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Case Report

Lower limb weakness: an unusual cause

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ABSTRACT

Osteochondromas are usually tumours of the long bones and occurs rarely in other bones. Osteochondroma of rib causing spinal cord compression is a rare presentation with only 8 cases reported in literature involving similar presentation in pediatric age group. Our case is a boy who presented with paraparesis of acute onset. All the usual suspects were ruled out. The cause was found to be a solitary lesion arising from the head of the 3rd rib causing cord compression. Laminectomy and decompression with excision of the lesion was done. On HPE it showed features of osteochondroma. Post operatively the child recovered well, the weakness and sensations improved.

Keywords: Costal osteochondroma, Laminectomy, Lower limb weakness, Paraparesis, Rare presentation, Spinal cord compression

INTRODUCTION

Osteochondromas are usually tumours of the long bones.[1] Costal osteochondromas are rare. A costal osteochondroma causing spinal cord compression is very uncommon.[2-4] These tumors can arise from the costovertebral or costotransverse joints and may pass through neural foramen and occupy the spinal canal, causing severe stenosis.[5] Most osteochondromas are solitary, but multiple lesions can occur, usually in association with hereditary multiple exostoses (HME).[2] When present in the spine, they have a special predilection for the cervical or thoracic spine.[2]

Osteochondroma of the spine usually arises from the neural arch of the cervical and thoracic vertebra and can cause compression of the spinal cord.

We describe such a case with paraparesis due to solitary osteochondroma of head of the 3rd rib causing spinal cord compression. Many osteochondromas are asymptomatic; however, complications can involve bone, nerve, and soft tissues via mass effect or intrinsic change.[5] Both Males and Females can be equally affected, but there is a slight male predominance.[6] The incidence of HME is approximately 1 in 50 000 live births.[1]

CASE REPORT

History and presentation

A 13-year-old boy with no family history of osteochondromas presented to us with a 2-week history of lower limb weakness, numbness of both feet, frequent falls, back pain in the lower thoracic dermatome radiating to upper abdomen.

The parents attributed all this to a trivial fall while playing in school, 2 days after which the symptoms started appearing. There was no loss of consciousness before or after the fall. h/o numbness involving both feet, no dysesthesia. No h/o breathing difficulty. No h/s/o upper limb weakness. No h/s/o raised ICT in the form of

headache or vomiting or visual disturbance. No h/s/o cranial nerve involvement. No h/s/o involuntary movements or abnormal eye movements. No h/o any fever, loose stools, rashes, or vaccination/IM injections.

No h/o any bowel or bladder disturbance. No h/o weight loss or chronic cough. No h/o contact with any active case of tuberculosis. No significant past history. Antenatal, Natal, Postnatal, Development and Family history were insignificant. General examination of the child was normal except for the BMI which was in the range of overweight (Figure 1a).

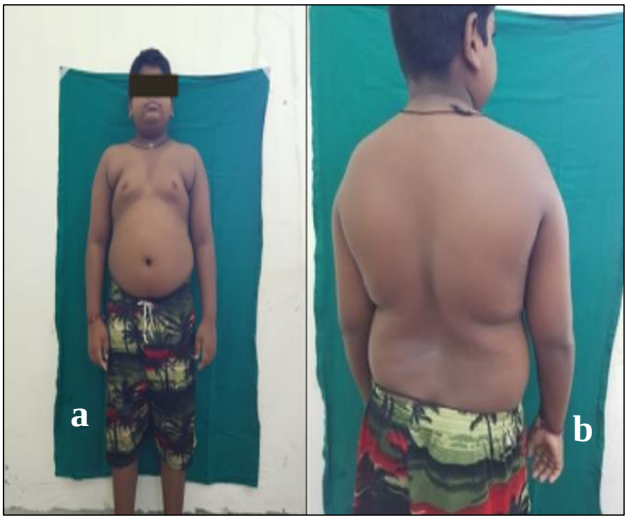

Figure 1: a) Overweight image of the child, b) Clinical picture of the child.

Romberg was positive, but there were no signs of cerebellar dysfunction. Gait was waddling. No signs of Meningeal irritation. Skull was normal. scoliosis of spine with curvature to right was seen. (Figure 1b). other systems were normal.

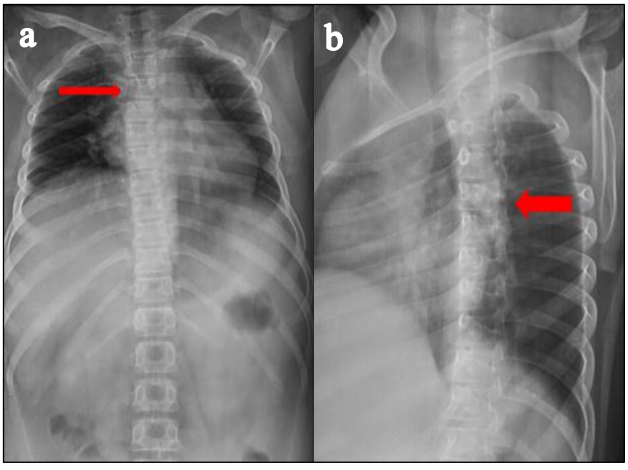

Figure 2: 2A) Right pedicle of D3 vertebra absent on the frontal radiography, 2B) Similar finding seen in lateral X-ray.

All blood investigations were normal, Mantoux negative, B12 levels normal Chest x-ray PA view showed the Right pedicle of the D3 vertebra to be absent (Figure 2a), similar finding was seen in lateral X-ray (Figure 2b).

Table 1: Motor system examination.

	Upper limb right	Left	Lower limb right	Left
Bulk	24/21 cm	24/21 cm	44/27 cm	44/27 cm
Tone	Normal	Normal	Normal	Normal
Power	5/5	5/5	3/5	3/5
Reflex				
Abdominal reflex	Absent			
Cremastric reflex	Absent			
Biceps	++	++		
Triceps	++	++		
Supinator	++	++		
Knee			+++	+++
Ankle			++	++
Plantar			Extensor	Withdrawal

Table 2: Sensory system examination.

Sensory system	Upper limb	Lower limb
Superficial sensations		
Touch	Present	Absent below T5T6 dermatome
Pain	Present	
Temperature	Present	
Deep pain	Present	Present
Tactile localisation	+	Inconsistent
Two-point discrimination	+	Inconsistent
Joint position sense	Normal	Absent in MTP and ankle Joint

CT scan showed a bony lesion arising from the head of the 3rd rib and compressing on the spinal cord (Figure 3A and 3B). MRI was done to assess the size of the lesion (Figure 4A and 4B) and tissue damage due to the pressure effects. Operation

Under GA, D2-3 laminectomy, excision of the lesion and decompression was done.

Tumour excised was sent for biopsy. 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 surgical intervention and risk of spinal destabilization were not considered.

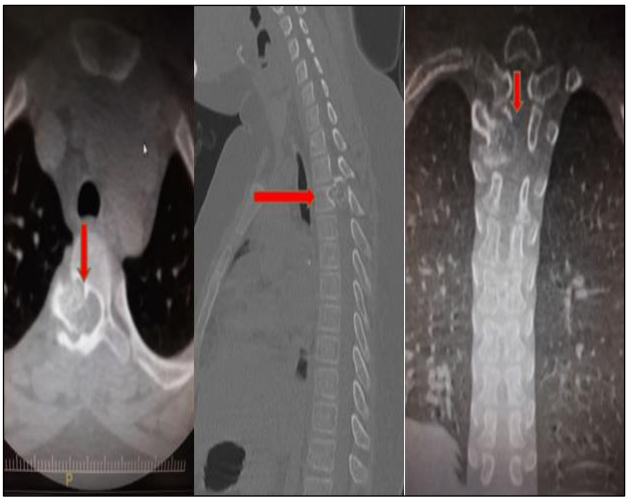

Figure 3: A) Well corticated bony lesion seen arising from medial aspect of the head of the right 3rd rib and extending medially into the spinal Canal, 3B) Cortical and trabecular continuity maintained between the lesion and the rib, 3C) The bony lesion is protruding into the adjacent neural foramina and the spinal canal. remodelling of posterior aspect of D3 vertebral body, pedicle, lamina noted on the right side.

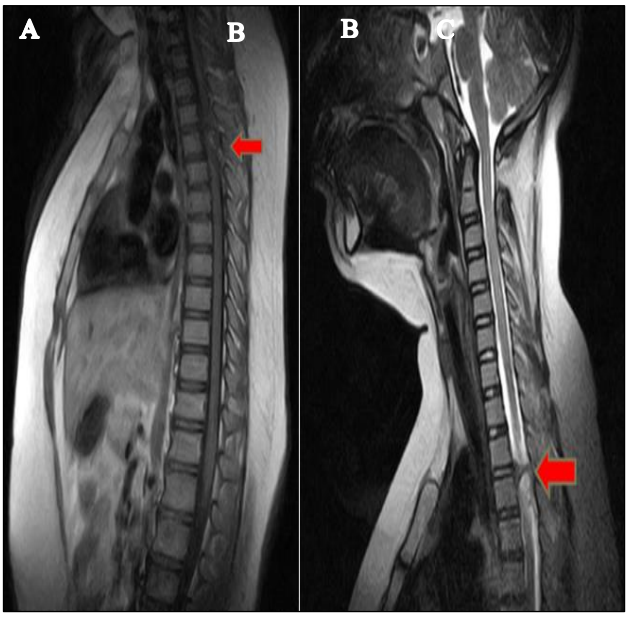

Figure 4A and 4B: Lesion measures-2 cm (cranio-caudal) X 1.4 cm (antero-posterior) X 1.6 cm (medio-lateral).

Postoperative course

The patient had an uneventful hospital course and was discharged on postoperative day 8. He developed no new neurological deficits and reported improvement in his pre-operative weakness in his 1-month follow-up examination. His sensations in the lower limbs also improved. Currently the patient is on regular followup.

DISCUSSION

Osteochondromas are the most common benign tumours of bone and may present as either a solitary lesion or as multiple lesions.[1,2] Solitary lesions are most common, but presentations of multiple lesions, usually with autosomal dominant inheritance, are termed hereditary multiple exostoses (HMEs).[1] These tumours commonly occur in the long bones, but seldom affect ribs.[3,4] Only 1.5% of all osteochondromas are costal osteochondromas, and spinal cord compression due to a tumour arising from the head of a rib is even more rare.[3,4]

The reported incidence of costal osteochondroma is very low.[3,4] However, the actual incidence is likely to be underestimated because costal osteochondromas mostly arise at or near the costochondral junction and are usually asymptomatic.[3,4,5] The type of osteochondroma described in this case, arising at the costovertebral junction with neural foraminal extension and spinal cord compression, is extremely rare. Only 15 such cases have been previously reported in the literature (Table 1).[3-18]

Of those only 8 were in the pediatric age group and only 2 have been reported in India.[7] were known to have HMEs, another 8 patients had solitary lesions. All the patients showed clinical improvement during the immediate follow-up period after complete tumour excision.

Presenting complaints

While previously reported symptoms are hemothorax, pneumothorax, hiccups, recurrent chest infections, and thoracic outlet syndrome.[9,10] 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 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.

Imaging modality

Although the location of the bony portion of the osteochondroma can be reliably determined using multiplanar CT reconstruction, the exact size of the tumour may be underestimated because the cartilage cap of the tumour is not detectable by CT.[3,4] MR imaging is the best radiologic modality for evaluating the hyaline cartilage cap. The non-mineralized portions of the cartilage cap have high water content, resulting in

intermediate-to-low signal intensity on T1-weighted images and high signal intensity on T2-weighted MR images. These features allow for accurate measurement of the thickness of the cartilage cap and distinction from overlying muscle on MR images.[19] In this case, the unique curvilinear high-signalintensity region covering the tumour, seen on axial T2-weighted images, represents the cartilage cap, which led to accurate preoperative diagnosis.

Malignant transformation, usually into a chondrosarcoma, occurs in approximately 1% of solitary osteochondromas and 10% of HMEs.[9,20]

Table 3: Previously reported cases.

Age , Gender	Rib of origin	Presenting symptom	Surgery done	Type of osteochondroma	Author
*17, Male	2,3	Brown-Sequard syndrome, Horner syndrome	Laminectomy	HME	Becker and Epstein
23, Male	5	Spastic paraparesis, Brown Sequard syndrome	Laminectomy	Solitary	Chanzo et al
*15, Male	10,11	Spastic paraparesis	Laminectomy	HME	Decker and Wei
19, Male	4	Spastic paraparesis	Laminectomy	Solitary	Faik et al
*17, Female	10	Myelopathy, pain, Spastic paraparesis	Costotransversectomy	Solitary	Kane et al
33, Male	3	Spastic paraparesis	Laminectomy	HME	Larson et al
*10, Male	8	Incontinence, Spastic paraparesis	Laminectomy	Solitary	Mannoji et al
21, Male	5	Incontinence, Spastic paraparesis	Thoracotomy	Solitary	Natarajan et al
21, Female	3	Pain	Thoracotomy	HME	Old and triplett
65, Male	6	Pain	Laminectomy	Solitary	Sener et al
*12, Male	5	Myelopathy, incontinence	Laminectomy	Solitary	Twerky et al
*11, Female	4	Spastic paraparesis, incontinence	Laminectomy	HME	Twerky et al
*12, Female	6	Spastic paraparesis	Costotransversectomy	Solitary	Rao et al
*16, Female	12	Pain	Costotransversectomy	HME	Tang et al
21, Female	3	Pain	Costotransversectomy	HME	Marcus et al
*13, Male	3	Pain, paraparesis	Laminectomy	Solitary	Present case

HME: Hereditary multiple Exostosis; *Pediatric age group

A sudden increase in lesion size or the development of new-onset pain suggests malignant transformation.[20,21] Bess et al. emphasised that preoperative radiographic evaluation should consist of MR and CT imaging in order to provide optimal information about the lesion, which aids in surgical planning.[3,22] Radiologic findings may show consistent growth of exostoses after closure of the growth plate, alterations in surface delineation in comparison with previous radiographic studies, internal lytic areas, erosion or destruction of adjacent bones, and the presence of soft tissue masses containing scattered or irregular calcifications.[22,23]

The size of the cartilaginous cap is the best indicator of malignancy.[24] MR imaging results showing a cartilage cap thickness exceeding 2 cm in adults and 3 cm in children should raise the suspicion of malignancy.[25] The use of gadolinium diethylenetriamine-pentacetate (Gd-DTPA)-enhanced MR imaging is an effective procedure for obtaining a differential diagnosis between malignant and benign lesions.[26] Generally, osteochondromas do not show contrast enhancement, but mild enhancement may be observed within the marrow.[20] In this case, Gd-DTPA-enhanced MR imaging was used, and the images did not show contrast enhancement. Because asymptomatic solitary osteochondromas have a low rate of malignant transformation, they can be followed up conservatively.[27,28]

Surgery

As osteochondromas may 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.[3]

If additional facetectomy is performed to remove the foramen and/or extraforaminal component, iatrogenic instability and kyphosis may occur during the follow-up period. In this case, only laminectomy was done and facectomy was not needed.

CONCLUSION

The type of osteochondroma described in the current case, which arose at the costovertebral junction with neural foraminal extension and spinal cord compression, is extremely rare. Only 8 such cases have been previously reported in the pediatric age group. Extra dural compression due to bone tumours should also be considered when evaluating a case with similar presentation. Surgical excision with conservative follow up is usually sufficient in case of solitary osteochondromas.

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