximately two liters was aspirated and samples from this fluid were sent for cytological examination. The tumor originated from the left ovary, it was solid, 8.5 x 6 x 4 cm in size, torsioned twice around both of the ovarian ligaments and had an irregular surface adherent to the parietal peritoneum (Figure 3). After adhesiolysis, unilateral salpingo-oophorectomy was performed. As the frozen section examination suggested a benign neoplasm of sex-cord stromal type, no further organ or tissue resection was required. To prevent premature uterine contractions in the patient, 250 mg of 17-hydroxyprogesterone caproate was administered via intramuscular injection, as well as 100 mg of indomethacin as a rectal suppository just before the surgery. Indomethacin was continued via the oral route after the surgery at a dose of 25 mg four times daily for a period of 48 hours. Clindamycin was used for the antibiotic prophylaxis.

Detailed pathological examination of the tumor revealed a thecoma containing cell nests with eosinophilic cytoplasm within minimally fibrotic stroma (Figure 4). Nuclear atypia was not encountered and mitotic activity was occasionally observed. Thus, the diagnosis was a luteinized thecoma. The ascitic fluid was free of any malignant cells.

After the surgery, the pregnancy continued uneventfully until term. A 2,500 g male infant was delivered via the vaginal route at the 37th week of gestation.

Discussion

Adnexal masses are observed in about one out of 600 pregnancies [5]. Mature cystic teratoma is the most commonly encountered benign ovarian tumor during pregnancy, whereas dysgerminoma and serous cystadenocarcinoma of low malignant potential are the most commonly observed malignant ovarian tumors [6, 7]. Malignant neoplasms account for only 2-5% of all ovarian tumors associated with pregnancy [6]. Unless there is a strong suspicion of malignancy or an acute complication such as rupture or torsion, immediate surgical exploration of an adnexal mass is not recommended, especially in the 1st

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Figure 1. — Transabdominal ultrasonography performed at the 13th gestational week. A left-sided solid adnexal mass 63 x 44 mm in size was observed.



Figure 2. — Transabdominal ultrasonography performed at the 16th gestational week. The adnexal mass was 88 x 46 mm in size.



Figure 3. — The findings at the surgery performed at the 19th gestational week. Midline laparotomy incision crossed the left side of the umbilicus. The left-sided ovarian tumor was 8.5 x 6 x 4 cm in size and torsioned twice around both of the ovarian ligaments.

trimester [6]. Most cystic masses observed in the first trimester are corpus luteum cysts which disappear spontaneously until the second trimester [8]. In our case, the left-sided ovarian mass was diagnosed in the first trimester and it was observed until the 18th week of ges-

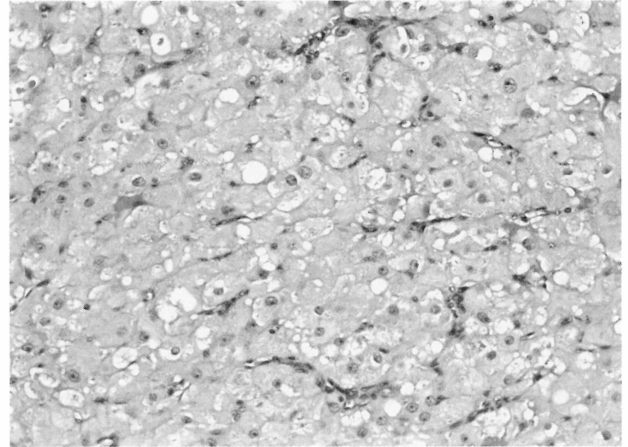


Figure 4. — Luteinized thecoma diagnosed at pathological examination. The tumor revealed cell nests within a minimally fibrotic stroma where the eosinophilic cytoplasm of the cells indicated luteinization.

tation. Although it was a solid mass, it only enlarged slightly during this observation period, but later on ascites developed. A small or moderate amount of ascites might accompany adnexal torsion [9], however in our case two liters of ascitic fluid had accumulated which was more than what was expected to develop secondary to the torsion. Hopkins *et al.* reported Meigs' syndrome associated with an ovarian thecoma in a pregnant women at the 32nd week of gestation [10]. That patient had pleural effusion in addition to ascites. Meigs' syndrome occurs in cases with benign solid ovarian tumors, especially fibromas. The ascitic fluid is thought to be secreted directly from the tumor, but the etiology of the pleural effusion is unclear [1]. Thecoma is classified within the same histologic group of ovarian tumors as fibroma and is considered to be virtually benign, similar to fibroma [3]. Malignant behavior is extremely rare [2].

In our case, the microscopic examination revealed occasional cells that had accumulated moderate to large amounts of lipids, and clusters of cells with eosinophilic cytoplasm similar to lutein cells. There were no crystalloids within the lipid-laden steroid cells. The histologic appearance of a luteinized thecoma, in fact, is quite similar to that of a stromal Leydig cell tumor except for the presence of crystalloids of Reinke in the latter [2]. Zhang *et al.* reported 46 cases of luteinized thecoma in their series. Fifty percent of these had estrogenic manifestations and 11% had androgenic manifestations, while the rest had no clinical evidence of steroid hormone secretion. Six patients were pregnant and two of those were virilized [2]. Our patient was also pregnant but she did not have any virilization. As there was an ongoing pregnancy, it was not possible to ascertain any clinical findings related to estrogenic activity of the tumor, even if there was.

Torsion of an ovarian tumor is a relatively common complication in pregnancy, occurring in about 15-20% of the cases [6, 8]. Most occur in the first trimester or in the puerperal period, related to the rapid growth or involution of the uterus [6, 8]. Hemorrhage or rupture, on the other hand, usually occurs during labor and is generally related with tumor size [1, 6]. In our patient, torsion of the tumor was diagnosed at the surgical exploration, but was not suspected beforehand. The patient had pelvic pain of moderate severity, however no findings indicating acute abdomen were revealed.

The best time to operate on a pregnant woman with an adnexal mass is around the 18th week of gestation [6, 7]. Besides the fact that many masses functional in origin disappear until this time, surgery at these gestational weeks is considered to be relatively safe for the fetus [7, 8]. During the surgery, 'hands off the uterus' is a rule and unnecessary manipulations must be avoided [6]. Close observation might be an alternative to antepartum surgery in some selected cases, especially in those with simple

cysts less than 5 cm in diameter [7, 8]. Normal or abnormal CA-125 levels should not change the approach to pregnant women [8].

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