The effect of simple local resection on pain and scoliotic curve in patients with scoliosis secondary to osteoid osteoma and osteoblastoma in the spine

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Objective: The aim of this study was assess the results of local resection without instrumentation in patients with scoliosis secondary to spinal osteoid osteoma and osteoblastoma.

Methods: The review of our database revealed 176 cases of osteoid osteomas and 18 of osteoblastomas. Painful scoliosis was seen in 5 out of 6 cases. The lesion was found on the posterior part of the apical vertebra in the concave side of the scoliotic curve. Surgical treatment consisted of simple en bloc excision. Mean period between diagnosis and operation was 2.6 years, mean age at the time of surgery was 12.5 years, and mean preoperative major Cobb angle was 37.2°.

Results: Four patients with a mean follow-up of 4.3 years were included in the study. At final follow-up, Cobb angle was 7.6°, and the average percentage of correction was 79.6%. Coronal decompensation was corrected by 87.7%. Pelvic tilt and shoulder imbalance were corrected by 15% and 74.5%, respectively. The preoperative mean Visual Analog Scale score was 9 before the treatment and 0 at the final follow-up.

Conclusion: Our results suggested that simple en bloc resection may be a safe and effective treatment option in patients with scoliosis secondary to spinal osteoid osteoma and osteoblastoma, if patient less than 16 years, with major Cobb angle less than 40°, and duration of complaint less than 22 months.

Keywords: En bloc excision; osteoblastoma; osteoid osteoma; painful scoliosis.

Level of Evidence: Level IV, Therapeutic Study.

Osteoid osteoma and osteoblastoma are benign primary bone tumors more frequently observed in young males, which may capture all bones such as the skull and clavicle sternum. Osteoid osteoma is a benign bone tumor with limited growth potential, which causes pain due to its small size (15–20 mm, typically <15 mm). [1–3] Osteoid osteoma accounts for 10%–12% of benign bone tumors and 3% of primary bone tumors. [3–9] Particularly, it accounts for approximately more than 50% of tumors identified in metaphysiodiaphysial sections of long bones such as the femur and tibia. [10] The backbone is affected by approximately 10%, and osteoid osteoma is mostly observed on the neural arc. [1] Osteoblastoma histologically resembles osteoid osteoma, but it is larger in size (>2 cm). Osteoblastoma accounts for 1%–5% of primary bone tumors. Most commonly, 28%–40% spine...
involvement is observed.\textsuperscript{[1,2,4]} It is reported that osteoblastoma (30\%–50\%) impacted the spine more than osteoid osteoma (10\%–25\%).\textsuperscript{[5–8,10–16]} Lesions are primarily observed on posterior vertebrae elements and are most frequently observed in the lumbar section, at least in the cervical section.\textsuperscript{[12,17]}

Lesions do not generally indicate evident clinical and radiologic symptoms other than local pain and slight scoliosis deformity. Average age of clinical diagnosis has been reported as 19 years old, with 80\% of patients applying before the age of 30, and it is observed more frequently in men than women.\textsuperscript{[6]} Being one of the most frequent reasons of painful scoliosis in the adolescent age group, osteoid osteoma is characterized with slight-to-medium pain which begins at night and which is recovered with salicylates. Especially when noted on the vertebral column, scoliosis may appear as a clinical table which accompanies it.\textsuperscript{[6,18,19]} It develops secondary to osteoid osteoma-related scoliosis muscle spasm, and although it is flexible in the early stage, it may become rigid scoliosis in time.\textsuperscript{[20]}

The nidus, a structure which is pathognomonic for osteoid osteoma, shows reactive bone sclerosis around an oval radiolucent osteoid structure smaller than 1.5 cm when monitored with computed tomography (CT).\textsuperscript{[6–8,10,20]} Thin section CT is recommended in early diagnosis.\textsuperscript{[21]} In osteoid osteoma treatment, non-surgical and surgical treatments have been defined, while nidus excision or radiofrequency ablation (RFA) constitutes the actual treatment.\textsuperscript{[22–26]}

The objective of the present study was the correction of scoliotic deformity secondary to osteoid osteoma and osteoblastoma by lesion excision, as well as assessment of the clinical results of the patients.

**Patients and methods**

Retrospective examination of the hospital archive from 1983 to 2014 found 194 osteoid osteoma and osteoblastoma patients. Of this group, 176 (90.7\%) had osteoid osteoma, and 18 (9.3\%) had osteoblastoma. Vertebrae involvement was noted in 6 (3.1\%) cases. Five of these 6 cases (3 male, 3 female) had osteoid osteoma (2.6\%), and 1 (0.5\%) had osteoblastoma. Painful scoliosis deformity accompanied 5 of the 6 cases, while scoliosis was absent in the remaining 1 case. In this case, the lesion related to osteoid osteoma was in the L2 vertebrae corpus, and it was excluded from the assessment. Radiologically, X-ray, CT, magnetic resonance imaging (MRI), and Tc99m bone scintigraphy were examined. The curve degree was measured with Cobb angle. Coronal decompensation, humeral balance, and pelvic tilt were calculated. Lesions were holding posterior elements of the apical vertebra on the concave side of the scoliotic curve. Limited surgical approval and en bloc excision were planned (Figure 1). Average time from onset of complaints to surgery was 2.6 years (range: 1.5–5 years), and average age at time of surgery was 12.5 years (range: 7.3–15.9 years). One case of osteoid osteoma with shorter follow-up period (1 month) was excluded from the study. Four cases with average follow-up of 4.3 years (range: 1.1–7.5 years) were examined in detail (Table 1).

These cases were assessed according to the Enneking system, which is used for primary benign spine tumors. The patients were accepted as phase II due to pain symptoms related with active lesion according to Enneking,\textsuperscript{[20]} and they were examined from a neurologic perspective.

**Results**

Measured from the region of most curvature, average preoperative anteroposterior Cobb angle was 37.2° (range:
27°–50°). At follow-up of 4.3 years on average (range: 1.1–7.5 years), Cobb angle was 7.6° (range: 1°–26°), and 79.6% correction (range: 48%–96.9%) was realized. Average coronal decompensation was 6.5 cm (range: 2.5–10 cm) preoperatively, while it was measured as 0.8 cm (range: 0.5–1.0 cm) at final follow-up, with a correction ratio of 87.7%. Average preoperative pelvic tilt was 2° (range: 0°–5°) and humeral tilt 4.7° (range: 1°–10°), while they were measured as 1.7° (range: 1°–3°) and 1.2° (range: 0.6°–2.5°), respectively, at final follow-up; correction achieved was 15% and 74.5% of cases, respectively (Table 1) (Figures 2–4).

Of the 4 cases which were assessed, in the 3 cases with curve degree ≤40°, average age was 11.5 years (range: 7.3–15.9 years), average Cobb angle was 33° (range: 27°–40°), and average period of complaint until surgery was 1.8 years (range: 1.5–2 years). After average follow-up of 5.4 years (range: 3.3–7.5 years) in these 3 cases, average Cobb angle was 1.5° (range: 1°–2°) and correction 96.5% (range: 92.6%–96.9%). Nevertheless, as observed in 1 case (male, age 15 years 8 months, Cobb angle 50°), aged ≤16 years with curvature degree ≤50°, correction was expected after removal of the lesion, though correction achieved was less than the other 2 cases (correction in this case was 48% at 14-month follow-up).

After the lesion was located with preoperative imaging methods, surgery was realized as limited (Figure 1). The pieces incised from the lesion during surgery were assessed by a pathology specialist, who confirmed their benign character. Average duration of surgery was 90 min (range: 45–120 min). During the early postoperative period, 3 cases were treated with temporary corset for an average of 4 months (range: 2–6 months). Pain subsided immediately after surgery, and patients were able to return to their normal activities in an average 1.5 months (range: 1–2 months). In only 1 case (osteoblastoma), 13° kyphosis increase was observed between the T12–L1 segments in the sagittal plan. Before surgery, visual pain scale average of the patients was 9, while it was 0 during the follow-up period.

**Discussion**

Osteoid osteoma is a rare benign primary bone tumor, which was first defined by de Jaffe in 1935.[27] It accounts for nearly 3% of primary bone tumors and is twice as prevalent in males compared females.[6,28] Age of 80% of patients is <30 years old, approximately 50% of tumors are observed on the femur and tibia, and 10%–20% occur on the spine. The lesions on the backbone are mostly observed on the posterior vertebrae, but only 10% affect
the vertebrae object.\textsuperscript{[6,11,15,12,29,30]} It is most frequently ob-
served in the lumbar section, particularly in the cervi-
cal section.\textsuperscript{[11,12,30,31]} Posterior vertebral involvement was
observed in the 5 cases in the present study and anterior
involvement in 1 case. Lumbar and dorsal spines were
impacted in 4 and 2 cases, respectively.

Osteoid osteoma differs from osteoblastoma with re-
spect to its size. McLeod et al. defined the lesions with a
diameter of \( \leq 1.5 \) as osteoid osteoma and those with di-
ameter of \( >1.5 \) cm as osteoblastoma.\textsuperscript{[32]} Primary benign
spine tumors may be classified by using the Enneking sys-
tem. Phase II and III lesions generally require treatment.
\textsuperscript{[20]} As a result of pathologic examination, 5 cases in the
present study were diagnosed with osteoid osteoma, as
the lesion was \( \leq 1.5 \) cm, and 1 case with osteoblastoma,
as the lesion was \( >1.5 \) cm. Thus, our cases were evaluated
as phase II according to Enneking classification.

In the literature, the frequency of scoliosis in osteoid
osteoma or osteoblastoma patients has been declared as
25\%–74\%.\textsuperscript{[12,19,30]} The tumor is typically on the apex or
near to the concave zone of the curve.\textsuperscript{[33,34]} If the fourth
and fifth vertebrae are impacted in the lumbar zone, then
the apex is typically seen on the cephalad of the lesion,
accompanied by pelvic obliquity.\textsuperscript{[35]} On the lesion side,
it is believed to cause scoliosis by stimulating asymmet-
tric painful muscle spasm.\textsuperscript{[34]} Ransford et al. reported
that if it is present for a sufficient period, it may lead to
asymmetric growth inhibition in the vertebral epiphysis
and result in rotational deformity, causing the curve to
become more structural.\textsuperscript{[35]} It is our belief that paraspi-
nal muscle spasm causes scoliosis. During our average
follow-up of 4.3 years (range: 1.1–7.5 years), no asym-
metric growth inhibition was observed in the vertebral
epiphysis.

In a meta-analysis, Saifuddin et al. reported scoliosis
in 293 (63\%) of 465 child and adult patients suffering
from spinal osteoid osteoma or osteoblastoma.\textsuperscript{[12]} Av-
erage age of scoliosis patients was 15.3 years, scoliosis
prevalence was 65\%, and osteoid osteoma and osteoblas-
toma prevalence 52\%. Scoliosis was identified more com-
monly in cases of osteoid osteoma than osteoblastoma
(\( p<0.0001 \)), and the lesion was identified significantly
higher in the thoracic and lumbar zone compared to the cervical zone in cases of osteoid osteoma. No significant relevance was found between age, gender, and period of symptoms. Ozaki et al. applied surgical treatment to 9 osteoid osteoma and 13 osteoblastoma patients. Four cases were identified in the cervical spine, 6 in the thoracic, 10 in the lumbar, and 2 in the sacrum; scoliosis was identified in 17 patients. In the present study, spine involvement was found in 1 of 18 osteoblastoma patients (5.5%), and 2.8% among osteoid osteoma patients.

The reasons of pain for children and adolescents who apply for lumbar pain may be spondylosis/spondylolisthesis, Scheuermann kyphosis, neoplasia, discitis/vertebra osteomyelitis, lumbar disc hernia, and etiologies with unknown causes. Osteoid osteoma was reported as the most common reason of painful scoliosis in adolescents. Bezer et al. identified that osteoid osteoma caused pain in 1 of 26 (3.8%) cases of child and adolescent patients with lumbar pain that were monitored.

Osteoid osteoma typically appears with an insidious onset with lumbar pain only or together with radicular symptoms without indicating any neurological findings during physical inspection of adolescent male patients between 10 and 20 years old. Pain typically becomes evident a few months following radiologic indication. It causes scoliosis in children and adolescents, especially when lying. The curve is rigid and typically advances quickly. Osteoid osteoma or osteoblastoma or related curve may lead to restriction in spine movements of all plans. The pain may become evident at night and may generally be reduced with salicylates. Night pain is typical, though it was identified in only 30% of the patients in 1 study. In rare cases, no pain (1.6%) was reported. The pain of osteoblastoma patients is relieved with non-steroidal anti-inflammatory (NSAI) drugs. Lesions are mostly associated with spinal deformities. Burn et al. observed that 97% of the symptoms of osteoid osteoma and osteoblastoma patients appeared

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Fig. 3. (a) Female patient aged 7 years 4 months old. Etiology is osteoblastoma. Cobb angle in coronal plan is 40° (T4-L2). (b) Expanded lesion with cortical erosion on left T10 vertebra pedicle and lamina on scintigraphy, CT and MRI T10. (c) Patient aged 5 years and 6 months. Postoperatively, Cobb angle is 1.5°, correction is 96.3%. [Color figures can be viewed in the online issue, which is available at www.aott.org.tr]
with pain, 23% of them had scoliosis, and 40% of the complaints were reduced with NSAIs. Our findings indicated that lumbar pain had an insidious onset. Night pain was observed in 5 patients. The pain was relieved or mitigated temporarily by the use of salicylates. Restriction of spinal movements and pain were particularly evident before surgery.

In a study by McLeod et al., 11 of 26 osteoid osteoma patients (42%) were not diagnosed with scoliosis for a minimum 15 months from the onset of symptoms. Ozaki et al. could not diagnose 2 of 9 patients with osteoid osteoma (22%) for 2 years from the onset of symptoms. The average period from the beginning of pain to surgery was 16.6 months in the 1980s, while it dropped to 8.6 months in the 1990s. The period between the onset of symptoms and diagnosis was reported as 13.7–26 months. The reason for this duration is believed to be the assessment of the cases as idiopathic scoliosis during onset. The average period of 2.3 years (range: 1–5 years) from the onset of complaints to surgical operation in the present study is due to the delay in the diagnosis of the patients.

Ozaki et al. reported neurologic disorders in 9 of 2 osteoid osteoma patients and 9 of 13 osteoblastoma patients before treatment. No neurologic deformities were found in our cases.

Plain radiography and technetium bone scintigraphy are the recommended investigations for diagnosis. Factors such as localization of the nidus, its size, position of internal organs, and abdominal gas complicate radiological imaging. Even if radiological findings do not support the diagnosis, CT and technetium bone scintigraphy are recommended for the evaluation of patients suspected of having osteoid osteoma and osteoblastoma. Evaluation with scintigraphy is always able to produce a positive result for osteoid osteoma and osteoblastoma; therefore, scintigraphic investigation in the early period would reduce the process of diagnosis after symptoms begin. Especially in cases of nidus <10 mm, which is difficult to observe with thin section (≤2 mm) CT, investigation may be identified here. Davies reported that correct diagnosis with 65% accuracy could be achieved with MRI, whereas Yamamoto was not able to diagnose 2 cases of osteoid osteoma with MRI. Ozaki et al. observed high signal intensity areas on 9 of 10 patients inspected in T2-weighted images. In the present study, no evident radiological nidus focuses were observed in the coronal and sagittal planes.

Fig. 4. (a) Male patient aged 15 years 11 months. Etiology is osteoid osteoma. Cobb angle in coronal plan is 27° (T7-L3). (b) The lesion is located between the left L4 lamina and facet on CT and MRI imaging. (c) En bloc resection site. (d) Patient aged 3 years and 4 months. Postoperatively, Cobb angle is 2°, correction is 92.6%. [Color figures can be viewed in the online issue, which is available at www.aott.org.tr]
were evaluated with CT and MRI to identify the etiology due to painful scoliosis deformities. In 2 cases, Tc99 bone scintigraphy produced positive results. In our cases with scoliosis, they were identified on the concave side of the curvature of the posterior elements of the nidus vertebra on CT. During MRI inspection, as shown by Ozaki,\textsuperscript{[17]} they were monitored as high-density impacted areas in T2-weighed sections.

When spinal deformity or compression of neural elements is not present, the treatment of osteoid osteoma may be started conservatively with anti-inflammatory drugs.\textsuperscript{[45]} During the natural course of osteoid osteoma, spontaneous remission potential was reported between 2 and 8 years. Long-term follow-up with Kneisl and Simon NSAIs has been shown to be as efficient as excision for the treatment of osteoid osteoma.\textsuperscript{[22]} However, long-term NSAI exposure of patients results in complications, thus restricting this mode of treatment. Surgical treatment is performed by curetting the lesion or historically by en bloc resection. In addition, resection by means of CT, CT-aided percutaneous RFA, or laser ablation of the nidus have been accepted treatment options since the 1990s.\textsuperscript{[25,31,42,49,46]} If medical treatment becomes unsuccessful, then the lesion may be effectively damaged with minimal invasive techniques such as percutaneous radiofrequency thermal ablation and laser photocoagulation. However, proximity of the lesion to neural structures may present a risk for the use of RFA, and the use of these techniques may cause a risk of thermal damage in the adjacent neural tissues.\textsuperscript{[49]} Furthermore, percutaneous treatment has certain restrictions due to its inability to completely destroy lesions larger than 1 cm. Particularly, it should not be used in the treatment of lesions which are adjacent to neurovascular structures with spine involvement.\textsuperscript{[47]} Vanderschueren et al. achieved 79% successful results with RFA treatment and 96% successful results with repeated RFA treatment; however, post-RFA treatment deformity re-occurred in 43% of patients with spine deformity.\textsuperscript{[49]} Failure to fix the curve completely despite surgical treatment connotes that the lesion could not completely be excised or that there may be another underlying pathology.

Osteoblastoma may be treated successfully if diagnosed when benign and aggressive. Excision and intralosional curettage may be implemented. The entire nidus may be excised with high-speed diamond burr. Fusion and instrumentation may be employed unless spinal stability can be obtained following excision of the lesion.\textsuperscript{[11]}

Spinal tumors are generally treated with en bloc excision due to compensatory scoliosis’ risk of being structural.\textsuperscript{[12,17,26,33,37,38]} En bloc excision is recommended for osteoid osteoma patients with fixed spine deformity, neurological compression findings, patients who are considered unsafe with RFA with respect to the anatomic location of lesion, or for patients who had been previously treated unsuccessfully. Excision is curative and removes the symptoms immediately. If resection causes instability, then instrumentation and arthrodesis are recommended.\textsuperscript{[44]} Ozaki et al. applied repeated resection due to recurrence caused by incomplete resection on 2 osteoid osteoma patients who were treated with open resection. They published that scoliosis was corrected with surgical resection on 16 of 17 patients with osteoid osteoma or osteoblastoma.\textsuperscript{[17]} Burn et al. attained 73% and 38% successful results on 30 osteoid osteoma or osteoblastoma patients treated with and without surgery, respectively.\textsuperscript{[41]} The period of symptoms may affect postoperative correction of the spine deformity. Non-surgical treatment of osteoid osteoma may result in the disappearance of symptoms in 30–40 months.\textsuperscript{[21]} However, invasive treatment may be required if the pain is strong and causes restriction. Complete treatment is expected with complete surgical excision. If the lesion is resected within 15 months from onset, then the concomitant spinal deformities may spontaneously be fixed.\textsuperscript{[19]} Pettine and Klassen reported that scoliosis was corrected or regressed in 11 of 12 patients who experienced symptoms for <15 months preoperatively, but the symptoms continued in patients with symptoms >15 months.\textsuperscript{[19]} Aydinli et al. found surgical excision of the nidus sufficient on 4 patients as a result of 27 months average follow-up of 5 osteoid osteoma cases with spine involvement and scoliosis.\textsuperscript{[40]} We treated surgically with en bloc excision as reported by MacLellan\textsuperscript{[28]} and Aydinli\textsuperscript{[48]} due to the risk of structurality of compensatory scoliosis curve of our cases and failure to correct lumbar pains with drugs (Figure 2). Asymmetric muscle spasm, which causes scoliosis, disappeared after the lesions were removed. Sufficient correction was obtained without applying additional fusion or instrumentation. Growth continued after the lesion was removed, and major curvature of scoliosis deformity was corrected by an average of 79.6% (range: 48%–96.9%).

As noted by Crist et al.,\textsuperscript{[38]} ensuring complete removal of the nidus during surgery is critical. Various methods have been described, such as preoperative radioisotope injection and intraoperative localization with gamma counter, CT scanning of the specimen, or as described by Lenke et al.,\textsuperscript{[49]} identification of the localization by means of Kirschner wire. Intraoperative radiographies or preoperative radiographies may be compared with preoperative CT.\textsuperscript{[19,28]} Fusion is recommended upon the
risk of instability.[3,19] Pain usually disappears within hours or days following excision.[26] Remaining pain indicates that the tumor is not completely removed.[26] We implemented en bloc excision with a limited surgical approach by locating the lesion fluoroscopically during surgery with Kirschner wire in our cases.[49] The CT image taken preoperatively was used intraoperatively for localization of the lesion.[3] We observed that secondary scoliosis to the pain was corrected by removing the lesion with only limited surgical approach, supporting reports in the literature.

The weakness of the present study is the assessment of clinical and radiological results of the surgical treatment implemented, due to the limited number of patients. During treatment, only surgical excision was used in our cases; RFA could also be preferred for treatment. Therefore, detailed meta-analyses are required to assess different treatment methods through extended clinical series.

As a result, we believe that en bloc excision is sufficient without requiring an extra instrument and/or fusion material to fix scoliosis curve in patients <16 years of age, with major curve of ≤40°, and <22 months from onset of complaints.

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