Case Report

Primary Ovarian Leiomyoma Associated with Multiple Uterine Leiomyomas

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Abstract

Ovarian leiomyomas are very rare. We report the case of a primary ovarian leiomyoma accompanied by multiple uterine leiomyomas. A 50-year-old woman was referred to our department for heavy menstruation, and a hot spot in the uterine lumen was observed on positron emission tomography–computed tomography (PET-CT). Cervical and endometrial cytology and tumor marker tests were negative. Pelvic magnetic resonance imaging revealed an endometrial polyp and submucosal leiomyoma in the uterine lumen and a 5-cm right ovarian tumor. Laparoscopic total hysterectomy, right salpingo-oophorectomy, and left salpingectomy were performed for radical treatment. Histopathology showed that ovarian tumors contained interlacing bundles of fusiform cells encircled by normal ovarian tissue. Immunohistochemical staining showed strong and diffuse positive staining for α-smooth muscle actin. We diagnosed the tumor as a primary ovarian leiomyoma because the leiomyoma was localized in the ovary and was larger than the size of uterine leiomyomas. No metastatic lesion was found on PET-CT. There was no tumor recurrence at the 6-month follow-up.

Keywords: Leiomyoma, ovarian tumor, tamoxifen

Introduction

An ovarian leiomyoma is a very rare tumor that accounts for only 0.5%–1% of all benign ovarian tumors.[1] Fewer than 100 cases have been reported. According to previous reports, ovarian leiomyomas are benign, unilateral, asymptomatic tumors that occur in premenopausal women.[2,3] They exhibit no specific symptoms and require discrimination from thecomas and fibromas on imaging.[2,3] Diagnosis is only confirmed by pathological tests. An ovarian leiomyoma is usually discovered during pelvic examination or surgery. We report the case of a primary ovarian leiomyoma discovered together with uterine leiomyomas on imaging.

Case Report

A 50-year-old gravida 2 para 2 woman presented at our hospital for a regular checkup for breast cancer. She had a family history of breast cancer on her maternal side and had undergone surgery for right breast cancer at the age of 43. For the past 6 years, she had taken tamoxifen and was followed up for cervical cytology. She also had childhood asthma and an appendectomy at the age of 12. She had complained of excessive menstruation, and a hot spot in the uterine lumen was detected using positron emission tomography–computed tomography (PET-CT) at the breast surgery department. She was referred to the obstetrics and gynecology department for further examination and treatment.

Physical and abdominal examination findings were normal. The results of the pelvic examination finding were also normal. Transvaginal ultrasonography revealed a 21.8-mm uterine mass and an enlarged right ovary. Cervical cytology was negative for malignant intraepithelial lesions. Endometrial cytology was negative. Her blood test showed a hemoglobin level of 8.7 g/dL and hematocrit of 29.2%. Laboratory findings such as platelet count and solidification

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systems were normal, and tumor markers including CA-125 and CA-19-9 were negative.

Pelvic magnetic resonance imaging (MRI) was performed to examine the uterine mass, which revealed an endometrial polyp and submucosal leiomyoma in the uterine lumen and a 5-cm right ovarian tumor [Figure 1]. The MRI signal pattern suggested that the right ovarian tumor was a fibroma or thecoma. Thus, the cause of anemia was thought to be due to the endometrial polyp and submucosal leiomyoma. Therefore, she underwent laparoscopic surgery under the clinical diagnosis of endometrial polyp, submucosal leiomyoma, and right ovarian tumor for polyp, leiomyoma, and tumor excision, respectively.

Laparoscopy showed that the right ovarian tumor was approximately 5 cm in diameter, a white solid tumor, suggestive of a fibroma or thecoma. The uterus was slightly enlarged. Laparoscopic total hysterectomy, right salpingo-oophorectomy, and left salpingectomy were performed. Tissues were excised out vaginally. Uterus and ovarian tumor weights were 276 g and 57 g, respectively. Surgery time was 163 min, and the amount of bleeding was 100 ml.

Histopathological examination revealed polyp-like ridges containing interlacing endometrial glands with mild alignment disorder and endometrial stroma in the uterine endometrium. There were ectopic endometrial tissues in the uterine muscle layer and serosal surface and some uterine intramural leiomyomas (maximum 6-mm diameter). The ovarian tumor measured 5.5 cm × 3 cm × 3 cm, and the cut section of the tumor showed a hard gray-white solid area which was well encapsulated [Figure 2]. Furthermore, the ovarian tumor contained interlacing bundles of fusiform cells with wide fibrosis and hyalinization. The cell density of the ovarian tumor was low. Nuclear atypia, pleomorphism, and necrosis were absent. The mitotic count was <1/10 high-power fields [Figure 2]. Immunohistochemical staining showed strong and diffuse positive staining for α-smooth muscle actin (α-SMA) [Figure 2].

The diagnosis was endometrial polyps, uterine adenomyosis, and intramural leiomyomas and an ovarian leiomyoma. The ovarian leiomyoma was larger than any other uterine leiomyomas, and there were no findings of metastatic leiomyomas on PET-CT. Therefore, the final diagnosis was a primary ovarian leiomyoma. The postoperative period was uneventful. Her blood test showed a hemoglobin level of 6.0 g/dL 1 day after surgery and 8.3 g/dL with iron administration 1 week after surgery. At the 6-month follow-up, the patient had no complaints and no signs of tumor recurrence.

Discussion

Ovarian leiomyomas account for 0.5%–1% of all benign ovarian tumors.[1] The majority of these tumors are unilateral and are discovered incidentally during pelvic examination, imaging, or surgery.[2,3] These occur in women between the ages of 20–65 years, especially in perimenopausal women.[2,3] If these tumors become large, ovary torsion, elevated CA-125 levels, and Meigs syndrome may occur.[1] Some leiomyomas grow rapidly in the context of high blood levels of estrogens such as during early pregnancy.[4] In the present case, the patient was treated with tamoxifen at perimenopausal phase, with unilateral tumor, and had no symptoms associated with the ovarian tumor that was discovered incidentally on imaging.

Histopathologically, an ovarian leiomyoma contains interlacing bundles or spindles of fusiform cells, and nuclei of these cells are oval and have poor atypia and mitosis similarly to uterine leiomyomas.[3] The same pathological histological findings were seen in the present case. The diffuse strong positive staining for α-SMA is valuable for the diagnosis of a leiomyoma.[3,5] In this case, we diagnosed an ovarian leiomyoma from pathological histological findings coincident with leiomyomas and positive staining for α-SMA and the location of the tumor being in the ovary.

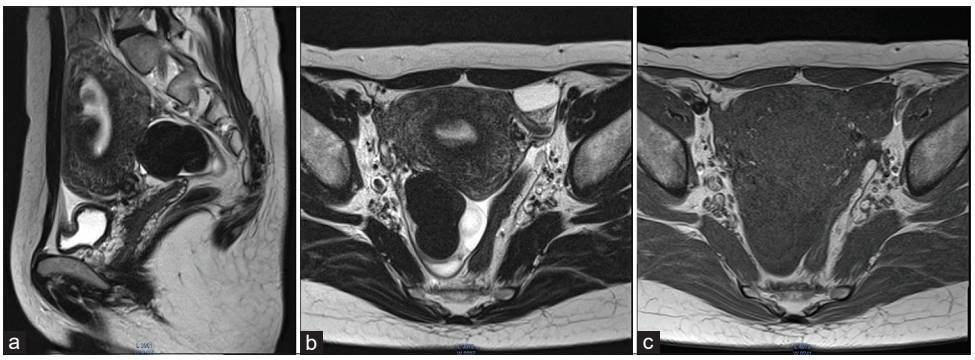

Figure 1: (a-c) Pelvic magnetic resonance imaging. (a) Sagittal T2-weighted imaging. (b) Axial T2-weighted imaging. (c) Axial T1-weighted imaging. Magnetic resonance imaging showing a slightly enlarged uterus due to a mass that projected into the uterine cavity. The mass is of low intensity (a-c). The mass in the right ovary is of low intensity (b and c) and 5.5 cm in diameter

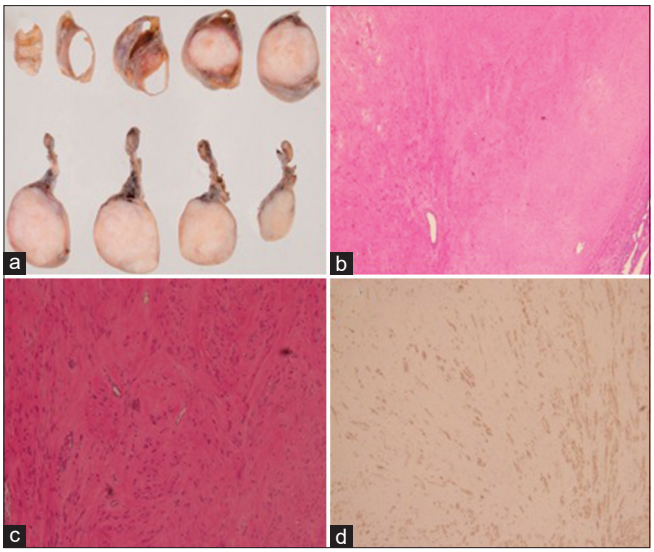

Figure 2: Continuous cross-sectional views of the right ovarian tumor (a), histopathological specimen of the right ovarian tumor stained using hematoxylin and eosin (b and c) or α-smooth muscle actin (d). (a) The tumor is completely encircled by the ovarian capsule. (b) The tumor has a clear border with ovarian stroma. (c) The tumor contains an increased number of spindle cells showing less atypia. (d) Marked immunoreactivity for α-smooth muscle actin

Ovarian leiomyomas show low signal intensity on T1- and T2-weighted MRI and early enhancement on contrast MRI.[6,7] Fibromas or thecomas – high-frequency, solid, benign, ovarian tumors – also show low signal intensity on T1- and T2-weighted MRI and poor enhancement on contrast MRI. In the present case, contrast MRI was not performed and the fibroma or thecoma was initially stated as a differential diagnosis. Contrast MRI may help to differentiate an ovarian leiomyoma; however, histopathological examination is necessary to definitively diagnose ovarian leiomyomas.

Furthermore, some sources suggest that tamoxifen, which is used as an adjuvant hormone therapy for breast cancer, causes growth of not only uterine leiomyomas but also extrauterine tumors.[4,8,9] Therefore, if an ovarian tumor is detected on imaging with findings suggestive of fibroma or thecoma, especially in patients treated with tamoxifen, an ovarian leiomyoma should be considered as a potential diagnosis, and subsequent imaging should be performed to examine for metastasis because leiomyomas have the potential of metastasis, unlike fibroma and thecoma.[10]

The coexistence of an ovarian and a uterine leiomyoma is rare. If coexistence is recognized, the uterus and ovaries should be examined for metastasis.[10] In our case, we recognized the coexistence. The ovarian tumor was clearly a leiomyoma histologically; it was completely encircled by the ovarian capsule and larger than any other uterine leiomyomas, with no metastatic findings on PET-CT or MRI; therefore, we diagnosed it as a primary ovarian leiomyoma.

In conclusion, we present a very rare case of a primary ovarian leiomyoma accompanied by a uterine leiomyoma. Ovarian leiomyomas are often discovered incidentally. If the findings on imaging are suggestive of fibroma or thecoma, especially in patients taking hormone therapies that may affect estrogen-dependent tumors, further examination should include the possibility of metastasis with the possibility of diagnosing ovarian leiomyoma.

Ethical approval

This case report was approved by the ethics committee of our university (approval number: 201877).

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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