CASE REPORT

Cervical osteochondroma with neurological symptoms: literature review and a case report

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INTRODUCTION: Osteochondromas are common benign tumors of bone and spinal involvement is uncommon. Solitary spinal osteochondromas may produce a wide variety of symptoms depending on their location and relationship to adjacent neural structures.

CASE PRESENTATION: Herein, we present a case of solitary osteochondroma arising from the posterior arch of C1, causing left-sided ascending numbness and paresthesia and difficulty walking. The lesion was totally resected through a posterior approach. Histopathological examination confirmed the diagnosis of benign osteochondroma.

DISCUSSION: Spinal cord compression is uncommon in spinal osteochondromas because in most cases the tumor grows out of the spinal column. To prevent neurological compromise, complete surgical removal is mandatory when an intraspinal osteochondroma with cord compression is diagnosed, which also helps to prevent recurrence. Our literature review of similar cases indicates that despite the old belief that C2 is the most commonly involved vertebra for osteochondromas, C1 is actually the most commonly involved vertebra in the cervical region.

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INTRODUCTION

Osteochondroma, or osteocartilaginous exostosis, is the most common skeletal neoplasm¹ and constitutes 20–50% of all benign bone tumors and 10–15% of all bone tumors.² It is commonly found in the appendicular skeleton but is very rarely seen in the spine.³ Osteochondromas may be solitary or multiple when associated with hereditary multiple exostoses or osteochondromatosis, an autosomal dominant trait.^{3,4} Only 1.3–4.1% of solitary osteochondromas originate in the spine.⁵ Rarely, spinal osteochondromas may present with nerve root or cord compression.^{4,6} Even symptoms due to vertebral artery occlusion have been described.⁷ When the tumor causes pain or neurological complications or when the diagnosis is unclear, it should be excised surgically.⁵ Herein, we present a case of osteochondroma arising from the posterior arch of C1 in a 48-year-old female with a favorable long-term surgical outcome and discuss the relevant literature.

CASE PRESENTATION

Methods

Using PubMed and Google search engines, we performed a thorough review of English language medical literature for reported cases of solitary cervical osteochondromas with neurological symptoms. Patient's characteristics, including age, sex, symptoms, the site of origin on the cervical vertebra, recurrences and the method of surgery were all reviewed.

Case Illustration

A 48-year-old woman presented with symptoms of left-sided paresthesia and weakness, ascending numbness and difficulty in

walking over 6 months. Her gait became steadily cumbersome during the last 2 months. Physical examination revealed no apparent sensory or motor impairment. Deep tendon reflexes were exaggerated, particularly on the left side and the left plantar reflex had an extensor response. Hoffman's reflex was positive in the left hand. Other neurological examinations were normal. MRI of the cervical spine revealed a hypointense mass lesion on parasagittal section with cord compression. (Figure 1a). Also, the compression due to spinal cord displacement was clear on axial sections (Figure 1b). Computed Tomography (CT) of the cervical vertebrae revealed the bony nature of the tumor and its origin from posterior arch of the first cervical vertebra (Figure 1c). The patient underwent a posterior cervical approach and laminectomy (without fusion) and radical resection of the tumor. The post-operative course was uneventful and the symptoms improved immediately after surgery, and the patient fully recovered without any residual deficit. Histopathological analysis of the specimen confirmed the diagnosis of osteochondroma (Figure 2). At 6-month follow-up, the patient had no clinical problems. On serial control radiography and CT scans, the lesion was found to have been completely removed without any evidence of recurrence at 5 years' long-term follow-up (Figure 1d).

DISCUSSION

According to the 2002 WHO definition, osteochondroma (osteocartilaginous exostosis) is a cartilage capped benign bony neoplasm on the outer surface of bone.⁸ Osteochondromas make up about 8.5% of all bone tumors and about 36% of benign lesions.⁵ These tumors are thought to arise in the peripheral portion of the growth plate. A focus of metaplastic cartilage forms

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Figure 1. Sagittal T2-weighted MRI demonstrating severe cord compression by an extradural hypointense lesion originating from posterior arch of c1 (a). Axial T1-weighted at the corresponding level shows cord distortion by the hypointense mass arising from posterior elements of C1 vertebra (b). Axial CT Scan depicting the relatively hyperdense lesion originating from c1 posterior arch with both intraspinal and extraspinal growth (c). Postoperative CT scan showing total resection of the tumor with sufficient cord decompression (d).

Figure 2. Hematoxyline and Eosine staining of the specimen at the junction of cartilaginous cap and underlying cancellous bone confirmed the diagnosis to be osteochondroma.

and grows through progressive endochondral ossification as a consequence of trauma or a congenital perichondral deficiency.³ These tumors develop as either solitary or multiple lesions when associated with hereditary multiple exostosis, an autosomal dominant condition.⁹

Most osteochondromas originate from the diaphysis of long bones, predominantly involving the distal femur, proximal tibia, proximal humerus and pelvis.¹⁰ Spinal involvement is rare.^{11–13} Only 1.3–4.1% of solitary osteochondromas arise in the spine,¹⁴ where they constitute up to 0.4% of intraspinal tumors or 3.9% of solitary spinal tumors.⁵ Unlike pedunculated long bone osteochondromas, vertebral osteochondromas are sessile.¹⁰

In terms of gender distribution, solitary lesions affect males more than females, and the common age of onset is in the second or third decade of life.^{12,15,16} The demographic pattern reveals the onset of these lesions at the mean age of 20 in the hereditary variety and 30 in the solitary subtype;¹⁶ however, a case as old as 84 years has been reported.¹⁷

These lesions can be found at any part of the vertebral column, but the cervical spine is the most common (50%).^{4,18} The propensity for exostoses to develop in the cervical spine has been attributed to the mobility of the cervical spine.^{3,19,20} Greater stress due to mobility could cause microtrauma and displacement of a portion of the epiphyseal growth cartilage^{9,20} with consequent displacement of a portion of physal cartilage and exostosis formation.¹⁹ C2 is the most common vertebrae to be involved in the cervical spine, followed by C3 and C6,¹⁸ but in the present case, the lesion is in C1 posterior arch, which is not the commonest site among cervical spine cases.

Cord compression is very unusual in patients with spinal osteochondromas^{11,18,21–23} because most of the lesions grow out

Table 1. Reported cases of Atlas osteochondroma

Case	Reference (year)	Age/sex	Chief complaint	Symptom duration	Neurological examination	Exostoses location	Surgery	Results
Mitsumori <i>et al.</i> Julien <i>et al.</i>	²⁵ (1975) ²⁶ (1978)	54/F 28/F	Hand pain Brown-Sequard syndrome	18 months 4 years	N/A Left hemiplegia & right hypaesthesia	C1–C2 C1	L L	N/A N/A
Wu <i>et al.</i>	²⁷ (1978)	54/F	Hand pain	7 months	N/A	C1	L	N/A
Lanzieri <i>et al.</i>	²⁸ (1985)	19/M	N/A	N/A	N/A	C1 posterior arch	L	N/A
Slavotinek <i>et al.</i>	²⁹ (1991)	57/F	Dysphagia	N/A	10–12 nerve palsy	C1 anterior arch	A	N/A
Calhoun <i>et al.</i>	³⁰ (1992)	43/M	Weakness	N/A	Hyper reflexia	C1 posterior arch	L	N/A
Lopez-Barea <i>et al.</i>	³¹ (1994)	31/M	Neck pain	N/A	Hyper reflexia	C1 posterior arch	L	N/A
Morikawa <i>et al.</i>	³² (1995)	21/M	Neck pain	N/A	Weakness	C1 posterior arch	L	N/A
Khosla <i>et al.</i>	³³ (1999)	39/M	Bilateral upper extremity weakness	N/A	Spasticity in lower limbs & paresis	Left C1 pedicle	HL	No recurrence after 35 months
Khosla <i>et al.</i>	³³ (1999)	45/M	Weakness in left leg & arm	2 years	N/A	Posterior arc of C1	HL	N/A
Sharma <i>et al.</i>	³⁴ (2002)	45/M	N/A	N/A	N/A	C1 posterior arch	L	N/A
Yoshida <i>et al.</i>	³⁵ (2006)	61/F	Sleep apnea	N/A	N/A	C1 lift anterior arch	N/A	N/A
Ozturk <i>et al.</i>	³⁶ (2007)	46/M	Pain & numbness in neck and left arm	N/A	Hypoesthesia in left C2 dermatome	C1 posterior arch	HL	N/A
Yagi <i>et al.</i>	³⁷ (2009)	77/F	Indurations in neck	N/A	Lower limb spasticity	C1 right side	HL	N/A
Schomacher <i>et al.</i>	¹⁴ (2009)	62/M	Neck & shoulder pain	1.5 years	Hypoesthesia	C1–C2 joint	HL	N/A
Wang <i>et al.</i>	³⁸ (2009)	16/F	Dysphagia & sleep apnea	N/A	N/A	Anterior C1 arch	A	N/A
Miyakoshi <i>et al.</i>	³⁹ (2010)	58/M	Weakness	N/A	Hyper reflexia & paresis	C1 posterior arch	L	N/A
Er <i>et al.</i>	⁴⁰ (2012)	42/F	Weakness & numbness in limbs	N/A	Spastic quadripareisis	C1	L	No recurrence after 2 years
Rahman <i>et al.</i>	²² (2012)	16/M	Headache & quadripareisis	6 months	Spastic quadripareisis	Posterior C1 arch	L	No recurrence after 1 year
Zhang <i>et al.</i>	⁴¹ (2014)	19/F	Headache, nausea and vomiting and loss of balance	N/A	Acute cerebellar infarct	C1	L	N/A
Lotfinia <i>et al.</i>	Present case (2016)	48/F	Left-sided paresthesia and difficulty in walking	6 months	Left side hyper reflexia	C1	L	Full recovery No recurrence after 5 years

Abbreviations: A, anterior cervical approach; A&P, two-stage posterior & anterior approach; F, facetectomy; HL, hemilaminectomy; L, laminectomy; L&PR, laminectomy and pedicle resection; N/A, not applicable; P, posterior approach and tumor resection (without laminectomy or facetectomy).

of the spinal canal.^{10,20} When there is compression of neural structures, this can cause various neurological symptoms and signs depending on the level and degree of cord and/or root compression. In our case, the tumor was projected inwards from the posterior arch of C1, leading to apparent cord compression. Because of the complex imaging of the vertebral bones, spinal osteochondromas are sometimes difficult to be readily detected on plain radiographs.⁹ CT Scan is the imaging modality of choice. Not only it can demonstrate the cartilaginous and osseous components of the tumor, but also clearly defines the tumor's extent and its relationship to the vertebral and neural elements of the spine.²¹ On MRI, osteochondromas are seen as isointense lesions with a low signal rim produced by the cortical bone. MRI provides excellent demonstration of the spinal cord and nerve root compression,⁹ so it is valuable for detecting evidence of neurological compression, which appears as low signal on T1-weighted sequences and high signal on T2-weighted sequences.¹

Asymptomatic lesions can be followed conservatively,^{10,24} but surgery should be considered whenever the diagnosis is not definite, or the patient is in pain or in the case of progressive neurological deficit.²⁴ Nearly all reported cases of spinal osteochondromas presenting with spinal cord compression are treated with laminectomy or hemilaminectomy, owing to the posterior origin of most osteochondromas.²¹ In this case, we planned for a radical excision of the tumor through C1 posterior arch removal to relieve the cord compression and to prevent recurrence. Incomplete resection may lead to recurrence, which occurs in 2–5% of lesions.^{15,18,23} Complete excision of the cartilaginous cap is essential to prevent recurrence.^{18,22} The mean time to recur has been reported to be 5 years.¹⁷

A thorough review of English literature using PubMed and Google found 80 reported cases of solitary cervical osteochondroma. The male-to-female ratio was 1.51:1. The youngest patient reported was 8 years old and the oldest one was 77 years old, and the mean age for solitary cervical osteochondroma in this review being 35.8 years.

The most common symptoms were arm radicular pain, limb weakness, neck mass and limb numbness. Other less common symptoms included dysphagia, headache, sleep apnea, hand atrophy, difficulty in walking, head tilt and dizziness. On examination, the most common signs were limb weakness, hyper reflexia, limb hyposthesia and neck mass. Other less common signs included restricted cervical range of motion, local tenderness, occipital neuralgia, limb atrophy, cranial nerve palsy, Horner's syndrome and torticollis.

It is believed that C2 is the most common vertebrae to be involved by solitary osteochondroma (24.4%), followed by C1 (18.83%) and C7 (15.15%).^{4,9,11,15,18,20,23} Our review of literature, including the present case, showed that C1 is the most involved vertebra in the cervical region (24%), followed by C2 (21%) and C5 (18%). The C3 body was the most uncommon site (5.4%). Although the exact origin of the tumor remains unidentified in most reports, the laminae were found to be the most common site, and less commonly involved structures were the pedicles, facets, spinous processes, transverse processes and anterior arch of C1 vertebra. Table 1 summarizes the characteristics of reported C1 osteochondromas.

Nearly all reported cases of cervical osteochondromas presenting with spinal cord compression have been treated with laminectomy or hemilaminectomy alone due to the posterior origin of most cervical osteochondromas. In 17 cases of this review, patients were followed up for signs of recurrence for a range of 6 months to 10 years (mean: 3/4 years). Two recurrences occurred 6 years postoperatively and one in 28 months. At 5-year follow-up of the present case, no signs of radiological recurrence have been witnessed.

CONCLUSION

Solitary C1 osteochondroma is rarely reported in middle-aged women. Gradual cord compression is a reason for delays in diagnosis; however, in most cases, the tumor is successfully treated by laminectomy with favorable outcomes. Complete surgical removal is necessary to prevent recurrence. Despite the old belief, C1 should be considered as the most commonly involved vertebrae.

COMPETING INTERESTS

The authors declare no conflict of interest.

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