

Original Investigation

Surgical Management of Osteoblastoma of the Spine: Case Series and Review of the Literature

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ABSTRACT

AIM: To describe the demographics, clinical and radiologic presentation, surgical considerations, and clinical outcomes associated with spinal osteoblastoma.

MATERIAL and METHODS: A spinal tumor database of 522 patients treated surgically at a single institution between January 2002 and June 2012 was analyzed and five patients with spinal osteoblastoma were identified and included in this study. Basic demographic and epidemiological data were recorded. Tumor characteristics, surgical parameters, and clinical follow-up data were noted.

RESULTS: The mean follow-up was 21.9 months. There were four males and one female, and the mean age at diagnosis was 28.4 years. There was a mean reported symptom length of 26.4 months prior to diagnosis. There were three cervical lesions, one lumbar lesion, and one sacrococcygeal lesion. One patient was Enneking stage III and four patients were Enneking stage II. Based on spinal instability neoplastic score criteria, two patients were stable and three patients were potentially unstable. Four patients had intralesional gross total resections and one patient had an en bloc marginal resection. All patients remained neurologically stable or improved postoperatively. A single patient had recurrence of a previously resected osteoid osteoma with progression to osteoblastoma.

CONCLUSION: Aggressive surgical resection of spinal osteoblastoma is suggested to minimize the risk of tumor recurrence, although this may still occur even with en bloc resection. Patients rarely present with preoperative spinal instability, but surgical fusion is often required due to removal of structural elements of the spine during resection. Of note, osteoid osteoma may progress to osteoblastoma in the spine despite prior resection.

KEYWORDS: Osteoblastoma, En bloc resection, Enneking stage

INTRODUCTION

Osteoblastoma is a rare primary tumor, accounting for around 1% of all bone tumors and up to 3% of benign bone tumors (13). They are located in the vertebral column in up to 40% of patients (18).

Surgical resection is the mainstay treatment for spinal osteoblastoma, with high recurrence rates with subtotal resection (12). Some have advocated intralesional gross total resections for Enneking stage II tumors and *en bloc* resections for Enneking stage III or "aggressive" variants (2, 12). As osteoblastomas typically involve the posterior elements of the spine (15), achieving a gross total resection or *en bloc* resection often involves removal of the facets or pedicles. The Spine Oncology Study Group (SOSG) recently developed the spinal instability neoplastic score (SINS) as a method to identify preoperative spinal instability in spinal oncology, though it has primarily been applied to metastatic disease (10). Although spinal insta-



Corresponding author: Benjamin D. ELDER E-mail: belder4@jhmi.edu bility is rarely encountered preoperatively, as is the case with many primary tumors of the spine, following excision of structural elements of the spine, spinal fusion may be required.

In this report, we present a retrospective review of five patients with surgically treated spinal osteoblastoma in order to describe the demographics, clinical and radiologic presentation, surgical considerations, and clinical outcomes associated these tumors. Additionally, a SINS analysis was undertaken to assess the degree of preoperative instability in these tumors. Additionally, we describe in detail the cases of an osteoid osteoma progressing to osteoblastoma and an osteoblastoma with a secondary aneurysmal bone cyst, which are two rarely reported events in the literature.

MATERIAL and METHODS

This study was approved by the Institutional Review Board at our center. 522 patients were operated on for primary or metastatic spinal tumors at our center between January 2002 and June 2012. Five patients underwent surgical treatment for osteoblastoma of the spine and were included in this study. The demographic information, preoperative characteristics, surgical treatment, and outcomes were retrospectively reviewed for each patient following protocols approved by the Institutional Review Board. Survival data was obtained from the Social Security Death Master File, accessioned online on 9/11/2014.

All patients included in the study presented with histologically confirmed osteoblastoma of the spine. Two patients were diagnosed with a computed tomography (CT)-guided biopsy, and five patients were diagnosed with open surgical resection. One patient with osteoid osteoma developed a recurrence and was diagnosed with osteoblastoma at the time of repeat resection.

For each patient, basic demographic and epidemiological data including age, gender, duration of symptoms preoperatively, pain at diagnosis, neurologic deficit at diagnosis, NSAID use prior to diagnosis, and scoliosis at time of diagnosis were recorded. Additionally, tumor characteristics including pathology, spinal location, Enneking stage (benign), Weinstein-Boriani-Biagini (WBB) staging (3), SINS (10), and tumor volume were obtained. Tumor volume was calculated using the approximated formula of an ellipsoid mass volume, [1/2 x height x width x depth]. Surgical characteristics were also recorded and included preoperative embolization, surgical approach, type of resection (intralesional versus en bloc), whether a fusion was performed, surgical duration, estimated blood loss (EBL), postoperative transfusion, length of hospital stay, postoperative complications and postoperative neurologic function. The EBL and transfusion information could not be located for one patient and the data was eliminated from the reported results for these parameters. Finally, follow-up data included duration of follow-up, tumor recurrence, neurologic function at last follow-up, mortality, and status at last follow-up.

RESULTS

There were five patients who were diagnosed with osteoblastoma. Table I displays the demographic information and preoperative characteristics of each patient. There were four males and one female diagnosed with osteoblastoma, and the mean age at diagnosis was 28.4 years (range 15-57 years).

All patients presented with regional pain at the time of diagnosis, but only 40% of the patients were using NSAIDs at the time of diagnosis. No patients presented with scoliosis. There was a mean reported symptom length of 26.4 months (range 6-60 months) prior to diagnosis. A single patient presented with a neurologic deficit preoperatively. This patient had a large sacrococcygeal osteoblastoma and presented with bowel, bladder, and sexual dysfunction, as well as left plantar flexion weakness (3/5 strength) (Figure 1A,B).

The tumor characteristics for each patient are found in Table II. There were three cervical lesions, one lumbar lesion, and one sacrococcygeal lesion. The cervical lesions all involved a single level, the lumbar lesion involved two levels, and the sacral lesion was multilevel. One patient was Enneking stage III and four patients were Enneking stage II. Based on SINS criteria, two patients were characterized as stable and three patients were characterized as potentially unstable. The mean tumor volume was 57.8 cc (range 2.2-265.5 cc).

| Patient # | Age at Diagnosis (yrs) | Gender | Length of Symptoms Preoperatively (months) | Pain at Diagnosis | Use of NSAIDs | Scoliosis at Diagnosis | Neurologic Deficit at Diagnosis |
|-----------|------------------------------|--------|--|----------------------|------------------|---------------------------|---------------------------------------|
| 1 | 57 | М | 24 | Yes | No | No | Yes* |
| 2 | 15 | F | 6 | Yes | No | No | No |
| 3 | 27 | М | 36 | Yes | No | No | No |
| 4 | 27 | М | 60 | Yes | Yes | No | No |
| 5 | 16 | М | 6 | Yes | Yes | No | No |

Table I: Demographics and Preoperative Characteristics of Patients

*Neurologic deficit included bowel, bladder, and sexual dysfunction, and left plantar flexion weakness (3/5 strength). **Abbreviations: #:** number; **yrs:** years; **NSAIDs:** non-steroidal anti-inflammatory drugs, **M:** Male, **F:** Female. Table II also lists the surgical approach and fusion characteristics. A posterior approach to the spine was used in all patients. A single patient had a preoperative embolization performed. Four patients had intralesional gross total resections and one patient had an en bloc marginal resection (sacrectomy) with sacrifice of the bilateral S2-5 nerve roots (Figure 1C). All patients had a posterior fusion though Patient 2 had an in situ non-instrumented fusion. For patient 1, an en bloc marginal resection was planned given concern for osteosarcoma and due to the apparent vascularity of the mass. The tumor appeared to involve S2 through the coccyx on imaging, and it was initially felt that an instrumented fusion would not be necessary. However, following osteotomies below the S1 nerve roots, it was apparent that the tumor had invaded the S1 vertebral body, requiring a more proximal cut between L5 and S1. Given that the S1-coccyx segments had been completely removed, it was felt that the patient would require lumbosacropelvic reconstruction with an instrumented fusion.

All patients remained neurologically stable or improved postoperatively. The mean estimate blood loss (EBL) was 2038 cc (range 300-6000 cc), and two patients required blood transfusions during the hospitalization. The mean hospital length of stay was 6.4 days (range 2-19 days).

No patients received postoperative chemotherapy or radiation. A single patient in the osteoblastoma series (20%) developed a postoperative complication. The patient with the large sacrococcygeal osteoblastoma developed a wound dehiscence requiring surgical revision on postoperative day 11.

The mean length of follow-up was 21.9 months. The patient with the large sacrococcygeal osteoblastoma developed a local recurrence 43 months postoperatively, with a 25.2 cc lesion noted at L5-S1 on the left (Figure 1D, E). A CT-guided biopsy was performed due to concern for osteosarcoma, but osteoblastoma was confirmed on the biopsy. A preoperative embolization was performed, followed by a repeat intralesional gross total resection with removal of the medial aspect of



Figure 1: Representative images for patient 1. Preoperative **A**) 3-D CT reconstruction and **B**) axial CT demonstrating large sacrococcygeal lytic, expansile lesion with cortical destruction, internal calcifications, and associated soft tissue component, depicted by the arrowheads. An *en bloc* marginal resection with a sacrectomy was performed and reconstruction with L3-pelvis instrumentation and a transiliac strut allograft was performed **(C)**. A recurrent lesion was noted 43 months later, with involvement of the remaining sacrum and at L5, depicted by the arrowheads **(D)**. A solid posterolateral fusion was noted **(E)**. Following a preoperative embolization, an intralesional gross total resection was performed and the medial aspect of the transiliac strut allograft was removed **(F)**. Pathology confirmed recurrent osteoblastoma.

| Patien # | t Pathology | Location | Enneking Stage (Benign) | SINS | WBB Class | Tumor Volume (cc) | Preoperative Embolization | Approach/ Type of Resection/ Nerve Root Sacrifice | Fusion? | Postoperative Neuro Status |
|---------------------------------|--|------------------------------------|----------------------------------|-----------------------|-------------------------------------|------------------------------|--|--|---|-------------------------------|
| - | Osteoblastoma, aggressive variant | S1-coccyx | ≡ | თ | AII A-D | 265.5 | No | Posterior sacrectomy/ En bloc marginal/ bilateral S2-5 roots | Yes, L3-pelvis, trans-iliac strut graft | Bowel/bladder dysfunction |
| N | Osteoblastoma, secondary aneurysmal bone cyst | C2 | = | 7 | 12,1,2,3 B-D | 7.3 | N | Posterior/ Intralesional GTR/No | Yes, in situ | Intact |
| e | Osteoblastoma | C7 | = | 7 | 10,11,12 B-C | 5.2 | Yes | Posterior/ Intralesional GTR/No | Yes, C5-T1 PCF | Intact |
| 4 | Osteoblastoma | L3-4 | = | 9 | 1,2,3 B-C | 8.8 | No | Posterior/ Intralesional GTR/No | Yes, L2-4 PLF | Intact |
| с, | Osteoid osteoma, osteoblastoma* | C2 | = | Q | 2,3,4 B-C | 2.2 | No | Posterior/ Intralesional GTR/No | Yes, C2-3 PCF | Intact |
| *Patient Abbrevi PLF: po: | 5 was initially diagnosed wi ations: #: number, SINS: sterolateral fusion. | :h osteoid oste spinal instabil | eoma, and a re ity neoplastic | ecurrence score, V | e one year lat VBB: Weins | ter was four tein, Boriar | nd to be osteoblas: ii, Biagini classific | toma. ation, GTR: gross total re. | section, PCF: poste | rior cervical fusion, |

the transiliac strut allograft (Figure 1F). He ultimately expired 5 months after the recurrence, but the cause of death was not known. Other than the single patient death, the remaining four patients were alive with no systemic evidence of local or systemic disease at last follow-up.

Patient 2 presented with neck pain and suspected to have an aneurysmal bone cyst (Figures 2A-C). A resection of the lamina and left lateral mass was performed (Figure 2D) and pathology demonstrated osteoblastoma with secondary aneurysmal bone cyst. Patient 5 was found to have a lesion in the left C2 lamina and lateral mass (Figure 3A, B), pathology demonstrated osteoid osteoma at the initial resection, at which time a C2-3 fusion was also performed (Figure 3C, D). However, he developed a recurrence 13 months postoperatively (Figure 3E-G), and a repeat resection was performed (Figure 3H). The recurrent lesion had transitioned to an osteoblastoma, confirmed by pathology. The original pathology was reanalyzed and still found to be consistent with osteoid osteoma suggesting progression to osteoblastoma.

DISCUSSION

Although several other larger series of spinal osteoblastomas have been reported (2, 8, 16, 17, 20, 21, 24), to the best of our knowledge, this is the first primary tumor series to assess preoperative spinal stability using the SINS criteria. Additionally, this series includes the first case to demonstrate pathology-confirmed progression of osteoid osteoma to osteoblastoma in the spine. Finally, an osteoblastoma with a secondary aneurysmal bone cyst is reported, which has been rarely described in the spinal clinical literature previously.

As described above, this manuscript presents the first case of pathology-confirmed progression of osteoid osteoma to osteoblastoma in the spine. Osteoid osteoma and osteoblastoma were originally felt to be variations, predominantly of size, of the same tumor type. However, current clinical consensus is that these are two separate entities with different clinical and radiologic characteristics as well as different risks of progression (14, 15). Two cases of progression of spinal osteoid osteoma to osteoblastoma were previously reported, though no pathology-confirmed diagnosis of osteoid osteoma was obtained. For instance, Bruneau et al. (4) reported the case of a 25-year-old patient with an 8 mm lesion in the left C1 lateral mass, concerning for osteoid osteoma. The patient initially refused surgery, but seven years later, he had increased pain and the lesion had grown to 16 mm. Surgical resection was performed with pathology demonstrating osteoblastoma. A similar case was reported in a 16-year-old patient in which a 6 mm lesion was identified in the right C6 pedicle concerning for osteoid osteoma. The patient originally refused surgery and one year later the lesion had increased in size and was found to be osteoblastoma following surgical resection (5). As such, there is controversy as to whether these "progressions" truly represent pathologic change, or if the original smaller lesions were simply incorrectly assumed to be osteoid osteomas without pathology confirmation (6, 19). The results reported here are interesting as biopsy samples were obtained at each resection and reviewed by the same senior musculoskeletal

Table II: Tumor Characteristics And Surgical Parameters

pathologist. Additionally, after the recurrent specimen was felt to be osteoblastoma, the original specimen was re-reviewed and still felt to be osteoid osteoma. However, this has previously been reported in the appendicular skeleton, as Bettelli et al. reported the case of a 24-year-old patient diagnosed with a femoral osteoid osteoma based on pathology, and later diagnosed with osteoblastoma following surgical resection for a recurrence 8 months later (1). Patient 2 in this series was found to have an osteoblastoma of C2 with a secondary aneurysmal bone cyst, which has been rarely reported in the literature. Although a retrospective review of 55 patients, with osteoblastoma in any region of the skeleton, reported a 14.5% rate of secondary aneurysmal bone cysts (7), there have otherwise only been anecdotal reports in the literature, with rare reports of this dual pathology in the spine. For instance, Tarantino et al. (25) presented a



Figure 2: Representative images for patient 2. Preoperative A) sagittal, B) coronal, and C) axial CT images demonstrate C2 lesion (arrows) in left lamina and lateral mass suspicious for aneurysmal bone cyst with an expansile osteolytic appearance with thin sclerotic margins. D) Resection of the lamina and lateral mass with curettage was undertaken and an *in situ* non-instrumented was performed.



Figure 3: Representative images for patient 3. Preoperative axial **A**) T2-weighted MRI and **B**) CT scan images demonstrate hypointense lesion extending to the left pedicle and lateral mass of C2 with hyperintense signal in the lamina (A), and a well-circumscribed lesion noted on CT (B), as depicted by the arrows. An intralesional gross total resection was performed with removal of the lamina, lateral mass, and pedicle on the left and skeletonization of the vertebral artery. A C2-3 posterior fusion was performed with a translaminar C2 screw on the left (**C-D**). A recurrent lesion was noted 13 months later surrounding the vertebral artery (**F-G**), depicted by the arrows, and solid fusion was noted on the right side (**E**). A repeat resection was carried out with further skeletonization of the left vertebral artery and removal of the C2-3 instrumentation on the left (**H**).

15-year-old patient with an osteoblastoma of the odontoid process with a secondary aneurysmal bone cyst, while Ramme and Smucker reported a 19-year-old patient with an osteoblastoma and secondary aneurysmal bone cyst in the posterior elements of C2 (22). Additionally, osteoblastoma with secondary aneurysmal bone cyst has been reported in the sacrum (26) and the posterior cranial fossa (11).

Histologically, they are composed of osteoid and woven bone, and frequently contain highly vascular regions (7) and numerous osteoblasts. They most commonly affect the thoracolumbar spine (15, 23), although frequent involvement of the cervical spine has also been reported (20). They have a greater incidence in males and are most commonly diagnosed in the second decade of life, with a prolonged duration of symptoms prior to presentation of up to an average of 20 months (12, 15). Patients typically present with localized pain in the neck or back, the pain may increase nocturnally. It typically improves with aspirin or non-steroidal antiinflammatory drugs (NSAIDs).

The majority of the demographic and clinical results of our series were not substantially different than those of prior reported series (2, 8, 16, 17, 20, 21, 24). According to a recent review (15), there is typically a male predominance, and this was observed in our series at a rate of 4:1. Additionally, the mean duration of symptoms prior to diagnosis was described as approximately 20 months (15), similar to the 26 months observed in this series, with 40% of the patients controlling their pain with NSAIDs. Additionally, the rate of neurologic deficit at presentation has ranged between 25-70% (15), with a 20% rate in this series, though only observed in the sacral lesion. In a series of 18 patients with sacral osteoblastomas, 4 patients (22%) were found to have neurologic deficits.

However, the mean age of diagnosis, at 28.4 years, with a patient in the sixth decade of life, was substantially older than has been reported in previous series, with the most common age of presentation in the second decade (15). Only one other series identified an older patient (60 years) with an average of 23 years of age (2), and most patients ranged between 15-30 years of age. The older age of the patients as well as cervical predominance in our series may explain why no patients presented with a deformity. Rates of scoliosis typically range between 4.5-100% at diagnosis, according to a recent review (15), though this is typically an issue more with thoracolumbar lesions in juvenile patients.

An area of controversy following resection of spinal osteoblastomas is whether spinal fusion should be performed following tumor resection. For instance, there is concern for postlaminectomy kyphosis especially after resection of C2 lesions in the posterior elements. However, the precise definition of spinal instability has been unclear in the clinical literature. The SOSG defined spinal instability as "a loss of spinal integrity as a result of a neoplastic process that is associated with movement-related pain, symptomatic or progressive deformity, and/or neural compromise under physiologic loads (10). The SOSG recently developed the SINS as a method to identify preoperative spinal instability in spinal oncology, though it has primarily been applied to metastatic disease. Based on SINS criteria, three patients were identified as potentially unstable and two patients were identified as stable, and this was the first time in the literature that a SINS analysis was applied to a primary spinal tumor. Although spinal instability is rarely encountered preoperatively, as is the case with many primary tumors of the spine, following excision of structural elements of the spine, spinal fusion may be required. The resections of the cervical and lumbar lesions in this series all involved removal of the facet joints or pedicles to allow for gross total resection. As such, all patients in the series were fused, although one patient had a non-instrumented fusion. The pediatric patients require further scrutiny, as they are at greater risk of postlaminectomy kyphosis (9), but there may be reluctance to limit the mobility of these younger patients with spinal fusion. Therefore, great care should be taken during surgical resection to minimize disruption of the facet joints and attempt to reattach the paraspinal muscles to the posterior elements, if fusion is not pursued.

The type of surgical treatment has also been debated and correlated with surgical grading (2, 12, 27). In our series, the patient with the aggressive variant. Enneking stage III, was treated with a marginal en bloc resection, while the remaining Enneking stage II patients were treated with intralesional gross total resections. According to a recent systematic review by Harrop et al. (12), the recurrence rates for intralesional subtotal resections or biopsies, marginal gross total resections, or en bloc resections were 93%, 15%, and 20% respectively. Additionally, the "aggressive" or Enneking stage III lesions have been reported to recur in up to 50% with subtotal resections. Therefore, they recommended intralesional gross total resection for Enneking stage II lesions, and en bloc resections for Enneking stage III lesions when "anatomically feasible," and this was the treatment strategy pursued in our series. Yin et al. (27) also identified tumor size >3 cm as predictive of increased risk of tumor recurrence on multivariate analysis. It is also important to at least obtain a biopsy of any recurrence as sarcomatous degeneration to osteosarcoma has been described (12).

With a recurrent lesion, as observed in the sacral mass in this series, radiation was not pursued, as it has not been shown to decrease the risk of recurrence following subtotal resection (12). Surgical resection remains the mainstay treatment. However, radiation can potentially be reserved as a treatment option for Enneking stage III tumors if there is no feasible surgical option (2). Similarly, a limited, if any, role for chemotherapy has been identified in the treatment of osteoblastoma.

A major limitation of this study is the retrospective nature with a small number of patients, as well as a single cohort with lack of a control group. However, even in this small series, rarely reported events in the literature including progression of osteoid osteoma to osteoblastoma and an osteoblastoma with a secondary aneurysmal bone cyst merit reporting.

Aggressive surgical resection should be considered for osteoblastoma to minimize the risk of tumor recurrence, although this may still occur despite *en bloc* resection. Patients rarely present with preoperative spinal instability, but surgical fusion is often required due to removal of structural elements of the spine during resection. Of note, osteoid osteoma may progress to osteoblastoma in the spine despite prior resection.

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