

Treatment of costal osteochondroma causing spinal cord compression by costotransversectomy: case report and review of the literature

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Abstract

In laminectomies for costal osteochondroma causing spinal cord compression, visualization of the extraforaminal part of the tumor is limited. The authors describe using a costotransversectomy to resolve spinal cord compression by a costal osteochondroma invading through the neural foramen. A 21-year-old woman with hereditary multiple exostoses presented with hand numbness and progressive neck and upper back pain. Plain radiographs identified a large lesion of the T2 and T3 pedicles, with encroachment on the T2-3 neural foramen causing ~50% spinal canal stenosis. Costotransversectomy was performed to resect the cartilaginous portions of the osteochondroma, debulk the mass, and decompress the spinal canal. A mass of mature bone was left, but no appreciable cartilaginous tumor. At five-year follow-up, the patient had improvement of neck pain, no new neurological deficits. a stable residual mass, and no new osteochondromas, indicating that appropriate surgical management can yield good results and no evidence of recurrence.

Introduction

Costal osteochondromas are a rare cause of spinal cord compression. These tumors can arise from the costovertebral or costotransverse joints and may pass through neural foramen and occupy the spinal canal, causing severe stenosis. Most osteochondromas are solitary, but multiple lesions can occur, usually in association with hereditary multiple exostoses (HME). We describe the use of costotransversectomy to resect a large costal osteochondroma arising from the third rib that caused spinal cord compression in a patient with HME.

Case Report

History and presentation

A 21-year-old woman with a family history of HME presented to a chiropractor with a 2-month history of bilateral hand numbness in the fourth and fifth digits and progressive neck and upper back pain. Plain radiographs of her neck identified a large thoracic lesion (Figure 1A), which prompted a neurosurgery consultation before any chiropractic manipulations were performed. The patient denied any loss of fine motor dexterity, bowel or bladder dysfunction, or gait difficulty. Physical examination revealed no weakness, sensory deficit, dysreflexia, or signs of myelopathy. Palpable bony protrusions were identified on her bilateral proximal metacarpals. A thoracic computed tomographic (CT) scan with bone windows showed a large expansile lesion with mature osseous architecture arising from the medial right aspect of the second and third ribs. The lesion enveloped the right T2 and T3 pedicles, with encroachment on the right T2-3 neural foramen causing approxi-

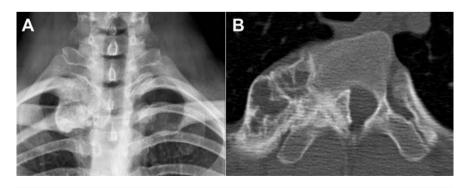


Figure 1. A) Plain radiograph demonstrating osteochondroma arising from the head of the right third rib. B) Axial computed tomography section at T3 demonstrating expansile lesion with mature osseous structure that arises from the third rib, envelops the pedicle, and extends through the T2-3 neural foramen to narrow the spinal canal by approximately 50%.

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mately 50% spinal canal stenosis (Figure 1B). A thoracic magnetic resonance imaging (MRI) study showed displacement of the thecal sac and the development of a small syrinx at this level but no abnormal signal change within the spinal cord (not shown). Imaging also revealed similar smaller osseous lesions arising from the costovertebral joints at T9 and T10, the vertebral body at L2, and the right scapula (not shown).

Operation

Surgical decompression of the spinal canal was performed in a posterolateral manner via costotransversectomy. A semilunar skin incision was used with the arc oriented toward the right overlying the origin of the exostosis. Dissection was carried down until the right T2 and T3 costotransverse joints, transverse processes, and lamina were exposed. Hemilaminectomy of T2 and T3 and costotransversectomy of T3 were performed. The right T3 costal head, proximal 4 cm of rib, transverse process, pedicle, and posterolateral vertebral body were removed to obtain the appropriate tangential angle to expose the anterior compartment of the spinal canal.

The tumor arose from the T3 costal head and extended both ventrolaterally to abut the pleura and medially through the T2-3 neural foramen and into the spinal canal (Figure 2A





and B). A cartilaginous cap surrounded by mature osseous architecture was visualized within the mass. The semilunar incision enabled excellent exposure both to resect the cartilaginous cap and decompress the intervertebral foramen and spinal canal. The prominence of tumor was debulked from the region between the second and third rib heads to the intraspinal space by using a high-speed drill with diamond bur (Figure 2C). The thecal sac was markedly displaced towards the left. Adhesions to the thecal sac were detached carefully so the dura was not violated. Intraoperative ultrasound was used to confirm adequate decompression of the spinal cord. After the decompression, a mass of mature bone was left between the T2 and T3 vertebral bodies, but there was no appreciable cartilaginous portion of the tumor remaining.

Intraoperative thoracic manipulation did not reveal abnormal motion at the affected levels so instability was not suspected. The tumor was not resected further so spinal instrumentation and fusion could be avoided. The decision was made to close the incision and wait for the pathological diagnosis, which, if it was malignant tumor, would guide further surgical and/or medical treatment. Histological examination of the operative specimen, however, confirmed the diagnosis of osteochondroma. Thus, given the patient's age and adequacy of the decompression, further osteochondroma removal and risk of spinal destabilization were not warranted.

Postoperative course

The patient had an uneventful hospital course and was discharged on postoperative day 3. She developed no new neurological deficits and reported improvement in her preoperative neck pain at her 1-year follow-up examination. Imaging obtained at 1, 3, and 5 years after the operation showed a stable 3×2 -cm residual mass at T2-3 that remained unchanged in size on each successive study, and no new osteochondromas were identified (Figure 3). Interval thoracic radiographs demonstrated normal anatomic alignment of the thoracic spine with no signs of instability.

Discussion

Osteochondromas are the most common benign bone tumors. They may be solitary or they may occur as multiple lesions, as in HME, a genetic disorder with autosomal dominant inheritance and incomplete penetrance in females. In both solitary osteochondromas and HME, cartilage-capped bony projections, also called exostoses, arise from the periosteum and grow exophytically by endochondral bone formation. They typically occur at the epiphyses of the long bones, such as around the knee and humerus, and are usually found during childhood and adolescence but may continue to grow as long as the parent bone epiphysis remains open. Involvement of the spine or posterior ribs is rare, accounting for less than 4% of cases,¹ most of which are asymptomatic.

Presenting symptoms

While previously reported symptoms include hemothorax, pneumothorax, hiccups, recurrent chest infections, and thoracic outlet syndrome,^{2,3} costal osteochondromas rarely cause spinal cord compression. In most cases, neurologic disease results as the mass slowly compresses neural structures as it expands; however, symptoms may occur acutely after sudden hyperextension of the spine or after a fall. Extradural spinal cord compression usually arises from an intraspinal osteochondroma originating from the pedicle, lamina, vertebral body, or facet. Spinal cord compression caused by expansion of extra-axial osteochondroma invading through the intervertebral foramen is much less common. Such costal osteochondromas may present as dumbbell-shaped lesions that originate from the rib head, extend from extraforaminal to intraspinal spaces, widen the neural foramen, and occupy the spinal canal.

Diagnosis and treatment

CT and MRI are useful for diagnosis and preoperative planning, allowing evaluation of the tumor origin, size, and extent of spinal canal invasion. CT allows optimal depiction of the point of attachment where the cortex and marrow of the parent bone flare in continuity into the osteochondroma, which is considered pathognomonic.⁴ MRI can best evaluate the cartilaginous cap of the osteochondroma as well as visualize the effect of the tumor on the spinal cord and nerve roots. To ensure an adequate resection, the cartilaginous cap of the tumor should be removed with clear margins. Hyaline cartilage cap thickness is dependent on skeletal maturity. In young patients, the cap is usually 1-3 cm, but it may be only a few millimeters thick or even appear absent in adults.⁴ Incomplete resection of the cartilaginous component can lead to recurrence, the rate of which is estimated to be 2-4%.⁴ Malignancy should be suspected in cases that recur after resection or if the thickness of the cartilage cap is greater than 3 cm in children or 2 cm in adults.5 Malignant degeneration occurs in about 1% of solitary osteochondromas and 10-15% in patients with HME.6

Given the risk of spinal cord injury and the risk of malignant transformation, all osteochondromas causing cord compression should be surgically resected. An appropriate surgical approach must account for the location of the cartilaginous portion of the tumor and the potential to compromise the stability of the vertebral column. Most cases of costal

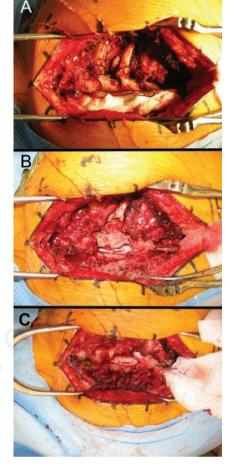
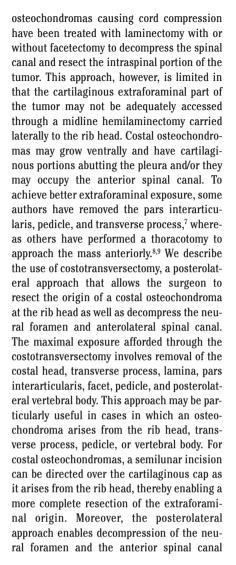


Figure 2. A) Intraoperative photograph of surgical dissection showing intact T2 (leftmost) and T3 lamina, spinous and transverse processes, and rib heads on the right side. The osseous tumor can be seen between the second and third rib heads. A surgical sponge is covering the left T2 and T3 lamina. B) Intraoperative photograph showing costal osteosarcoma extending into the spinal canal and causing leftward displacement of the spinal cord. C) Intraoperative photograph of spinal cord returning to anatomical location in spinal canal after resection of the intraspinal portion of the costal osteochondroma.



Figure 3. Axial computed tomography section at T2 obtained 5 years after osteochondroma resection. There is no evidence of tumor recurrence, and the spinal canal remains decompressed.



through a unilateral working space, so the risk of postoperative instability developing is decreased.

Review of reported cases

Only 15 cases of costal osteochondromas causing spinal cord compression have been reported, including ours (Table 1).7-19 Nine patients were male, 6 were female. Patients tended to be in their second or third decade of life (median age 17 years, range 10-65 years). Seven cases occurred in patients with HME, 8 cases were solitary lesions. Presenting neurologic symptoms included myelopathy,^{10,11} bowel or bladder dysfunction,^{8,11,12} spastic paraparesis,^{7,8,10,12-16} Brown-Sequard syndrome,^{7,17} Horner syndrome,¹⁷ and pain.^{9,10,18,19} All patients underwent surgical decompression, and all experienced improvement in preoperative symptoms after surgery. Most cases were managed by laminectomy to decompress the neural arch and debulk the tumor.^{11-13,15,17,18} In 1 case, laminectomy was augmented with removal of the pars interarticularis at the same level and removal of the pedicle and transverse process at the level below.7 In 2 cases, anterior resection was achieved through a thoracotomy.8,9 We found only three other reported cases of rib resection combined with laminectomy to resect a costal osteochondroma causing spinal cord compression.^{10,16,19}

Outcomes are excellent after decompression of neural elements for osteochondromas in the spinal canal. Marcove and Huvos²⁰ reported that in contrast to malignant tumors of the ribs, costal osteochondromas less than 4 cm in diameter may be treated by local resection,

even if incomplete, without recurrence. Sener et al.18 described a case of subtotal resection of a large solitary costal osteochondroma in which the remnant bulky mass passed through the neural foramen requiring another surgical resection within 3 years. Recurrence of a costal osteochondroma, however, has not been reported. This may reflect the very low prevalence of costal osteochondromas that are symptomatic and managed surgically. The residual cartilage cap from a costal osteochondroma would be expected to form exostoses in a similar manner to recurrent osteochondromas arising from other bones. Most osteochondromas are diagnosed in younger patients whose bones are still growing, so these lesions are unlikely to recur once a patient reaches skeletal maturity. Though isolated cases of late spinal cord compression in elderly patients have been reported, there is histological evidence that aged osteochondromas have diminished potential for endochondral ossification.4 In most elderly patients, osteochondromas remain asymptomatic until degenerative changes develop in the vertebral column and progressive spinal cord compression ensues.4

Conclusions

A costal osteochondroma invading through the neural foramen is a rare cause of spinal cord compression. Costotransversectomy provides an excellent approach for accessing the rib head, neural foramen, pedicle, and anterolateral spinal canal. The goal of surgery is to

Table 1. Reported cases of costal osteochondromas causing spinal cord compression.

Age, gender	Level	Rib of lesion origin	Presenting neurologic symptom	Operation	Osteochondroma type
17, M	T3	2, 3	Brown-Sequard syndrome, Horner syndrome	Laminectomy	HME
23, M	T5-6	5	Spastic paraparesis, Brown-Sequard syndrome	Laminectomy	Solitary
15, M	T10-11	10, 11	Spastic paraparesis	Laminectomy	HME
19, M	T4	4	Spastic paraparesis	Laminectomy	Solitary
17, F	T9-10	10	Myelopathy, pain, spastic paraparesis	Costotransversectomy	Solitary
33, M	T3	3	Spastic paraparesis	Laminectomy	HME
10, M	T6-8	8	Incontinence, spastic paraparesis	Laminectomy	Solitary
21, M	T4-5	5	Incontinence, spastic paraparesis	Thoracotomy	Solitary
21, F	T3-4	3	Pain	Thoracotomy	HME
65, M	T6-7	6	Pain	Laminectomy	Solitary
12, M	T4-6	5	Myelopathy, incontinence	Laminectomy	Solitary
11, F	T2-3	4	Spastic paraparesis, incontinence	Laminectomy	HME
12, F	T6-7	6	Spastic paraparesis	Costotransversectomy	Solitary
16, F	T12	12	Pain	Costotransversectomy	HME
21, F	T2-3	3	Pain	Costotransversectomy	HME
	17, M 23, M 15, M 19, M 17, F 33, M 10, M 21, M 21, F 65, M 12, M 11, F 12, F 16, F	17, M T3 23, M T5-6 15, M T10-11 19, M T4 17, F T9-10 33, M T3 10, M T6-8 21, M T4-5 21, F T3-4 65, M T6-7 12, M T4-6 11, F T2-3 12, F T6-7 12, F T6-7 16, F T12	origin 17, M T3 2, 3 23, M T5-6 5 15, M T10-11 10, 11 19, M T4 4 17, F T9-10 10 33, M T3 3 10, M T6-8 8 21, M T4-5 5 21, F T3-4 3 65, M T6-7 6 12, M T4-6 5 11, F T2-3 4 12, F T6-7 6 16, F T12 12	origin17, MT32, 3Brown-Sequard syndrome, Horner syndrome23, MT5-65Spastic paraparesis, Brown-Sequard syndrome15, MT10-1110, 11Spastic paraparesis19, MT44Spastic paraparesis17, FT9-1010Myelopathy, pain, spastic paraparesis33, MT33Spastic paraparesis10, MT6-88Incontinence, spastic paraparesis21, MT4-55Incontinence, spastic paraparesis21, FT3-43Pain65, MT6-76Pain12, MT4-65Myelopathy, incontinence11, FT2-34Spastic paraparesis16, FT1212Pain	origin17, MT32, 3Brown-Sequard syndrome, Horner syndromeLaminectomy23, MT5-65Spastic paraparesis, Brown-Sequard syndromeLaminectomy15, MT10-1110, 11Spastic paraparesisLaminectomy19, MT44Spastic paraparesisLaminectomy17, FT9-1010Myelopathy, pain, spastic paraparesisCostotransversectomy33, MT33Spastic paraparesisLaminectomy10, MT6-88Incontinence, spastic paraparesisLaminectomy21, MT4-55Incontinence, spastic paraparesisThoracotomy21, FT3-43PainThoracotomy12, MT4-65Myelopathy, incontinenceLaminectomy11, FT2-34Spastic paraparesis, incontinenceLaminectomy12, FT6-76Spastic paraparesis, incontinenceLaminectomy12, FT6-76Spastic paraparesis, incontinenceLaminectomy12, FT1212PainCostotransversectomy

HME, hereditary multiple exostoses.





decompress the spinal canal and resect the cartilaginous portions of the osteochondroma. The risk of recurrence is low, and a subtotal resection may yield excellent results and obviate spinal destabilization and the need for fusion.

References

- Albrecht S, Crutchfield JS, SeGall GK. On spinal osteochondromas. J Neurosurg 1992;77:247-52.
- Pham-Duc ML, Reix P, Mure PY, et al. Hemothorax: an unusual complication of costal exostosis. J Pediatr Surg 2005;40:e55-7.
- Alifano M, Morcos M, Molina T, Regnard JF. An unusual cause of hiccup: costal exostosis. Treatment by video-assisted thoracic surgery. Eur J Cardiothorac Surg 2003;23: 1056-8.
- Gille O, Pointillart V, Vital JM. Course of spinal solitary osteochondromas. Spine (Phila Pa 1976) 2005;30:E13-9.
- 5. Brastianos P, Pradilla G, McCarthy E, Gokaslan ZL. Solitary thoracic osteochondroma: case report and review of the liter-

ature. Neurosurgery 2005;56:E1379.

- 6. Dahlin D, Unii K. Bone tumors. Springfield, IL: Charles C. Thomas; 1986.
- 7. Chazono M, Masui F, Kawaguchi Y, et al. Dumbbell-shaped osteochondroma of the fifth rib causing spinal cord compression. J Orthop Sci 2009;14:336-8.
- Natarajan M, Balakrishnan D, Srinivasan V. Solitary osteochondroma causing spinal cord compression. Int Surg 1976;61:494-5.
- Old WL, Jr., Triplett JN, Jr. Osteochondroma with thoracic cord compression in hereditary multiple exostoses: a case report. Va Med 1979;106:302-6.
- Kane PJ, Coulthard A, Raghavan R, Jenkins A. Osteochondroma of the rib: an unusual cause of paraparesis. Surg Neurol 1994;41:414-7.
- 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.
- 12. Mannoji C, Yamazaki M, Kamegaya M, et al. Paraparesis caused by rib exostosis in a child with Down syndrome: a case report. Spine (Phila Pa 1976) 2008;33:E911-3.
- 13. Larson NE, Dodge HW Jr, Rushton JG,

Dahlin DC. Hereditary multiple exostoses with compression of the spinal cord. Proc Staff Meet Mayo Clin 1957;32:729-34.

- Decker RE, Wei WC. Thoracic cord compression from multiple hereditary exostoses associated with cerebellar astrocytoma. Case report. J Neurosurg 1969;30:310-2.
- Faik A, Mahfoud Filali S, Lazrak N, et al. Spinal cord compression due to vertebral osteochondroma: report of two cases. Joint Bone Spine 2005;72:177-9.
- Rao A, Abraham RG, Rajshekhar V. Osteochondroma of rib with neural foraminal extension and cord compression. Neurol India 2007;55:428-9.
- 17. Becker M, Epstein F. Case report 77. Skeletal Radiol 1978;3:197-9.
- Sener RN. Osteochondroma of the thoracic spine: a dumbbell mass associated with spinal cord compression. Comput Med Imaging Graph 1998;22:361-3.
- 19. Tang WM, Luk KD, Leong JC. Costal osteochondroma. A rare cause of spinal cord compression. Spine (Phila Pa 1976) 1998; 23:1900-3.
- 20. Marcove RC, Huvos AG. Cartilaginous tumors of the ribs. Cancer 1971;27:794-801.