Benign Metastasizing Leiomyoma of the Uterus

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Herein, we report on a well-characterized benign metastasizing leiomyoma, presented in an unusual site. Up to the knowledge of authors, so far, only 76 cases of benign metastasizing leiomyoma have been reported. The tumor presented as a retroperitoneal mass three years after a hysterectomy performed for leiomyomatosis of the uterus with extensive areas of hyalinization. Histopathologic and immunohistochemical studies of the resected mass were similar to the uterine leiomyoma, showing moderate cellularity of bland looking smooth muscle cells with minimal atypia, inconspicuous mitosis, and no necrosis in a hyalinized background.

Keywords: Benign metastasizing leiomyoma • retroperitoneum • uterus

Introduction

Benign metastasizing leiomyoma (BML) of the uterus is a rare albeit well-recognized entity, which has been reported in the medical literature up to 76 cases. Most of these cases were described with pulmonary nodules. Careful follow-up of these patients is recommended, since in spite of their benign appearance, they show a low-grade clinical malignant behavior. Herein, we present a patient with retroperitoneal involvement. Only a few cases of BML are reported in this site.

Case Report

A 50-year-old woman was admitted to our hospital for abnormal uterine bleeding. She underwent total abdominal hysterectomy and bilateral salpingo-oophorectomy for uterine myomas. On macroscopic examination, the myometrium contained multiple myomas, the greatest of which had a diameter of 15 cm. Areas of complete degeneration filled with blood and some papillary projections were also included. The clinical and macroscopic examinations suggested leiomyoma, although the final diagnosis was deferred for additional samples to rule out leiomyosarcoma.

Three years after hysterectomy, she was admitted again to our hospital for evaluation of an abdominal mass. On ultrasonography, a solid cystic retroperitoneal mass with mixed echo was identified on the upper pole of the right kidney, which was clinically and radiographically suspicious for malignancy. The patient underwent laparotomy. Grossly, the retroperitoneal mass was a large solid cystic structure which measured $13 \times 11 \times 8.5$ cm that was partially embedded in its surrounding fatty tissues and attached to the right kidney capsule and duodenal serosa. The external surface of the mass was grayish brown and smooth. On opening, it was a thin-walled multilocular cyst, the diameter of its largest chamber was 8 cm. The solid component was creamy, fleshy and demonstrated areas of cystic degeneration.

Pathologic findings

Microscopic examination of ten blocks prepared from the resected uterine mass showed leiomyomas. Extensive areas of hyalinization were interspersed between bundles of benign looking...
smooth muscle cells showing moderate cellularity, minimal atypia, inconspicuous mitosis, and no evidence of necrosis. Based on histopathologic criteria, it was considered as benign leiomyomata with hyaline degeneration.

Slides of the retroperitoneal mass revealed a spindle cell tumor that was histopathologically similar to the uterine myomas removed three years before (Figures 1 and 2). For definite diagnosis, a panel of immunohistochemical stainings (DAKO LSAB2 Kit/HRP) was requested for both the previous and the current masses, which revealed identical immunoreactivities including positive results for SMA, desmin, vimentin, NSE, ER, and PR, and negative results for S100, CD 34, CK AE1/AE3, EMA, and C-Kit. The immunohistochemical studies confirmed that the tumor was a mesenchymal derivation with smooth muscle differentiation and ruled out the possibility of epithelial origin and gastrointestinal or endometrial stromal tumors. The right kidney biopsy only demonstrated mild interstitial nephritis due to pressure effect. The clinician recommended a regular outpatient follow-up and no further treatment. Thereafter, she visited her clinician and had no additional problem for two years.

Discussion

BML is a rarely described condition of the women aged 35 – 55 years old, occurring years after hysterectomy. The most common site of metastasis is lung, although cases have been reported with involvement of lymph nodes, heart, skull, spine, and retroperitoneum. It appears that the tumor metastasizes to lungs and other extraterine tissues via hematogenous spread. Although some cases of benign metastatic leiomyomas have been reported, some authors still believe that they are metastasis of previously misdiagnosed low-grade uterine leiomyosarcomas. Definite diagnosis of BML should only be made after careful reviewing of the numerous samples of the primary uterine leiomyoma to exclude small foci of missing sarcoma, as we have done for our patient. The current criteria used for distinguishing benign, malignant, smooth muscle tumors of undetermined malignant potential in the uterus is the presence of necrosis, the mitotic index, nuclear atypia, cellularity, and the tumor border. Various histologic patterns such as myxoid differentiation coupled with enlarged and atypical cells and epitheloid feature in more than just a few foci of uterine smooth muscle are in favor of sarcoma.

Necessity of histopathologic and immunohistochemical studies to exclude other neoplasms, is obvious, especially when the site of metastasis is uncommon, as in our patient. These metastatic tumors reported in the literature tended to have greater mitotic index than the primary sites and revealed features of benign leiomyomas to low-grade leiomyosarcomas. Some researchers believe that only patients in whom no mitosis and nuclear atypia was present in their histologic samples should be placed in this category. They then, question the origin of the tumors: uterine metastasizing leiomyoma, multifocal hamartomas synchronous or metachronous leiomyomas. These tumors may be estrogen and progesterone receptor positive. Therefore, hormones may be used for their treatment. Regression of the tumor after hormonal manipulation through oophorectomy or medical treatment such as leuteinizing hormone...
releasing hormone (LHRH) analogue and tamoxifen has already been noted. Recurrence after treatment is common. Shrinkage of the tumor after menopause, pregnancy, and even spontaneously are reported.

References


