Serous Borderline Tumor of the Fallopian Tube Presenting as an Ectopic Pregnancy: Case Report and Review of the Literature

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Clinical Practice Points

- Borderline tumors located in the fallopian tube are an uncommon entity.
- The literature describes 12 cases, most of which are unilateral and serous.
- They usually appear in young women who often have not fulfilled their procreative desire.
- The clinical diagnosis is nonspecific and is usually based on postoperative histology studies.
- The prognosis seems to be good, so in women with unfulfilled procreative desires, we might consider less aggressive and more conservative treatments.
- We report a patient with a symptomatic ectopic pregnancy who underwent salpingectomy.
- Histological examination revealed a borderline fallopian tube tumor.
- She subsequently underwent assisted reproductive techniques with a successful outcome.

Introduction

Borderline tumors located in the fallopian tube (serous tumors of low malignant potential) are rare tumors. These cancers are characterized by the absence of stromal invasion. To date, there are only 12 cases described in the English literature (Table 1). The diagnosis of these tumors is usually based on postoperative histology studies. The clinical features, therapeutic management, and prognosis of these tumors are poorly understood.

Case Report

This case involves a 34-year-old patient with a history of a left ectopic pregnancy who was treated with a single dose of methotrexate in 2007. At that time, primary infertility was diagnosed based on a hysterosalpingography of the left tubal obstruction. The patient was admitted to the emergency service in February 2009 for severe abdominal pain, hypovolemic shock, and a positive pregnancy test. An ultrasound revealed the existence of a right tubal pregnancy and severe hemoperitoneum. An emergency laparoscopy was performed to confirm the ultrasound diagnosis; a bilateral salpingectomy was performed because of the patient’s prior left tubal obstruction and primary sterility. A histopathologic study revealed a significant left tube without evidence of damage, a coexisting right tube, a tubal pregnancy, and a borderline serous epithelial tumor (Figure 1). The abdominal and pelvic postoperative computed tomography and serum CA 125 were all normal.

The patient declined the surgery because of an unfulfilled procreative desire. With the diagnosis of bilateral tubal factor, in vitro fertilization was indicated as an assisted reproductive technique. In October 2010, after 2 cycles of in vitro fertilization, pregnancy was achieved by using 0.05 mg of gonadotropin-releasing hormone analogues and 225 U of follicle-stimulating hormone daily during 10 days.

Six clusters were obtained, and a sperm survival rate of 85% and a total motile sperm count of 12 were calculated. Three type 8A embryos were transferred (at the request of the patient), and 2 were cryopreserved blast. The luteal phase support was achieved with 600 mg per day of micronized progesterone. Thirteen days after the transfer, the beta human chorionic gonadotrophin level was 516.9 U/L. The existence of a twin-diamniotic dichorionic intrauterine gestation was confirmed on ultrasound. An ultrasound at the 12th gestational week showed aneuploidy markers in
the second twin: a nuchal translucency of 3.1 mm (<p90) and an 11-mm omphalocele.

The combined hazard ratio confirmed that the first twin was normal and predicted a high risk that the second twin would be affected by both trisomy 18 (1/1) and trisomy 21 (1/9). Chorionic villus sampling was performed, and normal results were obtained for the first twin, who was a “cytogenetically normal male 46, XY,” whereas the second twin was “a female with trisomy 18 (47XX), Edwards syndrome.” The patient chose not to pursue the selective disruption of the second twin.

At week 29, spontaneous amniorrhexis occurred and after antibiotic and corticosteroid treatment for fetal lung maturation, an emergency cesarean section was performed to reduce the risk of fetal loss and to maximize the well-being of the healthy twin. The first twin was a healthy male and the second twin was an affected female with Edwards syndrome, who died within the first hours after birth. Currently, the surviving twin is healthy, without defects or alterations in psychomotor development.

Thirty days after delivery, complete staging surgery was performed, including a laparoscopic peritoneal lavage for cytology, a right adnexectomy, peritoneal biopsies, and omentectomy. The histopathologic results did not show evidence of malignancy.

**Discussion**

Borderline tumors located in the fallopian tube are very rare tumors. Based on the few reported cases in the literature, these tumors usually appear in young women with a mean age of 32 years (range, 3-60 years); in many cases, the procreative desire of these women has

<table>
<thead>
<tr>
<th>Author</th>
<th>Year</th>
<th>Age</th>
<th>Presentation</th>
<th>Procedure</th>
<th>Gross (cm)</th>
<th>Micro</th>
<th>CA125 (U/ml)</th>
<th>Localization</th>
<th>Follow-Up (months)</th>
</tr>
</thead>
<tbody>
<tr>
<td>McCarthy⁵</td>
<td>1988</td>
<td>60</td>
<td>Constipation, Abdominal pain</td>
<td>LT: Subtotal hysterectomy, BSO</td>
<td>NR</td>
<td>Mucinous + Pseudomyxoma peritoneal</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
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<tr>
<td>Seidman⁷</td>
<td>1994</td>
<td>53</td>
<td>Pelvic mass</td>
<td>LT: TH, BSO</td>
<td>R: 2.8 L: 1.8</td>
<td>Bilateral Mucinous</td>
<td>NR</td>
<td>NR</td>
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<tr>
<td>Zheng⁸</td>
<td>1996</td>
<td>31</td>
<td>Abdominal pain</td>
<td>LC: SO, contralateral ovary biopsy, MB, node sampling, washings, PO</td>
<td>6</td>
<td>Serous papillary</td>
<td>108</td>
<td>All of the tube</td>
<td>72 DF</td>
</tr>
<tr>
<td>Alvarado-Cabrero²</td>
<td>1997</td>
<td>34</td>
<td>Incidental finding during surgery</td>
<td>NR</td>
<td>1.7 3</td>
<td>Serous Endometrioid</td>
<td>NR</td>
<td>Fimbriated end</td>
<td>28 DF recent case</td>
</tr>
<tr>
<td>Kayaalp¹⁰</td>
<td>2000</td>
<td>31</td>
<td>Adnexal mass on routine exam</td>
<td>LT: S</td>
<td>13</td>
<td>Serous</td>
<td>NR</td>
<td>All of the tube</td>
<td>NR</td>
</tr>
<tr>
<td>Haratz-Rubinstein¹¹</td>
<td>2004</td>
<td>28</td>
<td>Abdominal pain</td>
<td>LC: S</td>
<td>5</td>
<td>Serous FIGO IA</td>
<td>35</td>
<td>NR</td>
<td>NR</td>
</tr>
<tr>
<td>Villella⁹</td>
<td>2005</td>
<td>22</td>
<td>Abdominal pain</td>
<td>LC: Tubal cystectomy, LT: TH, BSO, O, A, PLND</td>
<td>3.4</td>
<td>Serous</td>
<td>Neg</td>
<td>NR</td>
<td>36 DF</td>
</tr>
<tr>
<td>Krasevic³</td>
<td>2005</td>
<td>34</td>
<td>Adnexal mass on routine exam</td>
<td>LC: S, washing</td>
<td>4.4</td>
<td>Serous</td>
<td>5.1</td>
<td>Ampular</td>
<td>55 DF</td>
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<tr>
<td>Seamon⁴</td>
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<td>12.5</td>
<td>Serous, FIGO IC</td>
<td>NR</td>
<td>Ampular</td>
<td>11 DF</td>
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<tr>
<td>Abreu¹</td>
<td>2011</td>
<td>25</td>
<td>Abdominal pain Adnexal mass</td>
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<td>5</td>
<td>Serous, FIGO IA</td>
<td>43.6</td>
<td>External 1/3</td>
<td>48 DF</td>
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<td>Stephens¹²</td>
<td>2010</td>
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<td>Abdominal mass palpable</td>
<td>LT: SO</td>
<td>7.3</td>
<td>Serous</td>
<td>NR</td>
<td>NR</td>
<td>DF</td>
</tr>
<tr>
<td>Ondic⁵</td>
<td>2011</td>
<td>41</td>
<td>Bad pain, dyspareunia, dysuria</td>
<td>LC: BSO, TH, PO, partial, Douglas peritoneectomy</td>
<td>7</td>
<td>Serous papillary with non-invasive implants</td>
<td>NR</td>
<td>Fimbria</td>
<td>6</td>
</tr>
<tr>
<td>Our Case</td>
<td>2012</td>
<td>34</td>
<td>Abdominal pain, hypovolemic shock for ectopic gestation</td>
<td>LC: BS</td>
<td>NR</td>
<td>Serous</td>
<td>NR</td>
<td>NR</td>
<td>34 DF</td>
</tr>
</tbody>
</table>

**Table 1 Literature review of borderline tumors of the fallopian tube**

Abbreviations: LT = laparotomy; LC = laparoscopy; SO = salpingo-oophorectomy; TH = total hysterectomy; BSO = bilateral salpingo-oophorectomy; MB = multiple biopsies; PO = partial omentectomy; O = omentectomy; A = appendix; S = salpingectomy; BS = bilateral salpingectomy; PLND = pelvic lymphadenectomy; PPALND = pelvic and para-aortic lymphadenectomy; RD = recently diagnosed; NR = not reported; DF = disease free; R = right; L = left.
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not yet been fulfilled. These tumors are usually unilateral and are located in the outer third of the tube (the ampullary region and the end of the fimbria).\(^1\)\(^-\)\(^5\) The patients usually present with abdominal pain, and an adnexal mass is discovered on routine examination or upon surgical exploration.\(^2\) In most cases, the diagnosis is postsurgical and histologic.

The most common histologic type is serous; however, mucinous tumors\(^6\) and peritoneal pseudomyxomas\(^6\) have also been reported. In another described case, the patient had bilateral involvement.\(^7\) The histologic differential diagnosis should include the following: pseudocarcinomatous hyperplasia, epithelial papilloma, papillary tumor, and metaplastic carcinoma.\(^8\) Because the diagnosis is made after surgery, the cancer antigen 125 level was evaluated in only 3 cases; it was normal in 1 case,\(^9\) and elevated in the other 2 cases.\(^1\)\(^,\)\(^8\) The elevation of this marker could be due to an increased epithelial tumor component, which would support a probable Müllerian origin.\(^8\)

There is no consensus on the optimal surgical treatment; surgical approaches have ranged from a simple salpingectomy, with or without surgical staging, to a complete surgery similar to that performed for tubal cancers. However, neither metastases nor invasive peritoneal implantations (1 case of noninvasive implantation)\(^3\) at the time of diagnosis have been described; in addition, no peritoneal recurrences have been reported. The disease-free interval is widely variable, with an average of 39 months. Because these tumors appear in young women of reproductive age, less aggressive and more conservative treatments should be considered.

We report a unique case that involved a woman with a symptomatic ectopic pregnancy who underwent emergency surgery, which consisted only of a salpingectomy; the histologic findings were surprising. The patient had not fulfilled her procreative desire, so we decided to begin treatment with assisted reproductive techniques, which were successful. One of the possible contraindications to these techniques is malignancy.

Borderline ovarian tumors have a different prognosis and overall survival than malignant tumors and, except for micropapillary serous borderline tumors, they rarely recur. Therefore, when indicated, it seems reasonable to use assisted reproduction techniques after the excision of the attached tumor and to defer staging surgery until pregnancy is achieved.

**Conclusion**

The natural history of borderline tumors located in the fallopian tube is still unknown, yet the unilateral salpingectomy seems to be the optimal treatment for women who still desire to have children. We believe that a greater number of published cases are necessary to better understand the treatment and prognosis of this rare entity.
Disclosure
The authors have stated that they have no conflicts of interest.

References