Spinal Exostoses

Analysis of Twelve Cases and Review of the Literature

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Study Design. Retrospective review of spinal exostoses treated at our institution and literature review.

Objectives. Review of 12 cases of spinal exostoses treated at our institution compared with 165 cases of spinal exostoses reported in the literature.

Summary of Background Data. Spinal exostoses are uncommon. Most reports consist of 1 to 3 cases. The relationship between solitary exostoses and those associated with multiple hereditary exostoses (MHE), as well as the incidence of intraspinal and extraspinal location, symptoms, and results of treatment are unclear.

Methods. The medical records, operative reports, and diagnostic imaging of 12 patients with spinal exostoses treated at our institution between 1972 and 2002 were reviewed. The literature was reviewed using MEDLINE search of English literature and bibliographies of published manuscripts.

Results. Solitary spinal exostoses were more common than those associated with MHE. Lesions were most common in the upper cervical spine and originated from the posterior elements. Patients with exostoses associated with MHE were significantly younger and had a higher incidence of symptoms consistent with neural structure compression than patients with solitary exostoses. Complete excision resulted in resolution of preoperative symptoms. Intralesional excision resulted in recurrence in all cases.

Conclusions. Spinal exostoses are more common than reported previously. Patients with MHE that present with back pain or neurological symptoms should produce a high index of suspicion. Evaluation should include both computed tomography and magnetic resonance imaging to define the origin of the exostosis and the presence of neural structure compression. Surgical excision should be preformed en bloc.

Key words: osteochondroma, exostoses, multiple hereditary exostoses, spine, neural structure compression.

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Spinal exostoses are reported as uncommon lesions, representing 4 to 7% of all primary benign spinal tumors.1–4 Spinal exostoses occur in two distinct clinical settings: as solitary lesions or in association with multiple hereditary exostoses (MHE).3 The incidence of solitary exostoses involving the spine ranges from 1 to 4%.2 The incidence of spinal exostoses associated with MHE varies from 3 to 9%.3,6 However, recent case reports suggest that spinal exostoses may be more common than reported previously.7–17 The purpose of this study was to review our clinical, diagnostic, and treatment experience with 12 cases of spinal exostoses and to contrast our results with 165 spinal exostoses reported previously.1,2,4–70

Materials and Methods

Between 1972 and 2002, 16 patients with spinal exostoses were evaluated at our institution. Four patients were referred from outside institutions for consultation regarding treatment options and ultimately received definitive management at their original institution. Twelve patients received definitive management at our institution. Medical records and diagnostic imaging of these 12 patients were reviewed (Table 1). Exostoses were classified as solitary or associated with a history of MHE. Medical records were evaluated for age, chief complaint, duration of symptoms, and symptoms indicative of neural structure compression (radicular pain, weakness, hyperreflexia, sensory deficit, and ataxia). Physical examination was reviewed for neurological deficit, hyperreflexia, palpable mass, and other pertinent findings. Method of treatment, surgical margins, histopathology, and complications were evaluated. All original radiographic studies were reviewed by one of us (M. R. R.). Lesions were evaluated for spinal location (cervical, thoracic, lumbar, or sacral), vertebral site of origin, and intraspinal or extraspinal location. Diagnosis was confirmed radiographically and, when available, by histopathology. Follow-up evaluations were reviewed for resolution, progression or recurrence of symptoms, and tumor enlargement or recurrence.

Literature Review. The literature was reviewed using MEDLINE search of English literature and bibliographies of published manuscripts. Cases were reviewed for clinical details including age, symptoms, method of treatment, resolution of symptoms, and tumor recurrence. Exostoses were classified as solitary or associated with MHE. The spinal location and site of origin on the vertebrae of the exostoses was recorded.

Statistical Analysis. Data were analyzed using a Student t test to compare the means between the two populations. Values of P ≤ 0.05 were considered to be statistically significant.

Results

Seven females and 5 males were treated at our institution. Five patients had MHE, and 7 patients had a soli-
tary exostosis. The mean age of all patients was 24.2 years (range, 7–52 years). Patients with MHE were significantly younger than patients with solitary exostoses, as the mean age for patients with MHE was 16.8 years (range, 7–34 years), compared with 29.5 years (range, 22–52 years) for patients with solitary exostoses ($P < 0.045$). All patients <20 years of age had MHE. The mean follow-up was 6.5 years (range, 0.5–15 years; Table 2).

**Patients With MHE**

Three of 5 patients with MHE (60%) had exostoses in the cervical spine, 1 patient had a lesion in the thoracic spine, and 1 had a sacral exostosis. All MHE patients with cervical exostoses had lesions at C2. All patients with MHE had intracanicular exostoses. Four of 5 lesions (80%) associated with MHE originated from the posterior elements, 1 originated from the vertebral body.

Three patients (60%) with MHE had local pain associated with their lesion. Three patients (60%) had symptoms or findings on physical examination consistent with neural structure compression. Two patients with intracanicular exostoses at C2 producing spinal cord compression had no symptoms or physical findings to suggest neural structure compression (Table 2).

Two of 5 patients with MHE were treated nonoperatively. Both patients reported resolution of symptoms and no tumor enlargement at average 2.2 year follow-up (range 2–2.5 years). Three of 5 patients (60%) were treated surgically with extralesional resection. There were no complications, no tumor recurrence, and complete resolution of symptoms and at mean follow up of 7.6 years (range 3–13 years; Figure 1).

**Patients With Solitary Lesions**

Three patients (42%) with solitary exostoses had lesions in the cervical spine, 3 (42%) had lesions in the lumbar spine, and 1 (14%) in the thoracic spine. Four patients (57%) had extracanicular exostoses. Six of 7 (86%)
solitary lesions originated from the posterior elements, 1 originated from the vertebral body (Table 2).

Four patients (57%) with solitary lesions had local pain. Three (42%) reported a palpable mass. Four patients (57%) had had symptoms or findings on physical examination consistent with neural structure compression (Table 2).

Two of 7 patients (29%) were treated nonoperatively. There was resolution of symptoms and no tumor enlargement at an average of 13 years follow-up (range 11–13 years). Five of 7 patients (71%) were treated surgically. Three patients were treated with extralesional resection. There was no tumor recurrence and resolution of presenting symptoms at mean follow-up of 4 years (range 0.5–7 years) in all patients. There was one early postoperative complication. An anterior compartment syndrome developed in the left lower extremity in a 23-year-old male after wide excision and stabilization of a large thoracolumbar osteochondroma. Despite emergent fasciotomies, postoperative follow-up at 4.5 years showed residual deficit in the left extensor hallucis longus muscle. Two patients underwent intraskeletal excision. Postoperative radiographic evidence of retained exostoses was confirmed in both cases after the initial excision. Both patients reported neck pain at follow-up evaluation. Both patients underwent repeat excision of the lesion. There was no tumor recurrence in either patient after the second procedure at a mean follow-up of 5.5 years (4 and 7 years).

**Literature Review**

Literature review identified 165 reported cases of spinal exostoses.1,2,4–70 Eighty-nine exostoses (53%) were solitary lesions, 56 exostoses (34%) were associated with MHE, and 20 exostoses (12%) were not specified. The location of the lesion was reported in 156 cases. The cervical spine was the most common site (85; 54%), followed by the thoracic (42; 27%), lumbar spine (26; 16%), and sacrum (3; 2%).

Solitary lesions occurred most commonly in the cervical spine (46; 51%), followed by near equal incidence in the thoracic and lumbar spine (20; 22% and 23; 26%, respectively). The most common site for exostoses associated with MHE was C2 (11; 19%; Figure 3).

Lesions associated with MHE also occurred most commonly in the cervical spine (32; 57%). However, thoracic lesions were more common than lumbar lesions (20; 36% and 2; 4%, respectively). The most common site for exostoses associated with MHE was C2 (11; 19%; Figure 3).

One hundred and six (88%) of the 121 exostoses whose origins were reported emanated from the posterior elements. The remaining 15 exostoses (12%) originated from the vertebral body. Solitary and MHE exostoses had similar distributions of vertebral origin.

<table>
<thead>
<tr>
<th>Total patients (male/female)</th>
<th>MHE</th>
<th>Solitary</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age, average years (range)</td>
<td>16.8* (7–34)</td>
<td>29.5 (22–52)</td>
<td>24.2 (7–52)</td>
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<tr>
<td>Palpable mass, total (%)</td>
<td>0</td>
<td>3 (43%)</td>
<td>3 (25%)</td>
</tr>
<tr>
<td>Neurological symptoms, total (%)†</td>
<td>3 (60%)</td>
<td>4 (57%)</td>
<td>7 (59%)</td>
</tr>
<tr>
<td>Intracanicular exostosis, total</td>
<td>5/0</td>
<td>3/4</td>
<td>6/4</td>
</tr>
</tbody>
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* P < 0.045 when compared to solitary.
† Symptoms or findings on physical examination consistent with neural structure compression.

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**Figure 1.** A-E, Seven-year-old male with MHE presented with intermittent clicking in his neck. The patient denied neck pain, weakness or other neurological symptoms. Physical examination showed normal, pain-free cervical range of motion. There were no palpable masses of the spine. Neurological examination showed no evidence of motor or sensory deficit. A and B, Anteroposterior and lateral cervical spine radiographs showed an osseous mass in the region of the C2 spinous process. C and D, Cervical spine CT and MRI revealed a 9 mm × 15 mm intracanicular exostosis arising from the anterior cortex of the central portion of the posterior arch of C2 occupying approximately one-half of the spinal canal with associated central spinal cord compression. E, Lateral cervical spine radiograph after tumor resection and spinal cord decompression through a posterior central laminectomy of C2 removing the mass in toto, followed by posterior arthrodesis from C2–C3. Histopathology was consistent with osteochondroma. Follow up at 7 years showed no evidence of tumor recurrence or symptoms.
Presenting symptoms were reported in 151 cases. Symptoms consistent with neural structure compression were reported in 105 patients (70%). Complaint of a mass (painful or nonpainful) was the second most common presenting symptom (26 patients; 17%). Eight (5%) patients presented with regional, nonradicular pain. Nine (7%) patients had other symptoms including dysphagia,4 cerebrovascular insufficiency,2 scoliosis (1), cough (1). There was 1 death. Spinal exostoses were diagnosed incidentally in 3 patients (2%).

The presenting symptoms were reported in 82 cases of cervical exostoses. Fifty-eight (70%) had neurological symptoms. A painful or painless mass was the most common non-neurological presenting symptom in the cervical spine (14 patients; 17%). The presenting symptom was recorded in 41 cases of thoracic exostoses. Thirty-five lesions (85%) were associated with neurological symptoms. The most common non-neurological symptom for patients with thoracic exostoses was a mass (9 patients; 7%). Twelve patients (46%) with lumbar exostoses presented with neurological symptoms. The most common non-neurological symptom for patients with lumbar exostoses was a mass (9 patients; 35%).

Solitary exostoses were associated with neurological symptoms in 26 patients (56%) with lesions in the cervical spine, 15 (75%) patients with thoracic exostoses, and 10 (43%) patients with lesions in the lumbar spine. Exostoses associated with MHE produced symptoms consistent with neural structure compression in 30 (94%) patients with exostoses in the cervical spine and 19 (95%) patients with thoracic lesions. Only 1 of 2 patients with a lumbar exostoses associated with MHE had neurological symptoms.

The mean age for all patients with spinal exostoses was 28.5 years (range, 2–69 years; Figure 4). The mean age of patients with MHE was 21.8 years (range, 7–48 years). The mean age of patients with solitary lesions was 32.8 years (range, 2 to 69 years). Patients with MHE...
were significantly younger than patients with solitary lesions ($P < 0.000001$). The mean age for all patients with neurological symptoms was 29.7 years (range, 2–68 years). The mean age of patients with MHE presenting with neurological symptoms was 22.3 years (range, 7–46 years) compared with 36.6 years (range, 2–68 years) for solitary lesions. Patients with MHE and neurological symptoms were significantly younger than patients with solitary lesions and neurological symptoms ($P < 0.000001$).

**Discussion**

Less than 10% of primary bone tumors occur in the spine.² Although, osteochondromas are the most common benign skeletal tumor, reportedly <3% occur in the spine.⁴,⁵¹,⁶⁰ The occurrence of spinal exostosis is likely under reported in relation to the true incidence because most are asymptomatic.¹,⁷¹ This study provides the largest clinical series of spinal exostoses from a single institution.

Conflicting reports exist as to whether solitary exostosis or lesions associated with MHE more commonly affect the spine. Roblot et al⁶⁰ reported that 72% (66 of 91 lesions) published cases of spinal exostoses were solitary. Similarly, Albrecht et al and Khosla et al reported that the solitary spinal exostoses were most common, composing 74% (72 of 97 lesions) and 75% (96 of 128 lesions) of cases reported, respectively.¹,⁹ However, other studies have reported a slightly increased incidence of spinal exostoses associated with MHE.¹⁸,²⁵,⁵⁵,⁶⁵ Our results indicate that the majority of spinal exostoses are solitary.

Although primary tumors of the cervical spine are uncommon, the majority of lesions treated in our series and identified in the literature review were located in the upper cervical spine, which is consistent with previous reports.¹,⁷,⁹,¹³,¹⁷,⁴⁷,⁴⁹,⁶⁸ Albrecht et al¹ reported 50% of lesions, both solitary and associated with MHE, were located in the cervical spine. Roblot et al⁶⁰ found that 36% of 92 lesions reported originated in the cervical spine compared with 29% and 28%, arising in the thoracic and lumbar spine, respectively. The propensity for exostoses to develop in the cervical spine has been attributed to the mobility of the cervical spine. Exostoses are classified as benign bone tumors; however, despite an approximately 2% incidence of malignant transformation, they are not true neoplasms. Exostoses are thought to develop from an epiphyseal growth disorder at the perichondral ring of Lacroix with lateral displacement of a portion of the physis through the perichondral fibrous ring resulting in subsequent aberrant epiphyseal development and growth at right angles to the long axis of the bone.³ The young age at diagnosis and the histopathology support this theory. Additionally, the mobility of the cervical spine may be associated with greater vertebral stress and microtrauma with consequent displacement of a portion of phyleal cartilage and exostosis formation, accounting for the greater incidence of these lesions in the cervical spine.¹,³

A number of previous studies have indicated that spinal exostoses are rarely associated with symptoms consistent with spinal cord or nerve root compression, reporting an incidence of 0.5 to 1% ⁷,¹³,³⁸,⁵¹ However, the majority of the patients in our series and cases identified in the literature had presenting symptoms consistent with neural structure compression. Additionally, several reports have found a higher incidence of symptomatic neural structure compression with lesions associated with MHE, especially in young patients, which is consistent with our findings.¹,⁶,⁹,⁴⁶,⁶⁰

O’Brien et al⁴ emphasized that intraspinal exostoses causing spinal cord compression must be excised, as the recovery of neurological function after surgical treatment is excellent, and the recurrence rate is low, whereas asymptomatic extraspinal lesions may be treated by observation. Albrecht et al³ also reported good results with surgical resection, finding that 89% of symptomatic patients treated operatively reported improvement of symptoms. All patients in our series surgically treated for intraspinal exostoses had eventual resolution of presenting symptoms after surgery. However, we have clinically followed two patients with intraspinal exostoses with no progression of symptoms.

Three complications occurred in our surgical treatment group. Two patients had symptomatic recurrence of exostoses after intralesional resection. Recurrence rates after resection of exostosis are approximately 2%.³,⁴¹,⁶² However, complete resection of the cartilaginous cap is necessary to prevent recurrence. This may necessitate wide laminectomy and partial facetectomy, and stabilization if the tumor originates from the facet or the lamina near the facet.¹,¹⁷ The third complication occurred as a result of lower extremity positioning after a prolonged excision and reconstruction of a presumed chondrosarcoma. Complete radiographic studies could have prevented all three complications. Spinal exostoses are best visualized on computed tomography (CT) than magnetic resonance imaging (MRI), as the bony nature

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**Figure 4.** Spinal exostoses patient age at presentation, literature review.
of the lesion is most clearly delineated. However, spinal cord compression is best visualized on MRI, as is the size of the cartilaginous cap, which assists in differentiating exostoses from a chondrosarcoma. We agree with previous recommendations that MRI and CT are necessary for preoperative assessment.

**Conclusions**

Spinal exostoses occur most commonly in the cervical spine. Neurological symptoms associated with spinal exostoses are more common than reported previously. Lesions associated with MHE tend to affect younger patients and have a higher incidence of neurological symptoms than solitary lesions. Preoperative radiographic evaluation should consist of MRI and CT to provide optimal information about the lesion and aid treatment options. Intralesional excision of the exostoses is associated with an unacceptably high recurrence rate and should be avoided.

**Key Points**

- Spinal exostoses are most commonly located in the upper cervical spine.
- Solitary spinal exostoses are more common than those associated with multiple hereditary exostoses.
- Compression of neural structures by spinal exostoses is more common than reported in the literature.
- Patients with spinal exostoses associated with multiple hereditary exostoses are younger and have a higher incidence of compression of neural structures than patients with solitary spinal exostoses.
- Intralesional excision has an unacceptably high rate of local recurrence.

**References**