CASE REPORT

Solitary thoracic osteochondroma presenting as Brown-Séquard syndrome

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SUMMARY

We discuss a rare case of solitary osteochondroma arising from D2–D3 vertebrae presenting as Brown-Séquard syndrome (BSS) in a 28-year-old man following a fall. MRI revealed cord compression by an extradural ventrolateral tumour projecting into the spinal canal at D2–D3 level. Intraoperatively, it was found to be a bony tumour arising from the left of the D2 and D3 vertebral bodies, left pedicles and adjacent articular facets. The neurological deficits of the patient improved after the surgery.

BACKGROUND

The vertebrae are infrequent sites of osteochondromas.¹ These tumours can manifest as solitary lesions or as part of a hereditary syndrome.^{2–4} In this report, we discuss a rare case of a solitary osteochondroma of D2–D3 vertebrae presenting as Brown-Séquard syndrome (BSS). Osteochondromas rarely cause BSS.⁵ The possible pathophysiology, clinical course and treatment outcome have been discussed.

CASE PRESENTATION

A previously healthy 28-year-old man noticed insidious onset progressive pain in his left shoulder blade for 1 year. He was advised analgesics by his local physician. The pain was replaced by hypoesthesia in the same region after few weeks. One month prior to presentation the patient developed insidious onset stiffness of his left lower limb. The patient ultimately presented to our emergency department with rapidly progressive weakness of the left lower limb and diminished sensation to pain on the right half of his body below the level of right scapula following a fall at home. There were no cutaneous stigmata or any significant family history to suggest a congenital lesion. On neurological examination, there was decreased pinprick and temperature sensation on the right half of the body up to the D5 dermatome level. Proprioceptive and vibratory sensations were impaired in the patient's left lower limb. Motor examination revealed spastic plegia of the left lower limb, whereas power in the right lower limb was +4/5. Plantars were extensors bilaterally.

INVESTIGATIONS

MRI demonstrated a sessile mass arising from the left posterior aspect of D2 and D3 vertebrae, with adjacent pedicles and articular facets of D2–D3 projecting ventral to the dorsal cord, causing severe compressive myelopathy. The tumour was isointense with the bone on T1-weighted and T2-weighted sequences. T2-weighted MRI demonstrated the

cartilaginous cap as hyperintense and the bony base as hypointense (figure 1A–C).

TREATMENT

Early surgery on the patient was planned as first case in the next elective operation theatre with peri-operative steroid coverage. The patient presented to us within 8 h of the fall, therefore an injection of methylprednisolone was given as bolus of 30 mg/kg of body weight followed by infusion at 5.4 mg/kg/h for 23 h. The patient underwent D2–D3 laminectomy. At surgery, the tumour appeared well-circumscribed, firm and calcified with a smooth cartilaginous surface. It also caused hypertrophy of adjacent ligamentous tissues. It had a cartilage cap that merged with the underlying bone. It was removed piecemeal with a bone cutting microdrill. A complete resection of the tumour was performed.

OUTCOME AND FOLLOW-UP

The patient's neurological deficits improved gradually after the surgery. He had a gradual improvement in stiffness and began to regain power in his left lower limb in the first week after surgery. On the 10th postoperative day the patient had normal power in his right lower limb with motor power of grade 3/5 in the left lower limb. Sensory symptoms showed gradual recovery to normal over the next 3 months. The patient regained normal power with rehabilitative physical therapy during that time. The postoperative scan demonstrated a complete excision with stable sagittal alignment (figure 1D). The histopathological examination was consistent with an osteochondroma (figure 2). The skeletal survey of the patient did not reveal similar lesions. At follow-up of 3 years the patient is leading a normal life.

DISCUSSION

Osteochondromas are the most common benign tumour of the bones.¹ Osteochondromas may be solitary or multiple, the latter being associated with the autosomal dominant syndrome known as hereditary multiple exostoses (HME).^{2–4} Only 1.3–4.1% of solitary osteochondromas arise in the spine; however, approximately 9% of patients with HME harbour spinal lesions.^{3 4 6} Approximately two-thirds of patients with HME have a positive family history.^{2–4} The majority of these lesions are asymptomatic due to growth out of the spinal canal.^{1 6} Osteochondromas are composed of cortical and medullary bone with an overlying hyaline cartilage cap and demonstrate continuity with the underlying parent bone cortex and medullary

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Figure 1 Postcontrast T1-weighted (A) and T2-weighted (B) sagittal MRI showing an extradural lesion arising from D2–D3 vertebral bodies, isointense to the bone, causing cord compression. (C) T2-weighted axial MRI showing the lesion arising from the left of the vertebral body, left pedicle and adjacent articular process. Diseased facets are hypertrophied as compared with opposite facets. (D) Postoperative sagittal MRI of the spine showing complete excision of the tumour.

canal.^{2 7} Malignant transformation is seen in approximately 1% of solitary osteochondromas and in 3–5% of patients with HME.^{4 8} Hyaline cartilage cap thickness is an important criterion in determining malignant transformation. A cartilage cap over 1.5 cm thick in a skeletally mature patient should be viewed with great suspicion.⁸ Men are affected more than women (1.5:1). Patients most commonly present with symptoms in the second and third decades of life, with a mean age of 20 years.^{1 3 4 6}

Osteochondromas are thought to arise through a process of progressive endochondral ossification of aberrant cartilage of a growth plate as a consequence of congenital defect or

trauma.^{7 9} Some authors consider it as a developmental bony lesion or hamartoma rather than a true neoplasm.^{1 7 9} Spinal osteochondromas are more common in the cervical spine, and C-2 is the most frequent site.^{1–4} Albrecht *et al*¹ have postulated that the predominance of cervical lesions is caused by micro-trauma inflicted on the epiphyseal cartilage (and displacement of a portion thereof), because of the greater mobility and flexibility of these vertebrae.

CT is the imaging of choice as it demonstrates the bony details accurately.^{2 10} Our patient presented in emergency with MRI already performed before referral, therefore we did not subject the patient to CT scan. The distinguishing feature of an osteochondroma is the continuity of its cortical margin with that of the surrounding bone, very well visible in our case.^{2 10}

About 14 cases of symptomatic solitary thoracic osteochondroma have been reported, none of which caused BSS.¹¹ Han and Kuh⁵ reported BSS due to posterior cord compression caused by an osteochondroma arising from the C7 lamina in a known case of HME. The pure BSS reflecting hemisection of the cord is not often observed. A clinical picture composed of fragments of the syndrome or of the hemisection syndrome plus additional symptoms and signs is more common. These less pure forms of the disorder are often referred to as Brown-Séquard-plus syndrome.^{12 13} Our patient fits into Brown-Séquard-plus syndrome due to the presence of bilateral extensor plantar reflexes and slight weakness of the right lower limb. Our case is peculiar for presentations such as Brown-Séquard-plus syndrome due to ventrolateral compression caused by a solitary thoracic osteochondroma. Acute deterioration over a history of chronic myelopathy may be explained by a sudden vascular event and oedema inflicted by minor trauma to the already compromised thoracic cord. Oedema of the spinal

Figure 2 Photomicrograph showing a cartilaginous cap with underlying new bone formation.

cord explains the presence of additional features such as weakness of the right lower limb and bilateral extensor plantar reflexes in our case. Surgical resection is the only form of treatment for such lesions. Complete resection of the lesion is reported to be curative. Recurrence is due to incomplete removal of the cartilaginous cap.^{1 6 14} During surgery, we found that the tumour was harder than the normal bone, making drilling and cutting extremely difficult. It also caused hypertrophy of adjacent ligamentous tissues. The proximity of the lesion to the thoracic cord made the surgery a formidable technical problem. We tried to minimise the removal of articular processes. We removed the medial third of hypertrophied diseased left-sided articular processes to minimise manipulation of the cord. Posterior fusion was not required in our case because we could remove the lesion without complete sacrifice of articular facets and pars interarticularis. Postlaminectomy kyphosis was not observed during the follow-up period. Posterior fusion should be done in cases that require sacrifice of pars and articular facets in addition to laminotomy or laminectomy.

Learning points

- Sporadic thoracic osteochondroma presenting with Brown-Séquard syndrome is extremely rare.
- Osteochondroma may manifest as acute exacerbation of neurological deficit over a background of chronic myelopathy after an episode of trauma.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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