

Osteoid Osteoma of the Atlas: Report of a New Case and Review of the Literature

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ABSTRACT

The upper cervical region and in particular the atlas remain very rare locations for osteoid osteomas. Herein a 30-year-old male with osteoid osteoma of the atlas arch and a history of severe neck pain for a year is presented. After establishment of the diagnosis by isotope and C.T. scan, tumor removal was accomplished through atlas arch laminectomy resulting in complete relief of pain. A literature review revealed that only four similar cases have been reported previously.

KEY WORDS: Atlas, cervical spine, osteoid osteoma, spine tumors

INTRODUCTION

Osteoid osteoma is the most common primary benign spinal tumor occurring in children and adolescents where more than 50% of the cases are seen in the second and third decade of life.

Osteoid osteoma was described for the first time by Bergstrand in 1930. Later, in 1935, Jaffe defined this tumor as a benign osteoblastic tumor of bone (3, 4, 10-13, 15). Osteoid osteomas are self-limited tumors and are small, ranging in size from 0.5 to 2 cm. This tumor is mostly found in the thoracolumbar spine followed by the cervical spine (3,4,10-13,15). The upper cervical spine remains the least common location (1,9). The tumor is typically located at the posterior elements of the spine.

Recently, in our practice, we were faced with a 31-year-old man suffering from severe nocturnal neck and occipital pain with a positive aspirin test. Cervical MRI was suspicious for a tumor at the arch of the atlas, but isotope scan clearly showed a hot spot at the lamina of C1, compatible with osteoid osteoma. Subsequent computerized tomography confirmed the diagnosis of osteoid osteoma of the atlas arch and clarified the details. Hemi-laminectomy of the atlas arch resulted in complete relief of the pain. With careful review of the literature, we could find only 5 cases of osteoid osteoma

of the atlas reported previously, 4 of which were located at the lamina and one on the lateral mass (2,5,8).

CASE REPORT

A 30-year-old man was admitted with a one-year history of the neck as well as occipital pain and spasm of the neck muscles. Examination revealed tenderness at the posterior aspect of the upper cervical spine with deep palpation whereas his neurological exam was normal.

Plain radiographs were normal. However, MRI was suspicious for pathology at the arch of the atlas. A technetium bone scan demonstrated a hot spot in the posterior arch of the atlas compatible with an osteoid osteoma. Subsequently, a C.T. scan of the upper cervical spine was performed with narrow slices that demonstrated a small right-sided tumor in the atlas arch with a sclerotic margin and a nidus inside the details of the tumor. Treatment with aspirin relieved the pain and ameliorated nuchal spasm. The risk-benefit of NSAIDs versus surgery was described and the patient chose surgery.

Via a mid-line incision, the paravertebral muscle was stripped off the laminae of C1. Subsequently, the tumor was removed along with the lamina of the atlas through an appropriate hemi-laminectomy.

Post-operatively, the patient's pain and neck spasm were completely relieved. Now, at 2 years follow up, the patient is normal and free of any pain.

DISCUSSION

Osteoid osteoma is a common primary tumor of the vertebral column. The series of Levine et al. included 41 patients treated during a 36-year period, and 18 cases were osteoid osteoma whereas none was located at the atlas (9). Furthermore, osteoid osteoma was found in 20 out of 61 primary tumors of the cervical spine reported by Abdu et al, but the atlas was not affected in any.1 In the current review, only 5 cases could be found in the literature (Table 1).

This tumor is composed of an osteolytic defect and sharp sclerotic margins. The osteolytic compartment contains osteoblasts that produce a soft osteoid and a nidus. The nidus consists of an irregular network of osteoid trabeculae with highly vascularized stroma and varying degrees of calcification presented as woven bones (3, 4, 10-13).

Osteoid osteoma usually measures from 0.5 to 2 cm in size and might be located in cancellous, sub-periosteal or cortical part of a bone (3, 4, 10-13, 15). The most common and classical form is the cortical type while sub-periosteal or cancellous ones remain less frequent types.

Males are more commonly affected than females with ratio of 3 to 1. Actually, more than 50% of the cases occur in young adults between the ages of 15 and 25 (3,4,10-13,15). However, it rarely occurs before the age of five and after the age of 40. The predilection site of osteoid osteoma in the vertebral column is the lumbar. The dorsal and the cervical spine are affected in decreasing frequency and the upper cervical region remains the least frequent site of formation (3, 4, 10-13, 15). Actually, osteoid osteomas are distributed throughout all levels except C1 and C2 (1,3,4,9-13,15). In careful review of the literature, we could find only 7 cases with axis involvement and only 4 cases affecting the atlas.

This tumor predominantly involves the posterior elements, such as the spinous processes, transverse

processes, facets, lamina, and pedicles while it rarely prefers the vertebral body. 3 out of 4 surveyed cases including the current case were in the lamina of atlas and the other two in the lateral mass (2, 5, 8).

The clinical picture of osteoid osteoma of the cervical spine is local neck pain that is maximum at night. Dramatic relief of pain by Aspirin can be used a screening test for the clinical diagnosis of this tumor (1, 3, 4, 9-13, 15). However in those with cervical spine involvement, varying degree of torticollis might be seen. Torticollis is a typical presenting feature in 10 to 100% of those with cervical involvement. 1,9Torticollis was seen in 2 out of 4 cases with atlas involvement (2, 5, 8).

Unfortunately, despite classical nocturnal pain, diagnosis of this benign tumor is often delayed. Conventional radiographs usually fail to show the lesion. Actually, these small tumors are easily obscured among the overlapping shadows of the cervical spine.

Bone scintigraphy is the most sensitive screening test and can provide accurate localization of the tumor (3, 4, 10-13, 15). Actually, early diagnosis of an osteoid osteoma can be achieved only by this tool in all cases with unexplained local pain. Thin axial slices and reformatted C.T. images help define both the accurate shape and the exact size of the tumor, but only after scintigraphy has shown the site of involvement (2, 5, 8). However, if the cuts are at the wrong level or too wide, the tumor might be missed completely.

GD-enhanced MRI focusing on the suspected site might clearly show and detect the tumor but is less sensitive than CT scan where scintigraphy remains the most sensitive test (2, 5, 8).

Nonetheless, after establishment of the diagnosis of osteoid osteoma of the atlas and localizing the site, the treatment in all cases has been surgical excision. In involvement of the neural arch, laminectomy of the corresponding lamina will ensure the surgeon that the tumor is completely removed (2, 5, 8). Other modes of surgery such as en bloc excision,

Table 1:

Author	Year	Sex	Age	Location	Surgery	Outcome
Jones	1987	M	8	Lamina	En-block resection	Good
De Praeter et al	1999	M	21	lamina	Hemilaminectomy	Good
De Praeter et al	1999	M	22	Lamina	Hemi-laminectomy	Good
Amirjamshidi et al	2007	M	14	Lateral mass	Drilling	Good
The Current case	2014	M	31	Lamina	Hemilaminectomy	Good

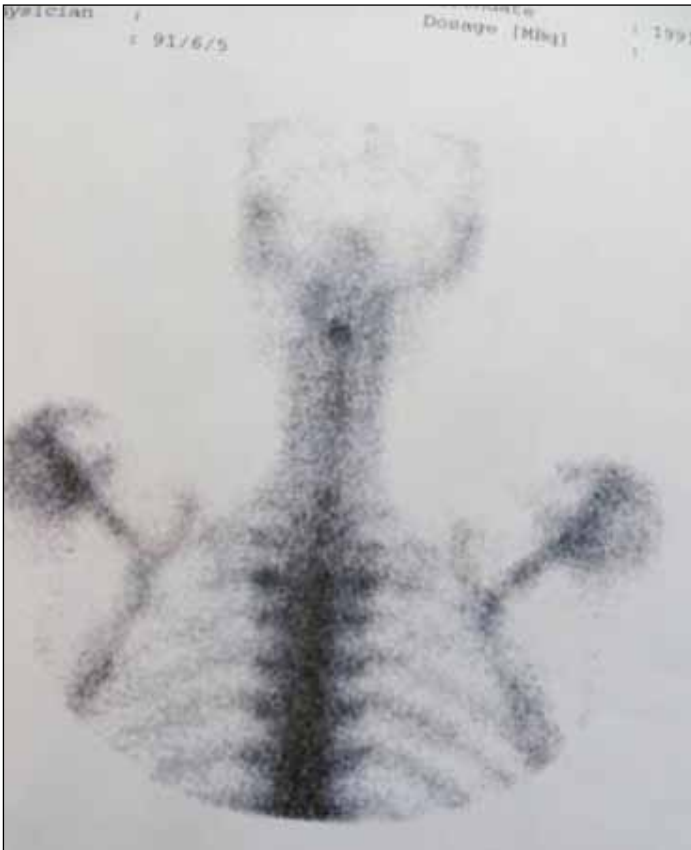


Figure 1: Isotope scans showing a hot spot in the upper cervical spine.

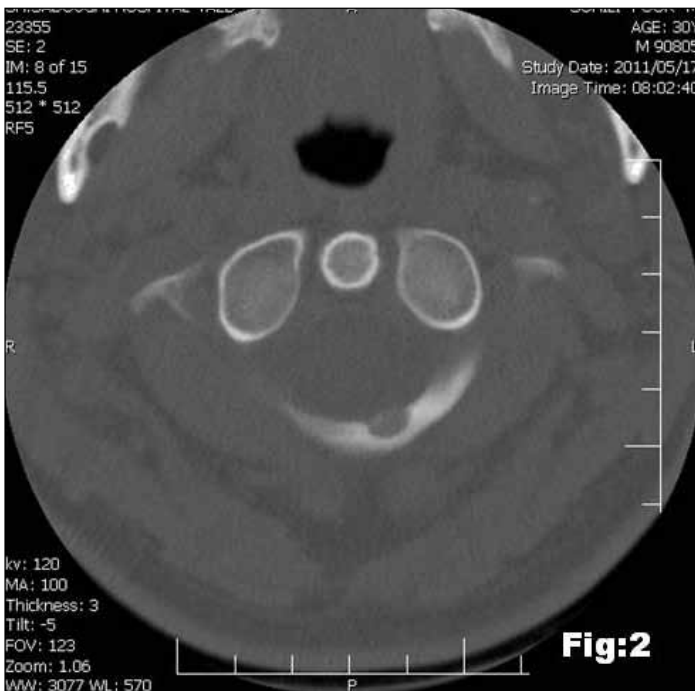


Figure 2: Axial C.T. scans through the atlas showing an osteolytic lesion in the lamina.

curettage and drilling may provide similar results, although the chance of recurrence exists. Laminectomy was applied in 4 out of 5 reviewed cases including the current case. The remaining one was managed by interlesional drilling (2, 5, 8). Regardless of the mode of surgery, tumor removal provides reliable relief of pain and the coexisting deformity disappears. Pain disappears soon after excision while torticollis resolves within a few days to a month (2, 5, 8).

Recently, with application of radiofrequency ablation of the osteoid osteomas located at the skeletal bones, success could be easily achieved (6,7). However, careful and safe application of this method in osteoid osteoma of the vertebral column is needed with regard to possible thermal damage to the neural structures. This method might be useful in tumors where the site of involvement is hardly accessible by surgery, but far enough from neural tissues (6, 7, 14). In those with comorbidities, or in those with a less accessible location, long-term conservative treatment with Aspirin or non-steroid anti-inflammatory drugs (NSAIDs) might result in disappearance of the tumor and its ultimate solidification (7).

In summary, the upper cervical region is rare location of osteoid osteoma. As imaging continues to evolve, the diagnosis of these lesions can become easier and made earlier. Involvement of the lamina of the upper cervical region is the simplest case that can be managed surgically either via tumor resection or through laminectomy. However, management of osteoid osteomas located at the pedicle or the body is a challenging issue that requires special consideration. Besides surgical intervention, recent evolution made by percutaneous CT guided radiofrequency has made the precise non-surgical resection of these tumors possible. Conservative treatment can be applied in those cases located at a non-accessible location or in those with serious comorbidities

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