Abstract

Osteoid osteoma is a small tumor of bone which affect spine in 10% of cases. The tumor has tendency to neural arc and the lumbar spine is most common site of presentation. Lesions of odontoid process are very rare. We present a 20-year-old man who had cervical pain for 8 month. The pain **responded** to medical therapy. After investigation there was a lytic lesion at the odontoid process with characteristic features of osteoid osteoma in CT-scan and MRI examination. At first medical treatment with nonsteroidal anti-inflammatory drugs was initiated but due to adverse effects and worsening of his pain the patient underwent surgical treatment **with intralesional curettage from transoral approach and** posterior C1-C2 fusion was performed. The pain disappeared and, after one year fallow up, the patient was symptom free.

Introduction

The osteoid osteoma (OO) is a benign tumor which 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 diaphysial bone (2). Ten percent of this bone forming tumor involve the spine (3-5), while 56.1% occurs in the lumbar spine, 26.8% in the cervical and the remaining in the thoracic (16%) and sacral (1.1%) regions (5). Neural arc is the most common site of involvement (3, 5-8) and the tumor occurs rarely in the vertebral bodies (9). Pain in the neck and back is the most common symptom which is aggravated with activity and usually is more severe at nights (9). Due to the complex anatomy of the spine, diagnostic delay is common and may last for 1-2 years (10).

The tumor is consisted of central nidus which is less than 15 mm in diameter often surrounded by sclerotic reactive bone (11). Medical treatment with non-steroidal anti-inflammatory drugs (NSAIDS) can relieve the pain but it necessitates long term administration (12). In patients who failed to respond to conservative treatment surgical excision by intralesional curettage and marginal or wide resection can be performed (13). Interstitial laser photocoagulation (ILP) and percutaneous radiofrequency coagulation (PRC) are treatments choices for OO outside the spine, but the possibility of neural damage limits using these new techniques in spinal lesions (14).
Limited cases of OO in the body and odontoid process of the axis have been reported in the literatures (5, 15-19). In current article, a case of OO in the base of the odontoid and its treatment was reported in addition to review of the literatures.

Case presentation

A 20-year-old man visited in the outpatient clinic with 8 month of neck pain which was radiated to the occiput. The pain is mild and intermittent and become worsen at nights. On physical examination there is no tenderness. Cervical range of motion was complete which was painful at the end points. During previous workup radiography of the neck was taken which was unremarkable for any pathologic lesion and he took indomethacin every day for relieving pain. At our clinic he 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 posterlateral region of the base of odontoid process (figure 1a, b). 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 (figure 1c, d). Bone scintigraphy was negative for any lesion in the cervical spine (figure 1e). Due to history and CT scan and MRI findings and despite negative bone scan, the diagnosis of osteoid osteoma of odontoid established. Because the surgical treatment was burden morbidity to patient and the benign nature of tumor medical treatment continued. After 4 month of conservative treatment gastrointestinal bleeding occurred and he could not intake NSAIDs and pain worsen and become persistent. At this time we decided to operate the patient. **The lesion was excised by transoral approach. At first we removed the anterior ring of atlas patially and anterior part of odontoid using micro drill then the nidus was removed by means of curette**. **Due to posterior location of nidus in odotoid process we don`t have access to the lesion without removing the anterior pa**rt (figure 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. C1-C2 fusion was performed using C1 and C2 pedicular screws construct posteriorly (figure 3). Histopathologic examination confirmed the diagnosis (figure 4). The patient symptoms **were** disappeared at the postoperative night completely. At one year fallow up the results was favorable without any significant disability.

Discussion

Osteoid osteoma is a benign tumor of the 2end decade of life with male are affected 2 time more than female (4). In spine the occurrence of tumor in the anterior elements is rare and in the

cervical spine the occipitocervical junction involvement is also uncommon (20). In the presented case the base of odontoid process is the site of lesion. The radiography was unremarkable for the lesion because of the complexity of spinal anatomy and overlapping of bony structures such as this case (21). Ct scan of OO is diagnostic and accurately define the extent of the tumor (22). OO can be managed conservatively with application of NSAIDS and aspirin but the duration of treatment regimen is long and the side effects of drugs preclude its use (12,23,24). In our patient the pain managed with indomethacin at first but gastrointestinal bleeding **excluded** medical treatment. The other aspect of our case is negative bone scan results without increase uptake in the tumor site. Some previous studies demonstrate that the sensitivity of bone scan for detection of OO is 100% (25-28) but others showed false-negative results and recommended, history and CT scan are enough for diagnoses of OO (29, 30).

Bucci reported an OO at the base of odontoid in 7 years old boy with atlantoaxial rotational limitation which is excised through transoral approach (15). Raskas in a case series of **11 patients** of spinal OO reported a 6 years old boy with tumor in the body of C2, which is resected without fusion (5). The other reported case was by Molloy in 15 years old boy. The tumor existed in the posterior body of C2, and excised through anteromedial approach to the upper cervical spine, and they avoid C1/C2 fusion and used halo jacket post-operative for 6 **months** (16). The Al-Balas also excised OO from odontoid process of C2 anteriorly in **16-year-old** male. The C1-C2 was auto fused preoperative because of intraarticular nature of lesion, and inflammatory reaction in atlantoaxial joint (17). There were two case of OO in the odontoid process who take NSAIDS for conservative management. 14-year-old girl who take celecoxib for as long as 2 years with disappearing of nidus after this period, and 18 years old boy who were without symptom after two years of follow up (18, 19). Gasbarrini in the review of 81 case of OO in the mobile spine found one case of tumor in the body of C2 which is underwent open biopsy and excision through an anterior minimally approach by percutaneous lead tunnel and C2-C3 fusion with minimal plate application (14).

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

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Legends

Figure 1a. There is a lesion at the odontoid process with calcified nidus and lytic rim, CT-scan coronal cut

Figure 1b. The lesion at the CT-scan sagittal cut

Figure 1c. In T1-waighted MRI the lesion has low signal intensity

Figure 1d. InT2-waighted MRI the lesion appear as intermittent signal central nidus and high signal peripheral rim

Figure 1e. The lesion does not increased uptake in bone scintigraphy

Figure 2a and b. **Post operation sagittal and coronal CT-scan show the lesion was completely removed.**

Figure 3. **Patient underwent C1-C2 fusion**

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