J Turk Spinal Surg 2024;35(3):157-161
DOI: 10.4274/jtss.galenos.2024.08769

WIDE RESECTION IN SACRAL OSTEOBLASTOMA: CASE SERIES

Albert Çakar[1], Saltuk Buğra Tekin[1], Mehmet Akif Aktaş[2], Esra Çirci[3], Barış Polat[4], Barış Peker[4], Yunus Atıcı[1]

[1]*University of Health Sciences Turkey, İstanbul Training and Research Hospital, Clinic of Orthopedics and Traumatology, İstanbul, Turkey*
[2]*Hakkari Yüksekova State Hospital, Clinic of Orthopedics and Traumatology, Hakkari, Turkey*
[3]*İstanbul Physical Therapy and Rehabilitation Training and Research Hospital, Clinic of Orthopedics and Traumatology, İstanbul, Turkey*
[4]*Dr. Burhan Nalbantoğlu State Hospital, Clinic of Orthopedics and Traumatology, Lefkoşa, Turkish Republic of Northern Cyprus*

ABSTRACT

Objective: The purpose of this study was to assess the outcomes of wide resection for sacral osteoblastoma (OB).

Materials and Methods: A review of our database revealed 6 cases of OB located in the sacrum. Localized pain in lesions that did not fully resolve although medical treatment was observed in all 6 cases. Surgical treatment consisted wide resection. The average time between diagnosis and surgery was 30 (24-36) months, and the average age at surgery was 14 (8-20) years.

Results: Postoperatively, the mean follow-up period was 74.3 months (24-110). At final followup, we did not encounter any complications, recurrence, spinal instability, and neural damage were not observed as a result of the removal of lesions in the sacrum area. The preoperative mean Visual Analog Scale score was 8 before treatment and 0 at the final follow-up.

Conclusion: Wide resection is a safe and effective treatment option for patients with sacral osteoblastoma.

Keywords: Wide resection, osteoblastoma, sacrum

INTRODUCTION

Osteoblastoma (OB) is a rare benign primary bone tumor with less sclerotic borders and no reactive perilesional bone formation. It grows slowly and is larger than 20 mm[1-4]. OB is commonly seen in adolescents under the age of 20[5]. OB is clinically divided into two types: conventional OB and aggressive OB (in older patients). Pain is the first clinical symptom. Pain is caused by the excessive production of prostaglandins. Moreover, OB is less responsive to non-steroidal anti-inflammatory drugs (NSAID)[6-10].

OB is seen more common in men. It is mostly located in the spine (30-50%), especially in the posterior elements (pedicle and lamina). But it can also develop from the vertebral body. It is less likely to be seen in the sacrum than in other spinal segments[9,11,12]. Due to the rarity of OB in the sacrum, there are few studies in the literature regarding its treatment[6,13-15]. Thus, this study aims to evaluate the clinical success of wide resection for OB located in the sacrum.

MATERIALS AND METHODS

This single-center retrospective study was carried out at the University of Health Sciences Turkey, İstanbul Training and Research Hospital in accordance with the Declaration of Helsinki guidelines (approval number: 66, date: 06.09.2024). An opt-out form, available on the hospital's website, was used to obtain informed consent. All participants received thorough information and gave their informed consent. Furthermore, all patients in this study provided informed consent prior to inclusion.

All patients were presented with pain in the lumbosacral and sacrococcygeal regions. Concurrently, one patient had pain localized at the S2-S3 level, another patient had pain localized at the S3-S4 level and 4 patients had pain localized at the S4 level. Patients preoperative and postoperative pain were evaluated with Visual Analogue Scale (VAS). All patients had no neurologic deficit, and their complaints were partially reduced with the use of NSAIDs. Local tenderness in the complaint area was present in 100% of the patients.

Address for Correspondence: Albert Çakar, University of Health Sciences Turkey, İstanbul Training and Research Hospital, Clinic of Orthopedics and Traumatology, İstanbul, Turkey
Phone: +90 532 281 57 22 **E-mail:** dralbertcakar@gmail.com **Received:** 26.07.2024 **Accepted:** 21.10.2024
ORCID ID: orcid.org/0000-0002-1422-2208

To define the lesions and prepare for preoperative planning, all patients were examined with pelvic and positron emission tomography (PET) computed tomography (CT) and magnetic resonance imaging (MRI) which were identified using the hospital's Picture Archiving and Communication System. A hundred percent of the lesions were larger than 2 cm. The patients went under the procedure of biopsy with the guide of the C-arm fluoroscopy and biopsy sample material was obtained. The samples were investigated in the pathology department, confirming the OB diagnosis. With the confirmed pathological diagnosis and correlated clinical evaluation, patients were staged as stage IIA and IIB according to the Enneking classification (Table 1).

Patients with no neurological deficits & no functional joint range of motion limitation and with the OB diagnosis confirmed by pathological assessment were included in our study. There was no strict exclusion criteria applied due to the rarity of OB in the sacrum.

All patients in this study also were thoroughly informed prior to surgical intervention and gave their informed consent. Under general anesthesia, with appropriate prepping and sterile draping on prone positioning, the posterior approach midline incision was used to expose both sides of the sacrum. Distally based V-shaped incision was made in the lumbosacral fascia, iliac crest was aimed and incision had ended when this landmark is reached. To achieve exposure the posterior aspect of the sacrum, subperiosteal elevation of the lumbosacral musculature was performed by releasing the multifidus distally and elevating it as a flap. Afterwards, OB lesion was widely resected within the proper resection margin area. The operation was concluded after gentle repositioning and repair of the erector spinae muscles and closure of the subcutaneous tissues and skin in layers. Dissected material was sent to pathology department for further confirmation. All patients that underwent wide resection through the posterior approach were without preoperative embolization. The interval between diagnosis and operation was approximately 30 months.

RESULTS

Between 2013 and 2020, wide resection was performed on 6 patients with Enneking classification stage 2 OB in the sacrum. The sample size of the study is total of 5 patients included; while 5 were male (83.3%), 1 was female (16.6%) and the mean age was 14 (8-20) years. The localized lesions in the patients were at the S2-S3 level in one patient, at the S3-S4 level in one patient, at the S4 level in 2 patients, and at the S4-S5 level in 2 patients.

The masses of all patients that were removed by wide resection using a posterior approach were confirmed OB diagnosis, with preoperative biopsy sample material and postoperative resected tissue material via pathology department. Reconstruction was not applied or required to any patient. There was no excessive bleeding during surgery in any of the procedures, further no significant hemogram abnormalities observed amongst all the patients during surgical follow-up. No postoperative complications were observed in any patient. Moreover, no recurrences occurred in any of the patients during follow-up.

After the wide resection procedure, the patient's pain complaints were evaluated with VAS. Between the comparison of ratings preoperative and postoperative, mean average of preoperative ratings were recorded as 8 and mean average of postoperative ratings were recorded as 0. The patients were not evaluated only according to their pain complaints in their clinical evaluation. Preoperatively, no functional joint range of motion limitation or no neurological deficits were observed in any of the patients. Postoperatively, there were no changes in their functional joint range of motion or neurological evaluation. Moreover, according to the clinical assessment of patients preoperatively and postoperatively using American Spinal Injury Association impairment scale, all the patients can be graded as E (sensation and motor function are graded as normal in all segments).

DISCUSSION

OB was first described as giant osteoid osteoma (OO) in 1954[16]. Lichtenstein[17] and Jaffe[18] defined OB as a separate clinical and morphological diagnosis from OO. Tumors with a diameter of ⩽1 cm were classified as OO, while those with a diameter of ⩾2 cm were classified as OB. Other criteria for diagnosing tumors between 1 cm and 2 cm included the relevant bone, site, presence of nidus, and presence of peripheral sclerosis. Compared to OO, radiographic features of OB are variable and non-specific, but they typically indicate a benign process. The lesion is typically oval or round, expandable, well-defined, and radiolucent. The central part can be completely lytic, but there

Table 1. Enneking staging for malignant musculoskeletal tumors; based on surgical grade, local extent, and presence or absence of metastasis

Enneking staging for malignant musculoskeletal tumors			
Stage	**Grade**	**Site**	**Metastasis**
IA	Low (G1)	Intracompartmental (T1)	No metastasis (M0)
IB	Low (G1)	Extracompartmental (T2)	No metastasis (M0)
IIA	High (G2)	Intracompartmental (T1)	No metastasis (M0)
IIB	High (G2)	Extracompartmental (T2)	No metastasis (M0)
III	Any (G)	Any(T)	Regional or distant metastasis (M1)

is usually some focal calcification. Furthermore, OB exhibits a distinct pain pattern, lacks reactive bone formation, and is larger[8-10]. OB is a slow-growing benign primary bone tumor made up of well-vascularized connective tissue that produces active osteoid and primitive woven bone[9].

In some of the cases, OB can break the cortical bone and can be aggressive, and differential diagnosis of these cases may be more difficult than low-grade osteosarcoma[19-21]. The diagnosis required for treatment is dependent on symptoms, imaging, and pathology[22]. MRI has a limited role in spinal OBs due to the potential for misleading images caused by adjacent inflammatory changes. The visual boundary between bone and soft tissues is less defined, leading to misdiagnosis of aggressive or malignant lesions[23]. For the diagnosis of spinal OB, imaging examinations such as radiography, CT, MRI, and PET have different value, non of them have specificity[24]. Therefore, combining CT, MRI and PET may be beneficial to optimize preoperative diagnosis and care of patients with OBs[25]. A preliminary diagnosis of OB was made clinically in our 6 cases. The diameter of the masses in all cases was larger than 2 cm. Biopsies were taken from the lesions. All pathology results were confirmed as OB. OB has no specific clinical presentation and the primary complaint is progressive pain, which largely depends on location and size. The tumor may enlarge and appear as a palpable mass with associated tenderness and swelling. Neurological symptoms may also be present in the spinal areas[11,26]. The complaints of our patients were pain in the lumbosacral and sacrococcygeal regions. Patients occasionally had complaints of nocturnal pain. Preoperatively, there were no neurological deficits in the patients. All patients stated that their pain was partially relieved when using NSAIDs. None of the patients had a palpable mass on physical examination.

Treatment options for OB include intralesional surgery, wide resection, radiofrequency ablation (RFA), radiotherapy, chemotherapy, and surgical intervention with radiotherapy or chemotherapy[13,14,26,27]. Radiotherapy and chemotherapy can be used as the main treatment or as an adjunct treatment method to surgery. Radiotherapy has been suggested for the treatment of OB in the sacrum, which is difficult to resect completely and carries a risk of complications[15,26]. However, debate continues as to whether it reduces recurrence or not. Radiotherapy has not been shown to improve local control to prevent recurrence after inadequate removal of OB. The disadvantages involve local side effects and the potential for leading to radio-induced sarcomas[26,28].

RFA may be preferred, especially in small lesions and in safe locations. RFA-treated spinal OB cases have been reported in the literature, but there are very few studies and several cases yet[27,29-32]. In RFA application, the minimum safe distance from the bone cortex around the tumor is 2 mm; however, more than 1 mm distance is needed for safety in case of proximity with cerebrospinal fluid and the lesion[33]. Thermal damage to the spinal cord and peripheral nerves is a risk that should be considered prior to RFA since more than 45 °C heating shown to be cytotoxic[27]. The temperature during intervention RFA decreases significantly only beyond the 1 cm distance from the active tip, as the study shows mean maximum temperatures of 69.1°, 51.3°, and 42.5 °C for 1-mm lamella; 59.2°, 46.5°, and 41.1 °C for 3-mm lamella; and 50.6°, 44.8°, and 40.0 °C for 5-mm lamella were measured 0, 5, and 10 mm, respectively, from the periosteum[34]. All in all, due to the risk of thermal damage to adjacent neurovascular tissues RFA has limited spinal application rate.

In the study of Rehnitz et al.[31], there were 2 OBs. One sacral lesion was located in the anterior left sacral ala, directly adjacent to the sacral nerve plexus. They recommend what they consider RFA as the treatment of choice for OB including spinal[31]. One sacral lesion was found in the anterior left sacral ala, right next to the sacral nerve plexus. They suggest RFA as the preferred treatment for OB, such as spinal. Wang et al.[32] also suggest that RFA can be considered as a safe and effective treatment for spinal S2 OB (3 cases). Arrigoni et al.[27] issued a set of 11 patients with OB of the spine who received RFA and achieved total relief in all cases. In another study, Beyer et al.[32] found the technical success rate to be 90.0% and the recurrence rate to be 44.4% after RFA treatment in 10 patients with spinal OB (2 cases in the sacrum)[32].

Considering the success of RFA appliance in some of the studies, the gold standard treatment for OB remains still as surgery. What should be considered in the surgical treatment of OB? First, the tumor should be completely removed, second, the sacral nerve and cauda equina should be preserved[11]. Primary benign spine tumours can be categorized by the Enneking system. Stage 2 and 3 lesions generally require treatment[35]. OB lesions can be evaluated as active OB lesions (Enneking stage 2, S2), and aggressive OB lesions (Enneking stage 3, 3, S3)[6,35]. Intralesional surgery is recommended for grade 2 lesions.

Wide resection is suggested for grade 3 lesions, more serious tumors, or lesions based in areas where a potential local recurrence could prove difficult to treat[6,21,35]. Boriani et al.[6] also recommended spinal S2 OB lesions intralesional curettage and S3 OB lesions block resection as treatment. Because S3-level lesions are aggressive and have a higher recurrence rate[6]. Zoccali et al.[21] nine out of eleven cases required intralesional surgery; wide resection was performed in the other 2 cases. No local recurrence was confirmed at 88 months of follow-up[21]. Intralesional curettage and incomplete resection can lead to recurrence. Ruggieri et al.[14] performed a high number of intralesional for sacral OB's. The recurrence rate was relatively high. They said that inadequate intralesional surgery was associated with a higher rate of local recurrence (40%, 2 local recurrences in 5 cases). Wide resection theoretically minimizes recurrence compared to intralesional resection. However, wide resection can increase the risk of morbidity, especially for lesions proximal to S3. Wide resection of the lesion can often lead to spinal instability and the spine cord or nerve root is as often at risk of damage. The risk of local recurrence in lesions

found in the sacrum is higher than in other areas because of complex anatomy and the existence of sacral roots[14].

All things considered, treatment options for OB shows variety [36-38]. For low stage or locally invasive lesions in Enneking classification, surgeons or clinics choice of treatment methodology seems to differ[1,5,13,15]. Due to rarity of the sacral OB, there is no clear consensus on the use of which treatment modality or their combinations[13,14,36]. However, for high grade lesions such as stage 3,wide resection treatment is the treatment modality of choice[37]. Keeping in mind that, treatment aim of OB is complete resection and avoidance of recurrence while preserving adjacent neurovascular tissues, preference of wide resection surgery should be considered in lower stage lesions for better postoperative prognosis[36]. Therefore, in our study, cases that were classified as stage 2 according to the Enneking classification were treated with wide resection surgery to avoid the risk of recurrence. We did not encounter any complications, recurrence, spinal instability or neural damage as a result of the removal of lesions in the sacrum area.

Study Limitations

Our study's major limitations include a retrospective design and a small number of cases.

CONCLUSION

Sacral OBs are rarely encountered. In our series, wide resection was successful in all of the patients. We recommend wide resection surgery in treatment of sacral OB.

Footnote

Ethics Committee Approval: This single-center retrospective study was carried out at the University of Health Sciences Turkey, İstanbul Training and Research Hospital in accordance with the Declaration of Helsinki guidelines (approval number: 66, date: 06.09.2024).

Informed Consent: Retrospective study.

Authorship Contributions

Surgical and Medical Practices: Y.A., Concept: A.Ç., Design: E.Ç., Data Collection or Processing: M.A.A., B.Pe., Analysis or Interpretation: S.B.T., B.P., Literature Search: S.B.T., M.A.A., Writing: A.Ç.

Conflict of Interest: The authors have no conflicts of interest to declare.

Financial Disclosure: The authors declared that this study received no financial support.

REFERENCES

1. Berry M, Mankin H, Gebhardt M, Rosenberg A, Hornicek F. Osteoblastoma: a 30-year study of 99 cases. J Surg Oncol. 2008;98:179-83.
2. Harrop JS, Schmidt MH, Boriani S, Shaffrey CI. Aggressive "benign" primary spine neoplasms: osteoblastoma, aneurysmal bone cyst, and giant cell tumor. Spine (Phila Pa 1976). 2009;34(22 Suppl):39-47.
3. Balioğlu MB, Albayrak A, Atıcı Y, Sökücü S, Tacal MT, Kaygusuz MA. The effect of simple local resection on pain and scoliotic curve in patients with scoliosis secondary to osteoid osteoma and osteoblastoma in the spine. Acta Orthop Traumatol Turc. 2016;50:330-8.
4. Loizaga JM, Calvo M, Barea FL, Tello FJM, Villanueva JP. Osteoblastoma and osteoid osteoma. Clinical and morphological features of 162 cases. Pathol Res Pract. 1993;189:33-41.
5. Versteeg AL, Dea N, Boriani S, Varga PP, Luzzati A, Fehlings MG, et al. Surgical management of spinal osteoblastomas. J Neurosurg Spine. 2017;27:321-7.
6. Boriani S, Amendola L, Bandiera S, Simoes CE, Alberghini M, Di Fiore M, et al. Staging and treatment of osteoblastoma in the mobile spine: a review of 51 cases. Eur Spine J. 2012;21:2003-10.
7. Li Z, Zhao Y, Hou S, Mao N, Yu S, Hou T. Clinical features and surgical management of spinal osteoblastoma: a retrospective study in 18 cases. PLoS One. 2013;8(9):74635.
8. Atesok KI, Alman BA, Schemitsch EH, Peyser A, Mankin H. Osteoid osteoma and osteoblastoma. J Am Acad Orthop Surg. 2011;19:678-89.
9. Lucas DR, Unni KK, McLeod RA, O'Connor MI, Sim FH. Osteoblastoma: clinicopathologic study of 306 cases. Hum Pathol. 1994;25:117-34.
10. Yalcinkaya U, Doganavsargil B, Sezak M, Kececi B, Argin M, Basdemir G, Oztop F, et al. Clinical and morphological characteristics of osteoid osteoma and osteoblastoma: a retrospective single-center analysis of 204 patients. Ann Diagn Pathol. 2014;18:319-25.
11. Lai Q, Wang Q, Liu H, Chen D, Wan Z, Yu X, et al. A rare case of giant osteoblastoma of the sacrum. Orthopade. 2019;48:343-7.
12. Galgano MA, Goulart CR, Iwenofu H, Chin LS, Lavelle W, Mendel E. Osteoblastomas of the spine: a comprehensive review. Neurosurg Focus. 2016;41:e4.
13. Reynolds JJ, Rothenfluh DA, Athanasou N, Wilson S, Kieser DC. Neoadjuvant denosumab for the treatment of a sacral osteoblastoma. Eur Spine J. 2018;27(Suppl 3):446-52.
14. Ruggieri P, Huch K, Mavrogenis AF, Merlino B, Angelini A. Osteoblastoma of the sacrum: report of 18 cases and analysis of the literature. Spine (Phila Pa 1976) 2014;39:e97-e103.
15. Rajkumar A, Basu R, Datta NR, Dhingra S, Gupta RK. Radiation therapy for sacral osteoblastoma. Clin Oncol (R Coll Radiol). 2003;15:85-6.
16. Dahlin DC, Johnson EWJR. Giant osteoid osteoma. J Bone Joint Surg Am. 1954;36-A:559-72.
17. Lichtenstein L. Benign osteoblastoma: acategoryofosteoid-andbone-forming tumors other than classical osteoid osteoma, which may be mistaken for giant-cell tumor or osteogenic sarcoma. Cancer. 1956;9:1044-52.
18. Jaffe HL. Benign osteoblastoma. Bull Hosp Joint Dis. 1956;17:141-51.
19. Dorfman HD, Weiss SW. Borderline osteoblastic tumors: problems in the differential diagnosis of aggressive osteoblastoma and low grade osteosarcoma. Semin Diagn Pathol. 1984;1:215-34.
20. Oliveira CR, Mendonca BB, Camargo OP, Pinto EM, Nascimento SA, Latorre Mdo R, et al. Classic osteoblastoma, atypical osteoblastoma, and osteosarcoma: a comparative study based on clinical, histological, and biological parameters. Clinics (Sao Paulo). 2007;62:167-74.
21. Zoccali C, Novello M, Arrigoni F, Scotto di Uccio A, Attala D, Ferraresi V. Osteoblastoma: When the Treatment Is Not Minimally Invasive, an Overview. J Clin Med. 2021;10:4645.
22. Kroon HM, Schurmans J. Osteoblastoma: Clinical and radiologic Şndings in 98 new cases. Radiology. 1990;175:783-90.
23. Shaikh MI, Saifuddin A, Pringle J, Natali C, Sherazi Z. Spinal osteoblastoma: CT and MR imaging with pathological correlation. Skeletal Radiol. 1999;28:33-40.
24. Si Z, Meng W. Multimodal Imaging Evaluation and Clinical Progress of Spinal Osteoblastoma: A Comprehensive Review. World Neurosurg. 2023;170:28-37.

25. Huang Z, Fang T, Si Z, Li Y, ZhAng L, Li S, et al. Imaging algorithm and multimodality evaluation of spinal osteoblastoma. BMC Musculoskelet Disord. 2020;21:240.
26. Berberoglu S, Oguz A, Aribal E, Ataoglu O. Osteoblastoma response to radiotherapy and chemotherapy. Med Pediatr Oncol. 1997;28:305-9.
27. Arrigoni F, Barile A, Zugaro L, Fascetti E, Zappia M, Brunese L, et al. CT-guided radiofrequency ablation of spinal osteoblastoma: Treatment and long-term follow-up. Int J Hyperthermia. 2018;34:321-7.
28. Merryweather R, Middlemiss JH, Sanerkin NG. Malignant transformation of osteoblastoma. J Bone Joint Surg Br. 1980;62:381-4.
29. Dupuy DE, Hong R, Oliver B, Goldberg SN. Radiofrequency ablation of spinal tumors. AJR Am J Roentgenol. 2000;175:1263-6.
30. Rehnitz C, Sprengel SD, Lehner B, Ludwig K, Omlor G, Merle C, Kauczor HU, et al. CTguided radiofrequency ablation of osteoid osteoma and osteoblastoma: clinical success and long-term follow up in 77 patients. Eur J Radiol. 2012;81:3426-34.
31. Wang B, Han SB, Jiang L, Yuan HS, Liu C, Zhu B, et al. Percutaneous radiofrequency ablation for spinal osteoid osteoma and osteoblastoma. Eur Spine J. 2017;26:1884-92.
32. Beyer T, van Rijswijk CSP, Villagrán JM, Rehnitz C, Muto M, von Falck C, et al. European multicentre study on technical success and long-term clinical outcome of radiofrequency ablation for the treatment of spinal osteoid osteomas and osteoblastomas. Neuroradiology. 2019;61:935-42.
33. Yamane T, Tateishi A, Cho S, Manabe S, Yamanashi M, Dezawa A, et al. The effects of hyperthermia on the spinal cord. Spine (Phila Pa 1976). 1992;17:1386-91.
34. Bitsch RG, Rupp R, Bernd L, Ludwig K. Osteoid osteoma in an ex vivo animal model: temperature changes in surrounding soft tissue during CT-guided radiofrequency ablation. Radiology. 2006;238:107-12.
35. Enneking WF. A system of staging musculoskeletal neoplasms. Clin Orthop Relat Res. 1986;204:9-24.
36. Dinçel Y, Arikan Y, Özer D, Can E, Dünki A, Kilinc S, et al. The Results of Wide Resection in Sacral Osteoblastoma. Int J Acam Med Pharm. 2022;4:31-5.
37. Wu M, Xu K, Xie Y, Yan F, Deng Z, Lei J, et al. Diagnostic and Management Options of Osteoblastoma in the Spine. Med Sci Monit. 2019;25:1362-72.
38. Jawad MU, Scully SP. In brief: classifications in brief: enneking classification: benign and malignant tumors of the musculoskeletal system. Clin Orthop Relat Res. 2010;468:2000-2.