Conservative management of decidualized ovarian endometriotic cyst during pregnancy mimicking malignancy: case report and a review of the literature

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Summary

We report here the case of a 30-year-old woman with a decidualized ovarian endometriotic cyst (DOEC) during pregnancy mimicking malignancy occurring after fertility-preserving surgery for ovarian carcinoma arising from an endometriotic cyst. Intracystic excrescences appeared in the left ovarian endometriotic cyst at five weeks and three days of gestation. The serum CA-125 level rose to 676.7 U/ml (normal, 0-35). Based on these findings, ovarian carcinoma arising from the left ovarian cyst was strongly suspected. Frequent sonographic examinations revealed that the sizes and quality of the intracystic excrescences remained essentially unchanged. The cyst was evaluated for DOEC during pregnancy. The patient eventually delivered a male infant by normal vaginal delivery. After the delivery, the intracystic excrescences in the left ovarian endometriotic cyst disappeared. Close observation may be a reasonable alternative to antepartum surgery in patients with a DOEC during pregnancy.

Key words: Decidualization; Ovarian endometriotic cyst; Pregnancy; Endometrioma.

Introduction

Management of an ovarian tumor during pregnancy is complex and troubling, especially after fertility-preserving surgery for ovarian carcinoma. We were able to conservatively manage a case of decidualized ovarian endometriotic cyst (DOEC) mimicking malignancy occurring during pregnancy after fertility-preserving surgery for ovarian carcinoma arising from an endometriotic cyst. The literature regarding DOEC mimicking malignancy during pregnancy was reviewed [1-13].

Case Report

This patient was a 30-year-old woman, gravida-4, para-1, who had undergone laparoscopic oophorocystectomy for a right ovarian endometriotic cyst at the age of 21. Histologic examination of the endometriotic cyst revealed that it was benign. Thereafter, she did not undergo gynecologic follow-up examinations. At the age of 24 years, about three years after the first surgery, she visited our hospital complaining of abdominal swelling. Transvaginal ultrasonography and magnetic resonance imaging (MRI) examination revealed enlargement of the right ovary to about 12 cm in diameter and an endometriotic cyst with intracystic excrescences. Laparotomy revealed the diagnosis of right ovarian carcinoma arising from an endometriotic cyst, and right salpingo-oophorectomy, retroperitoneal lymphadenectomy, partial omentectomy and appendectomy were performed. A diagnosis was made of endometrioid adenocarcinoma of the right ovary, Stage I c(b) (pT1cN0M0). Cytology of the ascetic fluid was negative. The uterus and left adnexa were conserved. Thereafter, she successfully completed three courses of monthly paclitaxel and carboplatin combination chemotherapy, and underwent regular follow-up medical examinations. Four years later, she got married, and an endometriotic cyst in her left ovary grew in size. There were no abnormal findings on CT or MRI examinations, and the serum CA-125 level was within normal range. We interpreted the signs as being suggestive of a potential pregnancy, and she soon was found to be pregnant. Intracystic excrescences appeared in the left ovarian endometriotic cyst at five weeks and three days of gestation (Figures 1-4). The serum CA-125 level rose to 676.7 U/ml (normal, 0-35) (Table 1). Based on these findings and the history, ovarian carcinoma arising from the left ovarian cyst was strongly suspected. There was concern about whether or not surgical intervention might be necessary. However, frequent TV-ultrasonographic examinations revealed that the sizes and quality of the intracystic excrescences remained essentially unchanged. Therefore, we decided to follow-up the patient to assess the changes in the cyst, and the diagnosis was finally made of decidualization of an ovarian endometriotic cyst during pregnancy. Thereafter, the patient delivered a male infant weighing 3,045 g with an Apgar score of 9 at 37 weeks and five days of gestation. After the delivery, the intracystic excrescences in the left ovarian endometriotic cyst disappeared (Figure 4), and the serum CA-125 values returned to within normal range (Table 1). At present, five months after the delivery, the patient remains well.

Discussion

Ovarian carcinomas are a heterogeneous group of neoplasms and are traditionally subclassified based on type and degree of differentiation. It is becoming evident that each major histological type exhibits characteristic genetic defects that deregulate specific signaling pathways in the tumor cells. Moreover, among the most common histological types, ovarian carcinoma associated with endometriosis has recently received increasing attention, and numerous reports about this type of cancer

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Fig. 1

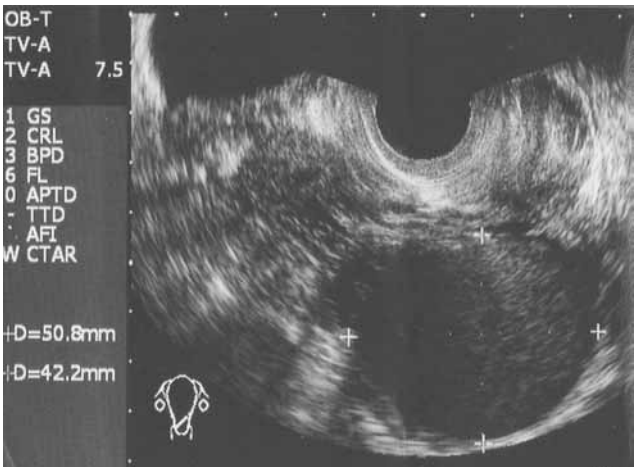

Fig. 2

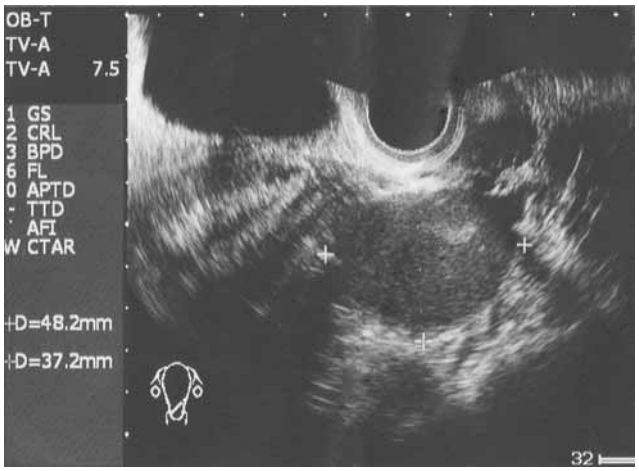

Fig. 3

Fig. 4

Figure 1. —TV-ultrasonographic findings of the left ovarian endometriotic cyst three months before pregnancy.
Figure 2. —TV-ultrasonographic findings of the left ovarian endometriotic cyst at 5 weeks/3 days of gestation; intracystic excrescences are observed.
Figure 3. — TV-ultrasonographic findings of the left ovarian endometriotic cyst at 12 weeks/3 days of gestation; intracystic excrescences are observed.
Figure 4: — TV-ultrasonographic findings of the left ovarian endometriotic cyst two months after the delivery; only one follicle is observed.

Table 1. — *Time-course of changes in serum CA-125 levels*.

CA-125 (U/ml) (normal value, < 35 U/ml)	
Before the operation for ovarian cancer: 210.7	
7 months before pregnancy:	33.4 (4 years after the operation for ovarian cancer)
3 months before pregnancy:	45.6
1 month before pregnancy:	67.6
5 weeks 3 days of gestation:	676.7
9 weeks 3 days of gestation:	173.2
12 weeks 3 days gestation:	129.4
16 weeks 3 days of gestation:	220.7
20 weeks 3 days of gestation:	255.8
27 weeks 3 days of gestation:	121.8 (5 years after the ovarian cancer operation)
31 weeks 5 days of gestation:	94.1
33 days after delivery:	39.8
2 months after delivery:	31.6
3 months after delivery:	26.9
4 months after delivery:	25.3

are being published in the literature. One of the characteristic findings strongly suggestive of ovarian carcinoma associated with endometriosis is the presence of intracystic excrescences in an ovarian endometriotic cyst. However, the detection of intracystic excrescences in an ovarian endometriotic cyst during pregnancy is often a difficult diagnostic issue. Malignant transformation of ovarian endometriotic cysts is well known. By contrast, DOEC during pregnancy mimicking malignancy is not well recognized, and is a challenging diagnostic entity. Adnexal masses (excluding the physiological corpus luteal cysts of early pregnancy) are seen in about 0.5% to 1.2% of all pregnancies, and 11% of these are endometriotic cysts [14]. Despite the relative high frequency of detection of ovarian endometriotic cysts during pregnancy, decidualization resulting in an appearance mimicking malignancy may be very rare. There are a few reports in the literature of DOEC during pregnancy [1-13]. In our literature review, we identified 13 studies

reporting on 25 cases of DOEC mimicking malignancy occurring during pregnancy [1-13]. The first case was reported in 1998 [1]. The subsequent reports were by Miyakoshi *et al*., 1998 (1 case), Tanaka *et al*., 2002 (1 case), Fruscella *et al*., 2004 (1 case), Sammour *et al*., 2005 (2 cases), Guerriero *et al*., 2005 (1 case), Iwamoto *et al*., 2006 (1 case), Asch and Levine, 2007 (1 case), Poder *et al*., 2008, (1 case), Takeuchi *et al*., 2008, (5 cases), Machida *et al*., 2008 (3 cases), Yoshida *et al*., 2008, (2 cases), Barbieri *et al*., 2009, (3 cases), and Ueda *et al*., 2010, (3 cases). Many such cases have undergone salpingo-oophorectomy during pregnancy on the basis of suspicious imaging findings, and the histologic examination of these ovarian cysts has revealed that the intracystic excrescences represented edematous vascularized decidualized endometrial tissue with abundant cytoplasm of the stromal cells. In retrospect, these surgical interventions might have been unnecessary. However, the differential diagnosis of a DOEC during pregnancy is difficult, and there are no definitive diagnostic guidelines. An ultrasonographic (US) guideline for DOEC during pregnancy will be necessary. We believe therefore that frequent US examinations before and during pregnancy are necessary in these patients. It is most important to have a high index of suspicion for this entity. Close observation may be a reasonable alternative to antepartum surgery in patients with a DOEC during pregnancy. The reporting of further cases of this entity should be encouraged, because analysis of even singular cases may provide important data for future research and development of treatment.

Serum CA-125 may be a useful marker for the diagnosis and monitoring of these cases. However, natural elevation of serum CA-125 during normal early pregnancy is a stumbling block. The present case may be the first of this entity occurring after fertility-preserving surgery for ovarian carcinoma arising from an endometriotic cyst.

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