

Case Report

A Rare Case of Primary Adrenal Tuberculosis Mimicking Metastasis in a Patient with Endometrial Stromal Sarcoma

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Keywords

- Primary adrenal tuberculosis
- High grade endometrial stromal sarcoma
- Mimic malignancy

Abstract

Background: To report a rare case of primary adrenal tuberculosis mimicking metastasis in a patient with endometrial stromal sarcoma.

Case presentation: A 50-year-old woman with advanced high grade endometrial stromal sarcoma (International Federation of Gynecology and Obstetrics stage IIIc) underwent staging operation and six courses of chemotherapy (cyclophosphamide, vincristine, doxorubicin, and dacarbazine). A whole-body Positron Emission Tomography-Computed Tomography suggested solitary right adrenal metastasis at 37 months after surgery. Laparoscopic right adrenalectomy was done and histopathological result showed primary adrenal tuberculosis.

Conclusion: In endemic areas for tuberculosis, a diagnosis of adrenal tuberculosis should be considered for adrenal nodule in women with gynecologic malignancies.

ABBREVIATIONS

ESS: Endometrial Stromal Sarcoma; CT: Computed Tomography; PET-CT: Positron Emission Tomography-Computed Tomography; PCR: Polymerase Chain Reaction

INTRODUCTION

Uterine endometrial stromal tumors are one of the three most common histologic variants of uterine sarcoma, divided into three types on the basis of mitotic activity, vascular invasion, observed differences in prognosis. High grade Endometrial Stromal Sarcoma (ESS), one of endometrial stromal tumors, is more aggressive clinical course and poorer prognosis than ESS, so 5-year disease free survival is less than 25% with frequent recurrence [1].

On the other hand, primary isolated adrenal tuberculosis is one of uncommon adrenal tumor and very rare disease entity, also the clinical features are nonspecific. So its diagnosis is difficult to obtain and often made on autopsy or after adrenalectomy [2].

This case report is on the primary adrenal tuberculosis mimicking metastasis in a patient with uterine high grade ESS.

To the best of our knowledge, primary adrenal tuberculosis concurrent with uterine sarcoma has not been previously reported.

CASE PRESENTATION

A 50-year-old woman was transferred to our institution due to suspected uterine malignancy after diagnostic laparotomy in November, 2012. On physical examination, a 20 weeks gestational age sized uterus was detected with fixation in pelvic cavity. Evaluation for uterine malignancy was done including chest X-ray, pelvis Magnetic Resonance Imaging (MRI), abdominopelvic Computed Tomography (CT), Chest CT, esophagogastroduodenoscopy, colonoscopy, whole-body Positron Emission Tomography-Computed Tomography (PET-CT). The distinctive thing of the preoperative tests, chest X-ray revealed small nodular increased opacities in left upper lung field, probable previous infection sequelae. But under the pulmonology consultation for differential diagnosis of active tuberculosis, this patient was needed to undergo chest CT, sputum Acid-Fast Bacteria stain, culture, Polymerase Chain Reaction (PCR) for

tuberculosis. All these tests were negative, so we can rule out of active tuberculosis.

On the imaging studies, the patient was diagnosed with uterine sarcoma with multiple peritoneal metastasis. Therefore, we performed staging operation and histological diagnosis was advanced high grade ESS with International Federation of Gynecology and Obstetrics stage IIIc. After surgery, she received cyclophosphamide 500 mg/m² intravenously bolus on day 1, vincristine 1.4 mg/m² intravenously bolus on day 1, doxorubicin (Adriamycin; Adria Laboratories, Columbus, OH) 50 mg/m² intravenously bolus on day 1, and dacarbazine 400 mg/m² by 1-hour infusion on days 1 to 3 cycles repeated every 28 days for six courses from December, 2012 to May, 2013.

During follow-up, a PET-CT scan at 37 months after surgery revealed newly fludeoxyglucose uptake only in the right adrenal gland, and additional adrenal CT revealed 1.4cm and 1.2cm sized right adrenal mass, suggesting adrenal malignancy (Figure 1). Laparoscopic right adrenalectomy was done by general surgeon after adrenal function test which was shown non-functioning adrenal tumor. Histopathological result showed primary adrenal tuberculosis, because histologic finding was suggestive of tuberculosis with PCR test for tuberculosis was positive (Figure 2). This woman was treated with an empiric antituberculous regimen and remained healthy.

DISCUSSION

The adrenal tuberculosis is almost always secondary to tuberculosis elsewhere, most often the lung but sometimes the genitourinary tract [3]. Primary tubercular adrenalitis is a rare

clinical entity and few cases are reported in the literature. In a systematic review, only 1 case of tubercular adrenalitis out of 370 reports of extrapulmonary TB was observed during a period of 10 years [4]. Adrenal tubercular infection has also been reported as cause of sudden death and as a cause of fever of unknown origin [5]. The diagnosis was often made on autopsy or after adrenalectomy, because most uncommon masses of the adrenal glands like tuberculosis do not have specific imaging features, so only histological examination can define the final diagnosis [6].

Sometimes, specific imaging features, including location, contour and presence of calcifications can be used in combination with clinical signs to help differentiate between tuberculosis and other lesions [7]. The specific findings of adrenal tuberculosis are bilateral adrenal involvement, a low attenuation center and peripheral enhancement on CT and MRI (47%) in acute phase, a calcification (59%) in later phase [2,7]. Despite of almost adrenal tuberculosis (69~90%) represents bilateral involvement [7,8], our patient represents a rare case of unilateral primary adrenal TB without any other organ involvement and there was no specific rim enhancement or calcification. So biopsy might be necessary to differentiate adrenal TB from an adrenal neoplasm or other infectious process.

Using CT-guided percutaneous fine-needle aspiration biopsy, the diagnosis of adrenal tuberculosis may be confirmed in suspected patients who presents with adrenal gland mass seen incidentally using CT scanning [9]. However, the aspiration of material from adrenal gland mass may be difficult while even exact sampling might not confirm the diagnosis because of the difficulty in the cytological definition of malignant disease from

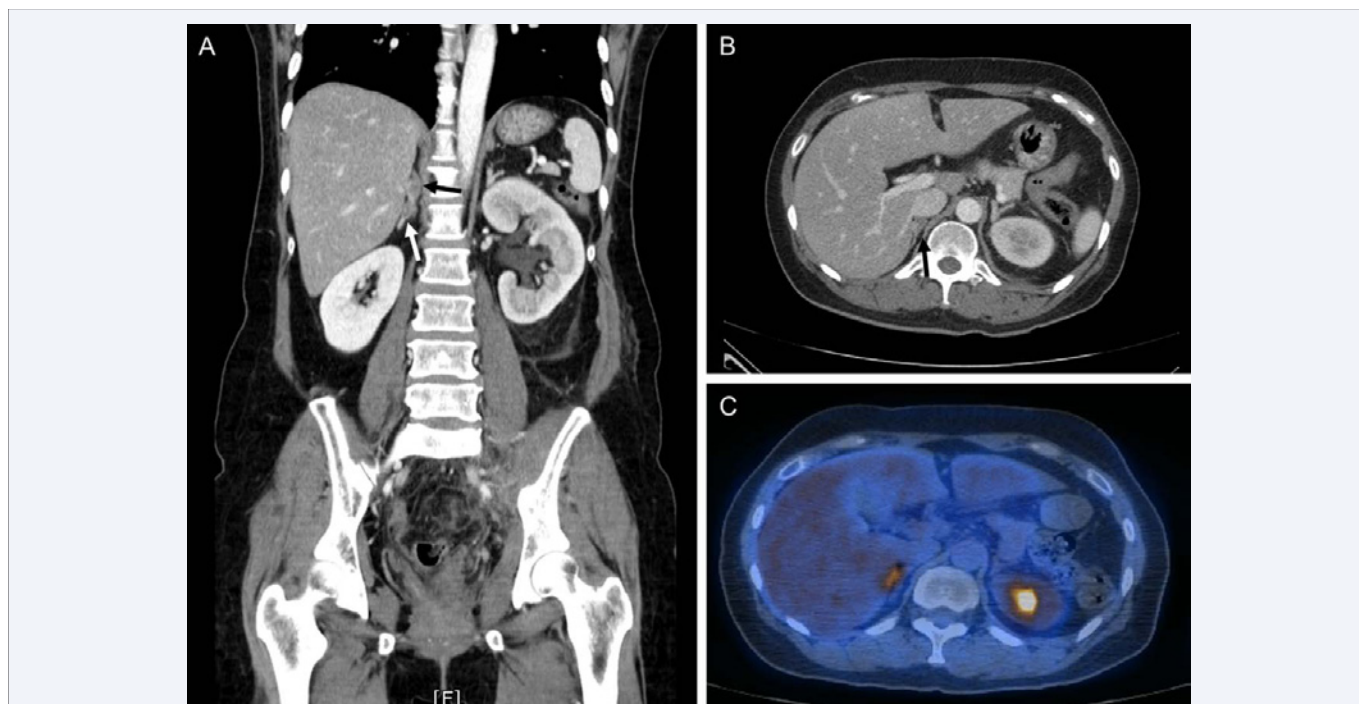


Figure 1 (A) The mixed density rounded 1.2cm, 1.4cm sized masses (arrows) lesion of the right adrenal gland on adrenal CT, (B) adrenal CT showed adrenal mass on the same site of PET-CT, and (C) PET-CT showed fludeoxyglucose uptake in the right adrenal gland.

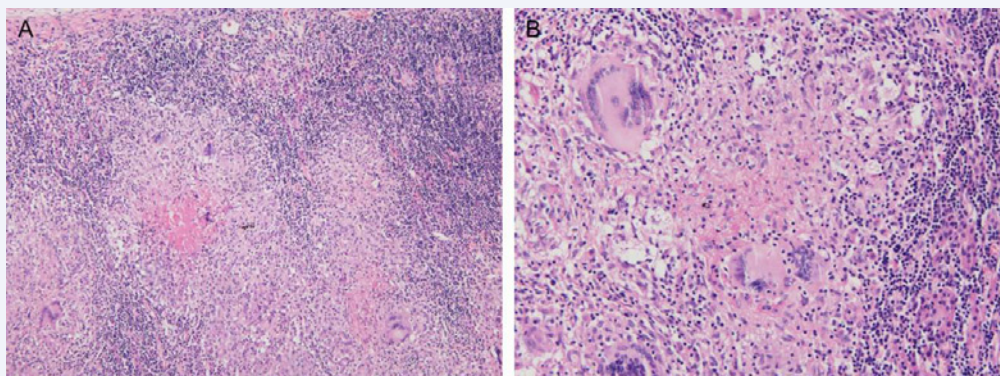


Figure 2 Adrenal tissue with extensive caseation necrosis, many epithelioid cells and Langhan's type giant cells. (A) hematoxylin and eosin stain, x 100 and (B) hematoxylin and eosin stain, x 200.

the aspirate. So, surgical exploration is advised in all solid adrenal masses with possible removal because of malignant potential. So our case was performed laparoscopic adrenalectomy rather than CT-guided aspiration biopsy, because our first impression was adrenal metastasis.

There was one possible report about pathophysiology of primary adrenal tuberculosis. Only rarely, in solitary lesion of the adrenal gland, the tuberculosis is due to the reactivation of small lesions which were produced during a bacteremic phase of a previous primary infection. One possibility in the previously reported case is that adrenal tuberculosis may be occurred originally with other tuberculous lesions and that adrenal gland remained involved while the other tuberculous lesions healed [10,11].

One thing in mind, tuberculosis can progressively destroy the adrenal glands. So, Addison's syndrome can characteristically appear when 90% of the tissue is destroyed, and can lead to adrenal crisis and become a life-threatening disorder especially during coincidental physiologic distress [8]. So clinician must take adrenal function test for suspicious with adrenal tuberculosis or all incidentaloma, like our patient.-

Our first impression for adrenal mass was adrenal metastasis because high grade ESS is characterized by extremely aggressive behavior leading to early metastatic disease and death [12], and the primary adrenal TB is very rare disease entity. For surgical difficulty and patient desire of no additional surgery, the patient might be taken unnecessary additional chemotherapy, unless the patient had fever. Even though, Primary adrenal tuberculosis with uterine sarcoma is uncommon, clinicians must consider the possibility of other differential diagnosis rather than cancer metastasis even if highly malignant tumor, especially patient that has only isolated metastatic lesion. There is no confident diagnostic tool for the differential diagnosis between metastatic mass of any organ, and surgery is frequently needed to make the diagnosis. . In the light of this case, clinicians must keep in mind of false positive of radiologic study and importance of biopsy confirmation, despite high sensitivity and specificity of radiologic evaluation including PET-CT and CT.

CONCLUSION

Adrenal tuberculosis should be considered in the differential diagnosis of adrenal mass, particularly in endemic areas, as it can closely mimic malignancy, even with negative sputum bacteriologic and cytologic studies. Particular radiological findings can help differentiate between tuberculosis and adrenal metastasis. And ascitic fluid adenosine deaminase levels and PCR can also aid in the diagnosis [12]. If these tests are negative and clinical suspicion remains, laparoscopy can confirm the diagnosis. Major unnecessary treatments like chemotherapy can therefore be avoided using biopsy confirm.

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