

Chapter VI

Case Report

The First Case of Solitary Recurrence of Mucinous Borderline Ovarian Tumor in Uterus

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Abstract

This is the first case of solitary recurrency of mucinous borderline ovarian tumor presented in uterus. There currency appeared 8 months after conservative surgery (unilateral oophorectomy left). In this case, the individual risk factors associated with recurrency could have been the laparoscopy complicated by intraoperative cyst rupture, bleeding after adhaesiolysis with reduced visibility and the presence of dense adhaesions between tumor and left uterine site which can hypotetically permit the implantation of tumor cells under perimetrium in the place of previous adhaesiolysis.

The early detection of recurrency was possible using ultrasound which is currently the method of choice in follow-up of patients after conservative treatment of ovarian malignancies. There current tumor was treated by hysterectomy and the patientis in complete clinical response.

Keywords: Recurrence; Borderline ovarian tumor; Mucinous borderline tumor; Ultrasound; Fertility; Conservative surgery

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Introduction

Borderline ovarian tumors comprise about 15-20% of all epithelial ovarian malignancies^[1,2]. BOTs differ significantly from ovarian carcinomas with regard to the percentile distribution of tumor histotypes, lower FIGO stage, the generally excellent overall prognosis for patients and the younger age distribution.

The majority of BOTs are serous tumors (50%), followed by mucinous tumors (30-50%), and less common histotypes like seromucinous tumors (4.5-7.5%) and others (4%)^[3]. In the past seromucinous BOTs have been considered a subset of mucinous tumors (endocervical type or Müllerian type). However, seromucinous BOTs reflect the morphologic and behavioural features that are shared with serous tumors, and both may present with implants.

Mucinous BOTs (M-BOTs) compose of mild to moderately atypical gastrointestinal-type, mucin containing epithelial

cells that show proliferation greater than that seen in benign mucinous tumors but stromal invasion is absent^[4]. Mucinous BOTs are usually unilateral and they do not present with peritoneal implants, therefore the advanced stage of this subtype does not exist. If the bilaterality or the advanced stage are detected, careful examination of the appendix, intestine, pancreas, biliary tract, or cervix is warranted to exclude an occult extraovarian primary tumor simulating the primary ovarian mucinous borderline tumor with intraepithelial carcinoma^[5]. Similarly, mucinous BOTs associated with *pseudomyxoma peritonei* usually represent metastases from primary low-grade (adenomatous) mucinous tumors of the appendix^[6]. Alternatively to explain advanced stage of disease, it is also possible that foci of destructive invasion representing existing primary ovarian carcinomas were un sampled^[4,7].

The prognosis of M-BOT is excellent after excluding patients with *pseudomyxoma peritonei* and with other metastat-

ic carcinomas to the ovary, which can mimic mucinous BOTs. Only a few cases of progression to carcinoma have been reported and these tumours were not adequately sampled.

On the other hand, it has been clearly demonstrated that a higher recurrence rate of M-BOTs is related to the radicality of surgical procedure (ovarian cystectomy versus salpingo oophorectomy, RR = 5.545, P = 0,01) and surgical removal of entire ovary is the treatment of choice^[8]. Unilateral salpingo oophorectomy is an option because of the low frequency of bilaterality.

Borderline ovarian tumors frequently affect women in their reproductive age, and a more conservative surgery in order to preserve subsequent fertility is preferred. Laparoscopy is more frequently used in the group of conservatively treated patients. Laparoscopic management of borderline ovarian tumors is associated with a higher rate of cyst rupture and incomplete staging^[9]. In a review conducted by du Bois et al., it was observed that the recurrency rate was higher after laparoscopically performed conservative surgery than after surgery performed using the laparotomic approach (14.9 vs. 7.7%)^[10].

In addition, some authors have questioned the role of hysterectomy during primary staging surgery where conservative surgery is not intended or in cases where definitive surgery after completing childbearing is planned as no solitary recurrences in uterus have been observed^[11].

We presented the first case of uterine recurrency of mucinous borderline ovarian tumor in a patient primarily treated with unilateral salpingo oophorectomy. This case shows us that rather than making assumptions, the prognostic factors for recurrency must be assessed each time individually. Although this tumor histotype is characterized by benign behavior and in this case cystectomy was avoided, there were additional risk factors potentially contributing to the development of recurrency, such as laparoscopy complicated by intracystic tumor rupture, intraoperative bleeding and the tumor adhaesions to the uterus.

Case Report

The 37 years old patient was referred for laparoscopy due to palpable well encapsulated cystic left ovarian mass of 13 cm in diameter at the widest point. The laparoscopy was made in October 2006, complicated by intraoperative tumor rupture with the evacuation of mucin in the pelvis, bleeding followed adhaesiolysis of dense tumor adhaesions between tumor and the left uterine wall, which required a conversion of laparoscopy into open laparotomy to complete the unilateral salpingo oophorectomy. The histological report revealed mucinous borderline ovarian tumor and the patient was referred to a Gynecological Oncology Center for close follow-up.

Eight months after the surgery, the suspected recurrent multilocular tumor of size 58.5 x 48.1 x 52.5 mm with perfused septa (color score 3) located under the uterine perimetrium was found using ultrasound (April 2007). Theserum tumor markers - Ca 125, CEA and CA 19-9 were negative. The salvage surgery was performed in June 2007, however the performing surgeon described the tumor under uterine perimetrium as being an subserous myoma in the left uterine site and abandoned the surgery. The second scan repeated the findings of the first scan and a suspected recurrency, which impressed the myometrium and filled the uterine wall between perimetrium and uterine cavity, was again noted. The complementary pelvic MRI revealed the

same findings. After a consultation with the patient, where she was thoroughly informed of the details, she agreed to complete the definitive surgery (hysterectomy). It was the patient's own wish to complete the right salpingo oophorectomy at the same time, as she was already 37 and not involved in a relationship at the time. The definitive surgery was undergone in December 2007. The histological report of tumor found in uterine myometrium revealed a mucinous tumor composed of gastrointestinal type epithelium with atypical architecture and cytological features more marked than those seen in cystadenoma corresponding with a diagnosis of recurrent mucinous borderline ovarian tumor. The patient is in complete clinical response and is currently followed in our centre, her last visit run in April 2017.

Discussion

Over the past several decades, surgical therapy has shifted from a radical approach to more conservative treatment; however, oncologic safety must always be balanced. This is the first report of solitary recurrency of borderline ovarian tumor in uterus. Firstly, this report gives evidence that there might be individual risk factors for recurrence (like the effect of the chosen surgical approach and surgeon's experience, intraoperative complication, dense adhaesions) and secondarily it emphasizes the role of close and detailed follow-up based on gynecological ultrasound. These individual risk factors must also be taken into account when recommending completion of definite surgery after the fertility plans are finished.

This case report has opened discussion regarding certain problems which often come up during the management of oncological patients. Firstly, there is the problem of insufficient preoperative diagnosis, which interferes with the correct referral of the patient to an experienced oncologic surgeon, and secondly, the problem of the preference of gross intraoperative findings in opposition to obtaining detailed imaging techniques preoperatively, which can postpone the recurrent tumor diagnosis.

Firstly, the preoperative diagnosis of ovarian mass based on the pelvic examination has known limitations and may not allow discrimination of benign masses from malignant ones, especially if the malignant ones manifest as encapsulated lesions with a smooth surface^[12]. Ultrasound is broadly accepted as a highly accurate preoperative method in discriminating between benign and malignant adnexal masses^[13-15]. Mucinous BOTs are typically unilateral (>95%), large (20 to 22 cm), multilocular tumors (with > 10 locules), with a smooth inner lining, echogenic cyst fluid (low level or ground glass) and a smooth outer surface^[16,17]. In this patient, ultrasound might be of help not only to make a specific diagnosis, but also to detect adhaesions, the precise size of the tumor and to allow the choice of adequate surgical approach performed by an experienced surgeon. Ultrasound plays also an invaluable role in the follow-up of conservatively treated patients. Sonographic features of recurrent BOT within the retained ovary resemble those described by others for different subtypes of primary BOT^[18]. However, in this case report the recurrent tumor was found under the uterine serosa and presented with the same sonomorphologic and doppler characteristics as primary ovarian mucinous borderline tumor and was detected correctly and in a timely manner on ultrasound scan performed by an experienced sonographer.

Secondly, the finding from gross intraoperative eval-

uation of ovaries and/or uterus is absolutely irrelevant in cases with intact serosa (i.e. visceral peritoneum). In these cases the salvage surgery was abandoned as a surgeon expected the presence of subserous myoma instead of recurrent tumor. Ultrasound is currently the most effective modern imaging method to detect discrete intraovarian and/or intrauterine abnormalities if it is performed by an experienced examiner. The close cooperation among the sonographer dedicated to gynecological oncology and the gynecological surgeon is crucial in the management of gynecological cancer patients.

Conclusion

This is the first case of uterine solitary recurrency of mucinous borderline ovarian tumor after unilateral oophorectomy. Although the tumor histotype and the performance of unilateral oophorectomy would normally lead to the presumption of an excellent prognosis for the patient, there was the presence of individual risk factors which require close follow-up preferentially based on ultrasound.

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