romanian NEUROSURGERY

Vol. XXXVI | No. 1 March 2022

Osteochondroma of axis vertebra treated with *en bloc* resection and arthrodesis. A case report and review of literature

> Deepak Kumar Singh, Diwakar Shankar, Vipin Chand, Mohammad Kaif

Deepak Kumar Singh, Diwakar Shankar, Vipin Chand, Mohammad Kaif

Dr. Ram Manohar Lohia Institute of Medical Sciences, Lucknow, INDIA

ABSTRACT

Osteochondromas are capped benign bony neoplasm that forms on the outer surface of the bone. They are most commonly found in appendicular skeletal but rare in the spine. The cervical spine is commonly involved in spinal osteochondromas. Here we discuss a case of a 24-year-old male with osteochondroma arising from the posterior arch of C2 (axis) vertebra presenting with myelopathy. Most of the cases are asymptomatic needing only observation. Surgery is needed in case of progressive neurologic deficit. It is recommended to do an en bloc resection of the lesion along with cartilage cap to prevent recurrence and spinal stabilization with arthrodesis to avoid postoperative kyphosis.

INTRODUCTION

Osteochondromas are the most common benign primary tumor of bone constituting 20-50% of all benign bone tumors and 10-15% of all bone tumors [5]. According to World Health Organization (WHO), osteochondromas are capped benign bony neoplasm that forms on the outer surface of the bone. Tumor originates from the ends of the bone near the growth plate where metaplastic cartilage undergoes endochondral ossification with subsequent proliferation. It is most commonly found in the appendicular skeleton but rare in the spine [6]. More than 50% of spinal osteochondromas are found in the cervical region [4]. Here we discuss a case of a 24-year-old male with osteochondroma arising from the posterior arch of axis vertebra presenting with myelopathy.

CASE REPORT

A 24-year-old male patient presented with complaints of progressively increasing difficulty in walking with weakness in the right upper and lower limbs for the last year. There was no history of trauma. There was no family history of MHE. His neurological examination revealed weakness (Medical Research Council, MRC muscle power grading - 4/5) of the right upper and lower limbs. Deep tendon reflexes across major

osteochondroma, C2 osteochondroma, axis osteochondroma, spinal tumour, posterior arch osteochondroma

Keywords

 \succ

Corresponding author: Diwakar Shankar

Dr. Ram Manohar Lohia Institute of Medical Sciences, Lucknow, India

dev123diwakar@gmail.com

Copyright and usage. This is an Open Access article, distributed under the terms of the Creative Commons Attribution Non-Commercial No Derivatives License (https://creativecommons .org/licenses/by-nc-nd/4,0/) which permits noncommercial re-use, distribution, and reproduction in any medium, provided the original work is unaltered and is properly cited. The written permission of the Romanian Society of Neurosurgery must be obtained for commercial re-use or in order to create a derivative work.

> ISSN online 2344-4959 © Romanian Society of Neurosurgery



First published March 2022 by London Academic Publishing www.lapub.co.uk





joints were brisk. Bilateral plantars reflex were extensor. Bilateral Hoffmann signs were positive. No involvement of bladder and bowel was seen. Sensory modalities were intact. The breath-holding time was 42s. The modified Japanese orthopedic association (mJOAS) score was 16/18.

Plain radiographs and computer tomography (CT) cervical spine revealed lobulated circumscribed bony growth along inner margin of right lamina of the axis (C2) vertebra with extension anteriorly into the spinal canal and producing marked canal stenosis (Figure 1). MRI cervical spine showed a large extradural mass lesion arising from axis vertebral lamina causing major compression at C2 vertebral level with myelopathic changes in the cord which was corelated to the clinical complaints and examination of the patient (Figure 2).

The patient was taken for surgery. Preoperative positioning was as per standard guidelines. Fluoroscopic guidance was used for surgery. Intraoperative en bloc resection of the lesion along with bilateral C2 vertebral lamina was done (Figure 3). Bilateral C1 and C3 vertebra lateral masses were fixed with titanium screws and rods system. Onlay autologous bone grafts were placed between C1 and C3 lamina. The postoperative CT cervical spine showed complete removal of the lesion with an increased effective canal diameter at the level of the C2 vertebral body (Figure 4). Pathologic examination was consistent with osteochondroma. The patient had significant improvement in his symptoms and was discharged with a hard cervical collar for three months. In six months, follow-up patient was improved.



Figure 1. Preoperative computer tomography (CT) images with the 3D reconstruction of the cervical spine showing

pedunculated circumscribed bony growth along the inner margin of the right lamina of axis (C2) vertebra with extension anteriorly into the spinal canal and producing marked canal stenosis.

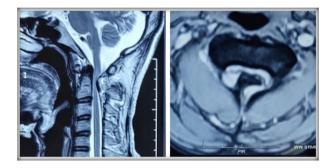


Figure 2. Preoperative magnetic resonance imaging (MRI) of the cervical spine showed a large extradural mass lesion arising from the right C2 vertebral lamina causing major compression at C2 vertebral level with myelopathic changes in the cord.

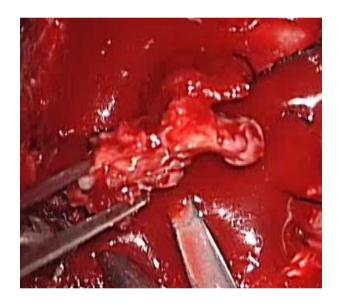


Figure 3. Intraoperative image showing en bloc resection specimen of the osteochondroma along with C2 (Axis) vertebral lamina.



Figure 4. The post-operative computer tomography (CT) of the cervical spine showed complete removal of the lesion with an

increased effective canal diameter at the level of the C2 vertebral body.

DISCUSSION

Osteochondromas are the most common benign primary tumor of bone constituting 20-50% of all benign bone tumors and 10-15% of all bone tumors [5]. It is twice more common in females. The usual age of presentation is 20-30 years. According to World Health Organization (WHO), osteochondromas are capped benign bonv neoplasm that forms on the outer surface of the bone. It arises from epiphyseal growth plate where microtrauma can lead to fibroblastic proliferation and the formation of new bone. Osteochondromas can present in two forms, Solitary Exostosis (SE) and Multiple Hereditary Exostosis (MHE). Solitary Exostosis is the sporadic form of the disease and constitutes 85% whereas MHE occurs as Autosomal Dominant syndrome which constitutes 15% of all osteochondromas. Osteochondromas can be sessile or pedunculated. Our patient had a solitary pedunculated lesion over the right lamina of the axis vertebra.

It is most commonly found in the appendicular skeleton including femur, proximal tibia and humerus, and pelvis but rare in the spine[1]. Spinal osteochondromas constitute 1-9% of all cases[4]. More than 50% of spinal osteochondromas are found in the cervical region[4]. This is due to increased mobility leading to the displacement of cartilage and resulting exostosis. Osteochondromas most commonly originate from the posterior element where ossification centers are present. The Axis vertebra is the most common site of osteochondroma in the spine as was in our case followed by the C3 and C6 vertebra [2].

Most of the cervical osteochondromas are asymptomatic as they commonly grow outwards away from the spinal cord. Inward growth causes myelopathy and radiculopathy which is seen in 0.5-1% cases[1]. Other symptoms include pain, weakness, numbness, neck mass, etc. As osteochondroma grows slowly, the patient may develop progressive symptoms. Sometimes it may present early after a trauma leading to compression of the spinal cord.

Plain skiagram and CT scan can be used for establishing the diagnosis. CT is the imaging modality of choice[9]. On CT scan there is a direct continuity of lesion with the cortical and medullary cavity of the underlying bone. Magnetic resonance imaging (MRI) is useful in assessing the degree of spinal cord compression as well as malignant transformation.

The most common treatment modality for asymptomatic cases is observation. Symptomatic patients with pain or progressive neurological deficit require surgery. Also, surgical excision may be done to establish the diagnosis[3]. Previously surgical decompression without fusion was popular but it associated with postoperative kyphotic was deformity, cervical instability, late neurological deficit. Hence, a shift towards en bloc resection with arthrodesis has been seen in recent times. Complete surgical excision of the cartilaginous cap is critical to prevent recurrence[4]. Sclubba et al in their study found that the recurrence rate increased from 11% after en bloc resection to 33% after intralesional resection. There is no role of neoadjuvant or adjuvant radiotherapy or chemotherapy.

Recurrence is seen in 2-11% of postoperative cases[4,8]. Malignant transformation, mostly into chondrosarcoma, is seen in 1% of SE and 3-15% of MHE cases. Features of malignant transformation are rapid growth, growth after skeletal maturity, cartilage cap thickness >3mm, recurrence after complete resection[7]. In our case, complete resection with arthrodesis was done which is found to be more effective than resection alone.

CONCLUSION

Osteochondromas are the most common benign primary tumor of bone. Most of them are found in the appendicular skeletal but rare in the spine. C2 vertebra is the most commonly affected vertebra. Most of the cases are asymptomatic needing only observation. Surgery is needed in case of progressive neurologic deficit. It is recommended to do an en bloc resection of the lesion along with cartilage cap to prevent recurrence and spinal stabilization with arthrodesis to avoid postoperative kyphosis.

REFERENCES

- 1. Brastianos P, Pradilla G, McCarthy E, Gokaslan Z. Solitary thoracic osteochondroma case report and review of the literature. Neurosurgery 2005; 56: E1379.
- Chatzidakis E, Lypiridis S, Kazdaglis G, Chatzikonstadinou K, Papatheodorou G. A rare case of solitary osteochondroma of the dens of the C2 vertebra. Acta Neurochir (Wien) 2007;149(6):637–8.

- Gul rkanlar D, Aciduman A, Gul naydın A, Kocak H, Clelik N. Solitary intraspinal lumbar vertebral osteochondroma: a case report. J Clin Neurosci 2004; 11:911–913.
- Lotfinia I, Vahedi A, Stefanaki K, Tubbs RS, Vahedi P. Cervical osteochondroma with neurological symptoms: literature review and a case report. Spinal Cord Ser Cases. 2017;3:16038.
- Murphey M, Choi J, Mark J, Kransdorf M, Flemming D, Gannon F. Imaging of osteochondroma: variants and complications with radiologic pathologic correlation. RadioGraphics 2000; 20: 1407–1434.
- 6. Quirini G, Meyer J, Herman M, Russell E. Osteochondroma of the thoracic spine: an unusual cause of spinal cord compression. AJNR 1996; 17: 961–964.
- Srikantha U, Bhagavatula ID, Satyanarayana S, Somanna S, Chandramouli BA. Spinal osteochondroma: spectrum of a rare disease. J Neurosurg Spine. 2008;8:561–6.
- Veeravagu A, Li A, Shuer LM, Desai AM. Cervical Osteochondroma Causing Myelopathy in Adults: Management Considerations and Literature Review. World Neurosurg. 2017;97,752.e755-752.e753.
- Victory F, Ali K. Solitary, asymptomatic, posterior, vertebral, intracanal, cervical spine osteochondroma. Radiology Case Reports. 2011;6:572.