CASE REPORT

Osteoid Osteoma of Odontoid: Case Report and Literature Review

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Abstract

Osteoid osteoma (OO) is a small tumor of bone that affects the spine in 10% of the cases. The tumor has a tendency to neural arc, and the lumbar spine is the most common site of presentation. Lesions of the odontoid process are very rare. We presented the case of a 20-year-old man who had cervical pain for 8 months. The pain responded to medical therapy. After investigation, there was a lytic lesion at the odontoid process with the characteristic features of OO in computed tomography scan and magnetic resonance imaging. Firstly, medical treatment was initiated with the administration of nonsteroidal anti-inflammatory drugs; however, due to adverse effects and worsening of his pain, the patient underwent surgical treatment with intralesional curettage from the transoral approach. In addition, the posterior fusion of the first and second cervical vertebrae was performed. The pain disappeared, and the patient was symptom-free after one year of follow-up.

Level of evidence: V

Keywords: C1-C2 fusion, Neck pain, Odontoid process, Osteoid osteoma

Introduction

Osteoid osteoma (OO) is a benign tumor that affects every bone in the body (1). The tumor accounts for 3% of all bone tumors (1). Half of the lesions are located in the tibia and femur and usually affect the cortical diaphyseal bone (2). Furthermore, 10% of this bone forming tumor involves the spine (3-5); however, 56.1%, 26.8%, 16%, and 1.1% of spinal OOs occur in the lumbar spine, cervical, as well as thoracic and sacral regions, respectively (5).

Neural arc is the most common site of involvement, and the tumor rarely occurs in the vertebral bodies (3, 5-9). Pain in the neck and back is the most common symptom that is aggravated by activity and usually becomes more severe at nights (9). Due to the complex anatomy of the spine, the diagnostic delay is common and may last for 1-2 years (10).

The tumor consists of central nidus lower than 15 mm in diameter often surrounded by sclerotic reactive bone (11). Medical treatment with nonsteroidal anti-inflammatory drugs (NSAIDS) can relieve the pain; nonetheless, it necessitates long-term administration (12). Surgical excision by intralesional curettage and marginal or wide resection can be performed on patients who failed to respond to conservative treatment (13).

Interstitial laser photocoagulation and percutaneous radiofrequency coagulation are treatment choices for OO outside the spine; however, the possibility of neural damage limits using these new techniques in spinal lesions (14). Limited cases of OO in the body and the

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odontoid process of the axis have been reported in the literature (5, 15-19). In the present article, a case of OO in the base of the odontoid and its treatment was reported in addition to a review of the literature.

Case presentation

A 20-year-old man was visited in the outpatient clinic with 8 months of neck pain that was radiated to the occiput. The pain was mild and intermittent and worsened at nights. On physical examinations, there was no tenderness. Cervical range of motion was complete and painful at the endpoints. During previous workup, the radiography of the neck was performed unremarkable for any pathologic lesions, and the case took indomethacin to relieve pain every day.

At our clinic, the patient underwent more workup with computed tomography (CT) scan, magnetic resonance imaging (MRI), and whole-body bone scan. In CT scan, there was a calcified nidus surrounded by lytic area and sclerotic rim in the right posterolateral region of the base of the odontoid process [figures 1A and 1B]. The MRI showed a low signal intensity in T1- weighted sequence and intermediate signal intensity at the central part with high signal intensity rim in T2-weighted sequence at the base of odontoid [figures 1C and D].

Bone scintigraphy was negative for any lesions in the cervical spine [Figure 1E]. Due to history, CT scan and MRI findings, and despite negative bone scan, the diagnosis of OO of odontoid was established. The medical treatment continued due to the surgical treatment as being a burden of morbidity to the patient and benign nature of the tumor. After 4 months of conservative treatment, gastrointestinal bleeding occurred, and the case could not take NSAIDs; therefore, the pain worsened and became persistent. At this time, it was decided to perform surgery on the patient.

The lesion was excised through the transoral approach. Firstly, the anterior ring of atlas and anterior part of odontoid were partially removed using a micro drill (Stryker Co., USA). Then, the nidus was removed

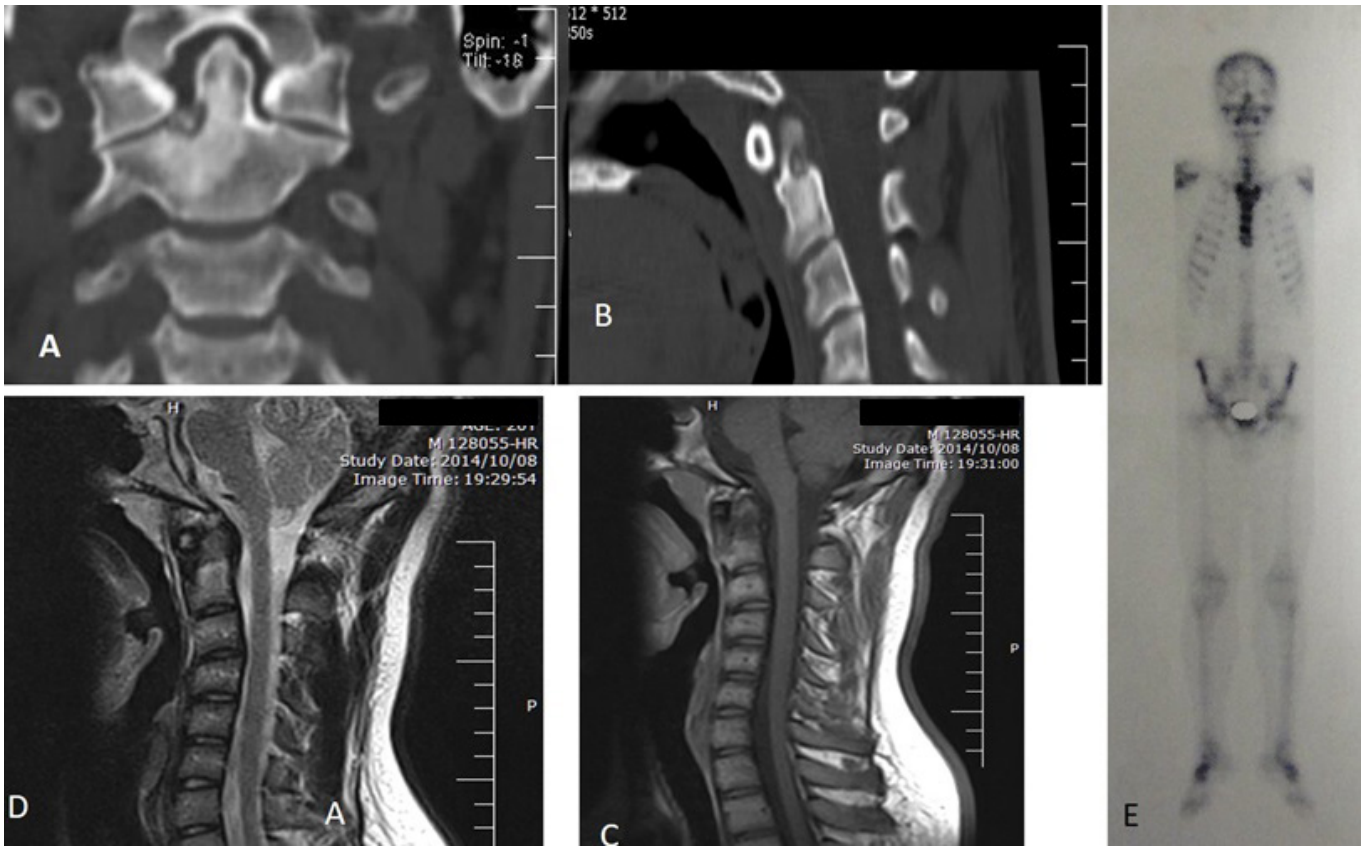


Figure 1A. Coronal computed tomography scan of cervical spine showing the presence of the lesion at the odontoid process with central nidus and sclerotic rim.
Figure 1B. Sagittal computed tomography scan of cervical spine showing the lesion.
Figure 1C. Lesion with low signal intensity in T1-weighted magnetic resonance imaging.
Figure 1D. Lesion appearing as intermittent signal central nidus and high signal peripheral rim in T2-weighted magnetic resonance imaging.
Figure 1E. Lesion with no increase of uptake in bone scintigraphy.

Figures 2a and 2b. Presentation of complete removal of lesion by postoperative sagittal and coronal computed tomography scans.

Figure 3. Patient undergoing the first and second cervical vertebrae fusion.

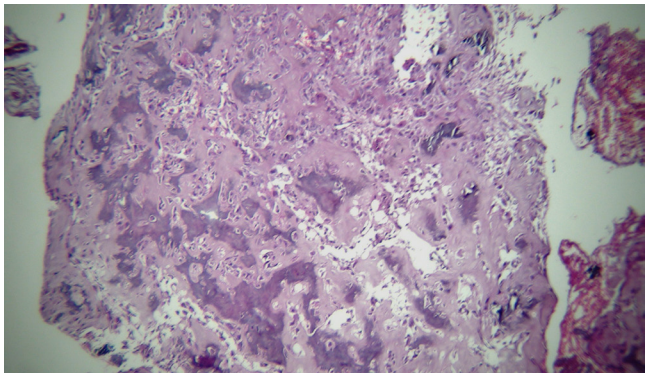
by means of a curette. Due to the posterior location of nidus in the odontoid process, there was no access to the lesion without the removal of the anterior part [figures 2a and b].

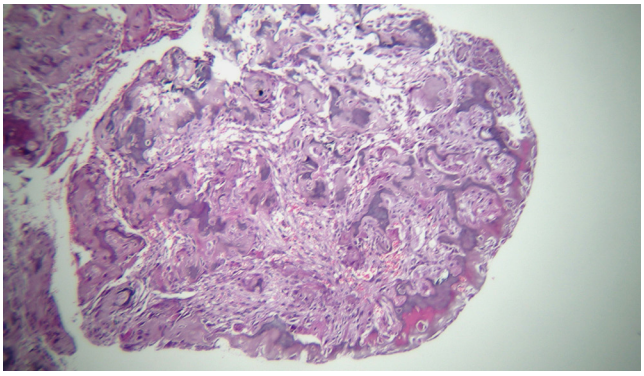
Because C1 anterior ring and base of the odontoid process were removed, the surgeons were concerned about the atlantoaxial instability and its catastrophic consequences. The first and second cervical vertebrae (C1-C2) fusion was performed using C1 and C2 pedicle screws construct posteriorly [Figure 3]. Histopathologic examination confirmed the diagnosis [Figure 4]. Symptoms of the patient disappeared completely at the postoperative night. During one year of follow-up, the results were favorable without any significant disabilities.

Discussion

The OO is a benign tumor occurring in the second decade of life with a twice-higher predilection in males than that in females (4). In the spine, the incidence of tumor in the anterior elements is rare, and the occipitocervical junction involvement is also uncommon in the cervical spine (20). In the present case, the base of odontoid process was the site of lesion. The radiography was unremarkable for the lesion due to the complexity of spinal anatomy and overlapping of bony structures, such as this case (21). The CT scan of OO is diagnostic and defines the extent of the tumor accurately (22).

The OO can be conservatively managed with the administration of NSAIDS and aspirin; however, the duration of the treatment regimen is long, and the side effects of drugs preclude its use (12, 23, 24). In the present case, the pain was firstly controlled with the administration of indomethacin; nonetheless, gastrointestinal bleeding medical treatment. Another aspect of the present case was the negative results of bone scan without the increase of uptake in the tumor site. Results of previous studies demonstrated that the





Figures 4a and 4b. Histopathologic images showing the presence of anastomosing trabeculae of osteoid and woven bone rimmed by a single layer of benign activated osteoblasts interspersed by fibrovascular stroma.

sensitivity of bone scan for the detection of OO is 100% (25-28). However, the findings of other studies showed false-negative results, and it is recommended that patient symptoms and CT scan are enough for the diagnosis of OO (29, 30).

Bucci reported an OO at the base of odontoid in a 7-year-old boy with atlantoaxial rotational limitation that was excised through the transoral approach (15). In a case series of 11 patients with spinal OO, Raskas reported a 6-year-old boy with the lesion observed in the body of C2 that was resected without fusion (5). Another reported case was a 15-year-old boy by Molloy. The tumor existed in the posterior body of C2, and it was excised through the anterior approach. In addition, C1-C2 fusion was avoided, and post-operative halo vest was used for 6 months (16). Al-Balas also excised OO from the odontoid process of C2 anteriorly in a 16-year-old male. The C1-C2 was preoperatively auto-fused due to the intraarticular nature of lesion and inflammatory reaction in atlantoaxial joint (17).

There were two cases of OO in the odontoid process who were administered with NSAIDS for conservative management. The cases included a 14-year-old girl who took celecoxib for 2 years with disappearing of nidus after this period and an 18-year-old boy who was reported without any symptoms after 2 years of follow-up (18, 19). Gasbarrini in the review of 81 cases of OO in the mobile spine observed a case of tumor in the body of C2 who underwent open biopsy and excision through a minimally anterior approach by percutaneous lead tunnel and C2-C3 fusion with minimal plate application (14).

The OO of odontoid can be conservatively managed; however, the duration of drug intake is extremely long, and the complication of medication may be the prevention of use. The operative approach is performed through an anterior approach with the complete excision of the nidus. Moreover, if there is atlantoaxial instability after tumor resection, C1 should be fused to C2 with the expense of cervical rotation.

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