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Osteosarcoma appearing in the uterus as malignant mixed Müllerian tumor

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Summary

Few cases of osteosarcoma appearing in the uterus as malignant mixed Müllerian tumor have been published in the literature to the authors' knowledge. They report a 59-year-old postmenopausal female that underwent total abdominal hysterectomy and bilateral adnexectomy and bilateral pelvic cavity and abdominal periaortic lymphadenectomy because of numerous irregular mass in the uterus. The histopathological diagnosis which was confirmed by immunohistochemical study was osteosarcoma appearing in the uterus as malignant mixed Müllerian tumor. Then for cycles of chemotherapeutic regimens (paclitaxel plus carboplatin, 21 days as one cycle) was started at 16 days after surgery. This was a rare case of osteosarcoma appearing in the uterine as malignant mixed Müllerian tumor.

Key words: Osteosarcoma; Malignant mixed Müllerian tumor; Uterine sarcoma.

Introduction

Cancer of the body of the uterus is associated with high morbidity among women in many countries [1, 2]. However, with the development of modern medical science, the mortality rates are consistently decreasing in most countries [3, 4]. Malignant mixed Müllerian tumors are uncommon neoplasms as of the uterus [5]. Osteosarcoma (OS), as a primary malignant bone tumor, often occurs in bone in young people with high morbidity and mortality [6]. Few cases of osteosarcoma appearing in the uterus as malignant mixed Müllerian tumor have been published in the literature to the present authors' knowledge. They report a rare case of osteosarcoma appearing in the uterus as malignant mixed Müllerian tumor.

Case Report

A 59-year-old postmenopausal female, ethnic Han, without a family history of gynecological malignant tumor, but with four days of vaginal bleeding, was admitted to Changxing People's Hospital (Huzhou, China). Pelvic MRI revealed numerous irregular masses at the junction of the corpus uteri and cervix, and it was considered to be a hysteromyoma. The uterine cavity was full of massive hematocele and the export was blocked (Figure 1). The patient chose the Huzhou Central Hospital for further treatment.The transvaginal ultrasonography inspection found the uterine cavity with accumulated fluid, calcification, and hysteromyoma in uterine body (Figure 2). The value of serum tumor marker CA125 and CA153 was 82.10 U/mL (normal value is less than 35 U/mL) and 28.50 U/mL (normal value was less than 25 U/mL), respectively. The examinations of routine blood count, liver and kidney functions, blood coagulation function, and electrolytes were normal. After discussion with several experienced gynecologist and patient's informed consent, total abdominal hysterectomy and bilateral adnexectomy was carried out. The frozen

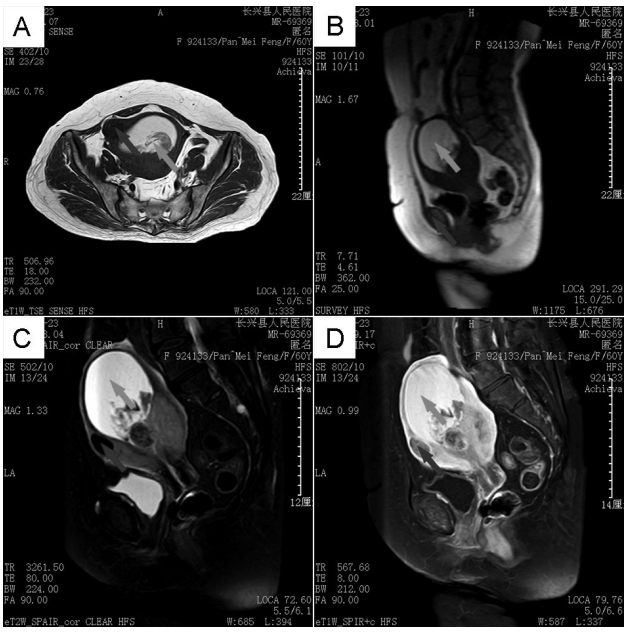

Figure 1. — Pelvic MRI. Figures A, B, C, and D represent axial T1 signal imaging, sagittal T1 signal images, sagittal T2 signal images, and sagittal diffusion weighted Imaging (DWI). The volume of uterine cavity is larger and the export is blocked (green arrow). The numerous irregular mass at the junction of the corpus uteri and cervix was considered to be a hysteromyoma (blue arrow).

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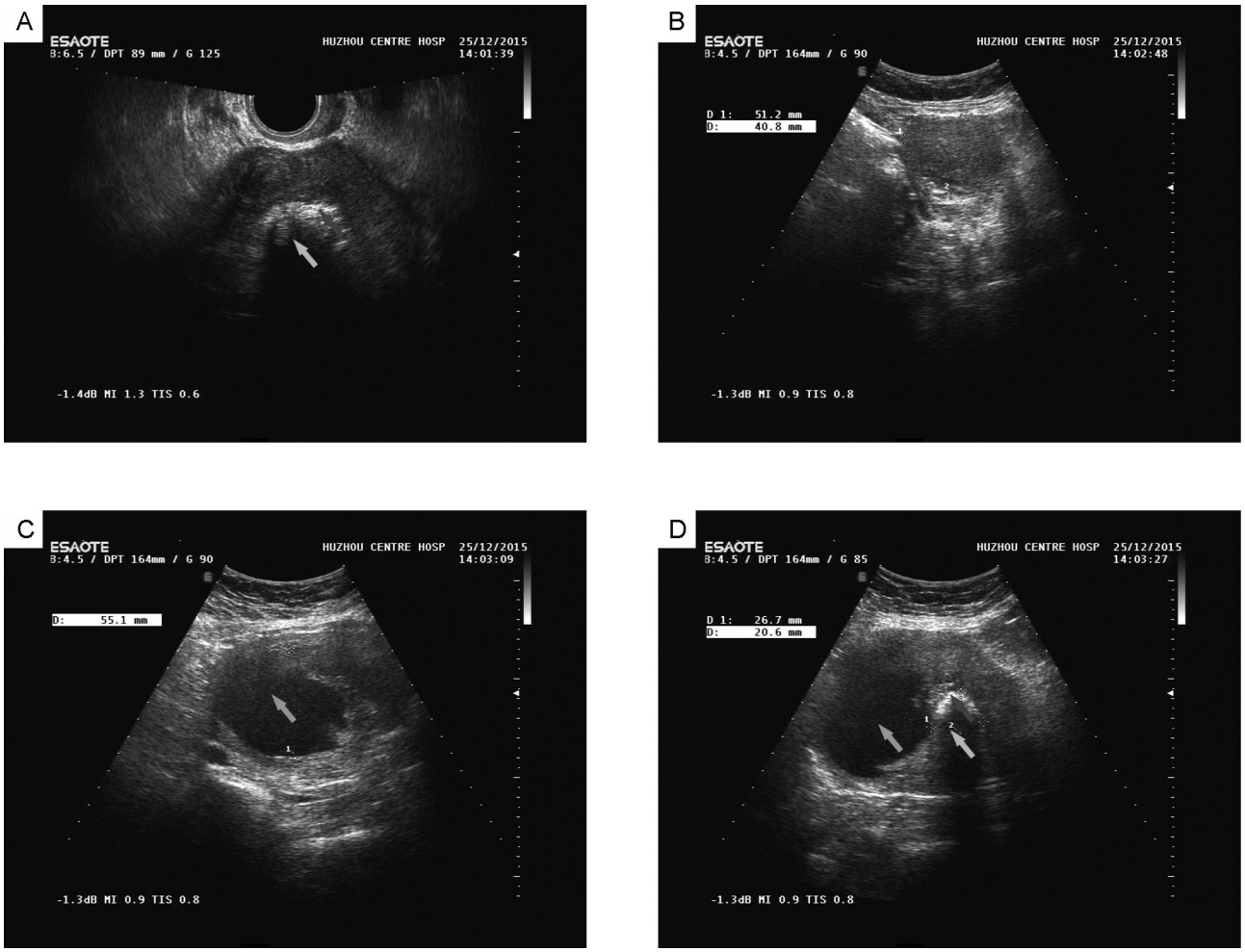

Figure 2. — Ultrasonography inspection imaging. An ultrasound instrumentation was used to examine the pelvic organs. Figure A is the transvaginal ultrasonic image. The number of ultrasonic probe is EC1239-5. Figures B, C, and D are the abdominal ultrasonic images. The number of ultrasonic probe is CA6217-3.m The volume of the uterus is 10.9×8.6×10.8 cm, The uterine cavity has accumulated fluid (green arrow). The hyperechogenic (blue arrow) with acoustic shadow (cyan arrow) are in the uterus. The ultrasonography was diagnosed as hysteromyoma.

section examination during the operation indicated the heterotypic cells in a leiomyoma and endometrioid adenocarcinoma. Further bilateral pelvic cavity and abdominal periaortic lymphadenectomy were performed after consultation with the relatives of patients. The operation went smoothly and no severe complications occurred. The results of the formal pathological diagnosis were as follows: The carcinosarcoma (diameter 5.5 cm) with squamous intraepithelial metaplasia was found in the fundus uteri and in the corpus uteri (Figure 3). The carcinosarcoma included 80% endometrial adenocarcinoma and 20% soft tissue sarcoma and osteosarcoma. Four leiomyomas with diameters from 1.3 to 5.5 cm were seen in the myometrium, with the largest being a cellular uterine leiomyoma. The cervical canal and ovarian and tubal tissue were not affected by malignancy. Bilateral cornua uteri, parametrium, and bilateral ovaries arteriovenous were neither involved by the malignancy (Figure 4). The immunohistochemical results confirmed the osteosarcoma, adenocarcinoma, and cellular leiomyoma was on the uterus (Figure 5). One of eight positive lymphatic metastasis on the left side of the pelvis cavity and one of five on the right were found. Two abdominal periaortic lymph nodes were positive (Figure 6). Carcinosarcoma staging was Stage IIIC2. The patient recovered well from surgery. Four cycles of postoperative chemotherapeutic regimens (270 mg paclitaxel plus 650 mg carboplatin, 21 days as one cycle) was started at 16 days after surgery.

Written informed consent was obtained from the patient for publication of this case report and accompanying images

Discussion

Malignant mixed Müllerian tumor (MMMT), which is also known as carcinosarcoma, is a particular type of bipha-

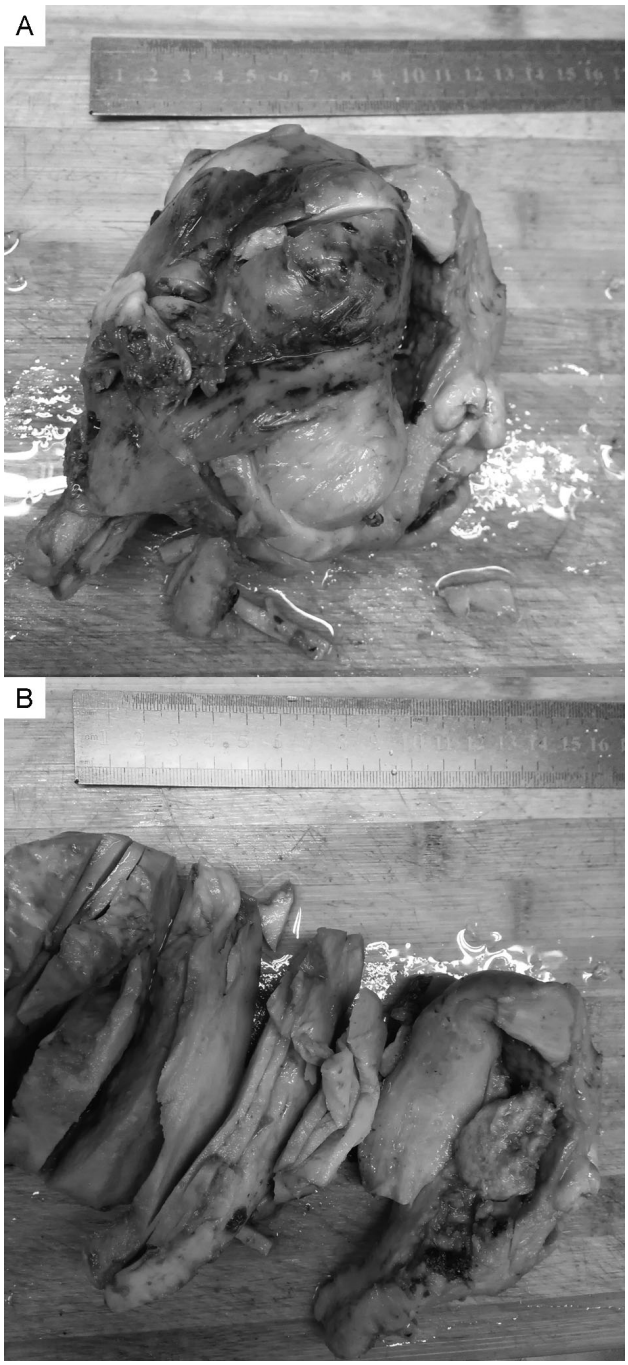

Figure 3. — Gross specimen. The volume of uterus is 12×14×11 cm, the uterine cavity volume is expanding and filled with blood. The surfaces of endometrium are uneven and resemble a cauliflower. A palpable nodule, about 3.5 cm in diameter, is right behind the uterine cavity. It was confirmed that the nodule contained adenocarcinoma (Grade III) and osteosarcoma (Grade II) under the microscope. It has a hard consistency. There are four leiomyomas with diameters from 1.3 to 5.5 cm in the myometrium. Cauliflower-like mass was mostly composed of endometrial adenocarcinoma (Grade III).

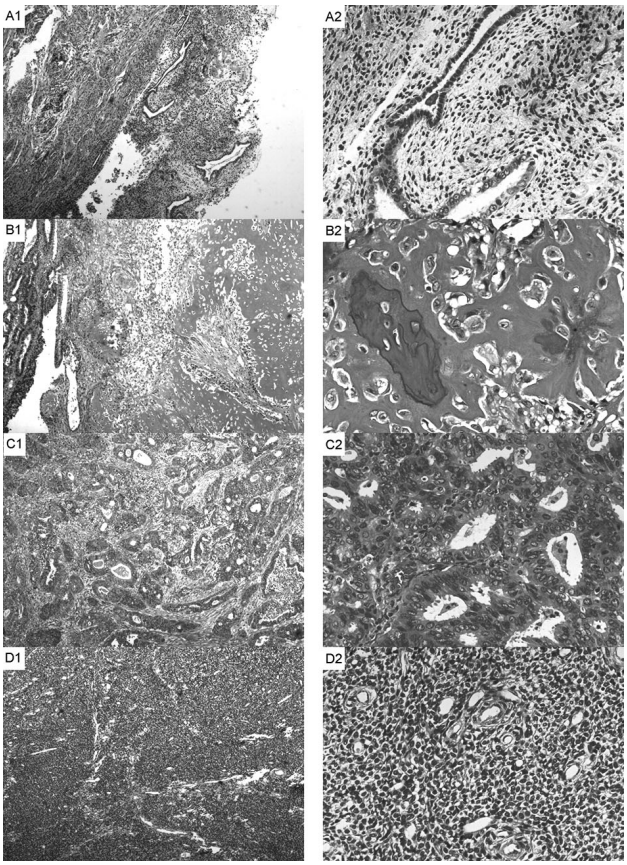

Figure 4. — Characteristic of pathological morphology. General HE staining are applied. Figures A1, B1, C1, and D1 are under 5×10 magnified visual fields with ordinary microscope, Figures A2, B2, C2, and D2 are under 10×20 magnified visual fields. Figures A1 and A2 are of the normal endometrium. Figures B1 and B2 are osteosarcomas. Figures C1 and C2 are adenocarcinomas. Figures D1 and D2 are cellular leiomyomas.

sic tumors which include both epithelial and mesenchymal components [7]. In the present patient the authors found at least three malignant tumors including osteosarcoma, cellular leiomyoma, and adenocarcinoma in the uterus. To their knowledge, osteosarcoma appearing in the uterus as malignant Müllerian mixed tumor occurs infrequently. King and Kramer reported 21 malignant Müllerian mixed tumors of the uterus in the New York Hospital from 1957 to 1977; only one case of osteosarcoma as the heterologous elements [8]. The more-common clinical symptoms and signs were vaginal bleeding, pain, weight loss, and abdominal mass [9]. In the present patient, four days of vaginal bleeding was the first symptom. Its origin, exact nature, prognosis, and treatment remain controversial [10]. Malignant Müllerian mixed tumors of the uterus are particularly deadly type of tumors, and they almost always are at advanced stage and have a poor prognosis once symptoms ap-

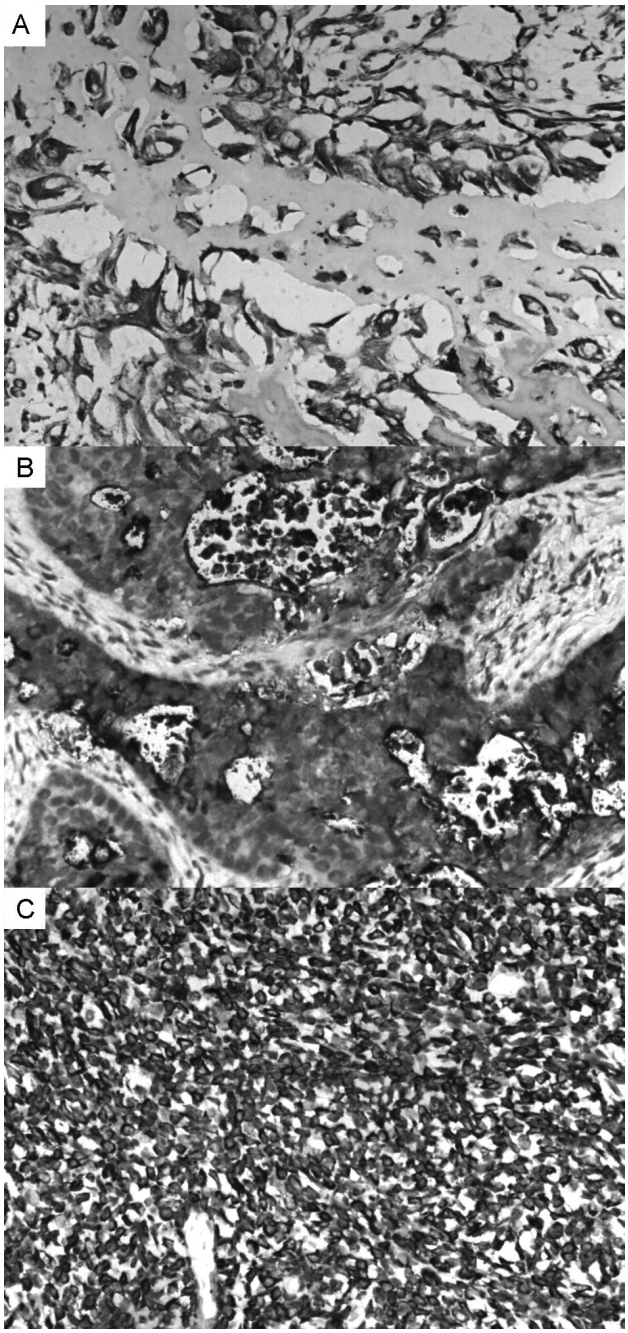

Figure 5. — Immunohistochemical features. Figure A shows vimentin protein positively stained and the cytoplasm is stained as brown. This verifies that this cell is an osteoprogenitor cell. Figure B shows the carcinoembryonie antigen (CEA) protein is positive and the cytoplasm is stained as brown. It verifies that this cell may be endometrial carcinoma of uterus. Figure C shows the desmin protein as positive and the cytoplasm is stained as brown, It verifies that this cell may be a leiomyoma cell.

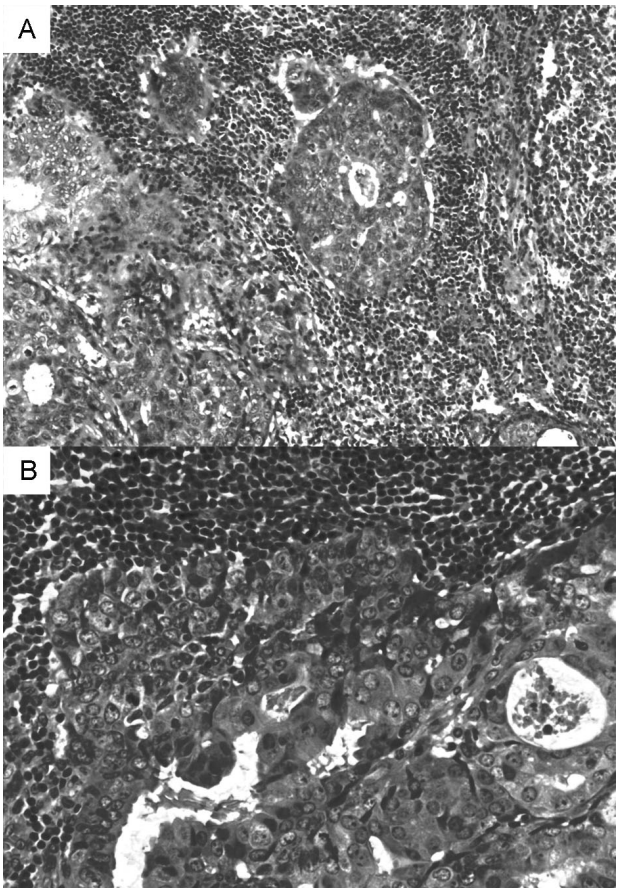

Figure 6. — Lymph node metastasis. Figures A and B are under 5×10 and 10×20 magnified visual fields with ordinary microscope, respectively. One of eight positive lymphatic metastases on the left side of the pelvis cavity and one of five on the right can be observed. Two abdominal periaortic lymph nodes are positive. The adenocarcinoma of endometrium in the lymph node is observed.

pear.[11] Gonzalez Bosquet *et al.* [12] reported that the sarcomatous component has a more aggressive biology compared with the carcinomatous component. Conversely, in the present patient, the adenocarcinoma of endometrium in the lymph node was observed. The osteosarcoma, as the sarcomatous component, were free of lymph node metastases.

Diagnostic methods of imaging have not yet definitely confirmed the tumor's nature. It is difficult to diagnose if the uterine mass is benign or malignant by various imaging methods such as with MRI and ultrasonography inspection. It is simple to be misdiagnosed as uterine fibroids and delay the treatment. This is an important reminder for clinicians.

Malignant Müllerian mixed tumors in the uterus have a poorer outcome and prognosis as compared to sarcoma [13]. Five-year overall survival rates of malignant Müllerian mixed tumors is only 16.3% [14]. Surgery and chemotherapy are the main treatment methods for these tumors [15, 16]. In the present patient, surgery was conducted at first and then she underwent adjuvant chemotherapy. Three main chemotherapeutic regimens including ifos-

famide plus cisplatin [17], adriamycin plus cisplatin [18], and paclitaxel plus carboplatin [19] were available for reference on the basis of the past literature, Eventually the present authors chose the paclitaxel plus carboplatin regimen. The patient currently shows no recurrence or metastasis and has a good living quality by far.

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