

Retrospective Study

## Osteoid Osteoma and Osteoblastoma of the Cervical Spine: The Cause of Unusual Persistent Neck Pain

Tuncay Kaner MD<sup>1</sup>, Mehdi Sasani, MD<sup>2</sup>, Tunc Oktenoglu, MD<sup>2</sup>, Sabri Aydin, MD<sup>3</sup>, and Ali Fahir Ozer, MD<sup>2</sup>

From: <sup>1</sup>Pendik State Hospital, Istanbul, Turkey; <sup>2</sup>American Hospital, Istanbul, Turkey; <sup>3</sup>Cerrahpasa Medical Faculty, Istanbul, Turkey.

Dr. Kaner is with the Department of Neurosurgery, Pendik State Hospital, Istanbul, Turkey. Dr. Sasani, Dr. Oktenoglu and Dr. Ozer are with the Department of Neurosurgery, American Hospital, Istanbul, Turkey. Dr. Aydin is with the Department of Neurosurgery, Cerrahpasa Medical Faculty, Istanbul, Turkey.

Address correspondence: Tuncay Kaner, MD  
Pendik State Hospital  
Department of Neurosurgery  
Pendik/ Istanbul, Turkey  
Email: tkaner2002@yahoo.com

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**Background:** The most important symptom in patients with osteoid osteoma and osteoblastoma is a resistant localized neck pain and stiffness in the spine.

**Objective:** To evaluate and analyze 6 cases of osteoid osteoma and osteoblastoma of the cervical spine that were surgically treated over a 7-year period and to emphasize the unusual persistent neck pain associated with osteoid osteoma and osteoblastoma of the cervical spine.

**Study Design:** Retrospective study.

**Methods:** Six patients, 3 male and 3 female, with a mean age of 21 years (range 16-31) diagnosed with osteoid osteoma or osteoblastoma during 2003 to 2009 were analyzed retrospectively. The preoperative neurological and clinical symptoms, neck pain duration, preoperative deformity, location of lesion, radiological findings, surgical technique and clinical follow-up outcomes of each patient were evaluated.

**Results:** The average follow-up duration was 40.5 months (range, 19 to 83 months). Three patients had osteoid osteoma (2 female and one male), and 3 patients had osteoblastoma (one female and 2 male). Two male patients had recurrent osteoblastoma. The locations of the lesions were as follows: C7 (2 patients), C3 (one patient), C2 (one patient), C3-C4 (one patient) and C5-C6 (one patient). The most common symptom was local neck pain in the region of the tumor. Among all patients, only one patient, who had osteoblastoma, had neurological deficits (right C5-C6 root symptoms). The other patients had no neurological deficits. All patients were treated with surgical resection using microsurgery. Two patients underwent only tumor resection, one patient underwent tumor resection and fusion, and the other 3 patients underwent tumor resection, fusion and spinal instrumentation. No perioperative complications developed in any of our patients. There was no tumor recurrence during the follow-up period.

**Limitations:** A retrospective study with 6 analyses of cases.

**Conclusion:** Surgical treatment of osteoid osteoma and osteoblastoma of the spine has been standardized. The most common symptom of osteoid osteoma and osteoblastoma of the cervical spine is local persistent neck pain in the region of the tumor. This symptom can be significant in the diagnosis of these tumors.

**Key words:** neck pain, osteoid osteoma, osteoblastoma, bone tumors, cervical spine

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**O**steoid osteomas and osteoblastomas are benign bone tumors that rarely settle in the cervical spine (1-4). Osteoid osteoma was first described by Jaffe (5) in 1935, and afterward Jaffe

(6) and Lichtenstein (7) independently defined benign osteoblastoma as a different entity in 1956. The vast majority of these tumors are observed in patients younger than 30 years of age (4,8,9). Ten percent of

all osteoid osteomas and 40% of all osteoblastomas occur in the spine (10-13). The majority of such tumors are caused by the posterior elements of the spine (2,9). The most important symptom in patients with osteoid osteoma and osteoblastoma is a resistant localized neck pain and stiffness in the spine (14). The pain is initially uncertain and nocturnal but progressively becomes constant and strong in nature. The pain is worse at rest, especially at night, and is noticeably relieved by nonsteroidal anti-inflammatory drugs (NSAIDs) (15,16). Persistent neck pain is observed in osteoid osteoma, whereas in osteoblastoma, pain is accompanied by bone destruction, a bigger tumoral mass, and neurological deficits (2,10,14,17,18).

In this clinical study, we retrospectively reviewed 6 cases of osteoid osteoma and osteoblastoma in the cervical spine. We emphasize the importance of the resistant neck pain, which is the most common symptom of such cervical spine tumors.

## PATIENTS AND METHODS

Six patients (3 females and 3 males) who had been diagnosed with osteoid osteoma or osteoblastoma were surgically treated in the neurosurgery clinic at Istanbul American Hospital from 2003 to 2009. The mean age of the patients was 21 (range, 16 to 31). Preoperative neu-

rological and clinical symptoms, preoperative duration of neck pain, preoperative deformities, localization of the tumor, radiological findings, surgical technique and clinical follow-up durations of all patients were evaluated and recorded (Table 1). Computerized tomography (CT) and magnetic resonance imaging (MRI) were used in the radiological diagnosis of the patients. Furthermore, a bone scan was used to aid the radiological diagnosis in all patients. Clinical and radiological evaluations of the patients were performed in the preoperative period, early in the postoperative period, 6 months after surgery, and regularly every year after that. Tumor resections performed using microsurgical techniques were carried out in all patients. Special attention was given to complete removal of the tumor during the surgical approach. The nidus was resected in all osteoid osteoma cases, and the tumor cavity was filled up to the normal bone tissue. Tumor invasion was higher in osteoblastoma cases, and tumor tissue was resected widely until the paravertebral soft tissues were reached. Surgical techniques including tumor resection, fusion, and anterior and posterior instrumentation were performed in the patients. Kyphosis was corrected and fusion plus posterior instrumentation was applied to one patient with a kyphotic deformity due to recurrent osteoblastoma.

Table 1. Patient characteristics.

Patient	Age(y)/ Sex	Diagnosis	Localization (Level, Side)	Pain Duration (months)	Symptoms	Preoperative Neurological Findings	Operation	Follow- up (months)
1	22 female	OO	corpus (C7)	3	neck pain	normal	corpectomy + anterior fusion with allograft + anterior instrumentation with plaque and screw	32
2	31 female	OO	lamina (C3, left)	12	neck pain	normal	laminectomy	83
3	26 male	recurrent OB	lamina, facet joint (C3-C4, right)	18	neck and right arm pain	normal	laminectomy, facetectomy, fusion	21
4	16 male	recurrent OB	lamina, facet, pedicle (C7, left)	7	kyphotic deformity, neck pain and arm pain	normal	laminectomy, facetectomy, pedicle excision, posterior instrumentation	23
5	26 female	OB	lamina, facet (C5-C6, right)	1	neck pain and right arm pain	C5,C6 root findings	laminectomy, facetectomy, partial corpectomy, fusion, anterior and posterior instrumentation	19
6	25 male	OO	lamina (C2, left)	24	neck pain	normal	laminectomy	65

OO: osteoid osteoma; OB: osteoblastoma

## RESULTS

### Patient features

Out of the 6 patients, 3 (2 females, one male) were diagnosed with osteoid osteoma and 3 (one female and 2 males) were diagnosed with osteoblastoma. The mean age of the patients was 21 years (range, 16 to 31), and the mean follow-up period was 40.5 months (range, 19 to 83 months). Two male patients had recurrent osteoblastoma and had previously undergone operations in other centers.

### Symptoms and neurological findings

The most common and obvious symptom in patients was neck pain. The only observed symptom in 4 patients was neck pain. This pain can be characterized as ongoing, resistant to acetylsalicylic acid and other NSAIDs, localized in a particular region of the neck, and more severe at night. In 2 of the patients, arm pain was present together with neck pain. Kyphotic deformity was present in one patient who was admitted with recurrent osteoblastoma. Neurological deficits were detected only in the C5-C6 osteoblastoma case, and right C5, C6 root findings were present.

### Neuroimaging Findings

A CT scan with a bone window in conjunction with an MRI was used in the diagnosis of all cases. Additionally, a bone scan was used to aid the radiological diagnosis for all patients. The lesions were localized in the C7 vertebrae corpus in one case; C2 lamina in one case; C3 lamina in one case; C7 pedicle in one case; C3, C4

lamina and facet joints in one case; and C5, C6 lamina and facet joints in one case.

### Surgical procedures

All patients were operated on by the same senior neurosurgeon. The lesions in 2 of the patients, with the C2 and C3 osteoid osteomas, a 25-year-old male and a 31-year-old female (Fig. 1), respectively, were in the lamina, and a laminectomy plus tumor resection with a posterior approach was performed in these patients. A C3, C4 laminectomy plus right C3, C4 facetectomy plus fusion with calcium phosphate was used for the 26-year-old male patient with the C3-C4 recurrent osteoblastoma (Fig. 2). C7 corpectomy plus anterior fusion (with allograft) plus anterior stabilization with plaque was performed for a 22-year-old female patient with an osteoid osteoma on the C7 corpus. The 26-year-old female with an osteoblastoma starting from the posterior right C5-C6 and extending to the corpus by a holding pedicle, was treated with total tumor resection plus fusion plus anterior and posterior instrumentation; first with a posterior approach and then with an anterior approach. Another patient, a 16-year-old male, had a recurrent osteoblastoma stemming from the C7 lamina that caused facet and kyphotic deformities after undergoing surgeries in other institutions in previous years (2006 and 2007). This patient was treated with tumor resection, kyphosis correction, and posterior instrumentation plus fusion. No complications developed during the surgical procedures. Complete tumor resection was performed in all patients using microsurgery.

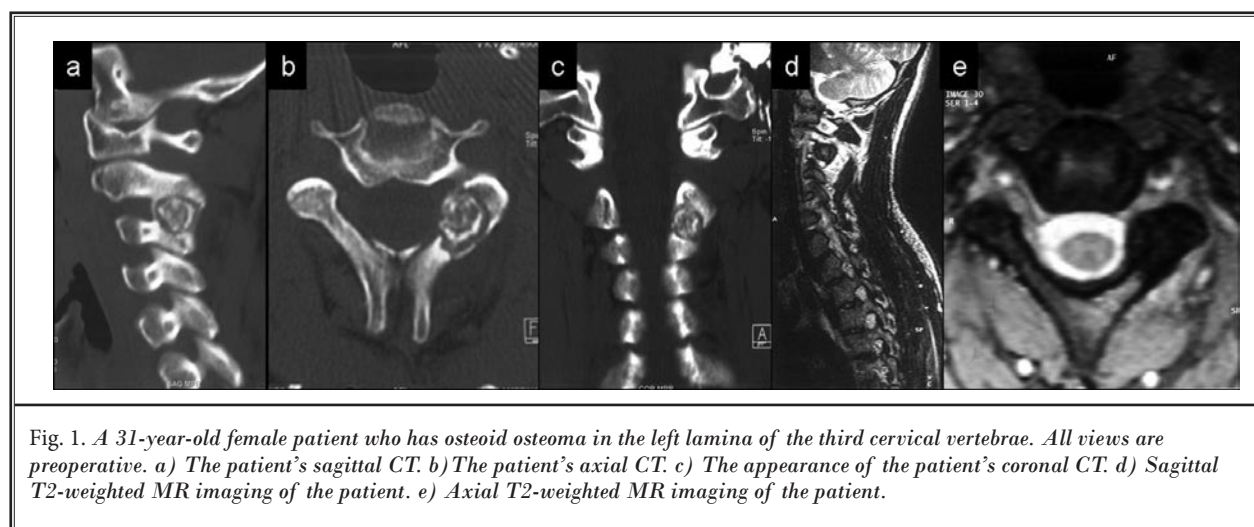


Fig. 1. A 31-year-old female patient who has osteoid osteoma in the left lamina of the third cervical vertebrae. All views are preoperative. a) The patient's sagittal CT. b) The patient's axial CT. c) The appearance of the patient's coronal CT. d) Sagittal T2-weighted MR imaging of the patient. e) Axial T2-weighted MR imaging of the patient.

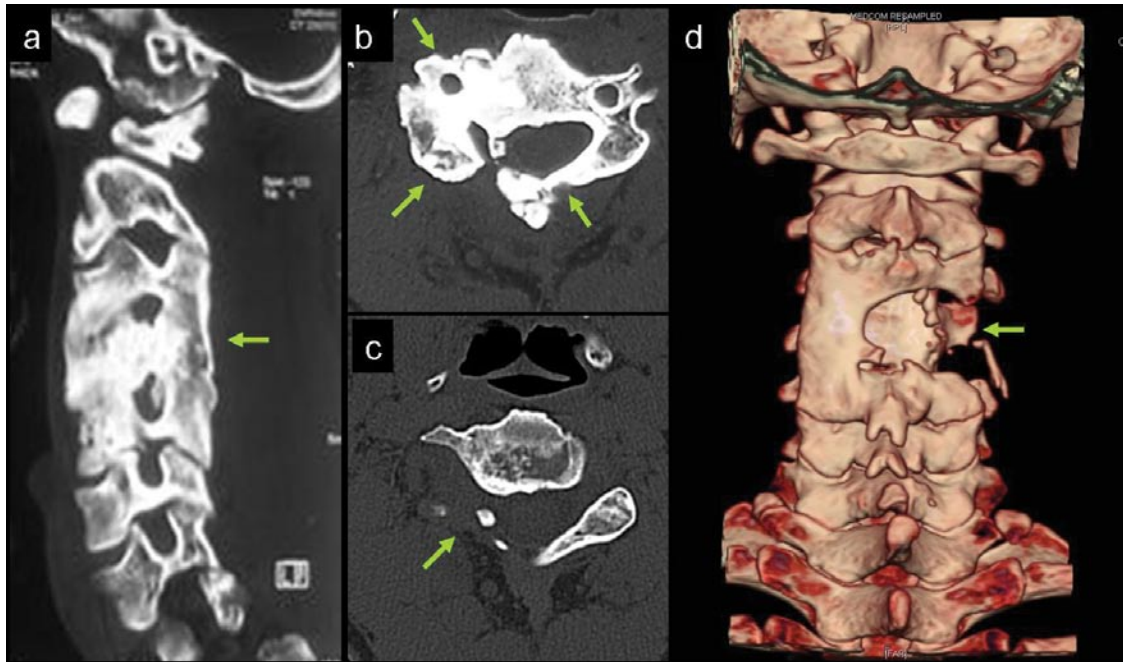


Fig. 2. A 26-year-old male patient who has recurrent osteoblastoma in the right C3-C4 lamina and facet joint. a) The patient's preoperative sagittal CT. b) The patient's preoperative axial CT. (In figures a and b the arrows represent recurrent osteoblastoma in the right C3-C4 lamina and facet joint.) c) The patient's postoperative axial CT. d) The patient's postoperative three-dimensional CT. (In figures c and d the arrows represent the region of resected recurrent osteoblastoma.)

### Follow-up

The patients were followed up in the early postoperative period, 6 months after surgery, and annually thereafter. An MRI and a CT scan were used in the radiological follow-ups of the patients. A highly significant decrease in pain was observed in all patients in the early postoperative period and at the last follow-up. No complications developed during our surgical procedures. We did not encounter any instrument failure or tumor recurrence during our follow-ups.

### Discussion

The differential diagnosis of patients, especially younger patients, with neck pain, painful scoliosis, spine stiffness, radicular pain in the arm, or pain extending toward the shoulder, should include cervical osteoid osteoma and osteoblastoma (2,10,14,17-21). Common symptoms with many other diseases might have considerably delayed the diagnosis of these diseases (21). Additionally, these tumors often cause pain before being visible on X-ray radiographs (13,19,22). In recent years, however, MRI or CT examinations have

often been used to diagnose patients with neck pain; as a result, delays in diagnosis have significantly decreased (21). Typical clinical symptoms of osteoid osteoma and osteoblastoma localized in the neck are similar. The earliest clinical symptom of osteoid osteoma and osteoblastoma of the neck is continuous local pain in the areas of the lesion, pain with increased activity, and increased pain which is particularly evident at night (2,3,8,14,19,23,24). Observed pain of this character usually responds well to aspirin and other NSAIDs (2,8,19,23). There might be moderate or severe sensitivity of the tumor tissue (19,25). Such tumoral lesions observed in the cervical spine might cause radicular-type lesions in the arms and shoulders (8,19,23,26-28). Since the symptoms of these tumors resemble those of lumbar and cervical disc herniation, such tumoral lesions are often treated as a disc herniation (19,29,30).

The most common and obvious symptom was local pain in the area of the tumor, and pain was continuous, as described by Zileli et al (2) for a series of 16 cases. Increased pain at night was observed in only 5 cases, and significant spine stiffness was determined in

12 cases. Bruneau et al (17) reported their surgical and clinical experiences in the removal of osteoid osteomas and osteoblastomas of the occipitocervical junction in 7 cases. All of their patients complained of persistent local pain that increased at night. They also reported spinal stiffness due to paravertebral muscle spasm in 3 of their patients. In their series of 42 cases with osteoid osteoma and osteoblastoma, Pettine et al (19) reported gradually increasing localized pain in almost all (41 cases) patients. Ninety-five percent of the cases exhibited increased pain with activity, and pain intensified at night. Twenty-nine percent of patients reported waking up with pain at night. Ninety percent of patients reported a reduction of pain with aspirin. In a series of 75 spine osteoblastomas cases, Nemoto et al (3) reported that 80% of the patients had pain and that 29% of the patients' pain was radicular in nature. Aspirin intake resulted in relief of pain in 27% of patients. The most obvious symptom was neck pain in our 4 cases, and pain was persistent and continuous even after intake of NSAIDs. Two of our patients had neck pain and pain extending to the arms. There were paravertebral muscle spasms and noticeable spinal stiffness in 3 of our patients. Four patients had increased pain at night.

As reported in previous publications, osteoid osteoma and osteoblastoma are usually seen in individuals under the age of 30 (2,4,8,9,17,19). The mean age of our patients was 21, and these patients had characteristics similar to those reported in the literature. Osteoid osteoma and osteoblastoma are more common in male patients, with approximately two-thirds of these tumors occurring in males (4,13). The male-to-female patient ratio was 1.6:1 in the study reported by Pettine et al (19), 2.5:1 in Nemoto et al and 2.5:1 in Bruneau et al (17). The ratio in our patients was 1:1.

Two important major symptoms of osteoid osteoma and osteoblastoma of the spine are scoliosis and neurological involvement (2). Scoliosis has been frequently reported in association with such tumors (10,14,19,21,24,31). Scoliosis occurs due to an inflammatory effect of the lesion in most patients (21). Ozaki et al (21) reported that scoliosis was a preoperative deformity in 17 of their 22 cases. Pettine et al (19) reported scoliosis in 2 of 12 patients with cervical lesions, out of 42 patients. Zileli et al (2) reported treating only one case for scoliosis among their 16 patients with osteoid osteoma and osteoblastoma. Bruneau et al (17) did not detect scoliosis in any of their 7 cases. Similarly, we did not detect scoliosis in any case. However, among our patients, one patient with recurrent osteoblastoma had a

kyphotic deformity. This deformity was corrected after tumor resection with posterior instrumentation during the operation. Neurological involvement is observed with osteoblastoma significantly more frequently than with osteoid osteoma (2,3,14,18,19,28). Neurological deficits up to 50% can be observed in patients with osteoblastoma (18). Among our patients, C5-C6 radicular involvement with neurological deficits was detected in only one patient with osteoblastoma; none of our cases with osteoid osteoma had neurological deficits.

Osteoid osteomas and osteoblastomas are bone-forming lesions. They are histologically similar to each other and often involve the posterior elements of the spine (2,3,9,10,18,21). The most important difference between these 2 tumors is the aggressive local expansion of osteoblastomas, which grow toward both the surrounding soft skeletal tissue and the neural arc (2,14,21). Osteoblastomas are diagnosed and differentiated from osteoid osteomas by size, as they are usually larger than 1.5 cm in diameter (3,9). In our series, one patient with osteoid osteoma had C7 corpus involvement; in all other cases, involvement was in the lamina, facet joints, and posterior elements containing the pedicle.

The preferred treatment option for osteoid osteomas and osteoblastomas is the complete removal of the tumor tissue (2,19,21). For osteoid osteoma, total resection including the nidus would ensure immediate pain relief in the postoperative period (21). Since the tumor mass in osteoblastomas is typically larger and more aggressively invasive, it should be treated more aggressively with wider bone resection (17). Particularly in osteoblastoma, if instability occurs after wide resection with removal of the facet joints, fusion should be added to the tumor resection (32). In a long-term follow-up study, Jackson reported an osteoblastoma recurrence rate of greater than 10% (33). Osteoblastoma recurrence is observed as a result of incomplete resection. The osteoid osteoma recurrence rate has been reported to be as low as 4.5% (13). Recurrence was not observed during our follow-up period, which was 40.5 months on average. We applied fusion in addition to tumor resection in our four cases exhibiting pedicle, facet joint, and vertebral corpus involvement.

## **CONCLUSION**

In cases of neck pain that are more severe at night and do not respond to medical treatment, accompanied by painful scoliosis and spinal stiffness, the existence of rare benign osteoid osteoma and osteo-



blastoma of the cervical spine should be considered, especially in young patients. The goal of surgical treatment of these tumors should be complete resection of the tumor. While tumor resection including the nidus might be sufficient in osteoid osteoma cases, a wider

tumor resection might be necessary to prevent recurrence in osteoblastoma cases. If structures that provide stabilization in the spine, such as facet joints and pedicles are included in the surgical resection, fusion should be included in the surgery.

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