Case Report

Extraskeletal Osteosarcoma of Penis: A Case Report

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ABSTRACT

Extraskeletal osteosarcoma (EOS) is rare and commonly arises in the retroperitoneum, limbs, head and neck. There is no significant difference between EOS and other malignant tumors in soft tissue. Localized pain and swelling are the common presenting symptoms. Clinical diagnosis of EOS is difficult, imaging techniques may be helpful and careful, and the histopathological analysis is necessary. The common histological variants of EOS include: osteoblastoma, chondroblastoma, and fibroblastoma, and other unusual subtypes were reported occasionally. It should be distinguished with myositis ossificans, malignant mesenchymoma, giant cell tumor and parosteal osteosarcoma. We present an EOS arising in the penis. The primary site and histological category of the tumor were extremely rare. We hope the case will be helpful to the recognition of clinical signs, iconography and histopathology of EOS.

Key words: Extraskeletal osteosarcoma; Penis; Giant cell

INTRDUCTION

Extraskeletal osteosarcoma (EOS) is a malignant mesenchymal neoplasm which is located in soft tissues. It is an extremely rare disease, accounting for only 4% of osteosarcoma and 1% of soft tissue sarcomas[1]. Clinical diagnosis of EOS is difficult, X-ray, CT and magnetic resonance imaging (MRI) techniques may be helpful to the detection of primary site, volume and relationship with the surrounding tissue of the tumor[2], and significant to the choice of operation. Careful histopathological analysis is necessary to final diagnosis. Here we report an EOS of penis.

CASE REPORT

Clinical examination: A 68-year-old man presented a tender subcutaneous nodule of the penis. The nodule had localized pain and grew from about 0.3 cm × 0.3 cm × 0.3 cm to 1.2 cm × 0.8 cm × 0.5 cm in a year.

Physical examination: A 1.2 cm × 0.8 cm × 0.5 cm mass can be touched 0.8 cm right to coronary sulcus. There was no red swelling of the skin and abnormal temperature. The edge of the mass was clear. The mobility of the mass was poor. The scrotum and orchis of the patient were normal, and there was no touched intumescent lymph node in inguen. The operation was performed in May, 2009.

Operation findings: The patient received 1% Lidocaine injection at the root of penis and the surgery was performed. The skin and subcutaneous tissue were dissected to segregate the mass. The tumor slightly adhered to the surrounding tissue but did not invade to tunica albuginea. Finally the mass was excised and a histological diagnosis was made.

Follow-up records: The patient was floating population. There was no other treatment except anti-inflammatory treatment after operation. The patient was followed up for 10 months, and then lost the connection with us.

Pathology

Gross: The neoplasm without envelope was grayish-white, grayish-pink and 1.5 cm×1 cm×1 cm. The cut surface of the mass was grayish-white and rigid, and had sense of grit and no weaving shapes.

Microscopy: The cells in the tumor were widespread and irregular, mainly spindle and ovoid. The cytoplasm of the cells is basophilic (Figure 1). The nuclei are obviously atypical, mostly clostridial form and polygons (Figure 2). Massive bone matrix coexisted with multinucleated giant cells can be found everywhere in the tumor. Tumor cells were in palisade arrangement and most commonly seen in and around the bone matrix

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(Figure 3). In some instances, 3.5 to dozen nuclei can be seen in a single multinucleated giant cell (Figure 4).

Immunological markers: Immunostaining was positive for vimentin, CD99, Bcl-2 and epithelial membrane antigen (EMA) (Figure 5), and negative for CK, S-100, desmin and CD34.

Pathological diagnosis: Penile primary EOS, giant cell-rich tumor.

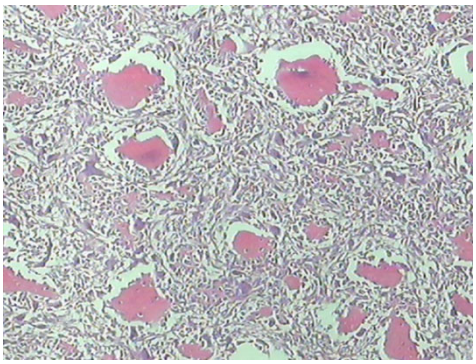

Figure 1. Bone formation can be seen in all parts of the tumor (×100).

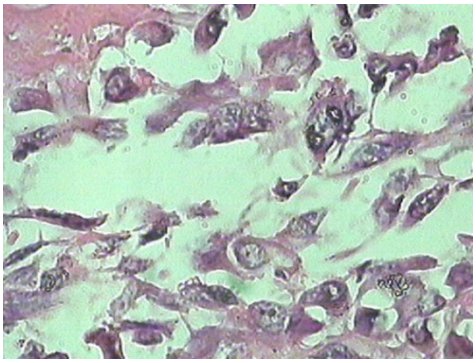

Figure 2. The nuclei in the spindle cells are obviously heterotypic (×400).

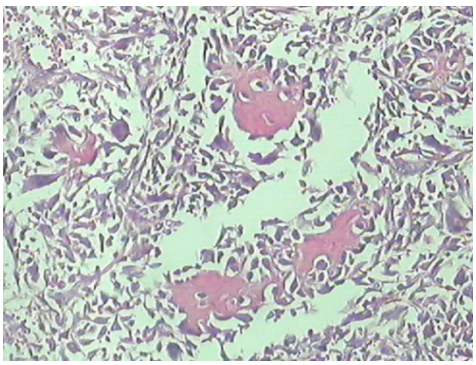

Figure 3. Tumor cells surrounding the interstitial substance are in palisade arrangement (×200).

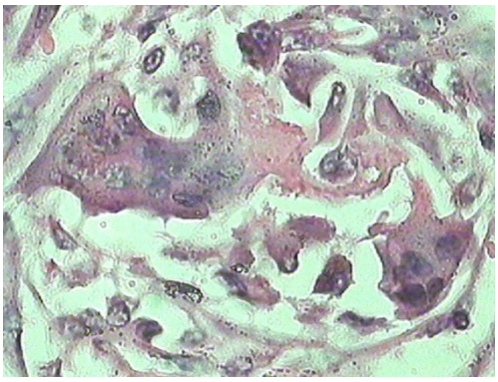

Figure 4. In a single giant cell, 3.5 to dozen nuclei can be seen (×400).

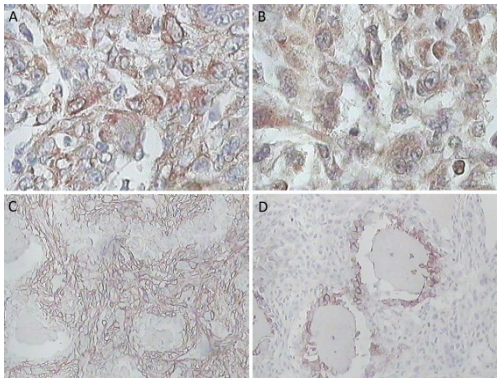

Figure 5. Immunostaining was positive for vimentin, CD99, Bcl-2 and EMA **A:** Vimentin (×400); **B:** CD99 (×400); **C:** Bcl-2 (×200); **D:** EMA (×200).

DISCUSSION

EOS is a malignant mesenchymal neoplasm that is located in soft tissues without direct attachment to skeletal system. EOS was first reported by Wilson in 1941[3]. It is extremely rare, accounting for only 1% of soft tissue sarcomas. Distinct to osteosarcoma usually afflicting young people, EOS mainly affect people after 50 years old, and the mean age was 54.6 years (range 16–87 years)[4,5]. Trauma and radiation are well-documented predisposing factors[6].

Most people consider that multipotential mesenchymal cells develop to allotypic osteoblasts, leading to the growth of EOS. The precisely origin is not clear now. EOS most commonly arises in the retroperitoneum and the muscles of thighs and limb girdles, rarely in lung, prostate, scalp, mammary gland, spermatic cord, pelvis and orbit. EOS of the penis is exceedingly rare; only six other well-documented cases have been reported in the

literature in English[7,8] and none in Chinese. The main types are osteoblastoma, chondroblastoma and fibroblastoma. The tumor which is full of giant cells is extremely rare. There was no significant difference in clinical manifestation between EOS and other soft tissue sarcomas. Localized swelling and pain are commonly seen. X-ray examination shows that scattered floccules or patchy high density in parenchyma, and the tumor has no connection with the adjacent bone tissue which is the characteristics of EOS. But the iconographic characteristic has no specificity. It is hard to distinguish with other malignant tumor, the final diagnosis must depend on histopathologic examination.

The volume of EOS ranged from 2.5 to 20 cm^3 and mostly lobulated, and 20% of the tumors were described as pseudoencapsulated masses and with satellite nodules surrounded. The cut surface ranged from gray-white to tan-yellow to dark-red, depending upon the degree of mucification, hemorrhage, and necrosis. Some tumors showed focal to cystic change. Except above-mentioned, there are some other subtypes such as: epithelioid osteosarcoma, clear-cell variant osteosarcoma, malignant fibrous histiocytoma and giant cells-rich osteosarcoma[9-12]. They are all short of unique biocharacteristics, and have no significance to therapy and prognosis. The distribution mode, volumn and number of nucleus of the giant cells we reported were similar to those of giant cell tumors, and the number of osteoclast-like multinucleated giant cells increased obviously. But massive bone trabecula and allotypic tumor cells have more density and uniformity than the giant cells. According to the result of immunohistochemistry, we can draw a conclusion that the tumor was the giant cell-rich type of EOS.

Lee, et al. introduced the diagnostic criteria of EOS: (1) in soft tissue and not attached to bone or periosteum; (2) osteosarcoma with the same image; and (3) produce osteoid or cartilaginoid matrix. The case of giant cell-rich EOS should be distinguished from the following diseases in pathology: (1) Myositis ossificans: Patients usually have a history of trauma. Patients often have masses with the construction of active proliferation fibrous tissue, irregular osteoid tissue and mature trabecular bone. (2) Malignant mesenchymal tumors: In addition to components of osteosarcoma, it should also find other malignant mesenchymal elements, such as rhabdomyosarcoma, and liposarcoma. (3) Giant cell tumor: They both have affluent multinucleated giant cells, but giant cell tumor has no formation of tumorous bone trabeculae in spindle cells. (4) Periosteal osteosarcoma: The mass is often located in the cortical bone surface, and closely integration and the formation of radial bone can be seen.

EOS is reported to carry an exceptionally poor prognosis. The disease is generally involved in invasion and metastasis. The recurrence, transfer and 5-year survival rates are 45%, 65% and 25% to 37%, respectively. Since the exact preoperative clinical diagnosis is difficult, so patients with newly diagnosed EOS were usually performed local mass excision, and often died of metastasis of lung, liver, lymph nodes, bone or soft tissue in 2 to 3 years.

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Disclosure of Potential Conflicts of Interest

No potential conflicts of interest were disclosed.

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