RESEARCH

The Results of Wide Resection in Sacral Osteoblastoma

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Abstract: The aim of this study was assess the results without instrumentation the clinical findings and treatment outcomes of wide resection in sacral osteoblastoma. A retrospective review was conducted in the hospital archive from 1983 to 2017. As a result of the examination, 238 osteoid osteoma and osteoblastoma patients were found. Osteoid osteoma was present in 210 (88.2%) patients and osteoblastoma was present in 28 (11.7%) patients. Five patients who had been operated for osteoblastoma of the sacrum were retrospectively evaluated. Preoperative and postoperative plain radiographs, MR, CT and scintigraphy scans of all patients were taken. The lesion was located at the S4-S5 vertebrae in two patients, at the S2-S3 in one, at the S1in one and at the S4 in the other. Diagnoses were made by either open or closed biopsy. The patients were treated with wide resection. The mean follow-up period was 31.6 (range: 18 to 50) months. One patient developed a superficial wound infection. No local recurrence was observed. All patients were pain-free in the postoperative period. Wide resection of sacral osteoblastoma proved successful results in the short follow-up period of 31.6 months, with no recurrence.

INTRODUCTION

Osteoblastoma is encountered in 1% of all bone tumors. In addition, it was found in 3% among all benign primary bone tumors[1,2]. The incidence of the neoplasm peaks in the second decade of life and 90% of these tumors are diagnosed before the third decade Its incidence in males is double that of females[3]. The tumor may have a vascular osteoid nature or may form bones with a vast number of osteoblastic cells[4].

Forty percent of osteoblastomas are located in the spine[5,6] . The sacrum is a rare location for the osteoblastoma to localize[7,8]. Only 7 to 17% of the primary sacral tumors are osteoblastic. Tumors in the sacrum may present with back pain, scoliosis and other neurological symptoms[5,9]. Osteoblastoma is a much-debated topic in the literature. One study suggests that differentiating between osteoblastoma and osteoblastic osteosarcoma is challenging[10.] Reported rates of local recurrence varies between 10 and 67%[10.] This situation may be related to the challenge in differentiating osteoblastoma from osteosarcoma or to the choice of treatment.

Varga[3] stated that nocturnal pain in the lower back or sacrum may be a warning symptom [3,11]. However, the authors also reported that sacral tumors might be diagnosed as non-specific lower back pain or disc hernia, due to difficulties in the evaluation of the radiographs. Plain radiographs of the sacrum often lead to delayed diagnosis, as the evaluation of these graphs are challenging and lack an established diagnostic method [3,11].

The optimal treatment method of sacral osteoblastoma is controversial due to the lesion's rare occurrence and challenges in diagnosis. The tumor may be misleading and can be diagnosed wrongfully. The literature reports of several treatment methods; radiotherapy (RT), intralesional surgery, intralesional surgery and RT, intralesional surgery and local adjuvants (phenol or cryotherapy techniques), and wide marginal resection[12-14].

The aim of our study was to evaluate the postoperative success of wide resection in sacral osteoblastoma in terms of treatment and recurrence.

A retrospective review was conducted in the hospital archive from 1983 to 2017. As a result of the examination, 238 osteoid osteoma and osteoblastoma patients were found. Osteoid osteoma was

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present in 210 (88.2%) patients and osteoblastoma was present in 28 (11.7%) patients. Our institutional database was queried and four patients [4 males (80%), 1 female (20%); mean age: 14 (range: 9 to 19) years] who had been operated with wide resection due to sacral osteoblastomas and followed up between the years 1983 and 2017 were included in the study. The mean follow-up period was 31.6 (range: 18 to 50) months. Medical data from the hospital records, surgical records, pathology reports, clinical notes and direct radiographs, magnetic resonance imaging (MRI), computed tomography (CT) and scintigraphy reports comprised our patient data. Staging was done based on the Enneking classification [15] for benign bone tumors and all lesions were classified as Stage 2.

Preoperative evaluations were made using standard radiographs, bone scintigraphs (BS), CT and MR images. Open or closed biopsies were performed before surgery under fluoroscopic guidance. The posterior approach was employed in all patients. None of the patients underwent embolization for preoperative bleeding control.

The patients were given non-steroidal anti-inflammatory drugs (NSAIDs) for three days in the postoperative early period. All patients were mobilized on Day 2.

Routine checks were made for all patients in the postoperative follow-up period. The patients were called for follow-up visits every six months in the first two years and once a year thereafter, and they were examined for local recurrence and complications.

The time between the time of diagnosis and time of the final follow-up was considered survival without progression.

RESULTS

The patients have had most of their symptoms before they presented to our clinic. The mean time to diagnosis was 9.2 (range: 2 to 16) months. All patients had the same symptoms of pain in the lower back and the sacrum and relief of pain with the use of NSAIDs. Postoperative findings of neurological examination of all patients were normal and no mass could be felt during physical examination. Local tenderness upon palpation was observed. The lesions were located at the level of S4-S5 vertebrae in two patients, at the level of S2-S3 in one and at the level of S4 in the other (Table 1) One of our patients developed superficial infection in the postoperative period, which was treated with antibiotics. The superficial infection was seen in our patient who was performed open biopsy (Case 4). All patients were treated with wide resection alone. (Figure 1, 2) This was the patient whose diagnosis was made after open biopsy and who developed superficial infection. The mean follow-up period of our patients was 31.6 months and no recurrence was seen within the follow-up period. In all patients, the lesion was reached via an incision from the posterior and no problems were encountered during the removal of the tumor via wide resection. As discussed and agreed by the tumor council before surgery, none of the patients underwent embolization for bleeding control due to low vascularity of the pathological tissue. Pain resolved in all patients in the postoperative period.

Table 1. Demographic and Clinical Characteristics in 5 Patients (Yr: Years; Mo: Months)

	Sex, Age (yr)	Enneking stage	Tumor Volume	Tumor Location	Abnormal Neurological Findings	Previous Treatment	Surgical Complication	Follow-up (yr, mo)
1	M,15	2	22*22*16 mm	S4-S5	No	No	No	2 yr 8 mo
2	M,9	2	25*24*16 mm	S4	No	No	No	2 yr 2 mo
3	M,19	2	23*18*16 mm	S2-S3	No	No	No	2 yr 8 mo
4	F,14	2	48*32*18 mm	S4-S5	No	No	Yes (superficial infection)	4 yr 2 mo
5	M,13	2	21*19*20	S1	No	No	No	1 yr 6 no

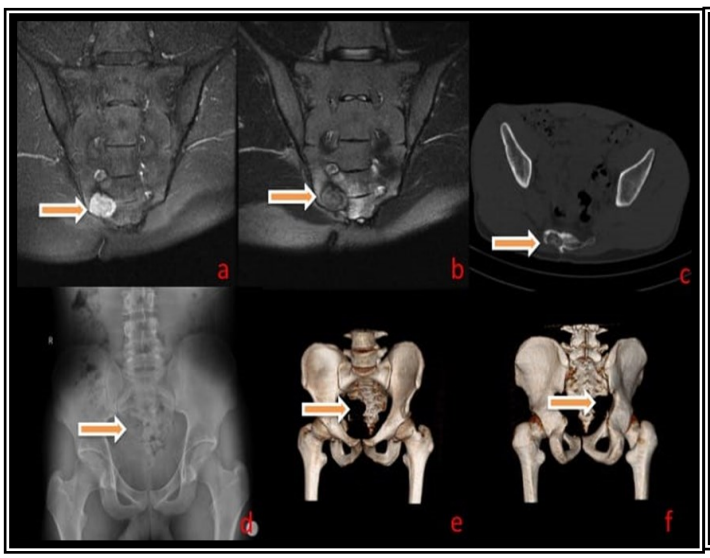

Figure 1. a) Preoperative MRI, b) Preoperative MRI, c) Preoperative CT, d) Preoperative standard x-ray, e) Postoperative CT AP, f) Postoperative CT PA

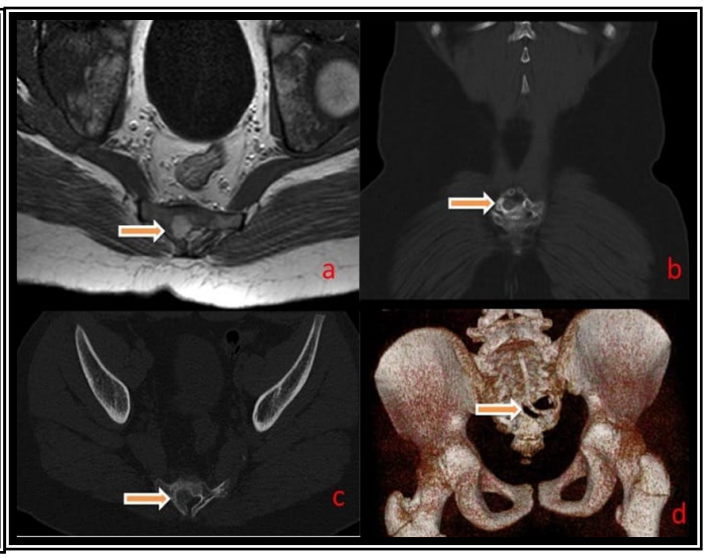

Figure 2. a) Preoperative MRI, b) Preoperative MRI, c) Preoperative CT, d) Postopertive CT

DISCUSSION

As a result of the literature review, 4 articles were obtained. These articles with publication dates between 1997-2017 were reviewed. Publications with relatively large patient series on sacral osteoblastoma (with two and more patients) were included in the literature analysis, whereas the other case reports (cases with one patient) were not considered (Table 2)

Osteoblastomas are rare, solitary, benign bone tumors comprised of well vascularized connective tissue and osteoids, where primitive woven bones are actively formed[2,4.] The tumor was first described by Jaffe [16-19] in 1932 and its characteristics was later defined by Jaffe and Lichtenstein[19-21]. Osteoblastomas are mostly seen in the posterior elements of the vertebra and rarely in the vertebral bodies. The reason for patients to apply to the outpatient clinic is usually local tenderness and pain. Symptoms have a wide range of variety and the diagnosis is delayed in most cases[22,23]. Although the growth progress of the benign neoplasm is slow, osteoblastomas are challenging when they are localized in the mobile spine and sacrum. The disorder is typically seen in the young adults, with a slightly higher incidence in males. In our study, 75% of our series were males. The mean age of the patients at the time of diagnosis was 14.2 years. In terms of age and gender, the findings in our series are compatible with those of the literature[24] .Patients usually show clinical signs of pain and neurological deficit. The time between the surgery and the onset of symptoms are usually delayed[25]. Our patients did not exhibit any neurological deficits in the preoperative and postoperative period. The mean time to diagnosis was 9.2 (range: 2 to 16) months.

Computed tomography can determine the localization of the lesion, bone involvement, as well as show the degree of sclerosis. Therefore, it is the preferred imaging method[26]. Because of inflammatory changes, findings in spinal osteoblastomas can be misleading, so MRI has limited use [27]. Edema is not a specific response to tumor-induced inflammation. Edema present challenges in visualizing the gaps between the bone edges and identifying the soft tissues. It also causes wrongful diagnosis of the aggressive and malign lesions[28,29]. Peritumoral inflammation is thought to occur secondary to prostaglandin production, and it has often been associated with osteoblastoma in the literature[30]. A non-specific inflammation, modulated by hypervascularity and hyperperfusion, occurs as a response[2,31]. Localized inflammatory response in MR images was first described as the "flare phenomenon" in 1990 [5]. Studies showing the importance of the inflammatory response in children were later seen in the literature [32]. COX inhibitors used in the treatment of osteoid osteoma, a lesion similar to osteoblastoma, can be an example for prostaglandin-mediated inflammation. Generally, the flare phenomenon often accompanies osteoblastoma, but MR images can be misinterpreted if CT and clinical data are ignored. Extensive bone marrow edema and soft tissue edema intuitively suggest a malign pathology as in the assessment of osteosarcoma, Ewing's or lymphoma sarcoma. Almost in all CT images or plain radiographs, these lesions show malign bone damages. The benign findings of osteoblastoma in CT outweigh the potentially alarming findings of MRI [33]. Preoperative assessment should be carried out using both CT and MR images. Although CT is necessary in exact assessment of the bone involvement, MRI is a complemental method. MRI is essential in the assessment of the medullary canal, nerve roots and soft tissues [24].

Following curettage, osteoblastomas may show locally aggressive behavior and recur. In addition, by definition they do not have the potential to metastasize and are benign. Pain is the most common clinical complaint in patients with osteoblastoma. Gait disturbances, swelling, increased temperature and tenderness are among other common symptoms. More severe pain is seen in aggressive osteoblastomas. Localized destruction zones can be considered as the reason for this. Pain in osteoblastoma, unlike osteoid osteoma, does not respond to NSAIDs and night pain is usually less severe. Paresthesia, paraparesis, scoliosis and torticollis may sometimes be seen with spinal osteoblastomas [8,34]. Our patients had temporarily benefited from NSAIDs and showed no signs of preoperative or postoperative neurological deficit.

Osteoblastomas can occur in a variety of ways. The area surrounding the primary tumor can be quite lytic or dense. The cortex of osteoblastomas can be thin or wide. They may also consist of fibrotic tissue without bone formation. Microscopically, osteoblastomas have a bony trabecular meshwork within a loose fibrovascular stroma, classically surrounded by a single row of benign osteoblasts. Minimal mitotic activity is seen in osteoblasts or stromal cells. In addition, irregular bone formation is often observed, accompanied by dense cortex [35,36].

Osteoblastoma may resemble lesions such as osteosarcoma, osteoid osteoma radiologically, and it also shows similarity with chondroblastoma, aneurysmal bone cyst, chondrosarcoma, and this causes difficulties in diagnosis. In addition, the pathological changes in osteoblastoma are usually different from other lesions. Although the radiographic appearance of osteoblastomas may seem aggressive, these tumors are benign histologically. It is a basic practice to take a biopsy before curettage or resection. Previous biopsy is very useful in choosing the type of procedure to be performed.

The mainstay of treatment is surgery and usually total block resection is employed [21]. This treatment is a radical one and does not allow for recurrence. Other procedures for the tumors localized in the vertebral column are curettage and marginal resection. The rate of relapse in patients treated with surgery alone is about 10%.

Radiotherapy is applied as an adjuvant treatment in unresectable or recurring tumors, aggressive forms of the lesion or after incomplete excision [37]. The rate of recurrence is high due to limitations on the surgical approach imposed by the neurological structures and anatomical structures like dura mater [38].

Table 2 : With 2 and more number of patients Results of Systematic Review from 1997 to 2017.

Number of patients	Authors	Year	Location	Symptoms	Treatment	Follow-up	Outcome
2	Sar et al[36]	2002	sacrum	not specified	Resection (wide margins)	65 and 51 mo	Local recurrens(-)
5	Berry et al[43]	2008	Sacrum	not specified	Curettage	not specified	Local recurrens(-)
2	Poleksic´ et al[13]	2010	Sacrum	Pain	Curettage	not specified	Not specified
18	Ruggueri et al[20]	2016	sacrum	pain	16 curatage, 1 curettage + RT,1 case Resection (wide margins)	8.4 yr	Local recurrens (-) 3 patients (17%)

Unfortunately, the success of RT in preventing recurrence after incomplete excision has not been shown. The disadvantages of this treatment are its local adverse effects and potential of causing radioactive sarcomas [39]. In their review of 197 osteoblastoma cases, Marsh [13] concluded that "RT does not change the course of the disease and is contraindicated". In our review of the literature, we found only one case of sacral osteoblastoma treated with RT; with a dose of 45 Gy, the lesion regressed and improvements in pain, motor and sensory functions were observed during the one-year follow-up period of an 18 -year-old male patient [40]. Due to the short period of follow-up in this study, one cannot advocate for the use of RT alone. Therefore, we did not prefer RT in our series.

Osteoid osteomas (OO) often cause pain and are treated for persistent pain. In addition, osteoblastomas increase the pain and also size of the bone and are therefore treated. Osteoblastomas cause destruction of bone and may pose a risk to various structures. These include the iliac vessels, hip, bony support of the pelvic ring, lumbosacral nerve roots, bladder, ureters, and rectum[7,11,25,36,41]. Compared to OOs, osteoblastomas are more aggressive, bigger and they have a higher tendency for recurrence. Cases with osteosarcomas mimicking osteoblastoma and malign degenerations have been reported and misleading or wrongful diagnoses have been made [7,42]. In some cases, the differential diagnosis of osteoblastoma from osteosarcomas may be histopathologically weak; molecular genetic testing may come handy in solving this problem [7,41]. In our series, the lesions could be diagnosed by pathology.

The treatment choices in osteoblastoma include RT, intralesional surgery, intralesional surgery and RT, intralesional surgery and local adjuvants phenol or cryotherapy, and wide resection. The decision for intralesional surgery usually depends on the relationship of the lesion with the nerve roots, pelvis and visceral structures. The disadvantage of intralesional surgery in aggressive diseases like this is the increased risk of recurrence [7]. Intralesional excision in the form of curettage provides good local control for common sacral osteoblastomas, whether or not supported by local adjuvants [36,43,44]. Theoretically, wide excision minimizes the risk of local recurrence; however, the lesion close to the S3 vertebra has a higher risk of surgical morbidity. We preferred wide excision in our patients and did not encounter surgical morbidity. 306 cases of osteoblastoma were reviewed by the Mayo clinic, and only 75 patients had full treatment and long clinical follow-up. In this study, intralesional surgery recurrence rate was 19%, marginal resection was 5.6%, and wide resection was 20% [36]. The exact localization of the tumor is one of the most important factors in successful removal.

Osteoblastoma is a hypervascular tumor. Therefore, preoperative embolization may be preferred to improve surgical conditions and reduce intraoperative bleeding [45]. The need for embolization was discussed in the oncology council and we concluded that embolization was not necessary.

Conclusion

We believe that the surgery of patients with sacral osteoblastomas should be performed in a specialized tumor center with high level of surgical and oncological expertise. We did not observe any recurrence or neurological deficit in our patients treated with wide resection. All our patients underwent a single surgery and received no additional treatment.

Conflict of interest

The authors declare that there are no conflict of interests.

Financial disclosure

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