Primary Osteosarcoma of the Spine: a case report

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Primary osteosarcoma of the spine is an uncommon disease. Here we report a 62-year-old female who suffered from progressive chest tightness, dizziness, urinary incontinence, and bilateral lower legs weakness for days. Chest and cervical spine radiographs revealed a large expansile mass with extensive ossifications and adjacent bone destructions in left superior mediastinum and lower neck. Chest CT and spinal MRI scans revealed a large mass with soft tissue components and extensive ossifications involving body, posterior element, paravertebral soft tissue, epidural space, and rib in C7, T1, and T2 levels. The intervertebral disc spaces were spared. Imaging studies are useful for detection, differential diagnosis, and staging of this disease. When a large mineralized tumor involves body and posterior element of the spine in a middle-age person, primary spinal osteosarcoma may be considered in the differential diagnosis.

Osteosarcoma is the most common nonhematologic primary malignancy of bone and has been extensively documented in the appendicular skeleton. Primary osteosarcoma of the spine is rare. It accounts for 1.5-4% of all osteosarcoma and 5% of all primary malignant tumors of the spine [1, 2, 3]. Here we report a case of primary osteosarcoma of the cervicothoracic spine. Clinical presentations, imaging findings, and differential diagnosis of this disease are discussed. Although primary osteosarcoma of the spine is rare, we should keep it in mind when diagnosing a spinal tumor.

CASE REPORT

A 62-year-old female presented to our hospital with the complaints of progressive chest tightness, dizziness, urinary incontinence, and bilateral lower legs weakness for days. Her medical history was significant for diabetes mellitus and hypertension with regular medical control. On neurologic examinations, she had muscle weakness of left upper and bilateral lower extremities. An increase of deep tendon reflex in both upper and lower extremities was also present. The laboratory examinations were unremarkable except for a rise of sugar level (126 mg/dl).

Chest radiograph and anterior-posterior cervical radiograph revealed a large expansile mass with extensive ossifications in left superior mediastinum and lower neck. Destruction of the C7 to T2 vertebrae and adjacent left 1st and 2nd ribs was seen (Fig. 1). A magnetic resonance imaging (MRI) scan of spine revealed a large soft tissue mass occupying in left side of the C7, T1, and T2 vertebral bodies, posterior elements, epidural spaces, paravertebral soft tissue, and adjacent ribs. Areas of low signal intensity in all pulse sequences were suggestive of matrix mineralization. The intervertebral disc spaces were relatively spared (Fig. 2). Post-operative computed tomography (CT) of the chest revealed extensive ossifications in this lesion. The left subclavial artery was stretched by the tumor (Fig. 3). The initial diagnosis was a chondrosarcoma of the spine with cord compression.
The patient received C7 to T2 laminectomy and epidural tumor excision. Pathologic examinations revealed an osteoblastic osteosarcoma, manifested as infiltrative spindle-like, epithelioid, or plasmacytoid cancerous cells, sometimes including multinucleated giant cells. The pathologic diagnosis was conventional osteosarcoma, osteoblastic type (Fig. 4).

There was no significant change in neurologic deficits after operation. Unfortunately, fever, dyspnea, and leukocytosis were present 2 weeks after operation. The following chest radiograph revealed extensive pneumonia. The patient was expired due to sepsis and respiratory failure.
Primary osteosarcoma of the spine

DISCUSSION

Primary osteosarcoma of the spine is an uncommon condition, accounting for 1.5-4% of all osteosarcoma and 5% of all primary malignant tumors of the spine [1, 2, 3]. There is no significant sex difference. The age of the patients with spinal osteosarcoma ranges from 8 to 80 years (median age, 34.5) [2]. Our case is 62 years old. The most common clinical presentations are pain, neurological symptoms, and palpable mass [1].

Osteosarcomas have been reported at all levels of the spine. The lumbar and sacral regions are most commonly involved, followed by thoracic and cervical [3].

The most common histologic subtype is osteoblastic; others include chondroblastic, telangiectatic, fibroblastic, and small cell subtypes. There are no low-grade intramedullary or other surface osteosarcomas [2]. Osteosarcomas are categorized as primary or secondary. Primary osteosarcomas are subcategorized as conventional, low-grade intramedullary, parosteal, periosteal, high-grade surface, telangiectatic, and small cell osteosarcomas. Conventional osteosarcomas are high-grade tumors begin in an intramedullary location but may break through the cortex and form a soft tissue mass. Histologically they may be primarily osteoblastic, fibroblastic, or chondroblastic. Periosteal osteosarcoma is an intermediate-grade malignancy that arises on the surface of the bone. The most common locations are the diaphyses of the femur and tibia. Low-grade intramedullary osteosarcoma is a rare type characterized by an indolent course with relatively benign features on roentgenogram. Parosteal osteosarcoma also is a rare, low-grade malignancy, but it arises on the surface of the bone. It has a peculiar tendency to occur on the posterior aspect of the distal femur. High-grade surface osteosarcoma is the least common type. It is an aggressive tumor arising on the outer aspect of the cortex. Telangiectatic osteosarcoma is a purely lytic lesion. It can have a ballooned appearance similar to an aneurismal bone.
cyst. Small cell osteosarcoma is a high grade lesion that consists of small blue cells that may resemble Ewing sarcoma or lymphoma. Secondary osteosarcomas occur at the site of another disease process. The most common factors include Paget disease and previous radiation treatment [4].

In imaging studies, radiographs and CT scans demonstrate mineralized matrix in the majority of the cases [2]. A pure lytic pattern is less common. The majority of the tumors involve the body alone or the body and posterior element. Isolated involvement of the neural arch is rare [3]. Invasion of the spinal canal secondary to a soft-tissue mass is common. Involvement of two vertebral levels is seen in 12% cases. Cross-sectional imaging is considered essential for staging and treatment planning for vertebral lesions. CT is useful in assessing the soft tissue extent of the neoplasm. An increase in the attenuation values of the tissue within the medullary canal is generally indicative of tumor extension or “skip” metastases [5]. CT will also aid demonstrating intrathoracic metastasis. MRI is superior to CT in the assessment of intraosseous tumor involvement and in the identification of neural compression [3]. The neoplasm is typically of low signal intensity on T1-weighted spin echo MR images and can be differentiated from normal fatty marrow. Inhomogeneous or homogeneous high signal intensity within the tumor is usually evident on T2-weighted spin echo MR images. Enhancement of the tumor is evident after the intravenous administration of gadolinium compounds [5].

The differential diagnosis for spinal osteosarcoma includes any other solitary lytic, sclerotic, or mixed lesion. The one producing the greatest concern is benign osteoblastoma. Several reported cases of primary spinal osteosarcoma are misdiagnosed initially as benign osteoblastoma, because there is a close histologic similarity between the two lesions [6]. Osteoblastoma is observed most frequently in persons younger than 30 years. When in the spine, osteoblastoma usually presents as a well-defined, expansile osteolytic lesion that is partially or extensively calcified or ossified and arises from the posterior elements, especially in the thoracic or lumbar spine [7]. Other than osteoblastoma, the differential diagnosis may include chondrosarcoma and sclerotic metastasis. The pattern of calcification in chondrosarcoma has typically been described as punctate, popcorn, or comma-shaped. However, in some cases, bone may develop within the cartilaginous lobules as a result of the occurrence of endochondral ossification. Chondrosarcomas with such areas of ossification differ from chondroblastic osteosarcomas in the absence of direct production osteoid by the malignant spindle cells and in the lack of tissue alkaline phosphates activity [5]. A few helpful differential points allow a fundamental differentiation of primary versus secondary malignant spinal tumor. The appearance of a soft tissue mass, presence of periosteal response, a long lesion (> 6 cm), expansion of bone, and solitary lesion are more commonly found in primary malignant tumors than with secondary ones [8].

Patient with primary spinal osteosarcoma tends to be diagnosed late. Metastases commonly occur at the time of presentation. Metastases to the lungs, pleura, and other bones, as well as liver and abdominal viscera have been reported [1].

The current treatment protocols for osteosarcoma include of neoadjuvant chemotherapy administered before surgery to decrease surgical morbidity, followed by nearly total tumor mass excision and postoperative adjuvant chemotherapy to increase systemic control rates [6]. Postoperative radiotherapy may be beneficial [9]. Our patient did not receive pre-operative chemotherapy due to an acute spinal cord compression. An emergency decompressive laminectomy was performed instead.

The prognosis is dismal. The median survival has a range of 6-23 months [1, 3, 8]. The prognosis is poor primarily because the lesions are usually large at presentation and cannot be completely excised [10]. Patients with metastases, large tumors, and rapid relapse after completion of the initial treatment have a poor prognosis [9].

In summary, we report a rare case of primary osteosarcoma of the cervicothoracic spine presented with clinical presentations and radiologic examinations. When a large mineralized tumor involves body and posterior element of the spine in a middle-age person, primary spinal osteosarcoma should be considered in the differential diagnosis.

**REFERENCES**

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原发性脊椎骨肉瘤：病例报告

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原发性的脊椎骨肉瘤是一种罕见的疾病。我們在此報告一個 62 歲的女性病人，因為近幾天越來越嚴重的胸悶、頭痛、尿失禁、與雙側下肢無力而求診。胸部及頸椎 X 光片發現在左側上縱膈腔與下頸部有一個骨化的腫塊合併周遭骨頭破壞。胸部電腦斷層和脊椎核磁共振掃描發現第七頸椎與第一、二胸椎有一個包含軟組織與骨化的腫塊，侵犯到脊椎體、脊椎後部、周圍軟組織、及硬膜外腔。椎間盤部分則沒有受到侵犯。影像學的檢查對此疾病的偵測、鑑別診斷、及分期是很有幫助的。如果一個中年人，在他的脊椎體及脊椎後部有一個大型的骨化腫塊，我們須將原癥性的脊椎骨肉瘤放入鑑別診斷之中。