Case Report

Rare Endobronchial metastasis from uterine leiomyosarcoma

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ABSTRACT

Uterine sarcomas are rare and represent approximately 3.2% of all invasive uterine cancers. The annual incidence rate is less than two per 100,000 women. The median age at which uterine sarcoma diagnosed is 56 years. The most common histologic pattern is leiomyosarcoma (LMS) which originates from the myometrium or myometrial vessels. Uterine LMSs are aggressive tumors with high rates of recurrence. The most common mode of spread is hematogenous, with lymphatic spread being rare. Recurrences of up to 70% are reported in stage I and II disease with the site of recurrence being distal, most commonly the lungs or the upper abdomen. But the intra bronchial spread is extremely rare. Here we are reporting a case of uterine LMS with endobronchial metastasis causing whole lung collapse.

KEY WORDS: Endobronchial metastasis, hematogenous, leiomyosarcoma

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INTRODUCTION

Uterine leiomyosarcoma (LMS) represents 25-36% of uterine sarcomas and 1% of uterine malignancies.[1,2] Although the disease is mostly confined to the uterus, the local and distant metastasis rates are high and long-term survival rates are between 20% and 60%. The possible sites of metastasis are the peritoneal cavity and omentum (59%), followed by the lung (52%), pelvic lymph nodes (41%), para-aortic lymph nodes (38%), and liver parenchyma (34%).[3] Lung metastasis is known to occur which present as single or multiple nodular lesions. We report a rare presentation of uterine LMS with endobronchial metastasis (EBM) causing left lung collapse.

CASE REPORT

A 38-year-old female patient presented with abdominal pain and irregular vaginal bleeding not controlled by medical management. Ultrasonography of pelvis revealed 12 × 9 cm uterine mass. She was planned for total abdominal hysterectomy for uncontrolled bleeding. But sub-total hysterectomy with bilateral salpingo-oophorectomy was done as there was massive adhesion and profuse bleeding during operation. Histopathology revealed it as a case of LMS of uterus FIGO stage II A. The patient received three cycles of chemotherapy with Doxorubicin-Ifosfamide-Mesna regimen 21 alternate days started 3 weeks after surgery. Post-operative CECT abdomen done 2 months after first cycle of chemotherapy showed a hypoattenuating space occupying lesion (3.8 × 2.8 cm) in residual uterine stump. So, the patient was given both radiotherapy and another three cycles of chemotherapy. Her preoperative chest X-ray was within normal limit.

After 4 months of completion of chemotherapy, the patient came with the complaints of dry cough and left-sided chest pain for 3 weeks and shortness of breath only on exertion for 7 days without any history of hemoptysis. General examination was within normal limit. Respiratory system examination revealed features suggestive of left-sided lung collapse. Chest X-ray [Figure 1] showed homogenous opacity on the left side with ipsilateral mediastinal shifting. CECT thorax [Figure 2] showed collapse of the left lung with abrupt narrowing of left main bronchus with compensatory hyperinflation of the right lung. Fibre optic bronchoscopy was done [Figure 3], which revealed smooth glistening mass occluding left main bronchus, 2 cm from carina.

Histology of the tissue [Figure 4] from the endobronchial mass showed small bits of tissue partly covered by

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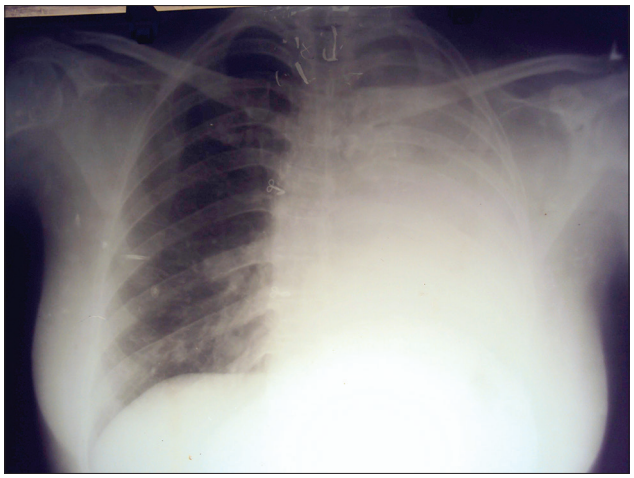

Figure 1: Chest X-Ray PA view: Homogenous opacity of the left side with same side mediastinal shifting—probably left whole lung collapse

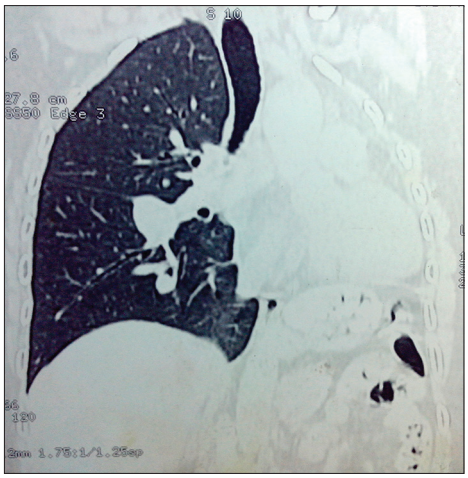

Figure 2: Contrast-enhanced CT scan of thorax (3-D reconstruction) showing complete collapse of the left lung

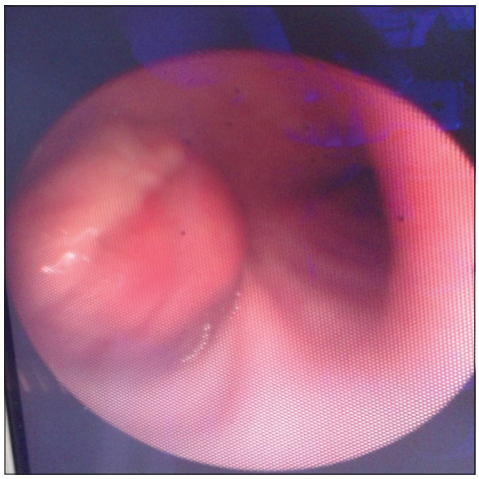

Figure 3: Fiber-optic bronchoscopy showing growth protruding from left major bronchus

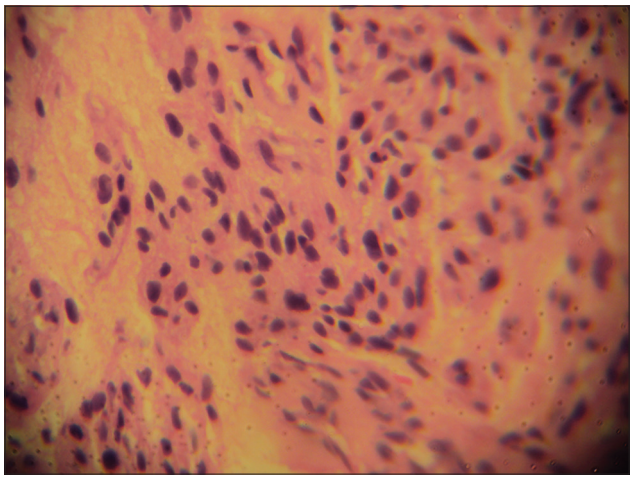

Figure 4: Histopathology showing tissue partly covered by bronchial epithelium. Subepithelial region showing a tumor composed of spindle-shaped cells arranged in interlacing fascicles. Tumor cells show nuclear pleomorphism and occasional mitotic figures. Tumor cells with bizarre hyperchromatic nuclei are present

bronchial epithelium. Subepithelial region showed a tumor composed of spindle-shaped cells arranged in interlacing fascicles. Tumor cells showed nuclear pleomorphism and occasional mitotic figures. Tumor cells with bizarre hyperchromatic nuclei were present. Immunohistochemistry showed tumor cells expressing DESMIN and SMA and immunonegativity for PgR.

So the final diagnosis was left-sided endobronchial pulmonary metastasis from uterine LMS.

The patient was given second-line chemotherapy with a single agent, Gemcitabine. Three cycles of chemotherapy were given 21 alternate days. But response rate was poor.

DISCUSSION

Uterine LMS represents 25-36% of uterine sarcomas and 1% of all uterine malignancies.[1,2] The annual incidence rate is less than two per 100,000 women.[4,5] The median age at which uterine sarcoma diagnosed is 56 years.[6]

Uterine LMSs are aggressive tumors with high rates of recurrence and metastasis, even when diagnosed at an early stage; recurrence rate has ranged from 53% to 71%.[7] Prognostic factors include tumor size > 5 cm and a high mitotic index, although they are highly aggressive even with a mitotic count of less than 2 per mm.[4,8,9] The most common mode of spread is hematogenous, with lymphatic spread being rare. Recurrences of up to 70% are reported in stage I and II disease with the site of recurrence being distal, most commonly the lungs or the upper abdomen.

EBM by definition is a bronchoscopically visible non-pulmonary tumor, involving the proximal central bronchus or subsegmental bronchi, with lesions histologically identical to the previously demonstrated primary tumor.[10] The frequency of EBM varies from 2% to 28% according to the definition.[10,11] Among

these non-pulmonary malignancies, breast, renal and colon neoplasms are most commonly responsible for EBM. Other rarely reported primary tumors include cancers from sarcoma, uterine cervix, skin tumors, thyroid gland, urinary bladder, and head and neck. EBM due to endometrial cancer is very rare. In 1996, Salud *et al.* reviewed 32 cases with EBM in a 9-year retrospective study and found that only one patient's primary cancer was endometrial carcinoma. In Sorensen's analysis of 204 patients with EBM diagnosed by bronchoscopy with biopsies, only 5 patient's primary tumor was from corpus uteri. The mean time interval from diagnosis of the primary tumor to the diagnosis of EBM has been reported to be 50 months.[12] According to the four developmental modes of EBM, our case was type I that is direct metastasis to the bronchus. The other three modes are: Type II, bronchial invasion by a parenchymal lesion; type III, bronchial invasion by mediastinal or hilar lymph node metastasis and type IV, peripheral lesions extending along the proximal bronchus. The identification of endobronchial histologic specimens together with the previous histologic specimens from the extrathoracic primary tumor is necessary in order to reach the correct diagnosis, because the treatment possibilities may be different. Immunohistochemistry may make it easier to differentiate between EBM and a new primary central airway tumor.

Treatment options for uterine LMS includes surgery, adjuvant radiotherapy and/or chemotherapy (docetaxel/gemcitabine), and hormone therapy, depending on the stage of disease.[13] Total hysterectomy with bilateral salpingo-oophorectomy is the preferred surgical option for the primary tumor. Lung metastasis can be managed with lobectomy or segmentectomy or pneumonectomy. But overall survival rate is very poor. However, for patients with uterine LMS who cannot be treated surgically because of multiple metastatic tumors or poor surgical risk-chemotherapy (paclitaxel and carboplatin) or stereotactic radiotherapy can be the alternative strategies.[14]

In conclusion, reports of EBM from endometrial cancer are extremely rare. Diagnosis of etiology remains challenging due to the absence of specific clinical characteristics. In this condition, an accurate diagnosis can only be made on pathological examination along with immunohistochemistry stain.

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