J Orthop Sci (2009) 14:336–338
DOI 10.1007/s00776-008-1323-6

JOURNAL OF
ORTHOPAEDIC SCIENCE

Case report

Dumbbell-shaped osteochondroma of the fifth rib causing spinal cord compression

Masaaki Chazono[1], Fumiaki Masui[1], Yasuhiko Kawaguchi[1], Hiromichi Hazama[1], Junko Ueda[1], Shigeru Saito[1], Yoshitaka Ito[1], Kentaro Kasama[1], Keisho Liu[1], and Keishi Marumo[2]

[1] Department of Orthopaedic Surgery, The Jikei University School of Medicine, Kashiwa Hospital, 163-1 Kashiwa-shita, Kashiwa-shi, Chiba 277-8567, Japan
[2] Department of Orthopaedic Surgery, The Jikei University School of Medicine, Tokyo, Japan

Introduction

Osteochondromas are the most common benign bone tumor, representing 45% of all benign bone tumors.[1] Most occur in the metaphyseal region of long bones such as the femur and tibia. Osteochondromas can also arise from flat bones and the spine.[2] Costal osteochondromas make up only 1.5% of all osteochondromas,[1] and compressive myelopathy due to a tumor arising from the rib is even rarer. We present an unusual case of a dumbbell-shaped osteochondroma causing spinal cord compression, and we review the literature concerning costal osteochondromas.

Case report

A 23-year-old man with a past medical history remarkable for nine prior surgeries for nonheritable multiple osteochondromas in the upper and lower extremities presented at a national cancer center 6 months prior to his current presentation, complaining of numbness in his left lower extremity. The etiology of his numbness was not identified, and he was referred to our hospital with gait disturbance and paresthesias originating from the abdomen and extending down both legs.

On physical examination, there was incomplete motor loss with 3/5 power in both lower extremities. Sensory examination showed diminished touch sensation on the right side and diminished pain sensation on the left, with the highest level of involvement at T7, suggesting symptoms of Brown-Sequard syndrome. The patient demonstrated hyperreflexia in both lower extremities with ankle clonus. The Babinski response was not evident.

Offprint requests to: M. Chazono
Received: June 3, 2008 / Accepted: October 14, 2008

Laboratory tests were within normal limits. Plain radiography showed a small lobulated calcified mass that appeared to arise from the right fifth rib. Multiplanar computed tomography (CT) revealed a dumbbell-shaped bony tumor arising from the right fifth rib at the costovertebral junction, creating an intraspinal and extraforaminal mass that widened the neural foramen at the T5/6 level (Fig. 1). In addition, CT myelography showed the bony mass compressing the spinal cord. Magnetic resonance imaging (MRI) demonstrated that the tumor, which had intermediate signal intensity on T1-weighted images and low signal intensity on T2-weighted images, pushed the spinal cord to the left side. The 2.5 mm thick rim of the intraspinal lesion demonstrated intermediate signal intensity on T1-weighted images and marked high signal intensity on T2-weighted images (Fig. 2). The clinical and radiological diagnosis was an osteogenic tumor, likely an osteochondroma that arose from the rib.

The patient underwent surgical resection of his tumor. With the patient in the prone position, a posterior midline incision was performed. Using a flexible ball-tipped wire, a diamond T-saw (modified threadwire saw coated with diamond particles on the surface of braided stainless steel wire, 0.54 mm in diameter; Medtronic Sofamor Danek Japan, Osaka, Japan) produced a fine cut of the pars interarticularis at T5 bilaterally. Next, the T-saw was introduced into the T6 foramen, and then the saw was pulled around the T6 pedicle ventral to the superior articular processes of T6 to produce a fine cut from the pedicle to the transverse process at T6 bilaterally.[3] Next, en bloc laminectomy of the pars interarticularis at T5 and the pedicle-transverse process at T6 was performed to expose the tumor fully. The dura was easily mobilized from the tumor. The visible tumor including the intraspinal and extraforaminal components were resected piecemeal using forceps and curettage, resulting in subtotal tumor removal. The T5 nerve root was preserved in its entirety during the procedure.

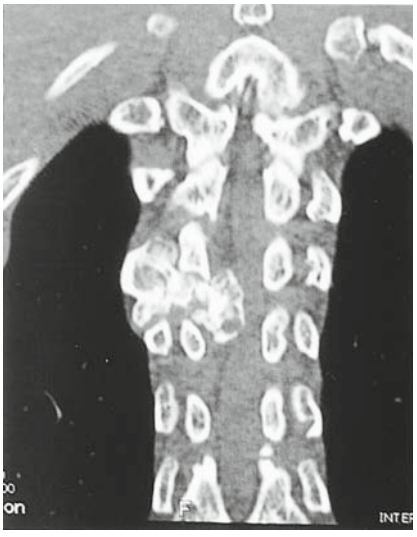

Fig. 1. Coronal reconstruction image of multiplanar computed tomography reveals a dumbbell-shaped bony tumor arising from the right fifth rib at the costovertebral junction, creating an intraspinal and extraforaminal mass that widened the neural foramen at the T5/6 level

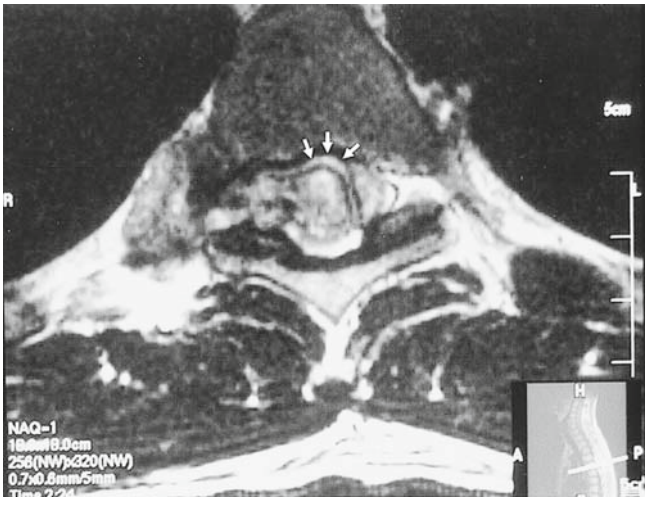

Fig. 2. Axial T2-weighted magnetic resonance image demonstrates a dumbbell-shaped lesion at the T5/6 level. The rim of the lesion demonstrated marked high signal intensity on T2-weighted images. The cartilage cap was well visualized on the image (*arrows*)

The complex of laminae and facets at T5 and T6 was recapped in the original anatomical position with sutures. Histopathological examination demonstrated a typical osteochondroma without evidence of malignant transformation (Fig. 3). Postoperatively, the patient's spine was immobilized in a thoracic orthosis for 6 months. The postoperative period was uneventful. Within a week, the patient's paraparesis gradually receded, and his numbness diminished. Two months

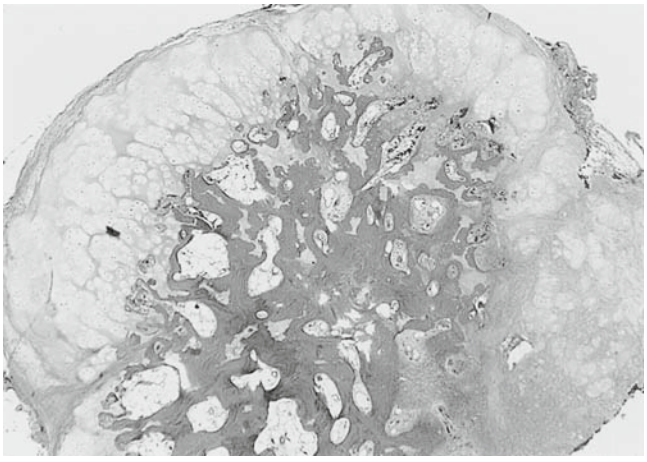

Fig. 3. Photomicrograph of the tissue specimen demonstrates a typical osteochondroma. A cartilage cap overlying the bony trabeculae was observed, and a cellular zone of chondroosseous transformation was seen at the interface. H&E × 5

postoperatively, the patient had fully recovered muscle strength in his bilateral lower extremities. At the 4-month-follow-up, CT revealed complete bone union at the osteotomy site of the left transverse process at T6. An additional CT scan obtained 6 months after surgery revealed no sign of tumor progression. The patient had no back pain, and there was no delayed instability or resultant deformity of the affected thoracic spine.

Discussion

The type of osteochondroma described in this case, with neural foraminal extension and spinal cord compression, is extremely rare. There have been only six cases previously reported in the English-language literature (Table 1).[4–8] All six patients were younger than 21 years (range 12–21 years), and there was a female predominance. Five of the patients had severe neurological deficits, and a posterior approach including laminectomy was chosen for surgical intervention. All patients showed clinical improvement during the recent follow-up period after the tumor was entirely excised.

Routine CT and MRI studies are valuable for the diagnosis and evaluation of costal osteochondroma, allowing determination of the tumor's origin, size, and extent of invasion into the spinal canal. Although the location of the bony part of the osteochondroma can be reliably demonstrated using multiplanar CT reconstruction, the exact size of the tumor may be underestimated because the cartilage cap of the tumor is invisible. Cartilage itself is best shown on MRI, where it demonstrates intermediate signal intensity on T1-weighted images and markedly high signal intensity on T2-weighted images. Cartilage caps with a thickness of

Table 1. Summary of six cases of costal osteochondromas causing spinal cord compression

Parameter	Twersky[8] (1975)	Twersky[8] (1975)	Natarajan[5] (1976)	Kane[4] (1994)	Tang[7] (1998)	Rao[6] (2007)
Age (years)	12	11	21	17	16	12
Sex	M	F	M	F	F	F
Original rib	5th	4th	5th	10th	12th	6th
Approach	L	L	T	L	L + F	L
Final follow-up (months)	ND	6	6	3	19	ND
Outcome	CR	PR	CR	CR	CR	PR
Recurrence	ND	ND	No	No	No	ND

L, laminectomy, T, thoracotomy, F, facetectomy; ND, no data available; CR, completely recovered; PR, partially recovered

>3 mm can be reliably detected on MRI.[9] Conversely, the cap may appear indistinguishable from adjacent soft tissue including epidural fat or cerebrospinal fluid on T2-weighted images when the tumor involves the spinal canal. The unique finding in this case is that the high signal intensity around the tumor, shown on axial T2-weighted images, may represent the cartilage cap, leading to the proper preoperative diagnosis.

Asymptomatic osteochondromas can be followed without intervention, whereas symptomatic lesions require surgical management. As a general rule, osteochondromas comprise a disease of growing bones and typically present in younger patients. Tumor growth occurs early during childhood and usually arrests after puberty when the epiphysis is closed. For symptomatic lesions, excision is recommended for the treatment. We believe that costal osteochondromas causing neurological symptoms should be excised as soon as possible, regardless of the patient's age.

As these osteochondromas can pass through the neural foramen and lead to cord compression, the surgical approach should include decompression surgery such as laminectomy and/or facetectomy at the corresponding level. Traditionally, laminectomy has been used to expose the spinal canal during surgery; however, it has a significant shortcoming in that the extraforaminal component is invisible. Even if additional facetectomy is performed, iatrogenic instability and kyphosis may occur during the follow-up period. To avoid tumor recurrence, carefully planned surgical strategy is critical for complete tumor excision.[10] In our case, en bloc laminectomy of the lamina–facet complex (pars interarticularis at T5 and pedicle-transverse process at T6) using a diamond T-saw provided wide exposure of the intraspinal and extraforaminal tumor originating from the rib. It also allowed reconstruction of the posterior element of the spinal canal and preserved stability of the affected spine. In this case, CT showed that the lamina–facet complex was restored at the original site, and bone union was achieved 4 months postoperatively. An additional CT scan obtained at 6 months after surgery revealed no sign of tumor progression. However, further clinical and radiological follow-up are required to monitor for tumor recurrence as well as any delayed instability and resultant deformity.

Acknowledgments. The authors thank the patient and his family for allowing us to present the clinical data reported here.

None of the authors of this manuscript has received any type of support, benefits, or funding from any commercial party related directly or indirectly to the subject of this article.

References

1. Uchida A, Otsuka T, Wada T, Okada K, Kawai A, Matsumoto S, et al. JOA Musculoskeletal Tumor Committee: Bone Tumor Registry in Japan. Tokyo: National Cancer Center; 2006 (in Japanese).
2. Brastianos P, Pradilla G, McCarthy E, Gokaslan ZL. Solitary thoracic osteochondroma: case report and review of the literature. Neurosurgery 2005;56:1379.
3. Kawahara N, Tomita K, Shinya Y, Matsumoto T, Baba H, Fujita T, et al. Recapping T-saw laminoplasty for spinal cord tumors. Spine 1999;24:1363–70.
4. Kane PJ, Coulthard A, Raghavan R, Jenkins A. Osteochondroma of the rib: an unusual cause of paraparesis. Surg Neurol 1994;41:414–7.
5. Natarajan M, Balakrishnan D, Srinivasan V. Solitary osteochondroma causing spinal cord compression. Int Surg 1976;61:494–5.
6. Rao A, Abraham RG, Rajshekhar V. Osteochondroma of rib with neural foraminal extension and cord compression. Neurol India 2007;55:428–9.
7. Tang WM, Luk KD, Leong JC. Costal osteochondroma: a rare cause of spinal cord compression. Spine 1998;23:1900–3.
8. Twersky J, Kassner EG, Tenner MS, Camera A. Vertebral and costal osteochondromas causing spinal cord compression. Am J Roentgenol Radium Ther Nucl Med 1975;124:124–8.
9. Lee JK, Yao L, Wirth CR. MR imaging of solitary ostechondromas: report of eight cases. AJR Am J Roentgenol 1987;149:557–60.
10. Albrecht S, Crutchfield JS, SeGall GK. On spinal osteochondromas. J Neurosurg 1992;77:47–52.