Atlanto-axial rotational limitation secondary to osteoid osteoma of the axis

Case report

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An unusual case of atlanto-axial rotational limitation secondary to an osteoid osteoma of the axis is presented. Transoral microsurgical resection followed by physical therapy improved the clinical symptoms. This case illustrates several unique problems within the cervical spine as well as the efficacy of the transoral approach to the axis.

KEY WORDS spine neoplasm cervical spine osteoid osteoma transoral approach

ACQUIRED torticollis refers to a rotational disorder of the upper cervical spine usually at the atlanto-axial joint. Etiologies include inflammatory processes, trauma, and neoplasia. A case of atlanto-axial rotational limitation secondary to an osteoid osteoma of the axis is presented. Conservative therapy failed to relieve the clinical symptoms, thereby requiring surgical resection via a transoral approach.

Case Report

This 7-year-old boy sustained three separate minor cervical hyperextension injuries over 1½ years, the most recent in February, 1987. Each trauma resulted in the patient turning his chin to the right and tilting his head to the left, with persistence of the deformity. His most recent episode lasted approximately 6 weeks and prompted neurosurgical consultation.

Examination. Neurologically, the boy was bright, alert, and appropriate, with normal mental status. He held his head in a position of rotation to the right with a slight left tilt. He complained of pain in the left paracervical region when the head was further tilted to the left of midline. The second through 12th cranial nerves were normal. Sensory examination was normal throughout with regard to pinprick, light touch, and position sense bilaterally. Reflexes were 2+ throughout and symmetrical, with flexor plantar responses bilaterally. Strength was normal in all four extremities. Gait and cerebellar testing were intact.

Cervical spine x-ray films were unremarkable. Dynamic x-ray computerized tomography (CT) through the upper cervical spine showed rotatory limitation as well as a sphere-shaped low-attenuation lesion with a high-attenuation central nidus and sclerotic edges. These imaging characteristics suggest an osteoid osteoma.

FIG. 1. Computerized tomography scans through C1–2 showing rotational limitation as well as a left-sided low-attenuation axis lesion with a high-attenuation central nidus and sclerotic edges. These imaging characteristics suggest an osteoid osteoma.
area of greatly increased activity in the upper cervical spine at the C1–2 region near the midline.

Operation. The patient underwent surgery in May, 1987. Halo traction was applied for intraoperative stabilization, and a tonsillectomy was carried out to facilitate exposure. Intraoperative bone scanning revealed an area of intense uptake in C-2. An inferiorly based pharyngeal flap was turned, and the anterior aspects of the C-1 and C-2 vertebral segments were exposed. With the operating microscope and high-speed drills, the junction of the dens and the C-2 vertebral body was explored. A soft vascular mass, 1 cm in diameter, was removed from the left superior aspect of the C-2 vertebral body. Repeat intraoperative bone scanning showed no persistent uptake.

Postoperative Course. No postoperative problems were encountered. Flexion-extension cervical spine x-ray films obtained 2 days postoperatively showed no instability. The patient was then removed from cervical traction and placed in a cervical collar. The range of motion of the cervical spine improved steadily. Aggressive physical therapy resulted in full range of motion within 3 months following tumor removal.

Discussion

Osteoid osteoma was first described in the English literature in 1935 by Jaffe. Although frequently located in the femur and tibia, approximately 10% of osteoid osteomas are located in the spine. This is in contradistinction to osteoblastomas, which occur approximately 35% of the time in the spine. The lumbar spine is the most frequently involved site, followed by the cervical spine. Approximately 75% of spinal osteoid osteomas involve the neural arch, while only 7% occur in the vertebral body.

Histopathologically, these lesions consist of trabeculae of osteoid containing varying amounts of mineralization supported by a vascular connective tissue stroma, together with newly formed bone. Some authors consider that osteoid osteoma is virtually indistinguishable from osteoblastoma histologically although there appears to be less osteoblastic differentiation with osteoid osteoma. 5,8,18

There is a 2:1 male to female predominance among patients with this disorder, and 90% of patients present at 30 years of age or less. Pain is the chief complaint and frequently antedates radiological findings by several months; in many cases the pain is relieved by aspirin intake. Often the pain is exacerbated at night. Additionally, there may be associated musculoskeletal deformities and growth disturbances such as scoliosis, torticollis, myelopathy, limb atrophy, and radicular pain. 1,11,18,20

Conventional radiographs are frequently normal, especially in the axial skeleton with multiple superimposed bone structures. A radiolucent nidus, under 1.5 cm in diameter with surrounding sclerosis, is pathognomonic of osteoid osteoma. Technetium pyrophosphate bone scans appear to be positive in all cases, showing intense photon activity in the lesion. Bone scans are particularly useful in patients with nonlocalizing symptoms or referred pain. 18,20

A CT scan most accurately demonstrates the characteristic radiographic features of osteoid osteoma: 1) a well-defined oval or round low-attenuation area (nidus); 2) a high-attenuation region in the center of the nidus surrounded by a low-attenuation halo; and 3) varying degrees of surrounding reactive bone sclerosis. The high-attenuation region in the center of the lesion represents mineralized osteoid. The reactive changes surrounding this region are secondary to cancellous sclerosis due to exuberant periosteal reaction, which subsequently results in thickening of the surrounding bone. 4

The natural history of osteoid osteoma is not readily discernible. In 1951, Dockerty, et al., 7 reported a suspected case of a femoral neck osteoid osteoma that disappeared over 7 years time without surgery. Vickers, et al., 10 reported an untreated femoral shaft osteoid osteoma that remained unchanged radiographically during a 15-year follow-up period. Interestingly, the patient's pain diminished progressively over the 15-year period. Similarly, in 1975, Simms 15 reported 20 cases of suspected osteoid osteoma that were not operated on. He also found that over time the pain decreased but the radiological features persisted. In 1956, Sabanas, et al., 13 proposed that suspected osteoid osteoma may be self-limiting and could be treated nonoperatively; however, recent authors have advocated surgical excision as definitive treatment. 3,5,6,8,10,20,21

Marginal resection either en bloc or by curettage appears to be the treatment of choice. Incomplete resection was associated with a 4.5% recurrence rate in 860 cases of osteoid osteoma. 8 No cases of malignant transformation have been reported in osteoid osteoma. This is in contradistinction to osteoblastoma, where cases of malignant transformation to sarcoma have been reported. 3,5,10 Total excision is curative; to date there have been no reported recurrences following complete resection. 3,20,21

The present case was unique in that the atlanto-axial rotational limitation was most likely secondary to an osteoid osteoma involving the axis, which has not been previously reported. Treatment with conservative therapy failed, and operative therapy was required. The most direct approach to the lesion was by the transoral technique, which has been previously described. 1,14,16 En bloc resection was successfully accomplished.

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