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Osteochondroma of Lower Dorsal Spine in Hereditary Multiple Exostosis Causing Spinal Cord Compression: Report of a Case and Review of the Literature

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Abstract
Introduction; *Osteochondroma or exostosis are the most common benign tumors of the bone that usually occur in the long bones and rarely found in the spine. When present in the spine, however, they have a predilection for the cervical region. They occur in two forms as solitary and multiple hereditary forms. We have reported here a case of spinal cord compression due to osteochondroma arising from the T-12 vertebral body and left pedicle in a 13-year-old female who presented to us with spastic paraparesis.*
Keywords: *Multiple Hereditary Exostosis; Osteochondroma; Spinal cord compression.*

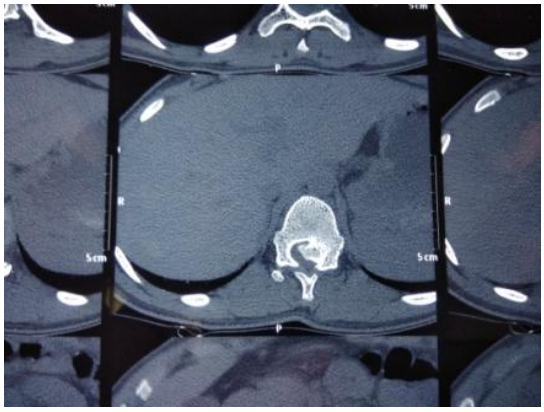
Introduction

Spinal osteochondromas are of two types: (1) Spinal osteochondromas in patients with multiple osteochondromatosis, and (2) solitary spinal osteochondromas.[1] Solitary tumors usually arise at the cervical spine, markedly the atlantoaxial joint, while in patients with hereditary multiple exostosis they usually appear in the lumbar or thoracic spine.[2] Most are asymptomatic and seen incidentally during radiographic examination. We describe an unusual case of Osteochondroma of lower dorsal spine causing spinal cord compression and spastic paraparesis in thirteen year old girl.

Presentation of case

An 13-year-old female patient presented with complaints of multiple bony hard swellings in thorax, thighs, lower abdomen and left shoulder since the past 2 years with back pain since 7 months and weakness of both lower limbs with difficulty in walking since the past 4 months. Weakness in both the lower limbs was insidious in onset and gradually progressive over a period of 4 months. There was no bladder and bowel involvement. There was no history of trauma or onset due to lifting of heavy weight. There was no history of fever, night sweats and weight loss. On physical examination multiple, non-tender, fixed, bony hard swellings of varying sizes were present in the lower extremities, left clavicle, multiple ribs and bilateral pelvis. The local examination of spine was normal. The neurological examination revealed spastic paraparesis with reduced

sensation below L2 dermatome on both sides. There was weakness, decreased pinprick sensation, and hyperreflexia of his lower extremity. Positive Babinski response was also elicited. Bladder and bowel functions were intact. Radiograph of the lower dorsal spine was normal. Magnetic resonance imaging of the whole spine showed 2cm x 1cm bony mass arising from the posterolateral aspect of D11 vertebral body and left with cortex and medulla in continuity with vertebral body compressing lower spinal cord. There was marked cord compression seen at this level without change in signal intensity [Fig-1]. Excision of the lesion in the D12 vertebra was undertaken by posterior approach. Tumour mass was removed including lamina and pedicle of the 12th thoracic vertebrae on the left side along with cartilaginous cap. The excised tissue was sent for histopathological examination which was suggestive of the diagnosis of osteochondroma.. Postoperatively patient was kept in ward for 1 week in which dressing was done on 2nd and 6th post-operative day. Staple sutures were removed after 2 weeks post-operatively. Wound was healthy. The tone of bilateral lower limbs became normal 2 weeks post-operatively. The weakness and numbness improved over 2 months. The patient was able to ambulate on her own without any support at 2 months after surgery and there was full neurological recovery after 1 year postoperatively.



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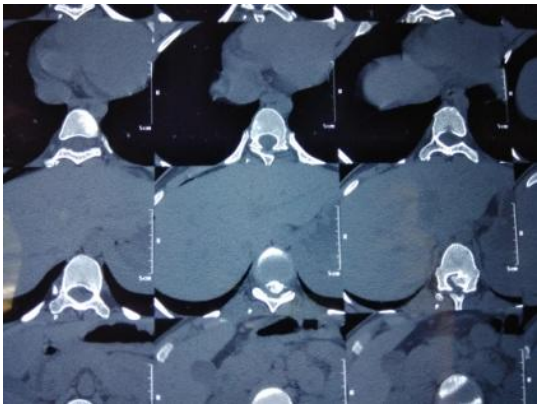

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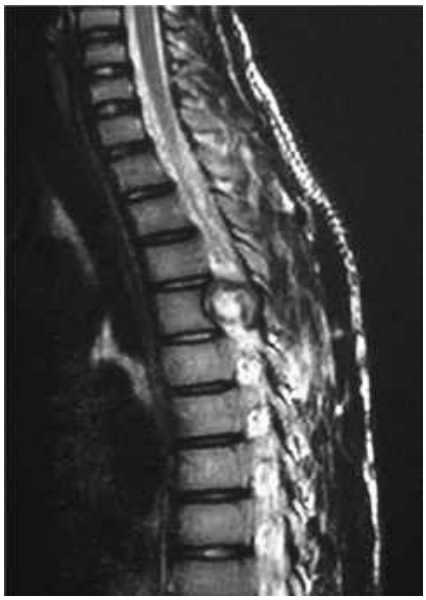

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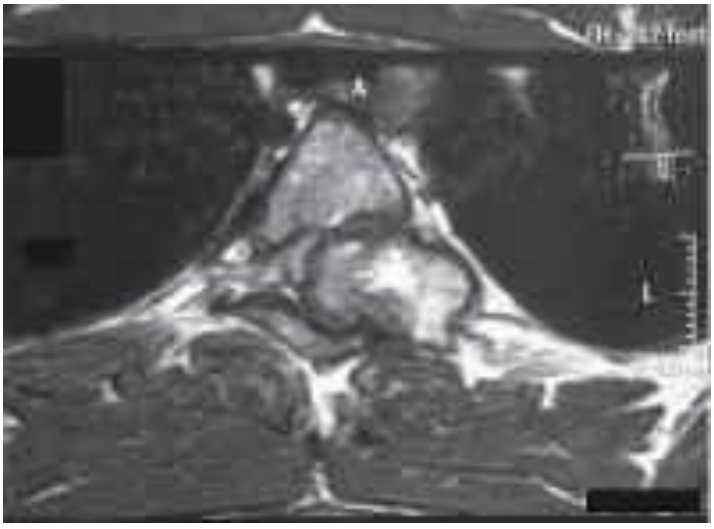

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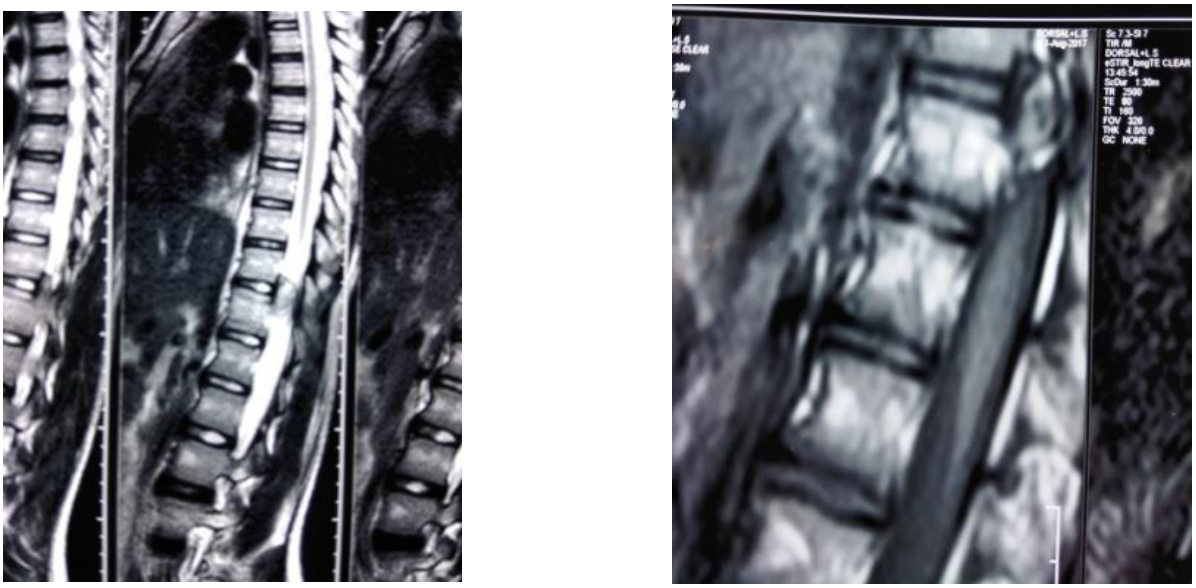

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Fig. 1 MRI image demonstrating the lesion at T-12 vertebra

Discussion

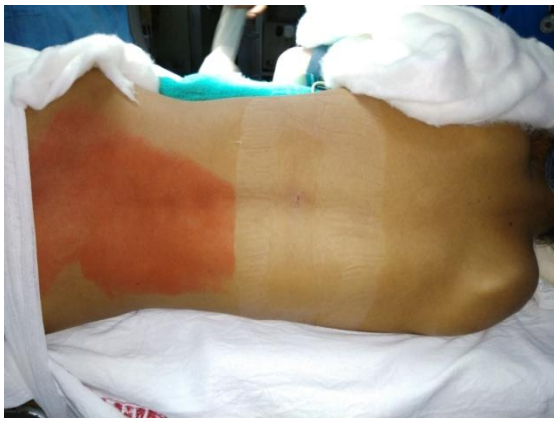
Osteochondromas are the most common benign bone tumours presented as solitary (90%) or multiple lesions (10%).[3] The involvement of the spine is very rare and comprises only 1.3%- 4.1% of all osteochondromas of the spine.[4] With the best of our knowledge there are about 27 cases of thoracic vertebral exostosis in patients with Hereditary Multiple Exostosis (HME) has been described in the literature [Table/Fig-2][2,4-27] The multiple osteochondromas are present in HME. HME is a genetic disorder with autosomal dominance pattern and are associated with mutations in tumour suppressor genes EXT1 or EXT2 or EXT3 located on chromosome 8q, 11p and 19p respectively.[28] OC are considered as developmental lesions rather than true neoplasms. Although aetiologically not clear, OCs are originated from the separation of epiphyseal growth plate cells followed by herniation through the periosteum adjacent the growth plate.[26] The vertebral OC are more common in younger male patients as seen in our case.[22] About 1% and 4% of solitary osteochondromas and 7% to 9 % in hereditary multiple exostoses develop a spinal lesion.[27] The spinal involvement and neurological Complications in multiple osteochondromas is higher than solitary variety.[28] In HME, thoracic and lumbar vertebrae are more commonly affected, while in solitary type cervical spine is commonly involved.[17] The involvement of sacrum is rare in both the types. A review of literature revealed about 27 cases of thoracic myelopathy due to spinal exostosis in HME [Table/Fig-4]. Mean age of the patients was 22.5 years. Nineteen patients were male while seven patients were female. D5 vertebrae (19%) were found to be most commonly affected. Though any part of vertebrae can be involved, the posterior arch is the most commonly affected.[24] In present case pedicle and lamina both were involved. Patients may present with back pain, cosmetic deformity and or a palpable mass. Very rarely vertebral OC may extend into the spinal canal causing cord compression and present with neurological compromise as occurred in our patient. Myelopathy is predominantly seen with multiple OCs.[17] The vertebral OCs are often small, sessile and easily missed on radiography. Computed tomography (CT) is useful to demonstrate spinal OCs which are small and have narrow stalk. In addition it is the best method to detect marrow, cortical continuity of vertebral OC though it was not done in our case. MRI of the whole spine should be performed in these cases to look for skip lesions or other masses and relation of

vertebral OC to the surrounding structures. Asymptomatic vertebral OC can be left as such and patients should be followed up. Review of literature showed surgery was done in majority of cases and resulted in good results in most patients.[2-5,9-19,21-25] Surgical excision and decompression of spinal canal vertebral OC is required in these cases. Similarly the decompression and excision of the mass was done in our case.

Table/Fig-2]: Previously reported cases of thoracic vertebral osteochondromas

Author	Year	Age	Sex	Family history	Level	Origin	Presenting complaint	Surgery	Outcome	Follow up	Remark
Cannon[4]	1954	23	F	Yes	D 10	NR	Weakness tingling numbness	laminectomy	Good	NR	No compli cation
Larson et al.,[5]	1957	33	M	No	D 3	CVJ	Paraplegia	yes	Good	NR	_
Decker & Wei[6]	1969	15	M	No	D10	CVJ	Parapareses	yes	Good	NR	Assoc iated Cereb
Blaauw[7]	1975	48	M	NR	D 1	CVJ	NR	yes	Good	NR	-
Twersky et al.,[8]	1975	53	M	No	D 5	CVJ	NR	yes	Worsened	NR	Associated costal
Becker & Epstein[9]	1978	17	M	NR	D 2	CVJ	NR	yes	Good	NR	-
Ho & Lipton[10]	1979	58	F	Yes	D 1 - D2	Lamin a inferop osterio	15-year gradual Progressive weakness, numbness,	laminectomy	Poor	12 Mo	No recovery
Old & Triplett[11]	1979	21	F	Yes	D 3	CVJ	NR	yes	Good	NR	_
Buur & Mørch[12]	1983	33	M	Yes	D 4	Pedicle	Spastic paraparesis	Laminectomy	Good	7 Mo	_
Moriwaka et al.,[13]	1990	9	M	NR	C 7 - D1	P e	Pain thigh, couln't	L a	Improve d/compl	3 Mo	_
O'Brien et al.,[14]	1994	14	F	NR	D 9 - D10,	P e d ic le	Decreased senastion with paresthesisa, spasticparaparesis with bladder	Wide lamin ectom y T11-	Good	1 Mo	_
Quirini et al.,[2]	1996	24	M	NR	D 8	VB endplate	Difficulty in walking,	yes, Excision	Good	NR	_
Govender & Parbhoo[15]	1999	14	F	NR	D 8	Neural arch	Weakness of both lower limbs and urinary	Po ste rio	Good	3 Mo	Misdiag nosed Tubercu
Mermer et al.,[16]	2002	15	M	Yes	D 5	VB	Weakness of right lower limb	Anterior decompress ion of T4–	Good	6 Mo	one or two clonus beats
Faik et al.,[17]	2004	17	M	NR	D2	Pedicle/ VB	spastic paraparesis	dec om	Good	NR	No complication
Bess[18]	2005	11	F	Yes	D 5	VB	Ataxia, Hyperreflexia	Observation	Good	29Mo	No complication
Roach et al.,[19]	2009	NR NR NR	M N R M	NR NR NR	D 9	N R N R	Quadriplegi a Progressive weakness	E x c i s	Partia lly resolv ed Good	N R N R	Associa ted cervical lesion
Ezra et al.,[20]	2010	4	M	Yes	C 7 - D1	L a m i	Pain in neck & B/L Leg Difficulty in walking urinary	L a m i	Improved	NR	Residual deficits included right arm and
Gunay[21]	2010	36	F	YES	D 12	Pedicle	Pain, Weakness,	Excision Laminect	Good	44 Mo	slight hypoesthesia
Lotfinia[22]	2010	31	M	YES	D 8	Facet	B/L Paresthesia,	Lami necto	Poor	NR	Partial Improvement
Tian et al.,[23]	2011	16	M	YES	D 6	VB endplate	progressive weakness and	L a m i	Good	12Mo	_

Zaijun[24]	2013	16	M	YES	D 5 - D6	SP & TP	Paraplegia, Hypesthesia	yes	Good	9 Mo	No complication
Bari et al., [25]	2012	16	M	NO	D 9 - D12	NR	Restricted movements and urinary incontinenc	Surgery	Good	NR	No complication
Al Kaissi et al., [26]	2013	9	M	NR	D3-5	Pedicle	NR	yes		NR	_
Calvo[27]	2013	9	M	NR	D 3	Pedicle	Inability to walk	La mi	Good	3Mo	_



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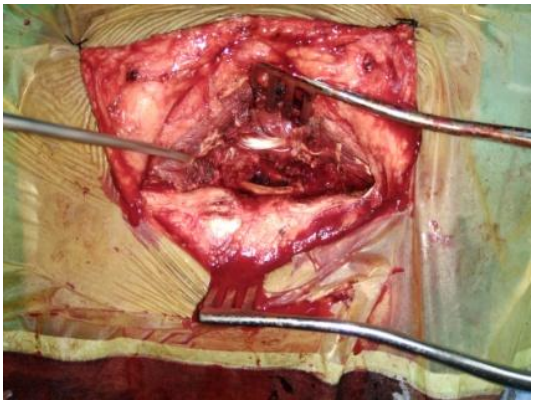

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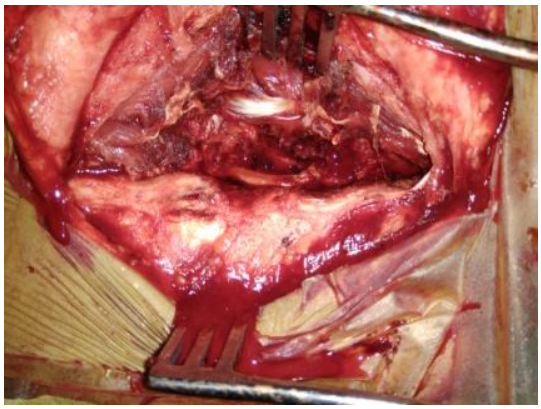

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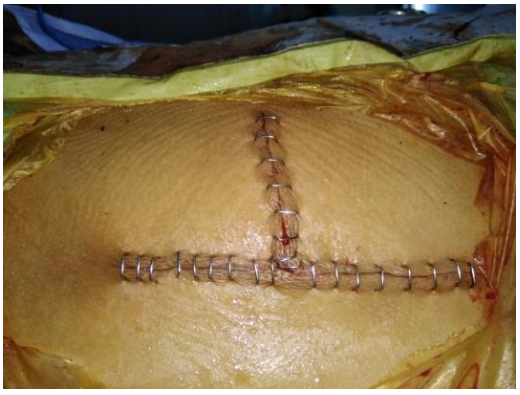

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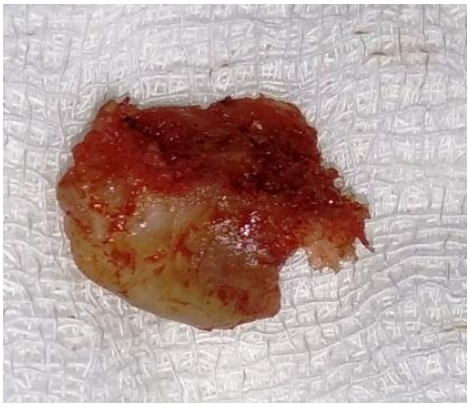

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Fig.3 Intraoperative findings: dorsal approach showing osteochondroma excision from the dorsal T-12 vertebra alongwith postion of the patient and the excised osteochondroma from the spine.

Conclusion

Though spine is very rare site for osteochondroma it can be seen in cases of Hereditary Multiple exostosis. A vertebral Osteochondroma should be excluded in all patients with hereditary multiple exostosis who presents with spinal pain and neurological deficit. Early imaging is helpful in diagnosing spinal Osteochondroma. Surgical intervention usually yeilds good outcome.

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Ethical approval: Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Author contributions:-All authors have made substantial contributions to the publication of this case report.

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