

Solitary Osteochondroma of the Spine—A Case Series: Review of Solitary Osteochondroma With Myelopathic Symptoms

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Abstract

Study Design: Case series and literature review.

Objective: There is a growing body of literature supporting that osteochondroma of the spine may not be as rare as previously documented. The purpose of this study was to perform an updated review and present our experience with 4 cases of solitary osteochondroma of the spine, including surgical treatment and subsequent outcomes.

Methods: A review of 4 cases and an updated literature review.

Results: All 4 cases were diagnosed as solitary osteochondroma of the spine based on clinical and histopathologic findings. Majority of the lesions arose from the posterior column with one case showing extension into the middle column with clinical neurologic sequelae. Treatment strategies for all cases included complete marginal excision of the lesions using a posterior approach. All 4 cases showed no radiographic evidence of recurrence. The literature review yielded 132 cases of solitary osteochondroma and 17 case associated with multiple hereditary exostosis. Out of the 132 cases, 36 presented with myelopathic symptoms.

Conclusion: Osteochondroma of the spine may not be as rare as previously reported. The best approach to treatment in almost all symptomatic cases include wide surgical excision of the tumor. This should include complete resection of the cartilaginous cap of the tumor in an effort to prevent recurrence. When excision is performed properly, the outcomes are excellent with very low recurrence of the tumor.

Keywords

osteochondroma, literature review, case series, exostosis, surgical excision

Introduction

Osteochondroma (exostosis) is the most common benign bone tumor, accounting for 36% of benign bone tumors.¹ Most often found in long bones, reports suggest osteochondroma of the spine to be relatively rare, accounting for only 4% to 7% of primary benign spinal tumors¹⁻³ and less than 3% of all osteochondromas.^{3,4} Osteochondroma can arise as a solitary lesion or as part of an inherited condition known as multiple hereditary exostosis (MHE).¹ Several studies have reported that solitary osteochondromas are more common in the spine when compared with osteochondroma associated with MHE.^{2,4,5} There is a growing body of evidence suggesting osteochondroma of the spine

may not be as rare as previously reported.⁶⁻⁸ In this article, we describe our experience with the diagnosis, treatment, and natural history of osteochondroma of the spine of 4 cases, and the most up-to-date literature review of this topic since 2003.

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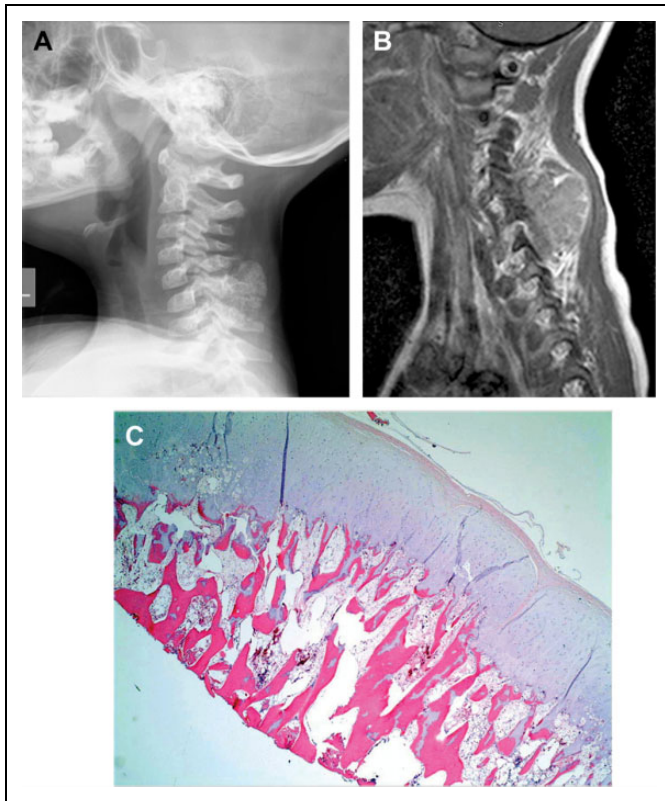


Figure 1. A 6-year-old female referred for a nonpainful mass in her neck noticed by her family. (A) Lateral radiograph showing ossified mass involving the C6 vertebrae and (B) sagittal magnetic resonance imaging showing the extent of the soft tissue involvement and the mass arising from the posterior cervical elements. (C) Hematoxylin and eosin slide of the cervical mass demonstrating a benign cartilage cap with subchondral bone, findings typical of an osteochondroma.

Case Series

The authors have obtained the patients' informed written consent for print and electronic publication of the case report.

Case 1

A 6-year-old female was brought to the emergency department for nonradiating neck pain that was localized to her left posterior neck. The patient had no neurological signs or symptoms. Radiographs demonstrated an osseous neck mass arising from the posterior cervical elements (Figure 1). Advanced imaging (magnetic resonance imaging and computed tomography scans) demonstrated an osseous lesion with a medullary cavity contiguous with the left C6 lamina. No signs of cord or root compression were seen. The patient has no known significant medical or family history of similar lesions.

Case 2

A healthy 35-year-old male complained of 2 weeks of persistent mid-back pain after riding a go-cart. The patient denied any neurological symptoms during this time and was

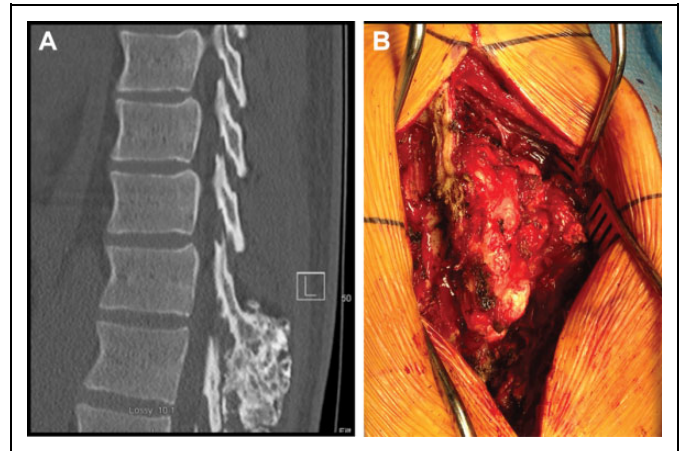


Figure 2. A 35-year-old male with mid-back pain following a minor injury. Plain radiographs demonstrated a mass arising from the posterior elements of T12. (A) Mid-Sagittal computed tomography scan of the thoracic spine demonstrating an osseous mass at the thoracolumbar junction. (B) Intraoperative clinical photograph of well-encapsulated thoracic mass.

neurologically intact on physical examination. Imaging studies showed an osseous lesion about the low thoracic and thoracolumbar junction (Figure 2) without neurological involvement. The patient denies any medical or family history of similar lesions.

Case 3

An 11-year-old male had progressive right posterior neck swelling for the past 10 months. A cervical computed tomography scan showed a mass at the C6 vertebrae with medullary continuity with the right lamina and spinous process. There was no evidence of cord or nerve root compression or vascular compromise. There was no significant medical or family history of similar lesions.

Case 4

A 36-year-old female had neck pain and progressive myelopathy (bowel and bladder dysfunction, gait abnormality, and progressive upper and lower extremity weakness). No history of antecedent trauma was reported. Plain radiographs showed an osseous lesion arising from the posterior column with significant canal compromise at the level of the C3 and C4 vertebrae (Figure 3). The patient had no medical or family history of similar lesions.

Literature Review

Ovid MEDLINE and other nonindexed citations database search engines were used with the assistance of a medical librarian. The terms "osteochondroma" and "spine" and/or proxy descriptors were used to query PubMed. No limit in publication year, country, or language of publication was used. This yielded a list of all reported cases of osteochondroma of

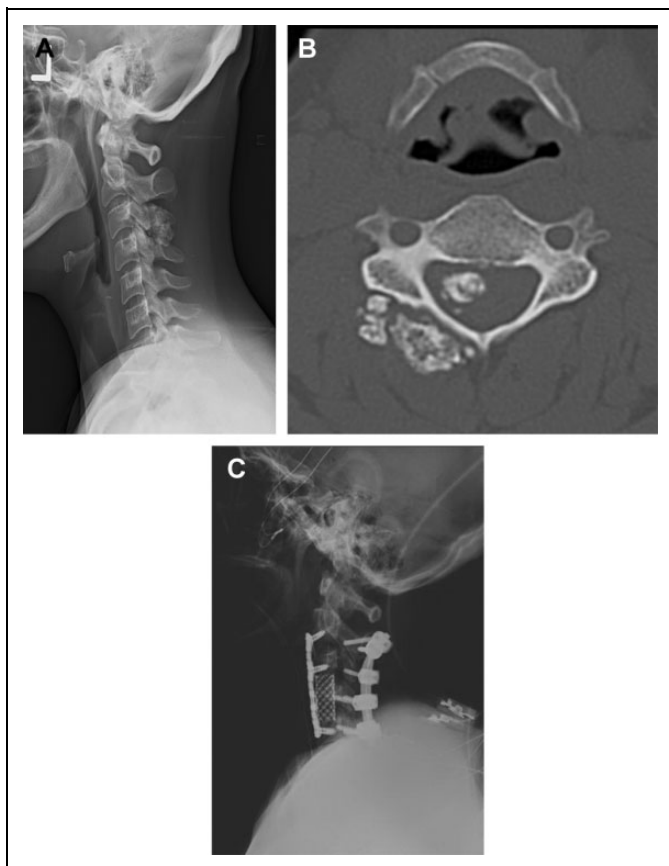


Figure 3. (A) Lateral radiograph and (B) axial computed tomography scan showing osseous mass arising from the right C3 lamina and invading the spinal canal causing cord compression. Biopsy revealed an osteochondroma. (C) Postoperative lateral radiograph showing C3 vertebrectomy, anterior reconstruction with a titanium cage and plate, and a posterior instrumented fusion from C2 to C5 required to stabilize the spine following a wide resection of the osteochondroma.

the spine since 1951. The list of articles was screened using the inclusion criterion—all reported cases from 2016 to 2004—and the following exclusion criteria: literature reviews, cases of primary tumor not arising from spine, non-case report accounts of cases, and nontumor processes (infection). Each case was reviewed for each parameter of clinical history and radiographic description whenever available in the case reports. Demographics, anatomic location of tumor, symptoms, treatment, and recurrence rates of tumor were almost always available and reported. A custom-built Excel database was used to organize and analyze the data. Descriptive statistics were used to summarize the results of the data.

Results

All 4 cases were diagnosed as solitary osteochondroma of the spine based on clinical findings and histopathologic features. All cases except for “Case 4” had no neurological symptoms—Case 4 was associated with cord compression and progressive

myelopathy. Three of the 4 cases involved the cervical spine (includes case with cord compression) and 1 of 4 from the thoracolumbar region. All cases of osteochondroma in this series appeared to arise from the posterior column, with one case showing extension into the middle column and clinical neurologic sequelae. Treatment strategies for all cases included complete marginal excision of osteochondroma lesions using a posterior approach. Additionally, Case 4 (osteochondroma with cervical retrovertebral lesion and cord compression) required anterior corpectomy with placement of an interbody cage, followed by posterior decompression and instrumented fusion. All patients had complete symptomatic relief at their latest follow-up (up to 2 years) and showed no radiographic evidence of recurrence.

The review literature yielded a total of 223 articles,²⁻⁹² of which 110 were from the 2016 to 2004 period. Twenty-six articles were excluded, leaving 84 articles in the final analysis. The 84 articles yielded 149 reported cases. One hundred and thirty-two (88.6%) were solitary osteochondromas and 17 (11.4%) were associated with MHE.

Table 1 lists all 132 cases of solitary osteochondroma of the spine from the literature. Table 1 highlights the interesting data from each case of solitary osteochondroma. The location, treatment, and outcome of the cases are shown, along with the demographic data. For solitary osteochondromas (Table 2), there was a female-to-male ratio of 1:1.6 and an average age of 35.2 years (range = 2-77). The most common spinal level involved was cervical, with 63 (52.2%) of the cases, followed by lumbar 35 (26.5%), thoracic 24 (18.2%), sacrum 9 (6.8%), and coccyx 1 (0.76%). The most frequent spinal anatomic column involved was the posterior column, with 85 cases (64.3%), followed by unknown 28 (21.2%), anterior column 19 (14.3%), and 0 in the middle column.

There were 36 (27.2%) cases that involved solitary osteochondroma with myelopathic symptoms (Table 3). This group had a female-to-male ratio of 1:2.6 and average age of 35.1 years (range = 8-77). The most common spinal level involved was cervical in 24 (66.6%) cases, followed by thoracic 8 (22.2%) and lumbar 4 (11.1%). The most frequent spinal anatomic column involved was posterior column, with 29 cases (80.5%), followed by anterior column 6 (16.6%), unknown 1 (2.7%), and middle column 0 (0%). The osteochondroma began in the posterior arch in 20 (55.5%) of the cases, followed by the lamina in 7 (19.4%), vertebral body in 5 (16.6%), spinous process in 2 (5.5%), and unknown location in 1 (2.7%) of the cases.

All 36 patients underwent surgery, of whom 29 (80.5%) underwent a posterior approach, 3 (8.3%) underwent an anterior approach, 2 (5.5%) underwent a combined anterior-posterior approach, and 2 (5.5%) approaches were unknown. The clinical outcomes showed improvement of symptoms in 34 (94.4%) of the patients, with 28 people showing a complete recovery and 6 with a partial recovery. Two cases showed worsening symptoms after surgery. There were 2 recurrences among all cases recorded, and none among solitary lesions with myelopathic symptoms.

Table 1. Reviewed Cases in Literature: Interesting Data^a.

Author	Journal	Year	Age	Sex	Tumor Level	Location	Presentation	Radiculopathy	Myelopathy	Treatment	Clinical Outcomes	Recurrence
Ramzan et al	<i>Pediatric Neurosurgery</i>	2016	8	Male	C1	Posterior C1 arch	Symptomatic	No	Yes	Complete excision	Complete resolution	N/A
Bauer et al	<i>Skeletal Radiology</i>	2015	19	Female	C1	C1 posterior arch	Symptomatic	No	Yes	Complete excision	Complete resolution	No
Michael	<i>Journal of Pediatric Orthopaedics</i>	2015	16	Female	L5-S1	Facet joints	Symptomatic	No	No	Complete excision	Complete resolution	No
Haque et al	<i>European Spine Journal</i>	2015	21	Male	S3-S4	Sacrum L of midline	Asymptomatic	No	No	Complete Excision	Complete resolution	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	35	Female	C7	N/A	N/A	N/A	N/A	En bloc resection	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	48	Male	S1	N/A	N/A	N/A	N/A	En bloc resection	N/A	Yes
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	46	Male	C7	N/A	N/A	N/A	N/A	Intralesional excision	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	46	Female	T9-T10	N/A	N/A	N/A	N/A	Intralesional excision	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	61	Male	L2	N/A	N/A	N/A	N/A	Intralesional excision	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	65	Male	C3-T2	N/A	N/A	N/A	N/A	En bloc resection	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	48	Male	S1	N/A	N/A	N/A	N/A	En bloc resection	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	43	Female	C6-C7	N/A	N/A	N/A	N/A	En bloc resection	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	76	Female	T11-T12	N/A	N/A	N/A	N/A	Intralesional excision	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	21	Male	S1	N/A	N/A	N/A	N/A	En bloc resection	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	49	Female	C5-C7	N/A	N/A	N/A	N/A	Intralesional excision	N/A	No

(continued)

Table 1. (continued)

Author	Journal	Year	Age	Sex	Tumor Level	Location	Presentation	Radiculopathy	Myelopathy	Treatment	Clinical Outcomes	Recurrence
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	17	Male	S1	N/A	N/A	N/A	N/A	En bloc resection	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	32	Male	L4-L5	N/A	N/A	N/A	N/A	Intralesional excision	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	60	Male	T12	N/A	N/A	N/A	N/A	Intralesional excision	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	68	Female	L2	N/A	N/A	N/A	N/A	Intralesional excision	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	13	Female	T1	N/A	N/A	N/A	N/A	En bloc resection	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	19	Male	L5	N/A	N/A	N/A	N/A	En bloc resection	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	38	Male	L4-L5	N/A	N/A	N/A	N/A	En bloc resection	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	20	Male	L5	N/A	N/A	N/A	N/A	En bloc resection	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	36	Female	T11-T12	N/A	N/A	N/A	N/A	En bloc resection	N/A	Yes
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	26	Male	C4-C5	N/A	N/A	N/A	N/A	Intralesional excision	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	33	Female	L1	N/A	N/A	N/A	N/A	En bloc resection	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	21	Male	T7	N/A	N/A	N/A	N/A	En bloc resection	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	18	Male	T1-T2	N/A	N/A	N/A	N/A	Unknown	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	13	Male	L5-S3	N/A	N/A	N/A	N/A	En bloc resection	N/A	No

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Table 1. (continued)

Author	Journal	Year	Age	Sex	Tumor Level	Location	Presentation	Radiculopathy	Myelopathy	Treatment	Clinical Outcomes	Recurrence
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	17	Female	C1-C3	N/A	N/A	N/A	N/A	Intralesional excision	N/A	No
Sciubba et al	<i>Journal of Neurosurgery: Spine</i>	2015	2	Female	T8-T11	N/A	N/A	N/A	N/A	En bloc resection	N/A	No
Kang et al	<i>JAMA Otolaryngology—Head and Neck Surgery</i>	2015	60	Female	C2	N/A	Symptomatic	No	No	Complete excision	N/A	N/A
Eren et al	<i>The Spine Journal</i>	2015	24	Female	L4	Spinous process	Symptomatic	No	No	N/A	N/A	N/A
Neal et al	<i>Military Medicine</i>	2015	40	Male	L5	Right anteriosuperior endplate of L5	Symptomatic	No	No	Complete excision	Complete resolution	No
Dormont et al	<i>Clinical Neuroradiology</i>	2014	59	Male	C4	Posterior C4 arch	Symptomatic	No	No	Complete excision	Near complete resolution	No
Mahore et al	<i>BMJ Case Reports</i>	2014	28	Male	D2-D3	Posterior arch	Symptomatic	No	Yes	Laminectomy	Complete resolution	No
Boucetta et al	<i>Pan African Medical Journal</i>	2014	48	Male	C6	Posterior arch	Symptomatic	No	No	Complete excision	Complete resolution	N/A
Kantarsi et al	<i>The Spine Journal</i>	2014	24	Female	C3	Lamina	Symptomatic	Yes	No	N/A	N/A	N/A
Fumiaki et al	<i>Neurologia Medico Chirurgica</i>	2014	57	Male	L4	Inferior articular process	Symptomatic	Yes	No	Laminectomy	Complete resolution	No
Fumiaki et al	<i>Neurologia Medico Chirurgica</i>	2014	63	Female	S1	Superior articular process of	Symptomatic	Yes	No	Hemilaminectomy	Complete resolution	No
Fumiaki et al	<i>Neurologia Medico Chirurgica</i>	2014	48	Female	L4	Inferior articular process	Symptomatic	Yes	No	Hemilaminectomy	Near complete resolution	No
Fumiaki et al	<i>Neurologia Medico Chirurgica</i>	2014	32	Male	L4	Inferior articular process	Symptomatic	Yes	No	Hemilaminectomy	Complete resolution	No
Fumiaki et al	<i>Neurologia Medico Chirurgica</i>	2014	62	Male	L4	Inferior articular process	Symptomatic	Yes	No	Hemilaminectomy	Complete resolution	No
Barbagallo et al	<i>European Review for Medical and Pharmacological Sciences</i>	2014	68	Male	C4-C5	Anterior arch	Symptomatic	No	No	Complete excision	Complete resolution	No
Parekh et al	<i>BMJ Case Reports</i>	2014	20	Male	C7-T1	Posterior arch	Asymptomatic	No	No	Complete excision	N/A	N/A
Hopper et al	<i>Journal of Belgian Society of Radiology Orthopedics (Healto)</i>	2014	68	Female	T9-L3	Posterior arch	Symptomatic	No	No	Complete excision	Complete resolution	No
Mont et al	<i>Orthopedics (Healto)</i>	2014	11	Male	L2-L4	Inferior articular process	Symptomatic	Yes	No	En bloc resection	Complete resolution	No

(continued)

Table 1. (continued)

Author	Journal	Year	Age	Sex	Tumor Level	Location	Presentation	Radiculopathy	Myelopathy	Treatment	Clinical Outcomes	Recurrence
Jameel et al	<i>Journal of Clinical and Diagnostic Research</i>	2014	14	Female	C5-C6	Transverse process	Asymptomatic	No	No	Total excision	Complete resolution	No
David et al	<i>Asian Spine Journal</i>	2014	52	Male	C2-C6	Transverse process	Symptomatic	Yes	No	Laminectomy	Complete resolution	No
Chow et al	<i>Pediatric Neurology</i>	2013	9	Male	C1-C2	Inner surface of C2 arch	Symptomatic	Yes	No	C2 hemilaminectomy, resection of posterior C1 arch	Complete resolution	N/A
Scuotto et al	<i>BMJ Case Reports</i>	2013	56	Female	L2	Lamina	Symptomatic	Yes	No	En bloc resection	Complete resolution	No
Garg et al	<i>Kulak Burun Bogaz Ihtis Derg</i>	2013	22	Male	C3-C4	Vertebrae and pedicles	N/A	N/A	N/A	N/A	N/A	N/A
Jianru et al	<i>Journal of Spinal Disorders Tech</i>	2013	43	Male	L4	Spinous process	Symptomatic	Yes	No	Complete excision	Complete resolution	N/A
Jianru et al	<i>Journal of Spinal Disorders and Techniques</i>	2013	26	Male	C1-C2	Lateral mass	Symptomatic	No	Yes	Complete excision	Complete resolution	N/A
Jianru et al	<i>Journal of Spinal Disorders and Techniques</i>	2013	11	Male	T1	Laminar mass	Symptomatic	Yes	No	Laminectomy, complete excision	Complete resolution	N/A
Jianru et al	<i>Journal of Spinal Disorders and Techniques</i>	2013	60	Female	C1	Lateral mass	Symptomatic	No	Yes	Complete excision	Worsening of symptoms	N/A
Jianru et al	<i>Journal of Spinal Disorders and Techniques</i>	2013	34	Female	C1-C2	Lateral mass	Symptomatic	No	Yes	Laminectomy, complete excision	Complete resolution	N/A
Jianru et al	<i>Journal of Spinal Disorders and Techniques</i>	2013	17	Female	C1	Transverse process	Symptomatic	Yes	No	Complete excision	Complete resolution	N/A
Jianru et al	<i>Journal of Spinal Disorders and Techniques</i>	2013	63	Female	C5-C7	Lamina	Symptomatic	Yes	No	Complete excision	Complete resolution	N/A
Jianru et al	<i>Journal of Spinal Disorders and Techniques</i>	2013	17	Female	T6	Pedicle	Symptomatic	Yes	No	Laminectomy, complete excision	Complete resolution	N/A
Jianru et al	<i>Journal of Spinal Disorders and Techniques</i>	2013	49	Female	C2-C3	Vertebral body	Symptomatic	No	Yes	Laminectomy, complete excision	Worsening of symptoms	N/A
Jianru et al	<i>Journal of Spinal Disorders and Techniques</i>	2013	68	Female	L2	Lamina	Symptomatic	No	Yes	Laminectomy, complete excision	Complete resolution	N/A

(continued)

Table 1. (continued)

Author	Journal	Year	Age	Sex	Tumor Level	Location	Presentation	Radiculopathy	Myelopathy	Treatment	Clinical Outcomes	Recurrence
Jianru et al	<i>Journal of Spinal Disorders and Techniques</i>	2013	56	Female	T5	Vertebral body	Symptomatic	Yes	No	Laminectomy, complete excision	Partial functional resolution	N/A
Jianru et al	<i>Journal of Spinal Disorders and Techniques</i>	2013	57	Female	C5	N/A	Symptomatic	Yes	No	Laminectomy, complete excision	Complete resolution	N/A
Madan et al	<i>South Asian Journal of Cancer</i>	2013	9	Male	T1	Vertebral body, posterior arch	Symptomatic	No	Yes	Partial resection	Near complete resolution	No
Rai et al	<i>Global Spine Journal</i>	2013	65	Male	C2	Vertebral body	Symptomatic	No	No	Complete excision	Complete resolution	N/A
Ghasemikhah et al	<i>Iranian Journal of Radiology</i>	2013	19	Male	T9	Posterior arch	Symptomatic	No	Yes	Laminectomy	Complete resolution	N/A
Temiz et al	<i>Acta Orthopaedica et Traumatologica Turcica</i>	2012	62	Female	L2	Inferior articular process	Symptomatic	No	Yes	Complete excision	Complete resolution	No
Chang et al	<i>Skeletal Radiology</i>	2012	39	Male	L4	N/A	Symptomatic	No	No	N/A	N/A	N/A
Hussain et al	<i>BMJ Case Reports</i>	2012	16	Male	C1	Posterior arch	Symptomatic	No	Yes	Laminectomy	Near complete resolution	No
Temiz et al	<i>Turkish Neurosurgery</i>	2012	48	Male	L3	Inferior articular process	Symptomatic	Yes	No	Hemilaminectomy, complete excision	Near complete resolution	No
Kettner et al	<i>Spine</i>	2012	21	Female	C5	Spinous process	Symptomatic	No	No	Laminectomy w/ en bloc resection	Complete resolution	N/A
Mamindla et al	<i>Asian Journal of Neurosurgery</i>	2012	14	Male	C3	Lamina	Symptomatic	No	Yes	Laminectomy w/ en bloc resection	Complete resolution	N/A
Shin et al	<i>Journal of Korean Neurosurgery</i>	2012	32	Male	C4-C5	Lamina and facet joint	Symptomatic	No	Yes	Hemilaminectomy	Near complete resolution	No
Kars et al	<i>Asian Spine Journal</i>	2012	42	Female	C1	Lamina	Symptomatic	No	Yes	Laminectomy	Complete resolution	No
Nakamura et al	<i>Skeletal Radiology</i>	2011	69	Male	C7-T1	N/A	Symptomatic	No	Yes	Laminectomy	Complete resolution	No
Rousseaux et al	<i>Orthopaedics & Traumatology: Surgery & Research</i>	2011	23	Male	C4	Posterior arch	Symptomatic	No	Yes	Laminectomy	Complete resolution	No
Kettner et al	<i>Journal of Manipulative and Physiological Therapeutics</i>	2011	24	Male	C4	Vertebral body	Symptomatic	No	No	Nonsurgical—Spinal manipulation	Complete resolution	No

(continued)

Table 1. (continued)

Author	Journal	Year	Age	Sex	Tumor Level	Location	Presentation	Radiculopathy	Myelopathy	Treatment	Clinical Outcomes	Recurrence
Saglik et al	<i>Archives of Orthopaedic and Trauma Surgery</i>	2011	26	Male	L1	Spinous process	Symptomatic	No	No	Complete excision	Complete resolution	No
Saglik et al	<i>Archives of Orthopaedic and Trauma Surgery</i>	2011	9	Male	C3/C4-T1	Spinous process, posterior arch	Asymptomatic	No	No	Complete excision	Asymptomatic	No
Saglik et al	<i>Archives of Orthopaedic and Trauma Surgery</i>	2011	36	Female	T11-L1	Lamina	Symptomatic	Yes	No	Laminectomy, complete excision	Near complete resolution	No
Saglik et al	<i>Archives of Orthopaedic and Trauma Surgery</i>	2011	65	Male	C4	Vertebral body	Symptomatic	Yes	No	Anterior excision, followed by anterior cervical fusion	Complete resolution	No
Saglik et al	<i>Archives of Orthopaedic and Trauma Surgery</i>	2011	19	Male	C5-C6	Spinous process	Symptomatic	No	No	Complete excision	Complete resolution	No
Saglik et al	<i>Archives of Orthopaedic and Trauma Surgery</i>	2011	32	Female	L3-L4	Lamina	Asymptomatic	No	No	Nonsurgical	Asymptomatic	No
Schneider et al	<i>Ethiopian Medical Journal</i>	2010	7	Male	Coccyx	Coccyx	Symptomatic	No	No	En bloc excision	Complete resolution	N/A
Shimada et al	<i>Neurologia Medico Chirurgical</i>	2010	58	Male	C1-C2	Spinous process	Symptomatic	Yes	Yes	En bloc excision	Complete resolution	No
Meshkini et al	<i>Journal of Neurosurgery: Spine</i>	2010	29	Male	L4	Pedicle	Symptomatic	Yes	No	Laminectomy	Complete resolution	N/A
Meshkini et al	<i>Journal of Neurosurgery: Spine</i>	2010	58	Male	L5	Vertebral body	Symptomatic	Yes	Yes	Laminectomy	Complete resolution	N/A
Meshkini et al	<i>Journal of Neurosurgery: Spine</i>	2010	60	Male	C5	Lamina	Symptomatic	No	Yes	Hemilaminectomy	Complete resolution	N/A
Meshkini et al	<i>Journal of Neurosurgery: Spine</i>	2010	34	Male	C5-C6	Lamina	Symptomatic	No	Yes	Laminectomy	Near complete recovery	N/A
Meshkini et al	<i>Journal of Neurosurgery: Spine</i>	2010	55	Male	T9	Vertebral body	Symptomatic	No	Yes	Complete excision	Complete resolution	No
Meshkini et al	<i>Journal of Neurosurgery: Spine</i>	2010	17	Male	L3	Inferior facet	Symptomatic	Yes	No	Hemilaminectomy	Complete resolution	N/A

(continued)

Table 1. (continued)

Author	Journal	Year	Age	Sex	Tumor Level	Location	Presentation	Radiculopathy	Myelopathy	Treatment	Clinical Outcomes	Recurrence
Meshkini et al	<i>Journal of Neurosurgery: Spine</i>	2010	34	Female	C7	Pedicle	Symptomatic	No	Yes	Hemilaminectomy	Near complete resolution	N/A
Meshkini et al	<i>Journal of Neurosurgery: Spine</i>	2010	31	Male	T8	Superior facet	Symptomatic	No	Yes	Laminectomy	N/A	N/A
Kim et al	<i>The Spine Journal</i>	2010	54	Female	S1	Sacral ala— anterior surface	Symptomatic	No	No	Complete excision	Complete resolution	No
Cha et al	<i>Journal of Korean Neurosurgery</i>	2010	57	Female	L3	Lamina	Symptomatic	Yes	No	En bloc resection, laminectomy, facetectomy	Complete resolution	N/A
Horiuchi et al	<i>Journal of Neurosurgery: Spine</i>	2009	77	Female	C1	Posterior arch	Symptomatic	No	Yes	Hemilaminectomy w/t en bloc resection	Complete resolution	No
Horiuchi et al	<i>Journal of Neurosurgery: Spine</i>	2009	72	Male	L4	Inferior facet	Symptomatic	No	No	Marginal resection and facetectomy	Complete resolution	No
Horiuchi et al	<i>Journal of Neurosurgery: Spine</i>	2009	69	Male	L4-L5	Inferior facet	Symptomatic	No	No	Intraarticular injection, biopsy, and ablation of articular facet joint	Complete resolution	No
Tian et al	<i>Orthopedics</i>	2009	38	Male	L5	Lamina	Symptomatic	No	Yes	Laminectomy	Complete resolution	No
Jakheria et al	<i>Journal of Pediatric Orthopaedics B</i>	2009	8	Female	C2-C6	Spinous process	Symptomatic	No	No	En bloc resection	Complete resolution	No
Chou et al	<i>Case Reports/ Journal of Clinical Neuroscience</i>	2009	16	Female	C1-C2	Vertebral Body	Symptomatic	No	No	Complete Excision	Complete resolution	No
Wenyuan et al	<i>SAS Journal</i>	2009	28	Male	T8	Transverse process	Symptomatic	No	No	Radial excision	Complete resolution	N/A
Hassankhani et al	<i>Cases Journal</i>	2009	16	Female	L3	Spinous process	Asymptomatic	No	No	En bloc resection	N/A	No
Srikantha et al	<i>Journal of Neurosurgery: Spine</i>	2008	17	Male	C3	Spinolaminar junction	Symptomatic	No	Yes	En bloc resection	Complete resolution	No
Srikantha et al	<i>Journal of Neurosurgery: Spine</i>	2008	23	Male	C4-C5	Transverse processes, lamina, pedicles	Symptomatic	No	Yes	Partial resection, C4-C5 corpectomy, C3-C5 fusion	Complete resolution	No
Srikantha et al	<i>Journal of Neurosurgery: Spine</i>	2008	40	Female	C6	Superior articular facet	Symptomatic	Yes	No	Medial facetectomy	Complete resolution	No

(continued)

Table 1. (continued)

Author	Journal	Year	Age	Sex	Tumor Level	Location	Presentation	Radiculopathy	Myelopathy	Treatment	Clinical Outcomes	Recurrence
Byung-June et al	<i>Joint Bone Spine</i>	2007	23	Male	L5-S1	Facet	Symptomatic	Yes	No	Partial laminectomy	Complete resolution	No
Song et al	<i>European Journal of Pediatric Surgery Spine</i>	2006	11	Male	T4	Superior articular process	Symptomatic	No	Yes	Laminectomy (T2-T3)	Complete resolution	No
Zhao et al	<i>Spine</i>	2007	23	Female	C7	Transverse process	Symptomatic	No	No	En bloc resection	Complete resolution	No
Chatzidakis et al	<i>Acta Neurochirurgica</i>	2007	22	Male	C2	Dens of C2	Symptomatic	No	No	N/A	N/A	No
Ozturk et al	<i>Acta Orthopaedica Belgica</i>	2007	46	Male	C1	Lamina	Symptomatic	Yes	No	Hemilaminectomy	Complete resolution	No
Maheshwari et al	<i>Orthopaedic Surgery</i>	2006	20	Male	C7	Pedicle	Symptomatic	No	Yes	Laminectomy	Complete resolution	No
Moon et al	<i>Pediatric Neurosurgery</i>	2005	16	Male	C5-C7	Spinous process	Symptomatic	Yes	Yes	Hemilaminectomy, complete excision of tumor	Complete resolution	No
Samartzis et al	<i>Spine</i>	2006	11	Male	S2	Lamina	Symptomatic	Yes	No	Laminectomy S1-S4	Complete resolution	No
McCall et al	<i>Journal of Neurosurgery</i>	2006	13	Female	C3	Lamina	Asymptomatic	No	No	Complete excision	N/A	N/A
Yoshida et al	<i>Acta Oto-Laryngologica</i>	2006	61	Female	C1	Anterior arch	Symptomatic	No	No	Complete excision	Complete resolution	No
Grivas et al	<i>European Spine Journal</i>	2005	46	Female	C7	Pedicle	Symptomatic	Yes	No	Complete excision	Complete resolution	No
Brastianos et al	<i>Neurosurgery</i>	2005	26	Female	T12	Vertebral body	Symptomatic	No	Yes	Complete excision, T12 corpectomy	Complete resolution	No
Agrawal et al	<i>Pediatric Neurosurgery</i>	2005	14	Male	L5-S1	Iliac crest	Symptomatic	Yes	No	Laminectomy	Complete resolution	No
Faik et al	<i>Joint Bone Spine</i>	2005	19	Male	T4-T5	Costovertebral angle, T4-T5 foramina	Symptomatic	Yes	No	Laminectomy, complete excision	Complete resolution	No
Miyamoto et al	<i>Spinal Cord</i>	2005	23	Male	C2	Pedicle	Symptomatic	No	Yes	L hemilaminectomy, partial excision	Partial functional recovery	No
Kouwenhoven et al	<i>European Spine Journal</i>	2004	42	Male	C1-C2	Neural arches	Symptomatic	Yes	No	Laminectomy, en bloc resection	Complete resolution	No
Gu rkanlar et al	<i>Journal of Clinical Neuroscience</i>	2004	35	Male	L4	Lamina	Symptomatic	Yes	No	Complete Excision	Complete resolution	No
Schrot et al	<i>Journal of Neurosurgery</i>	2004	15	Male	C8	Dermatome	Symptomatic	Yes	No	Hemilaminectomy, pedicectomy w/t complete excision of tumor	Complete resolution	No
Kulkarni et al	<i>Neurologia Medico Chirurgica</i>	2004	15	Male	T10-T11	Facet	Symptomatic	No	Yes	Laminectomy	Complete resolution	No

(continued)

Table 1. (continued)

Author	Journal	Year	Age	Sex	Tumor Level	Location	Presentation	Radiculopathy	Myelopathy	Treatment	Clinical Outcomes	Recurrence
Gille et al	<i>Spine</i>	2004	18	Female	C4	Transverse process	Symptomatic	Yes	No	Cervicotomy	Complete resolution	No
Gille et al	<i>Spine</i>	2004	15	Male	C5	Vertebral body	Symptomatic	No	Yes	Laminectomy and cervicotomy	Complete resolution	No
Gille et al	<i>Spine</i>	2004	73	Male	C2	Posterior arch	Symptomatic	No	Yes	Laminectomy	Complete resolution	No
Gille et al	<i>Spine</i>	2004	18	Male	T11	Pedicle	Asymptomatic	No	No	Laminectomy	Complete resolution	No
Gille et al	<i>Spine</i>	2004	28	Female	L4	Posterior arch	Symptomatic	Yes	No	Laminectomy	Complete resolution	No
Gille et al	<i>Spine</i>	2004	45	Female	S1	Vertebral body	Symptomatic	Yes	No	Lumbotomy	Complete resolution	No

^aAll 132 cases reviewed from literature are presented. Age, sex of the patient, location of lesion, type of surgery, symptoms, and recurrence are shown. If symptomatic w/o myelopathic or radiculopathic symptoms, symptomatic due to pain. N/A, data unavailable in the literature.

Table 2. Demographic Data of Solitary Osteochondroma of the Spine, 132 Cases, Without a Known Hereditary Genetic Disorder.

Sex, males	61.3%
Age, years (mean, range)	35.2 (2-77)
Spinal level of tumor	
Cervical	65 (52%)
Thoracic	24 (18%)
Lumbar	35 (27%)
Sacrum	9 (7%)
Coccyx	1 (1%)
Involved spinal column	
Posterior	85 (64%)
Anterior	19 (14%)
Middle	0 (0%)
Unknown	28 (21%)

Table 3. Demographic Data on Solitary Osteochondroma With Spinal Cord Compression^a.

Sex, males	72.2%
Age, years (mean, range)	35.1 (8-77)
Spinal level of tumor	
Cervical	24 (66.6%)
Thoracic	12 (22.2%)
Lumbar	5 (11.1%)
Sacrum	0 (0%)
Involved spinal column	
Posterior	29 (81.5%)
Anterior	6 (16.6%)
Middle	0 (0%)
Unknown	1 (2.7%)
Origin of tumor	
Pedicle	3 (8.3%)
Laminae	7 (19.4%)
Spinous process	2 (5.5%)
Posterior arch other than pedicle, laminae, spinous process	17 (47.2%)
Vertebral body	6 (16.6%)
Unknown location	1 (2.7%)
Treatment	
Anterior approach	3 (8.3%)
Posterior approach	29 (80.5%)
Combined anterior-posterior approach	2 (5.5%)
Unknown approach	2 (5.5%)
Patients requiring excision	36 (100%)

^aA case series of 27 patients with unknown locations of osteochondroma and unknown symptoms, which was part of the data, had to be excluded from the results due to lack of data.

Discussion

The first solitary osteochondroma was reported in 1843 by Reid.⁶⁴ Many reports in the literature show that solitary osteochondroma is more common than lesions associated with MHE. The prevalence of osteochondroma in the spine is likely higher than previously thought. There seems to be a rise in the amount of case reports of osteochondroma published in the recent years (2004 to 2016). When Albrecht et al² reviewed the relevant English literature from 1843 to 1992, it yielded

96 cases of solitary spinal osteochondroma. When Gille et al⁸ updated the review, they identified 54 additional cases of solitary spinal osteochondroma from 1992 to 2003. Our study yielded 132 new cases reported from 2004 to 2016, representing a 2.4-fold increase since 2003.

This increase in the number of cases in a smaller period of time is likely due to a higher rate of case reports being published on the topic rather than an actual increase in the incidence of these tumors. Nevertheless, the higher number of reported cases in the past decade likely underestimates the true prevalence of osteochondroma, because a significant portion of these tumors/lesions remain asymptomatic and, thus, may not be seen by a health care provider and/or require surgical treatment.

The review and analysis of the reported cases corroborate some of the trends seen in the literature, such as cervical spine being the most common site for a solitary osteochondroma of the spine, complete surgical excision being the most common method of surgical treatment, and the good outcomes and low recurrence rates after excision.

Additionally, the review of literature indicated that 27.2% of the cases with solitary osteochondromas of the spine had myelopathic features. This is in concordance with previous reports of 30% by Albrecht et al.² It is proposed that the myelopathic symptoms seen in osteochondroma are due to progressive compression of the spinal structures, but may include a potentiated effect as the tumor grows over several years; likewise, the onset of age-related degenerative changes seen with spinal stenosis may also contribute.⁸

Osteochondroma is a form of exostosis that can be seen in any age group. It is generally reported that the age range for symptomatic presentation for solitary osteochondroma is between 10 and 30 years for peripheral lesions, but it appears that spine patients develop symptoms at an average age of 32, distinctly different from the peripheral lesions seen in children. By definition, osteochondroma has a characteristic cartilage cap on histology and a medullary continuity with the host bone, and can be sessile or pedunculated. MHE involves many exostoses in a single patient, unlike in the case of solitary osteochondroma, which is more common. An incidence of 1.3% to 4.1% has been reported as the percentage of solitary osteochondromas that affect the spine; however, 9% of MHE lesions are found in the spine.² In the current review, 11.4% of all the cases of osteochondromas of the spine reviewed were associated with MHE.

Malignant transformation is low in solitary osteochondroma (<3%), but can be as high as 10% when associated with inherited genetic mutations as seen with MHE. MHE has an autosomal dominant inheritance pattern and involves mutations in the EXT 1, 2, and 3 genes on chromosome 8, 11, and 19, respectively. Malignant degeneration leads to a low-grade peripheral chondrosarcoma, which is managed with complete surgical resection. Malignant transformation of solitary osteochondroma is most frequently reported in the pelvis and rarely occurs in the spine.

A treatment algorithm for these lesions should begin with a thorough history and physical examination, to evaluate for genetic inheritance of similar lesions and to rule out neurovascular compromise that will necessitate surgery. Moreover, the majority of these lesions remain benign and are painless. In benign cases, observation with radiographic surveillance (computed tomography and magnetic resonance imaging and other advanced imaging may be used as indicated to better characterize the lesion and its local effects). Osteochondromas do have a tendency to increase in size and, depending on its location, may be associated with neurologic sequelae. In cases where unrelenting pain and/or evidence of neurovascular compromise (radiculopathy, myelopathy, or vascular compression) exists, surgical management may be warranted. Surgical treatment may include in situ marginal or wide excision, via a posterior, anterior, or combined approach, with or without instrumentation. In some cases, that is, Case 4, a need for cord or nerve root decompression along with instrumented stabilization with or without fusion may be required. Tumor excision may sometimes require both an anterior approach and a posterior approach. Of paramount importance during surgical excision is complete resection of the characteristic cartilage cap seen with these tumors. Incomplete resection of the cartilage cap may increase the risk of recurrence, and the pediatric population is more susceptible to tumor recurrence given their higher growth potential/age at presentation.

The recurrence rate in the review of the literature was 1.3% for all cases, and 0% for solitary spinal osteochondromas with myelopathic symptoms. Nevertheless, the current review of literature demonstrates a lower recurrence rate than previously reported (4%).⁸ However, there may be a number of unreported recurrences, given that not all cases in the literature explicitly reported this parameter. There is also the impact of a better understanding of the biology of the tumor, advanced imaging, and surgical techniques allowing for more expedient treatment in the recent years.

Conclusion

Osteochondroma is a relatively common bone tumor, accounting for 36% of all benign bone tumors,¹ but occurs infrequently in the spine accounting for less than 3% of all osteochondromas.^{3,4} The solitary lesions in the spine may cause neurologic symptoms including radiculopathy and myelopathy, 29.5% and 27%, respectively, as reported in this review. The best approach to treatment in almost all symptomatic cases is marginal excision of the tumor. Meticulous surgical excision, with complete resection of the cartilaginous cap of the tumor, is important in preventing recurrence. When tumor excision is performed adequately, the outcomes are excellent with very low recurrence rates.

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employee of Norton Healthcare; board member, Scoliosis Research Society; receives consulting fees from Medtronic and DePuy; receives payments for lectures from Medtronic, DePuy, and Norton Healthcare; holds patents and receives royalties from Medtronic; is on the editorial review board of *JBS Highlights*, *Spine*, *Spine Deformity*, *JAAOS*, and *Global Spine*. Nuvasive provided funds directly to database company. No funds are paid directly to individual or individual's institution 06/2012 to 04/2015. LYC is an employee of Norton Healthcare; member, Editorial Advisory Board, *Spine* and *Spine Journal*; institutional review board member, University of Louisville Institutional Review Board; research committee member, Scoliosis Research Society; receives research funds from the Orthopedic Research and Educational Fund, 2013 to present; receives Scoliosis Research Society Research Funding, 2013 to present; received funds for travel for Study Planning Meetings from the Center for Spine Surgery and Research of the University of Southern Denmark; received funds for travel for annual required Continuing Education for Institutional Review Board Members, University of Louisville Institutional Review Board; Nuvasive provided funds directly to database company. No funds are paid directly to individual or individual's institution 06/2012 to 4/2015.

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