Case Report

Destructive osteoblastoma of the cervical spine with complete neurologic recovery

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Study design: Case report.

Objective: To describe a patient with a large tumor lesion of the 6th vertebrae affecting surrounding soft tissue, and symptoms of cord compression. Histologic diagnosis indicated a destructive osteoblastoma following dorsal and anterior resection and internal fixation.

Setting: University Hospital, Germany.

Methods: A 23-year old male patient was admitted with a 2-month history of increasing upper extremity weakness and pain. X-ray and MRI indicated massive involvement of the anterior and posterior elements of the 6th vertebrae with a large soft tissue mass. Following emergency decompression and dorsal stabilization, the pathologic investigation revealed a destructive osteoblastoma. Subsequent dorsal and anterior resection with internal fixation were performed.

Results: The patient initially presented with symptoms of beginning paraplegia of C6/7. According to the neurologic classification of spinal cord injury, motor function score was 56 and sensory function score 83. After emergency dorsal decompression and internal fixation with Luque-Instrumentation[®] he showed increasing neurological recovery. Complete neurological recovery was achieved at 2 and 12-months postoperatively, following secondary dorsal and anterior resection of the tumor and internal fixation with bone cement (PalacosR[®]) and Harms-cage[®]. Radiologic signs of local recurrence were identified 1 year postoperatively.

Conclusion: Osteoblastoma of the cervical spine is rare. Patients often present with severe neurological symptoms due to significant tumor mass. Complete resection is necessary to regain full recovery, to prevent recurrence and, in some cases, malignant transformation.

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Introduction

Benign osteoblastoma was independently described by Jaffe (1956)¹ and Lichtenstein (1956).² It is a vascular, osteoid and bone-forming tumor, cytologically characterized by osteoblasts. Radiological signs include osteolytic lesions larger than 2 cm, showing little or no evidence of perifocal sclerosis.³ Osteoblastomas account for approximately 1% of all primary bone tumors.^{3–5} About 30% to 40% of all cases involve the spine^{3,5–7} and of those about 20% to 40% the cervical spine.^{5,7–9} Most often the osteoblastoma is confined to the posterior elements.^{7,10–12} The main clinical feature is pain, following neurological symptoms and scoliosis.^{5,7,8} Frequently, there is an invasion of the epidural space, surrounding of nerve roots and cord compression.^{7,8,13,14} Recurrence rates after resection are described up to 10%.^{9,14–17} Some authors

have reported the possibility of malignant transformation.^{18–20} Surgery is aimed at complete resection and protection of the sensitive neuroanatomic structures.

Case report

A 23-year-old male patient presented with acute neurological symptoms, after complaining of muscle weakness in the upper extremity for 2 months. No ambulatory X-ray investigation had been performed up to this point.

The physical examination indicated a loss of superficial pain in the lower extremity, the trunk and the right digiti quintus. Superficial sensory function was decreased in the lower extremity and the trunk. Muscle stretch reflexes of the upper extremity were hyperactive, and normal for the lower extremity. Muscular status results indicated grade 0 for the lumbrical muscles, grade 3 for C7 and C8 innervated

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muscles on both sides and grade 4 for C4–6 innervated muscles. C1–3 innervated muscles showed normal power on the right side. The patient's motor function score was 56 and his sensory function score reached 83 according to the standard neurologic classification of spinal cord injury. The ASIA/IMSOP scale was classified grade B. Standard cervical X-ray (anteroposterior and lateral view) showed a sclerotic tumor lesion involving the vertebrae with no sign of instability (Figure 1).

MRI of the spine indicated a local destructive tumorous lesion of the 6th cervical vertebrae with compression of the spinal cord and paravertebral involvement, both anteriorly and posteriorly (Figures 2 and 3).

Figure 1 Sclerotic osteoblastoma of the 6th cervical vertebrae

According to the Enneking score of benign musculoskeletal lesions the tumor was classified grade 3.²¹

Emergency dorsal decompression of C5–7 combined with Luque-Spondylodesis® C4–Th1 was performed initially. Thereafter the patient presented with normal power, decrease of pain and an increase of sensory function. Biopsy results indicated a local destructive osteoblastoma. Postoperative angiography of the vertebral arteries produced no sign of compression. Complete dorsal extirpation of the tumor was performed 3 weeks later. The patient continued to present with normal power and reduction of sensation

Figure 2 Involvement of the anterior and posterior parts of the vertebra with signs of spinal cord compression

Figure 3 Tumorous destruction of the posterior lamina and the vertebra with involvement of the surrounding soft tissue

of the upper extremity. Light touch, pin prick and vibration were decreased, especially on the right side, of C7 and C8 dermatomes, where temperature was normal. During a 3rd stage operation the tumor and the 6th vertebrae were resected anteriorly and segment stabilization was performed with bone cement (Palacos®), Orion-plate® and Harms-cage® implantation (Figure 4).

Macroscopic and histologic results indicated disappearance of the tumor. However, postoperative X-ray showed a sclerotic area at the dorsal side of the preserved part of the body.

Philadelphia-Orthesis® was applied for postoperative mobilization during the following 6 weeks. Before discharge the patient still showed reduced sense of light touch, pin prick and vibration of C7 and C8 on both sides. ASIA/IMSOP scale was classified grade D. The motor function score increased to 89 points and the sensory function score to 216 points. Two months post surgery the patient presented with full neurologic recovery and functional scoliosis due to muscular weakness of the cervical spine. The X-ray still indicated a small sclerotic lesion on the dorsal side of the vertebral body and with stable internal fixation. At 4-months post surgery, CT-scans showed no evidence of recurrence and normal neurologic function. Changes in the sclerotic area were visible in the 1-year postoperative X-ray (Figure 5).

A subsequent MRI and CT-scan found high rates of metal artefacts, indicating signs of local recurrence paravertebrally at the right transverse process of the 6th vertebrae (Figure 6).

The patient maintains normal sensory and motor functions (100 points motory function score and 224 points sensory function score).

Therefore, current surgical treatment is not necessary, and the next follow-up is scheduled 4 months later for clinical, X-ray and MRI investigation.

Figure 4 Internal fixation with Orion-plate®, Harms-cage® and Palacos® anterior and Luque-Spondylodesis® C4-Th1 posterior

Discussion

Osteoblastoma is rare. It accounts for approximately 0.5% to 1% of all primary bone tumors.^{3–5} Spinal involvement is described in 30% to 40% of all cases.^{3,5–7} In 20% to 40% of those, the cervical spine is affected.^{5,7–9} Osteoblastoma is most frequently found in the posterior elements.^{7,10–12} Involvement of the cervical spine when compared to other spinal regions is related to a higher rate of anterior destruction, probably due to reduced volume of the posterior elements.¹¹ Pain is usually the main clinical symptom, followed by neurologic symptoms, scoliosis and torticollis.^{5,7,8} Compared to osteoid osteoma there is a higher rate of severe neurological deficit.^{7,8,13,14} The large tumor mass can be associated with compression of the vertebral arteries.

Angiography in the current case showed no sign of compression. Zambelli *et al.* recommended that the vertebral arteries should be preserved in all cases.²² Classical X-ray investigation shows radiolucent destructions with a perifocal sclerosis.³ In the current case the X-ray showed a sclerotic lesion of the vertebra and the pedicles, indicating a departure from the classic radiographic picture.

Delay of diagnosis is very common, and seems to occur on average 6–12 months or later following the

Figure 5 One year postoperative with changes of the dorsal sclerotic area

Figure 6 Signs of local recurrence at the right transverse process of the 6th vertebra

initial presentation.^{5,8,23} In the current case, the patient was correctly diagnosed after 2 months due to his significant neurological symptoms. Symptoms of

increasing muscle weakness and pain were evident even though no prior X-ray examination had been performed.

The treatment of choice for osteoblastoma is complete surgical resection. Preoperative interdisciplinary cooperation is necessary between radiologist, neurosurgeons, vascular surgeons and orthopaedic surgeons. In our case macroscopic and histologic results did not point to a residual tumor mass, but the postoperative X-ray still showed a sclerotic area at the dorsal column of the left vertebral part. Recurrence rates are described up into 10%, especially in Enneking Grade 3 lesions.^{9,14–17} One year post surgery, this case showed radiologic signs of paravertebral local recurrence without any neurological symptoms. In some cases there is a possibility of malignant transformation.^{18–20} If complete resection is not possible, radiotherapy and in some cases chemotherapy seem to be alternative treatment options.^{24–27}

Osteoblastoma should be ruled out if patients present with neurological symptoms and pain over extended periods of time, especially at night. Delay in diagnosis is common and effective treatment is important to prevent neurologic complications, recurrence and malignant transformation. In cases with large tumor masses it is difficult to perform complete resection.

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