Unusual presentation of more common disease/injury

Solitary osteochondroma of the atlas causing spinal cord compression: a case report and literature review

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Correspondence to Dr Asifur Rahman, bijoun14@yahoo.com**Summary**

Solitary osteochondroma (SOC) of the spine is very rare, though OC is the commonest benign tumour of the bone. Only about 1–4% of SOC occur in the spine. And solitary osteochondroma of spine causing cord compression is even rarer. These tumours slowly enlarge, creating insidious but progressive symptoms of myelopathy or radiculopathy or both. Clinical histories, routine radiographs, CT studies, MRI studies and histopathological studies are the adjuncts for a definitive diagnosis. Surgical intervention can lead to functional and neurologic improvement with very little chance of recurrence. The authors present a case of a C1 posterior arch intraspinal SOC with cord compression who recovered very well after surgery and was doing well without recurrence after 1 year of surgery. The authors are reporting this case with English language medical literature review as it is quite rare among the SOC of the cervical spine.

BACKGROUND

A new case of spinal cord compression by an isolated osteochondroma (OC) of the spinal canal arising from the posterior arch of the atlas is added to the 15 previously reported cases in the English-language medical literature.

CASE PRESENTATION

A 16-year-old male, with no significant family history, presented with a history of suboccipital headache and progressively increasing quadripareisis for duration of over 6 months with no bladder or bowel dysfunction. Physical examination revealed spastic quadripareisis grade 4.

INVESTIGATIONS

Cervical CT scan with 3D reconstruction (figure 1), following a questionable plain radiograph, showed an intraspinal OC arising from the posterior arch of the atlas in the midline protruding into the spinal canal. MRI (figure 2) showed significant compression of the spinal cord with high signal intensity of the cord at C1 level.

TREATMENT

The patient was taken up for surgery and a C1 laminectomy with excision of the bony mass in toto was performed. Intraoperatively, dense fibrous adhesions were found between the dura and the mass which had to be meticulously dissected. Complete excision of the tumour was ascertained with a postoperative CT scan (figure 3) and the histopathology (figure 4) confirmed the diagnosis of OC.

OUTCOME AND FOLLOW-UP

The patient was discharged on the 7th postop day with significant improvement. On follow-up after 1 year the patient was found to have minimal residual deficit.

DISCUSSION

OCs constitute 0–15% of all bone tumours and 20–50% of all benign bone tumours.^{1 2} Only about 1–4% of solitary OCs (SOC) occur in the vertebral column, and this tumour is only slightly more common in the hereditary form.^{1 3–8}

Figure 1 Preoperative 3D CT scan of cervical spine lateral (A) and superior (B) views showing an intraspinal osteochondroma arising from the posterior arch of the atlas in the midline protruding into the spinal canal (white arrows).

A disease of growing bone, affecting males more frequently than females (2.5:1), OCs typically present in younger patients during the 2nd to 3rd decades of life; however, it has also been described in the 8th decade.^{4 9 10} Our patient presented in his second decade which is the commonest age of presentation.

We performed a thorough review of English language medical literature using PubMed and Google search to check the literature for reported cases of SOC of the atlas (C1) causing spinal cord compression (table 1). Patient's characteristics including age, sex and the site of origin on the vertebra were reviewed.

These lesions are more common in the appendicular skeleton.²² Spinal involvement in SOC is between 1–4%.^{4 14} Of

the OCs with spinal involvement, 56% are in the cervical spine, 38% are dorsal and 6% are lumbar.²³ The propensity for exostoses to develop in the cervical spine has been attributed to the mobility of the cervical spine.⁶ In the cervical spine, the C-2 vertebra is the most frequent site, followed by C-3 and C-6.^{4 8 9 15} Although the cervical spine is the commonest location among the SOC of the spine, in the present case the lesion is in C-1 posterior arch, which is not the commonest site among cervical spine cases.

OC typically arises from the pedicles or lamina, but can also arise from the vertebral body.³ The predilection for the neural arch could stem from the fact that it has numerous secondary ossification centers (for example, spinous, transverse and articular processes) that appear between the ages of 11 and 18 years.²⁴ Though the C-1 does not have a typical spinous process, which is substituted with the slender posterior arch, it is possible to have origin of a SOC from the secondary ossification centre, as we feel what happened in our case.

Spinal cord compression by an OC is an unusual and extremely rare phenomenon, because the majority of these lesions grow out of the spinal canal and only 0.5–1% of spinal OC may present with neurological dysfunction.^{14 25–27} In our case the OC was projected

Figure 2 MRI of cervical spine T2WI showing compression of the spinal cord from behind by the protruding osteochondroma (black arrow) and signal change of the cord at C1 level (white arrow).

Table 1 Literature review of cases of SOC of the atlas presenting with spinal cord compression

Authors	Year	Age (years)	Sex	Origin of lesion
Miyakoshi <i>et al</i> ¹¹	2010	58	M	Posterior arch
Wang and Chou ¹²	2009	16	F	Anterior arch
Ozturk <i>et al</i> ¹³	2007	46	M	Posterior arch
Chooi <i>et al</i> ¹⁴	2005	23	F	Posterior arch
Sharma <i>et al</i> ¹⁵	2002	45	M	Posterior arch
		18	M	Posterior arch
		40	M	Posterior arch
Kroppenstedt <i>et al</i> ¹⁶	2002	61	M	Posterior arch
Khosla <i>et al</i> ³	1999	45	M	Posterior arch
		39	M	Posterior arch / left lamina of C1
Calhoun <i>et al</i> ¹⁷	1992	43	M	Posterior arch
Slavotinek <i>et al</i> ¹⁸	1991	57	F	Anterolateral arch
Lanzieri <i>et al</i> ¹⁹	1985	19	M	Posterior arch
Julien <i>et al</i> ²⁰	1978	28	F	Posterior arch
Mitsumori <i>et al</i> ²¹	1975	54	M	Posterior arch

Figure 3 Postoperative 3D CT scan lateral (A) and superior (B) views showing cut margins of posterior arch of atlas after complete removal of the osteochondroma (white arrows).

inwards from the posterior arch of C-1 compressing on the spinal cord giving rise to cervical myelopathy.

Spinal OC may be difficult to detect and interpret on plain radiographs because of the many superimposed structures found on various projections.²⁸ X-ray cervical spine in the lateral view in flexion showed a well-demarcated oval osseous lesion arising from the under surface of the C-1 posterior arch protruding into the spinal canal (figure 5) which was not very obvious in the extension view (figure 6). CT findings may be characterised by a paraspinal, dumbbell, or eccentrically located, round, sharply outlined mass in the spinal canal with bonelike density

Figure 4 Photomicrograph (H&E stain, X40) showing bony trabeculae (black arrow) with cartilage cap (white arrow).

Figure 5 X-ray cervical spine lateral view in flexion showing a well-defined osseous mass arising from the under surface of C1 posterior arch and protruding into the spinal canal (white arrow).

with scattered calcifications and osteosclerotic changes in neighboring bone and lacking contrast enhancement.²⁹ CT scan of the cervical spine in our case exhibited an irregular shaped lobulated osseous mass with sclerotic margin that projected from the inner aspect of the exostotic posterior arch of C-1 into the spinal canal. MRI is useful in demonstrating the level and the extent of neural compression, along with the marrow content and the cartilaginous cap, which gives the appearance of a prominent peripheral rim of low intensity corresponding to ossification, and a small central core of intermediate signal similar to that of bone marrow.⁹ Both the T1WI and T2WI sagittal sections showed a definite compression on the spinal cord at C-1 and C-2 levels. The MRI of the patient also demonstrated the signal intensity of bone marrow in the centre of the mass and peripheral hypointense rim suggestive of ossification.

Due to a low rate of malignant transformation, asymptomatic SOC can be followed conservatively.¹⁰ When the tumour causes pain or neurological complications due to compression, or the diagnosis is indefinite, it should be excised at its base. When feasible, complete excision is recommended, because incomplete removal of the cartilaginous cap may lead to tumour recurrence.⁵⁻⁸ Intralesional excision of the exostoses is associated with an unacceptably high recurrence rate and should be avoided.⁶ Our patient was having clinical symptoms of cervical myelopathy which was progressive in nature and we planned for a total removal of the mass to relieve the cord compression as well as to prevent recurrence. We excised the lesion enmass with partial C-1 laminectomy.

Figure 6 X-ray cervical spine lateral view in extension where the lesion is not very discernible.

Figure 7 Excised osteochondroma showing bosselated, bluish-gray glistening outer surface (white arrow).

On gross examination, OC has a bosselated, glistening cartilage cap which has a bluish-gray surface, reminiscent of cauliflower (figure 7). On microscopic examination, a cartilage cap appears to merge with the underlying bone and is covered with a thin layer of fibrous capsule that functions as a perichondrium (figure 4).^{30 31}

Recurrence rates after resection are approximately 2% with a highly variable disease-free interval, ranging from 1 to 26 years.^{2 4} At 1 year follow-up the patient showed clinical improvement with no signs of recurrence.

Learning points

- ▶ Preoperative radiographic evaluation should consist of CT and MRI to provide optimal information about the lesion and aid treatment options.
- ▶ Appropriate surgical intervention can lead to functional and neurologic improvement with no evidence of recurrence.
- ▶ Given the diagnostic difficulty of these spinal lesions, the difficulty of clinical and radiologic surveillance, and the risk of malignant transformation, we recommend a complete surgical resection of all diagnosed SOC in the vertebral column.
- ▶ Spinal cord compression caused by a SOC is improved by surgery in majority of patients.

Competing interests None.

Patient consent Obtained.

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