An Unforeseeable Diagnosis in a Pregnant Woman with a Suspicious Adnexal Mass

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Abstract
Hereby, we present a rare case of 22 year-old pregnant woman with a suspicious adnexal mass coexisting multiple nodules on ovary, peritoneum and omentum diagnosed as disseminated peritoneal leiomyomatosis.

ABBREVIATIONS
DPL: Disseminated Peritoneal Leiomyomatosis

INTRODUCTION
Disseminated Peritoneal Leiomyomatosis (DPL) in the name of leiomyomatosis peritonealis disseminata is a rare disease that usually occurs in the reproductive aged women [1]. Hormonal, genetic and iatrogenic theories were defined as possible mechanisms in etiology [2]. An animal study revealed that, the influence of estrogen or both estrogen and progesterone promotes the differentiation of subcoelomic mesenchyme to smooth muscle-like and decidua-like cells and these cells were observed as subperitoneals nodules in macroscopic view [3]. Because of the prolonged elevation of estrogen and progesterone levels in pregnancy, pregnancy was suggested as a causation of DPL and this entity was identified in case reports in the literature [4,5]. Although the name of the disease connotes malignancy, it acts as benign except some cases developing malignancy [6].

Because of the rarity of disease and coexistence of DPL with other gynaecological conditions, we aimed to present a case of DPL in a pregnant woman with leiomyomatosis uteri and a suspicious adnexal mass.

CASE PRESENTATION
A 22 year-old pregnant woman with a complaint of persistent adnexal mass admitted to our outpatient clinic. She had no complaint in terms of abdominal swelling, abdominal pain or acute abdomen. In her history, an adnexal mass had been diagnosed at the sixth weeks of gestation and sonographic follow-up had been performed until the 13th weeks of gestation.

On physical examination an enlarged uterus and a right sided adnexal mass measuring 6 centimeters in diameter were palpated. Transvaginal ultrasonography revealed, a 13 weeks aged live fetus, a heterogenous adnexal mass on the right adnexa measuring 66x38x44 millimeters in diameter and a fundal leiomyoma measuring 42x32 millimeters in diameter were revealed. The mass was suspicious of malignancy because of the morphology and the low resistance blood flow on Doppler sonography. There were no ascites in the pelvic cavity. Hemotological, biochemical and tumor marker analysis were all in normal range.

Patient was advised to have magnetic resonance imaging examination but she declined and wished to learn the definite diagnosis as soon as possible. Therefore, the patient underwent a laparotomy with midline incision because of an adnexal mass that is suspicious for malignancy. During the exploration, the uterus was enlarged a size of a 14 weeks aged gestation. There were multiple of uterine nodules measuring nearly two centimeters in diameter in the biggest one. On the right ovary, 6 centimeters cyst with well surrounded multiple nodules above the cyst capsule was identified. The right pelvic peritoneum was also containing multiple nodules similar to the ones on the cyst capsule. These nodules were smooth surfaced and egg-shaped and the biggest one was measured 7 millimeters in diameter. There were no presence of free fluid in the pelvic cavity and the omentum was in normal appearance except adhesions. Peritoneal washings, a right salpingo-oophorectomy, removal of the peritoneal nodules on the right pelvic cavity and partial omentectomy were performed. On macroscopic view after removal, the cyst was containing a 3x2 centimeters in diameter solid nodule and on frozen section analysis, no malignant findings were found. On final microscopic view, there were well-defined smooth muscle nodules on peritoneum, ovary and omentum. All of the nodules were stained by desmin. The final pathology of the nodules on ovary, peritoneum and omentum revealed DPL. The cyst was diagnosed to be a follicular cyst. The patient had an uneventful...
recovery and was discharged on the second day of the operation. The follow-up was uneventful and after birth the patient and newborn were both healthy.

DISCUSSION

The gynaecological problems appearing in pregnancy cause trouble both for the pregnant and the clinician. Because of the enlarging uterus and in order to not to harm the fetus preserving the safety of the fetus, most of the clinicians may postpone the surgical interventions to dates after delivery. It is important to distinguish malignant lesions from benign ones before making a decision about a surgical intervention [7]. Because of the rarity of the disease and information confirmed by case reports, there is a challenge about management and follow-up of DPL in pregnancy.

There is not a perfect imaging modality to distinguish DPL from peritoneal carcinomatosis or benign conditions especially for the pregnant women. Therefore, the definitive prognosis should be confirmed by pathology [8].

In the previously reported papers, the proper treatment for reproductive aged patients was defined as removal of the nodules and omentectomy followed by cessation of the hormonal exposure [2]. Because of the lack of data for pregnant, the management remains unclear. Although there is data confirming spontaneous regression of disease after pregnancy, the development of malignancy in DPL has also been identified [6,9,10]. For the follow-up, the suggested technics after curative resection were computed tomography imaging and tumor marker testing annually for three years [11]. The influence of alteration in estrogen and progesterone levels during pregnancy on DPL was not shown and the follow-up during pregnancy was not defined.

In conclusion, clinicians should keep in mind the possibility of rare benign conditions coexisting with adnexal masses. Benign conditions may act as malignants and the use of frozen section analysis avoids unnecessary interventions during surgery.

REFERENCES