

Case Report

Solitary osteochondroma of the pediatric cervical spine: a case report

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Abstract: Osteochondroma of the cervical spine is rare. This is a report of an affected 9-year-old boy with no known familial predisposition for osteochondromas. Imaging revealed a sessile osteochondroma arising from the spinous process and a portion of the left lamina of the seventh cervical vertebrae. The patient was successfully treated with an en-bloc excision of the tumor. Histopathology confirmed that the tumor was an osteochondroma. A follow-up of five-year and five-month confirmed the positive outcome of the treatment with no signs of recurrence and resulting in patient satisfaction.

Keywords: Osteochondroma, cervical spine, pediatric

Introduction

Osteochondroma is the most common benign tumor of the appendicular skeleton characterized by abnormal periphyseal ectopic endochondral ossification [1]. The tumor can be solitary or present itself as a manifestation of multiple hereditary exostosis (MHE). Solitary or multiple lesions seldom occur along the vertebral column with a representation of less than 5% of all osteochondromas [2]. This report presents a case of a 9-year-old boy with a solitary, sessile, and cauliflower-like osteochondroma arising from the spinous process and the left lamina of the seventh cervical vertebrae.

Case report

A previously healthy 9-year-old boy with no family history of osteochondroma presented to the Pediatric Orthopedics Outpatient Clinic at the Shengjing Hospital of China Medical University in Shenyang, China, with a hard bony mass in the posterior aspect of the neck. The tumor did not increase in size over the following two-month period. On examination the lump was readily visible and tender to palpation. The mass was hard, regular and fixed to the vertebral column. The movements of the cervical

spine were not restricted. The patient denied clumsiness of the hands or feet, paresthesias, or neck pain. No motor weakness, sensory disturbance, or hyperreflexia were noted. There was no neurological deficit in the extremities or in the cervical spine. The other comprehensive skeletal surveys of this patient were normal.

The anteroposterior and lateral plain radiographs demonstrated a calcified mass arising dorsally from the sixth and seventh cervical vertebrae (**Figure 1A, 1B**). Computerized tomography (CT) scans confirmed that a solitary, sessile, and cauliflower-like mass was arising from the spinous process and left lamina of C-7 (**Figure 2**). The parents of the patient had a concern regarding a sarcomatous degeneration of the osteochondroma and the persistence of the tenderness of the mass. They strongly requested that the tumor be removed. The patient underwent surgery with a complete resection of the tumor via a posterior midline cervical incision. At surgery, the tumor appeared lobulated, well circumscribed, firm and calcified (**Figure 3**). The tumor, the spinous process and a portion of the left lamina of C-7 were removed including the periosteum and cartilaginous cap. The excised bony tissue had a cauliflower shape



Figure 1. The preoperative anteroposterior and lateral plain radiographs demonstrate a calcified mass arising dorsally from the cervical vertebrae (arrow).

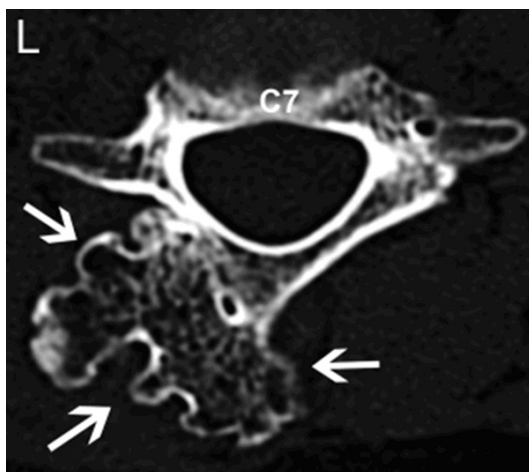


Figure 2. The preoperative CT scans verified that a solitary, sessile, and cauliflower-like mass was arising from the spinous process and the left lamina of C-7 (arrows).

with a cartilaginous cap and measured 3.0 × 3.0 cm.

The histological evaluation revealed a zone of enchondral ossification and trabecular bone structure in the center of the mass surrounded by fibrous tissue. There were no signs of malignancy. The final histopathological examination of the resected specimen confirmed the diagnosis of osteochondroma.

No immobilization or physiotherapy of the neck followed postoperatively. The five-year and five-month follow-up showed a healthy cervical spine with a totally normal range of cervical movement. The postoperative anteroposterior and lateral plain radiographs (Figure 4A, 4B) and CT scans (Figure 5) demonstrated a gross-total resection with a stable sagittal alignment and no evidence of recurrence. The patient was very happy and satisfied with the outcome.

Discussion

Osteochondroma accounts for 34.3% of benign cartilage tumors and for 8.5% of all bone tumors [3]. They occur less frequently in the axial skeleton and only 1.3%-4.1% of solitary osteochondromas originate in the spine [2]. Osteochondroma accounts for 12.9% of primary bone tumors in the pediatric spine [4]. Schomacher et al. [5] reviewed the English literature from 1970 up to 2008 with regard to osteochondroma of the cervical spine and found that the mean age at clinical presentation was 31.6 years with a range from 8 to 66 years. The ratio between males and females was 30 to 21. Approximately 50% of solitary spinal osteochondromas originate in the cervical spine and most frequently from C2 [2]. Solitary spinal osteochondromas also originate in the thoracic and lumbar spine but rarely in the sacrum [3]. The multiple adjacent osteochondromas often fuse and restrict mobility. Three tumor morphologies have been described: sessile, polypoid and cauliflower-like [3].

The location and origin of solitary osteochondromas of the cervical spine may vary. Osteochondromas arise most frequently from the posterior elements of the spine; however, any portion of the vertebral body can be affected [6]. They can arise from the articular facet, the spinous process or the arch of all cervical vertebral bodies [7]. The tips of the spinous pro-

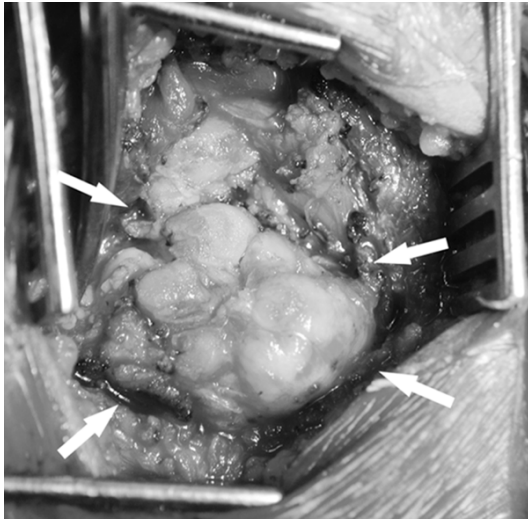


Figure 3. The appearance of the mass under surgical exposure (arrows).

cesses are the preferred sites involving more than two levels [3]. The development in the posterior vertebral elements can be due to the abundance of secondary ossification centers within the neural arch [8]. Predilection for the cervical spine, specifically the atlantoaxial joint, may be related to the varying mobility within the divisions of the spine. The increased flexibility of the cervical spine predisposes the spine to greater stress on these vertebrae, thereby increasing the risk of micro trauma to the epiphyses and promoting exostotic growths [9, 10].

The majority of spinal osteochondromas is asymptomatic [11]. The most common physical finding is a non-tender palpable mass in the posterior aspect of the cervical spine. The anterior cervical osteochondromas are rare and may present as a pharyngeal mass leading to hoarseness, dysphagia or vascular compression [8]. In early stages, the cervical mass may be difficult to discern because of obstruction caused by the surrounding bony column. Neurological compromise from osteochondroma is rare (0.5%-1%) because most lesions lie outside the lumen of the spinal canal [9]. As in the presented case, most spinal osteochondromas do not cause neurological symptoms; however myelopathy and radiculopathy from neural compression have been reported [3]. Spinal cord impingement should be suspected when there is lower extremity discomfort associated

with decreased balance, impaired coordination or other central neurologic dysfunctions.

Plain radiographs may reveal an osseous projection arising from the affected bone. But sometimes, spinal osteochondromas are difficult to detect on radiographs due to the complexity of the vertebral column [10]. CT and magnetic resonance imaging (MRI) are the appropriate methods for detecting and evaluating osteochondromas [3]. The use of CT simplifies preoperative planning because the margin of the osseous and cartilaginous components of the tumor can be well defined. The presence and extent of the lesion are best delineated with CT. MRI demonstrates compression of the spinal cord or nerve root and the relationship of the mass to the surrounding tissue [8].

Surgery is required when neural compression or chronic pain is suspected, or when there are concerns regarding sarcomatous transformation. Surgical technique requires an extra capsular or marginal type of resection that includes the entire cartilaginous cap and overlying periosteum and perichondrium [12]. In the present case an en-bloc excision of the osteochondroma was performed involving the spinous process and the left lamina of the seventh cervical vertebrae including the periosteum and cartilaginous cap.

Excision of the entire osteochondroma and its cartilaginous cap is usually curative. The risk of recurrence after a complete resection is less than 2% [9]. The recurrence risk is proportional to the thickness of the cartilage. Incomplete resection of these lesions predisposes the reemergence of the osteochondroma due to the continued growth of the cartilaginous cap. Curettage will serve to remove the bulk of the lesion but may be a poor surgical technique for obtaining definite tumor-free margins. Because large osteochondromas may include foci of grade I chondrosarcoma they should be resected en-bloc [13].

Sarcomatous degeneration occurs in 1% and 5%-15% of the solitary and multiple forms, respectively and seems to be related to the EXT family of tumor suppressor genes [14, 15]. Axial skeleton osteochondromas may have a somewhat higher rate of malignant degeneration.

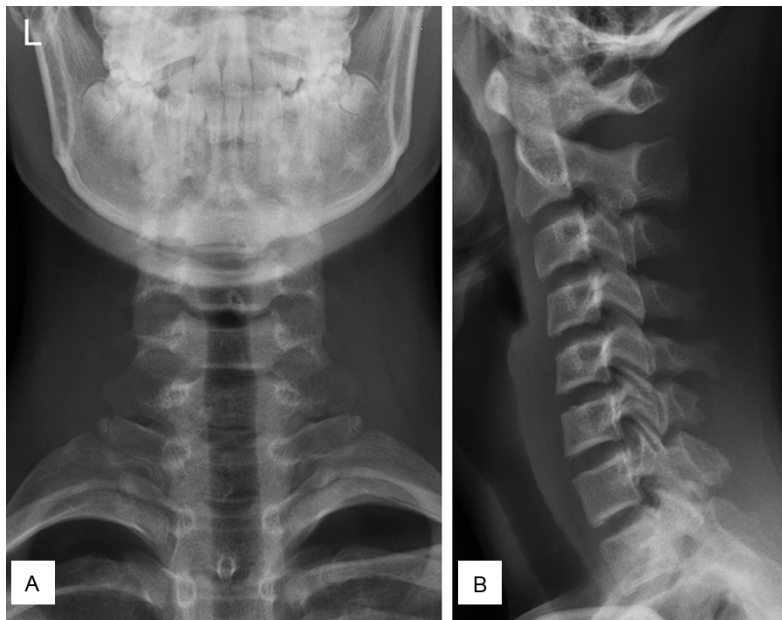


Figure 4. At the five-year and five-month follow-up the postoperative antero-posterior (A) and lateral plain (B) radiographs demonstrate a stable sagittal alignment of cervical spine with no evidence of recurrence.

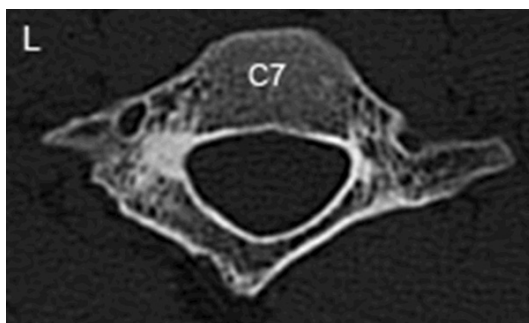


Figure 5. The postoperative CT scans demonstrate no evidence of recurrence at the five-year and five-month follow-up.

tion than the osteochondromas of the appendicular skeleton.

Disclosure of conflict of interest

None.

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References

- [1] Brien EW, Mirra JM, Kerr R. Benign and malignant cartilage tumors of bone and joint: their anatomic and theoretical basis with an emphasis on radiology, pathology and clinical biology. I. The intramedullary cartilage tumors. *Skeletal Radiol* 1997; 26: 325-353.
- [2] Prasad A, Renjen PN, Prasad ML, Bhatta GB, Madan VS, Buxi TB, Agarwal SP. Solitary spinal osteochondroma causing neural syndromes. *Paraplegia* 1992; 30: 678-680.
- [3] Albrecht S, Crutchfield JS, SeGall GK. On spinal osteochondromas. *J Neurosurg* 1992; 77: 247-252.
- [4] Weinstein JN, McLain RF. Primary tumors of the spine. *Spine (Phila Pa 1976)* 1987; 12: 843-851.
- [5] Schomacher M, Suess O, Kombos T. Osteochondromas of the cervical spine in atypical location. *Acta Neurochir (Wien)* 2009; 151: 629-633; discussion 633.
- [6] Quirini GE, Meyer JR, Herman M, Russell EJ. Osteochondroma of the thoracic spine: an unusual cause of spinal cord compression. *AJNR Am J Neuroradiol* 1996; 17: 961-964.
- [7] Sakai D, Mochida J, Toh E, Nomura T. Spinal osteochondromas in middle-aged to elderly patients. *Spine (Phila Pa 1976)* 2002; 27: E503-506.
- [8] Morard M, de Preux J. Solitary osteochondroma presenting as a neck mass with spinal cord compression syndrome. *Surg Neurol* 1992; 37: 402-405.
- [9] Khosla A, Martin DS, Awwad EE. The solitary intraspinal vertebral osteochondroma. An unusual cause of compressive myelopathy: features and literature review. *Spine (Phila Pa 1976)* 1999; 24: 77-81.
- [10] Maheshwari AV, Jain AK, Dhammi IK. Osteochondroma of C7 vertebra presenting as compressive myelopathy in a patient with nonhereditary (nonfamilial/sporadic) multiple exostoses. *Arch Orthop Trauma Surg* 2006; 126: 654-659.

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- [11] Gille O, Pointillart V, Vital JM. Course of spinal solitary osteochondromas. *Spine (Phila Pa 1976)* 2005; 30: E13-19.
- [12] Lewis MM, Sissons HA, Norman A, Greenspan A. Benign and malignant cartilage tumors. *Instr Course Lect* 1987; 36: 87-114.
- [13] Scot DL, Pedlow FX, Hecht AC, Hornicek FJ, Tumours A. Primary benign and malignant extradural spine tumours. In: Frymoyer JW, Wiesel SW, editors. *The adult and paediatric spine*. 3rd edition. Vol. I. Philadelphia: Lippincott Williams and Wilkins; 2004. pp. 204-205.
- [14] Stickens D, Clines G, Burbee D, Ramos P, Thomas S, Hogue D, Hecht JT, Lovett M, Evans GA. The EXT2 multiple exostoses gene defines a family of putative tumour suppressor genes. *Nat Genet* 1996; 14: 25-32.
- [15] Wise CA, Clines GA, Massa H, Trask BJ, Lovett M. Identification and localization of the gene for EXTL, a third member of the multiple exostoses gene family. *Genome Res* 1997; 7: 10-16.