A Rare Case of Pulmonary Metastasis from Uterine Smooth Muscle Tumor of Uncertain Malignant Potential (STUMP): A Diagnostic Dilemma

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Abstract

Uterine smooth muscle tumor of uncertain malignant potential (STUMP) is a rare disease and commonly diagnosed in women of reproductive age. Metastasis from uterine STUMP are extremely rare. We report a case of 44 years old, premenopausal asymptomatic woman with incidental finding of multiple lung nodules on a CT scan KUB carried out for evaluation of left renal colic. Initial investigations including CT scan chest, abdomen and pelvis (CAP) and Bronchoscopic biopsy were inconclusive. Subsequently she underwent video-assisted thoracoscopic Surgery (VATS) and histopathology reported benign metastasizing leiomyoma of the uterus. Follow up imaging showed progression of lung disease and a repeat lung biopsy through VATS was arranged. Histopathology review of repeat biopsy and archival tissue confirmed the final diagnosis of metastasis from uterine STUMP, Estrogen receptor positive. She remains asymptomatic. She declined hysterectomy and was commenced on tamoxifen. We plan to evaluate treatment response with CT scan in 6 months interval.

Keywords: Uterine STUMP, Pulmonary metastasis from STUMP, Metastatic STUMP

Introduction

Uterine STUMP arises from smooth muscle cells of the uterus. The clinical and pathological characteristics of these tumors are unclear.[1] Uterine STUMP has heterogeneous characteristics with malignant potential qualifying between leiomyoma and leiomyosarcoma.[2] Usually affects premenopausal women with menstrual abnormalities and lower abdominal pain. Pelvic ultrasound and sometimes CT-scan are needed for diagnosis while histopathology remains a gold standard for final diagnosis. Histopathology based upon finding one out of three criteria; coagulative tumor cell necrosis, cytological atypia and increased mitosis.[3] Hysterectomy is treatment for local disease control, however, myomectomy remains an option for women who wish to preserve their fertility. Its known for higher risk of recurrence and transformation to leiomyosarcoma.[4,5]

Case Report

A 44 years old, premenopausal woman presented to the Emergency department with one-week history of intermittent colicky left flank pain radiating to the lower abdomen with associated difficulty in micturition. She denied any history of fever, breathlessness, night sweats or weight loss. Past history included hysteroscopic-myomectomy and ovarian cystectomy for recurrent uterine-fibroids. She is a non-smoker and lives with her husband and 4 children. There was no family history of malignancy. Clinical-examination revealed suprapubic tenderness. ultrasound scan was unremarkable. CT-scan kidney and

urinary bladder reported no evidence of renal/ureteric calculus. Multiple small sub-centimeter sized basal pulmonary soft tissue nodules were identified. She was managed conservatively and referred to Pulmonology for further assessment.

CT-scan chest showed multiple bilateral variable sized pulmonary nodules, some with calcification, suggestive of granulomatous disease (Figure 1). PET-CT scan reported hypermetabolic lymph nodes in the upper mediastinum measuring 10 mm with low grade avidity, SUVmax 3, along with bilateral small non-avid pulmonary nodules without metabolic activity. Bronchoscopy, transbronchial aspiration of paratracheal lymph node and broncho-alveolar lavage were negative for malignancy. CT-guided right lung nodule biopsy showed fragments of necrotic tissue with no evidence of malignancy. Respiratory culture, Acid fast Bacilli smear and culture excluded evidence of infection including Tuberculosis. The patient was advised close clinical follow-up.

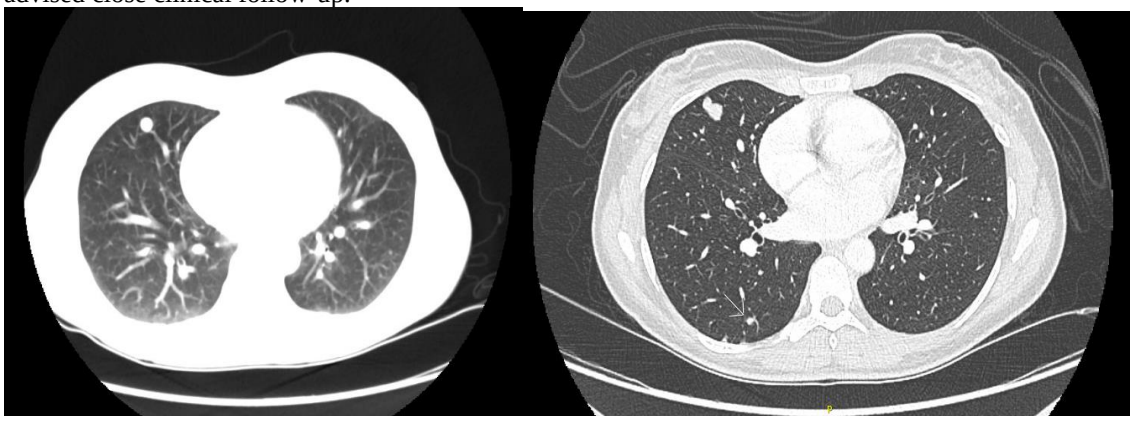

Figure 1: Initial CT scan chest with contrast shows bilateral multiple lung nodules.

Six months later, CT-scan showed Increase in size of most of the nodules in both lungs while persistent stable appearance of mediastinal lymph nodes. She underwent Video-assisted thoracoscopic surgery (VATS) and histopathology revealed mesenchymal tumor composed of spindle cells with cigar-shaped nuclei forming interlacing fascicles (smooth muscle tumor). There was no necrosis or significant cytological atypia with 4/10 high-power fields (HPF). Immunohistochemistry showed strongly positive Desmin, Caldesmon and Estrogen and Progesterone receptors in the mesenchymal elements. Pancytokeratin, HMB45, CD10, CD34, CD117& CD 31 were negative. Ki-67 proliferation index was low. In view of known mitotically active leiomyoma of the uterus in the past, it was concluded that the lung lesions likely represent benign metastasizing leiomyoma. She remained asymptomatic and continued follow-up with the pulmonologist and Gynecologist.

Three years later, follow-up chest x-ray showed an increase in the number and size of multiple small pulmonary nodules while she had no respiratory symptoms. Subsequent CT-scan reported an interval increase in the number and size of pre-existing bilateral pulmonary nodules; the largest nodule measured 3x2x4 cm (previously 1x1x1 cm) and newly seen calcified right perihilar, bronchial and subcarinal lymph nodes (Figure 2a). MRI-scan Pelvis showed two enlarging heterogeneous polypoid submucosal lesions suggestive of adenomyomas (Figure 2b).

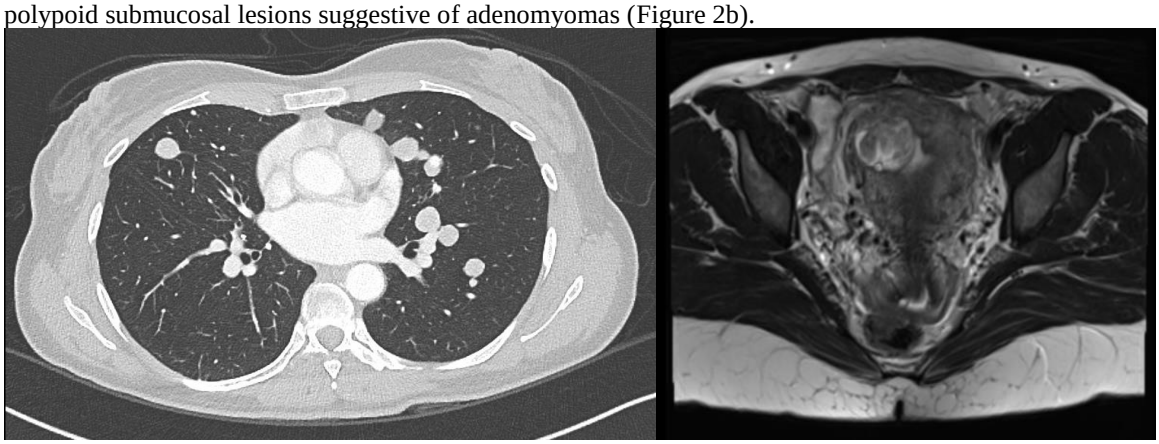

Figure 2: a) Follow up CT scan chest shows progression of bilateral lung nodules. b) MRI scan pelvis shows uterine polypoid adenomyosis.

Repeat biopsy of lung nodule showed the same morphological appearance as the previous lung nodule; suggested diagnosis of benign metastasizing leiomyoma. Re-review of recent and previous lung and uterine biopsies was requested. The previous myomectomy specimen showed a cellular spindle cell tumor with diffuse mild to moderate atypia and a mitotic rate up to 18/10 HPF. It was concluded that the lung lesions are STUMP and most likely originated from the STUMP in the uterus (Figure 3). Immuno-histochemistry confirmed estrogen receptor positive disease. Options treatment were discussed including hysterectomy and anti-estrogen therapy. Patient declined hysterectomy and was commenced on tamoxifen 20 mg daily. We plan to evaluate treatment response with CT scan in 6 months interval.

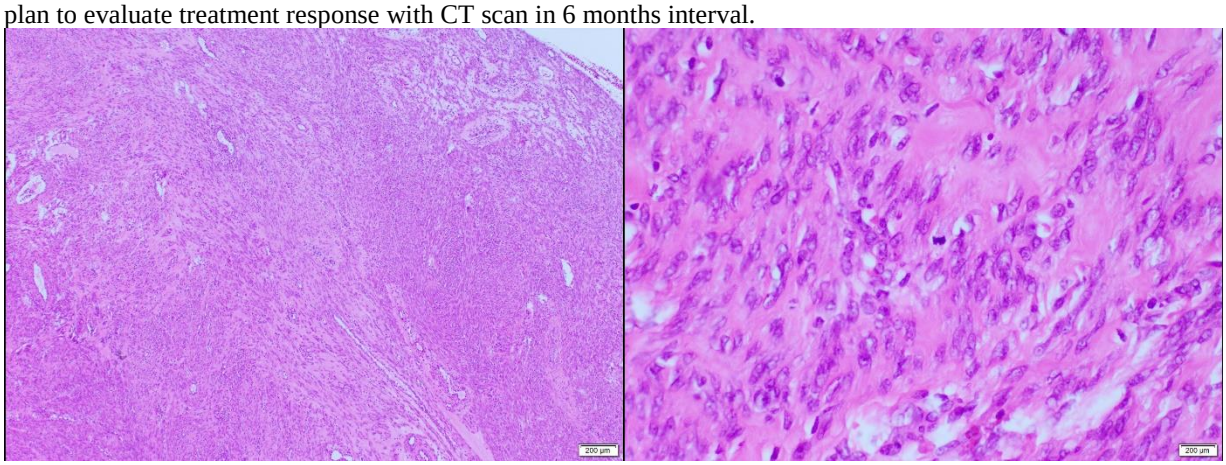

Figure 3: Histopathological examination of the uterine lesion showed a well-defined spindle cell tumor with some areas of degenerative change (H&E stain 40x magnification). High power magnification of the uterine lesion showing mild cytological atypia and increased mitotic activity (H&E stain 400x magnification).

Discussion

Uterine Stump is a rare disease and to date just over 100 cases have been reported in the literature.[5] Lung secondaries from uterine STUMP are extremely uncommon and literature search revealed only few cases of lung metastases from uterine STUMP.

The first case of pulmonary secondaries from "primary uterine myoma" was reported by Steiner in 1939[6] and the term STUMP was first described by Kempson in 1973.[7] There is no clear information available regarding metastatic behavior of uterine STUMP. Uterine STUMP exhibits heterogenous characteristics with malignant potential lying amidst leiomyoma and leiomyosarcoma.[2] STUMPs are morphologically challenging, requiring extensive sampling of the lesion for microscopic diagnosis. General diagnostic criteria are the presence of one out of three criteria for leiomyosarcoma; coagulative tumor cell necrosis, cytologic atypia, elevated mitotic activity. Other useful parameters include atypical mitoses, vascular involvement and infiltrative or irregular margins.[8]

Despite modern imaging methods, Pre-operative diagnosis on imaging does not usually predict accuracy.[9-12] In a systematic review by Giuseppe JD et al, the risk of recurrence after resection of uterine disease is approximately 20% and most were local recurrences.[4] Recurrent tumors may have identical histology to prior disease or may convert into leiomyosarcoma[13]

To the best of our knowledge, there are only few cases with lung secondaries reported in recent literature. Three of them had bilateral small nodular lesions while the latest one reported huge mass occupying the left thoracic cavity treated with pneumonectomy.[14] In contrast, our case presented with asymptomatic multiple, small, nodular soft tissue lesions in both lungs with a past history of recurrent uterine fibroids, treated with myomectomy as the patient wished to preserve her fertility.

There is paucity of data regarding systemic treatment of metastatic uterine STUMP. However, some authors report response to anti-estrogen therapy in patients with estrogen receptor positive disease especially in postmenopausal women. Options include GnRH analogues, tamoxifen or aromatase inhibitors.[15]

Our patient has progressive pulmonary metastases and remains asymptomatic. The options of systemic therapy were discussed and in view of estrogen receptor positive disease, commenced on tamoxifen.

Conclusion

Metastatic Uterine STUMP is a rare condition and poses a diagnostic dilemma. Multidisciplinary approach including careful and thorough histopathological examination is often required to reach the diagnosis. Overall prognosis is good. Treatment should be individualized based on patient preferences and symptoms.

Conflicts of interest and acknowledgements

None

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