Ultrasound Diagnosis of Serous Surface Papillary Borderline Ovarian Tumor: A Case Series with a Review of the Literature

Manuela Ludovisi, MD, PhD, Xulin Foo, MD, Sara Mainenti, MD, Antonia Carla Testa, MD, PhD, Rupali Arora, MD, Davor Jurkovic, MD, FRCOG

1 Gynaecology Diagnostic and Treatment Unit, University College Hospital, London, UK
2 Department of Obstetrics and Gynecology, Catholic University of the Sacred Heart, Rome, Italy
3 Department of Hystopathology, University College Hospital, London, UK

Received 13 December 2013; accepted 16 November 2014

ABSTRACT: Serous surface papillary borderline ovarian tumors (SSPBOTs) are a rare morphologic variant of serous ovarian tumors that are typically confined to the ovarian surface, while the ovaries themselves tend to appear normal in size and shape. In this report, we describe the findings from five premenopausal women diagnosed with SSPBOTs, in whom ultrasound showed grossly normal ovaries that were partially or wholly covered with irregular solid tumors. In all five cases, histologic examination showed evidence of borderline serous tumors. These findings demonstrate that SSPBOTs can be diagnosed on a preoperative sonographic examination, which could facilitate conservative, fertility-sparing surgery in young women affected by this condition.

INTRODUCTION

Borderline ovarian tumors (BOTs) account for approximately 15% of all epithelial ovarian tumors. One-third of those occur in women younger than 40 years old, who often wish to preserve their reproductive capacity. Serous surface papillary BOTs (SSPBOTs) are a distinct subtype of serous ovarian tumors. They are typically confined to the ovarian surface, while the ovaries themselves tend to appear normal in size and shape.

Serous BOTs have a generally good prognosis, and conservative fertility-sparing surgery needs to be considered in all women of fertile age, preferably by using a laparoscopic approach. It is sometimes difficult to differentiate between borderline and invasive epithelial tumors intraoperatively, so an accurate preoperative diagnosis is essential for optimizing patient management.

Ultrasound (US) is routinely used for the assessment of women with adnexal tumors. Various studies have described the morphologic characteristics of serous borderline tumors on US, which typically appear as cystic ovarian lesions with papillary projections. In comparison, only a few reports have described SSPBOTs using different imaging modalities.

In this report, we describe the cases of five women with SSPBOTs that were all correctly classified as serous BOTs on preoperative US examinations.

CASE REPORTS

Over a period of 3 years, we identified five women with a diagnosis of SSPBOT. The indications for examination were suspected ovarian tumors on previous US or MRI examinations in three women, oligomenorrhea and clinical suspicion of polycystic ovary syndrome in one
woman, and abdominal distension and pain in the fifth woman. The patients’ clinical symptoms are listed in Table 1. All women were premenopausal, nulliparous, and in good general health, and none of them had a family history of ovarian, breast, or other cancers. Three of the women had CA125 concentrations >35 kIU/l (Table 1).

In all cases, the transvaginal US examination of the pelvis was performed by using a 5–7-MHz transvaginal transducer and a scanner with three-dimensional facility (Voluson E8; GE Healthcare Ultrasound, Milwaukee, WI). US revealed bilateral adnexal tumors in four women and a unilateral lesion in one woman. All women had at least one adnexal lesion manifesting the following features: solid tumor with irregular surface surrounding a normal ovary or an ovary containing a small cystic lesion with papillations. These small cystic lesions were suggestive of, although not typical of, small borderline lesions (Table 1). All tumors were poorly to moderately vascular on Doppler US examination (Figure 1A and 1B). Two women (cases 4 and 5) also had large cysts in the ovary contralateral to that containing the lesions just described. These had the typical appearance of serous BOTs, and normal ovarian parenchyma was seen adjacent to the cyst in both cases (ie, positive ovarian crescent sign).

All women underwent surgery, but none of them required adjuvant chemotherapy (Table 1). Two women attended for follow-up visits. One of them (case 1) had no signs of tumor recurrence for 4 years, and she then conceived spontaneously. Her pregnancy was uncomplicated, and she had a vaginal term birth. Another woman (case 5) had been followed up for 6 months and was found to have a recurrent right serous borderline tumor. She underwent a laparoscopic right salpingo-oophorectomy. At surgery, her left ovary was found to be normal, with no signs of a recurrent SSPBOT.

DISCUSSION

In this series, we present the cases of five women who were diagnosed with SSPBOTs. In all cases, at least one ovary appeared normal grossly but was covered with tumor deposits confined to its surface. These features are specific to this type of tumor, and they facilitate its correct preoperative detection on US. The appearance of SSPBOTs differ from those of the majority of borderline serous tumors, which tend to grow within the ovaries, causing their enlargement. However, in the two women in our series who had bilateral lesions, one of the ovaries manifested typical features of borderline tumor, which helped to achieve a correct preoperative diagnosis. On US examination, the SSPBOTs appeared as irregular solid lesions, which were poorly to moderately vascular on Doppler US examination and surrounded normal ovarian parenchyma. The appearance of the solid tumor component was nonspecific, and the only finding that facilitated its differentiation from other malignant lesions was the presence of normal ovaries—or at least some normal ovarian tissue—completely or partially surrounded by solid tumor (Figure 1A and 1B). On histologic examination, four of our patients had a few small microscopic foci suggestive of low-grade serous invasive cancer, but these findings did not have a significant effect on the management of the disease (Figure 1C).

Kim et al reported a case of SSPBOT that manifested with bilateral ovarian masses that had irregular, nodular, or papillary margins and internal branching patterns on MRI examination. Within the tumors, normal-appearing ovaries containing multiple follicles could be clearly discriminated. Another MRI study described six cases of SSPBOT that showed similar features of solid masses covering normal ovaries. In all of their cases, those authors found a “sea anemone–like papillary architecture and internal branching pattern.” These features are signs of an orderly hierarchical pattern of papillary formation that reflects the well-differentiated nature of the tumor. They are considered typical and helpful diagnostic findings.

We found only two single case reports that describe the US findings in women with SSPBOTs. Burkholz et al described the transabdominal US findings in one case of SSPBOT in a 14-year-old girl, which showed multiple solid and cystic areas and prominent vascularity. The appearance of this tumor differed from those we found, which could reflect some limitations of a transabdominal US examination for the assessment of pelvic tumors. In the other single report, Kwon et al described the case of a patient with lobulated and polypoid exophytic tumors on the surface of seemingly normal ovaries, which is very similar to our findings.

Identification of normal ovarian tissue adjacent to ovarian tumors (ie, the ovarian crescent sign) has been shown to facilitate the differentiation between BOT and invasive ovarian cancer. Our case series shows that this finding is also helpful...
<table>
<thead>
<tr>
<th>Case No.</th>
<th>Age (years)</th>
<th>Symptoms</th>
<th>Sonographic Findings</th>
<th>CA125 (kIU/l)</th>
<th>CT Findings</th>
<th>Operation</th>
<th>Histology</th>
<th>Follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>29</td>
<td>Oligomenorrhea</td>
<td>Normal ovary surrounded by a 49 × 43 × 43-mm solid tumor</td>
<td>N/A</td>
<td>N/A</td>
<td>Laparoscopic right oophorectomy</td>
<td>Serous borderline micropapillary tumor</td>
<td>No evidence of recurrence</td>
</tr>
<tr>
<td>2</td>
<td>32</td>
<td>Primary infertility</td>
<td>A 15 × 15 × 11-mm unilocular/solid cyst in the lateral pole; entire ovary surrounded by a 53 × 53 × 44-mm solid tumor</td>
<td>Yes</td>
<td>Yes</td>
<td>Peritoneal deposits</td>
<td>Bilateral borderline serous tumors with invasive and noninvasive peritoneal deposits</td>
<td>No evidence of recurrence</td>
</tr>
<tr>
<td>3</td>
<td>35</td>
<td>Abdominal distension and pain</td>
<td>A 14 × 13 × 12-mm unilocular/solid cyst in the lateral pole; entire ovary surrounded by an 87 × 71 × 44-mm solid tumor</td>
<td>Yes</td>
<td>No</td>
<td>Small-volume peritoneal deposits</td>
<td>Bilateral borderline serous tumors with microscopic foci of low-grade serous carcinoma, invasive and noninvasive peritoneal deposits</td>
<td>No evidence of recurrence</td>
</tr>
<tr>
<td>4</td>
<td>26</td>
<td>Abdominal pain</td>
<td>Normal ovary surrounded by an 80 × 66 × 51-mm solid tumor</td>
<td>Yes</td>
<td>Yes</td>
<td>Open left ovarian cystectomy, resection of right adnexal tumor</td>
<td>Bilateral borderline serous tumors with microscopic foci of low-grade serous carcinoma</td>
<td>No evidence of recurrence</td>
</tr>
<tr>
<td>5</td>
<td>36</td>
<td>None</td>
<td>A 68 × 54 × 42-mm unilocular solid lesion with multiple papillary projections</td>
<td>No</td>
<td>No</td>
<td>Laparoscopic right ovarian cystectomy, resection of left adnexal tumor</td>
<td>Bilateral borderline serous tumors</td>
<td>Recurrence of right serous borderline tumor after 6 months of follow-up</td>
</tr>
</tbody>
</table>
in cases of SSPBOT that manifest with superficial ovarian tumors.

The preoperative detection of SSPBOT could have a considerable positive effect on the management of the disease in younger women in whom the preservation of reproductive capacity is critical. At surgery, SSPBOTs tend to appear as solid, irregular lesions that are indistinguishable from invasive epithelial and metastatic tumors (Figure 2). Prior knowledge of the likely tumor type and the presence of normal ovaries—or at least some normal ovarian tissue—in its center is likely to facilitate attempts to conserve the ovaries at surgery. Although three women in our series had evidence of microscopic invasion, the bulk of the tumor was borderline, and the presence of microinvasion did not change the management of the disease in these women. However, there are no data on reproductive and other outcomes following fertility-sparing surgery in cases of SSPBOT.

The five cases of SSPBOT in our series were selected because of their unique presentation on US examination. For all five women, we recorded their operative and histologic findings. In view of that, we are unable to comment on the sensitivity of US diagnosis of SSPBOT, but our series indicates that the findings are likely to be highly specific. Prospective studies are needed to confirm that and to assess the sensitivity of US diagnosis of SSPBOT. Long-term follow-up data are also needed to obtain information about reproductive and other health outcomes after fertility-sparing surgery.

REFERENCES


FIGURE 1. (A) Gray-scale sonogram shows a normal ovary (O) surrounded by solid tumor (arrows) in a woman with SSPBOT (case 2). (B) On Doppler US examination, the same tumor (arrows) appeared relatively avascular in comparison with the ovary (O). (C) Histologic examination of the tumor shows a serous surface papillary borderline tumor (arrows) on the surface of a normal ovary (O) (hematoxylin-eosin stain; original magnification, ×200).

FIGURE 2. Photograph shows the intraoperative findings in a case of serous borderline tumor (case 5).