Treatment of osteoblastoma at C7: a multidisciplinary approach. A case report and review of the literature

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Abstract Osteoblastoma is a rare benign bone tumor that presents with back pain and occurs in the spine approximately 40% of the time. The time from onset of symptoms to diagnosis is typically several months because it is a rare entity and radiographic studies are often negative early in the course of the disease. These highly vascular and locally aggressive tumors require complete and precise resection. The patient presented is a 15-year-old boy with a 14-month history of right-sided neck and shoulder pain. Computerized tomography and magnetic resonance imaging demonstrated a lesion in the posterior elements of C7 which extended through the pedicle and into the body. Preoperative angiography confirmed a hypervascular lesion which was successfully embolized. He subsequently underwent piecemeal tumor resection and instrumented fusion. Immediate postoperative imaging demonstrated

complete resection. At 18 months follow up the patient has maintained resolution of preoperative symptoms and demonstrates evidence of solid fusion on CT. This multidisciplinary approach markedly decreased blood loss and improved visualization to help achieve complete surgical resection and resolution of clinical symptoms.

Keywords Osteoblastoma · Embolization · Spinal fusion · Spinal tumor · CT · Bone scan

Introduction

Osteoblastoma is a rare, benign bone tumor that occurs in the spine approximately 32–46% of the time, has a 2:1 male to female ratio, and usually presents with pain in the 2nd or 3rd decades of life [10, 24]. Osteoblastoma is histologically similar to osteoid osteoma, but osteoblastoma occurs in slightly older patients, has a greater propensity for the spine and is more biologically aggressive, often eroding through spinal cortical bone and creating large soft tissue masses [1, 11, 13, 15]. Some distinguish the two based on size with lesions less than 1.5 cm classified as osteoid osteoma, and those larger than 1.5 cm considered osteoblastoma [13].

Patients typically present with progressive focal or radicular pain exacerbated by movement. These symptoms are often present for greater than 12 months prior to diagnosis, and many patients will go on to manifest neurological sequelae or a painful scoliosis [1, 15–17, 19]. Delayed diagnosis occurs because initial symptoms are often nonspecific and osteoblastoma accounts for less than 1% of all bone tumors [11]. Radiographically the tumor is often radiolucent, almost always involves the posterior elements and the adjacent bone may or may not exhibit

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reactive sclerosis [1, 13, 22]. Vertebral body involvement rarely occurs alone but may be involved via tumor extension through the pedicle [11, 22, 24]. When these lesions are small and do not involve the transverse or spinous processes, they can be missed on plain films [13, 25]. With the utilization of CT scans in the workup of back pain and the utilization of bone scans in painful scoliosis, the sensitivity for detecting these tumors will increase substantially. The treatment goal is complete surgical resection which avoids radiotherapy and decreases the risk of recurrence. However, complete surgical resection is often difficult in these highly vascular tumors.

In this report we describe the treatment of a 15-year-old boy who presented with a C7 osteoblastoma. The patient underwent preoperative embolization followed by piecemeal tumor resection and spinal fixation. This treatment paradigm markedly decreased blood loss and improved visualization to help achieve complete surgical resection and resolution of clinical symptoms.

Case report

History

A 15-year-old boy presented with a 14-month history of severe right-sided neck and shoulder pain. The pain was constant and exacerbated when abducting his right upper extremity. Although more characteristic of osteoid osteoma, the pain was described as worse at night and would frequently wake him from sleep. Medications, including aspirin, offered little relief. He only attained relief by keeping his arm adducted. On examination, his right deltoid muscle was severely atrophied, and he had point tenderness in the midline in his lower cervical spine. His motor exam was severely limited by pain, but best effort showed 2/5 power in his deltoid and 4/5 power in his triceps and hand intrinsics. The rest of his motor exam was unremarkable. Radiographic work up included a computerized tomography (CT) (Fig. 1), and magnetic resonance imaging (MRI). The CT showed an expansile lytic lesion

centered in the right C7 pedicle causing moderate narrowing of the spinal canal and effacement of the C6/7 and C7/T1 neural foramen.

Preoperative embolization and operative procedure

Osteoblastomas are very vascular tumors and the patient underwent a preoperative angiogram and embolization one day prior to surgery. A right subclavian angiogram demonstrated a hypervascular tumor at the level of C7 with arterial feeders arising mostly from the thyrocervical trunk (Fig. 2a). A micro catheter was used to select the supra-scapular and thyrocervical branches and polyvinyl alcohol particles (PVA) of 150–250 microns were used to embolize using fluoroscopic guidance. In addition, platinum 0.018 in. straight and helical micro coils were deployed in branches of the thyrocervical trunk to achieve complete stasis. A post embolization angiogram (Fig. 2b) demonstrated significant reduction in tumor vascularity. The feeders from the left thyrocervical trunk were then embolized (Fig. 2c, d). Smaller branches were visualized of the vertebral arteries that were not embolized.

Post embolization the patient had a substantial increase in his neck and shoulder pain secondary to tumor ischemia that required high dose intravenous fentanyl. On the following day the patient underwent a C7 laminectomy, right C6/7 and a C7/T1 facetectomy with removal of the pedicle and a portion of the C7 body. The tumor was adherent to the dura and required careful dissection of the C8 nerve root. During the final stages of the resection a large embolized artery was visualized. Tumor resection was followed by a C5-T2 posterolateral instrumented fusion (Fig. 3a–d) and the patient was then placed in a Miami J collar (Jerome Medical, Moorestown, NJ). The total blood loss for the procedure was 600 mL, and the patient did not require transfusion.

Postoperative course

A CT scan was obtained post operative day 1 and demonstrated a complete surgical resection. Four weeks after

Fig. 1 Sagittal (a), axial (b), and coronal (c) CT images demonstrating an expansile, lytic lesion (*broad arrow*) at the level of the right C7 pedicle with moderate spinal canal impingement and foraminal narrowing

Fig. 2 Digital subtraction angiography of (a) the right thyrocervical trunk (*arrow head*) demonstrating hypervascularity and tumor blush (*broad arrow*) at the C7 pedicle level. **b** Post coil (*thin arrow*) and PVA embolization demonstrate almost complete absence of tumor vascularity. **c** The left thyrocervical trunk (*arrow head*) showing tumor blush (*broad arrow*) from the contralateral side. **d** Post coil (*thin arrow*) and PVA embolization demonstrate almost complete absence of tumor vascularity

Fig. 3 Sagittal (a), axial (b), and coronal (c) CT images after right C6/7, C7/T1 fascetectomies and partial C7 corpectomy show complete resection of osteoblastoma and posterolateral fusion. **d** Plain

anterior/posterior X-ray of the cervicothoracic fusion. *Small arrows* are pointing to embolization coils

surgery a plain film showed that the hardware was unchanged and the Miami J collar was discontinued. At his 3 month follow up the patient was pain free, with increased muscle bulk in his right shoulder girdle and 4/5 deltoid motor strength and 5/5 power in his triceps and hand intrinsics. At his six month follow up he was pain free with normal muscle bulk and 5/5 strength in his deltoid and no evidence of recurrence. At 18 months follow up the patient has complete resolution of preoperative symptoms and CT imaging has revealed solid fusion with no evidence of

recurrence (Fig. 4a, b). The patient will receive annual CT scans for the next several years to screen for recurrence.

Discussion

Osteoid osteomas and osteoblastomas were first described by Jaffe in 1935[8, 9]. These tumors arise in both cortical and cancellous bone and are highly vascular [11, 25]. 20% of osteoid osteomas and 40% of osteoblastomas are found

Fig. 4 Sagittal reconstruction CT at 18 months follow up demonstrating solid fusion and no evidence of tumor recurrence

in the spine [7]. Histologically, the two are similar, but pathologically osteoblastomas demonstrate more aggressive characteristics than osteoid osteomas, often invading extraskeletal tissue and have been reported to transform into osteosarcoma [7, 13, 20]. Because of this locally aggressive behavior, neurological deficit is seen in 43–70% of patients with vertebral osteoblastoma [1, 15, 25].

The most common presentation for both entities is back pain. For osteoid osteomas this pain is usually more severe at night and is relieved with aspirin. In contrast, the pain with osteoblastoma is classically not as severe at night and does not respond as well to aspirin [25]. These patients are more likely to present with scoliosis or neurological deficit. In a report by Boriani et al., 40% of patients (12/30) presented with painful scoliosis [1]. Ozaki et al. reported scoliosis in 77% (17/22) of their patients, with an improvement seen in 16 of the 17 after tumor resection [15]. The tumors are found on the concavity of the scoliosis with a proposed etiology being muscle spasms secondary to inflammatory factors [18].

Imaging modalities used to diagnose osteoid osteoma and osteoblastoma are primarily CT and MRI. Additional studies include plain X-ray and bone scan. Plain X-rays are more useful in the detection of osteoblastomas, but may miss small neoplasms [1, 13, 25]. CT scan will delineate the location and osseous involvement of the mass and MRI will provide appreciation of the effect on soft tissues and neural elements [15]. Recently, Hosalkar et al. [6] reported the alarmingly high rate of misdiagnosis of osteoid osteoma based on MRI alone (81% misdiagnosed). For greater sensitivity technetium bone scanning may be utilized [16]. If the diagnosis is uncertain or the CT and MRI are negative and there is clinical suspicion for an osteoblastoma, we obtain a technetium bone scan.

This case illustrates the successful management of a patient with an osteoblastoma involving the C7 posterior elements and body utilizing preoperative embolization followed by surgical resection and fixation. In 1979, Dick et al. [3], reported on the use of embolization as an adjunctive therapy for benign bone tumors. This treatment modality has been successfully applied to highly vascular spinal tumors including spinal metastasis and aneurysmal bone cysts [4, 5]. Most recent case series on osteoblastoma acknowledge the potential utility of preoperative embolization, however, only a few reports document the efficacy for osteoblastomas of the spine. Silva and Brunelle [21] described two patients whom they successfully embolized prior to surgical resection. More recently, Meyer et al. reported a 15-year-old girl who underwent successful preoperative embolization for a thoracic osteoblastoma with a secondary aneurysmal bone cyst. There were no signs of recurrence at 18 months follow up [14]. In 2006 Trubenbach et al. [23] reported on three patients, two were recurrence free at one and two years, respectively. One patient experienced a local recurrence at 6 months and was subsequently treated with repeat resection and salvage radiation therapy. In 2007 Denaro et al. [2] described 9 cases of cervical osteoblastoma treated with preoperative embolization, with no signs of recurrence at an average of 39 months follow up.

In summary, the highly vascular nature of osteoblastomas and the importance of complete resection strongly favor preoperative embolization. Without it, the recurrence rate has been reported between 9.8–15% [1, 11, 25]. Radiotherapy can be utilized with incomplete resections or recurrences, but carries potential risks including that of malignant transformation into post radiation sarcoma [12]. The number of documented preoperative embolizations in

the treatment of spinal osteoblastomas remains small. Consequently, assessment of statistical significance with regard to tumor recurrence and long term outcome when compared with resection alone has not been possible. However, a growing number of reports, including our experience exemplify the utility of preoperative embolization. Anecdotally, only one report of local recurrence in 16 cases (6.25%) has been described [2, 14, 19, 21]. The use of preoperative embolization in our patient decreased bleeding and improved visualization. Enhanced visualization allowed careful, meticulous, and safe tumor curettage leading to a complete resection thereby lowering his risk of recurrence and the need for subsequent surgeries or radiotherapy.

Conflict of interest statement None of the authors has any potential conflict of interest.

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