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CASE REPORT

Case Report

Osteoid Osteoma of the C2 Pedicle

Surgical Technique Using a Navigation System

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Abstract An osteoid osteoma of the cervical spinal pedicle is rare and carries a high surgical risk because of the close anatomic relationship to the spinal cord, nerve root, and vertebral artery. We report the case of a 12-year-old girl with an osteoid osteoma of the C2 pedicle. Computed tomograms showed an oval nidus and marked sclerosis around this lesion at the right C2 pedicle. There also was expansion of the medial and inferior cortical bone of the C2 pedicle. After failure of nonoperative treatment, we planned surgery. Owing to concerns regarding thermal damage to the spinal cord, nerve root, and/or vertebral artery using computed tomography (CT)-guided radiofrequency ablation, we curetted the nidus using a navigation system. Twenty-eight months after surgery, her pain was relieved with no limitation of cervical movement and there has been no evidence of recurrence. Navigation allowed safe curettage of the nidus through a small hole while maintaining spinal stability.

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Each author certifies that his or her institution has approved the reporting of this case report, that all investigations were conducted in conformity with ethical principles of research, and that informed consent for participation in the study was obtained.
This work was performed at Tottori University.

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Introduction

An osteoid osteoma is a benign bone tumor that comprises approximately 3% of all bone tumors and approximately 10% of all benign bone tumors [10]. The spine is affected in 10% to 25% of all cases, with 26.8% of these cervical, 16% thoracic, 56.1% lumbar, and 1.1% sacral [26]. An osteoid osteoma in the spine shows a 70% to 100% propensity for posterior element involvement [6, 8, 16]. Excluding the posterior elements, cervical spinal osteoid osteomas have been reported in the dens axis [22], the lateral mass of C1 [4] or C5 [7], the vertebral body of C2 [4, 20] or C3 [28], the transverse process of C2 [2] or C4 [21], and the pedicle of C3 [29], C5 [25, 29], or C6 [18]. To our knowledge, however, there has not been any report of an osteoid osteoma in the pedicle of C2.

Surgery generally is recommended for patients who do not respond to treatment with antiinflammatory drugs [14]. However, surgery for an osteoid osteoma of the cervical spine, especially the pedicle, carries a risk of damage to the spinal cord, nerve root, and/or vertebral artery [29].

We report a patient with an osteoid osteoma of the C2 pedicle treated by computer-assisted surgery to minimize the risks of neural and/or arterial injury and to maintain spinal stability.

Case Report

A 12-year-old girl was referred to our hospital with an 8-month history of neck pain. There was no history of injury. For the first 4 months, she had intermittent neck pain, but thereafter severe neck pain developed during the nighttime that sometimes kept her awake. Her neck pain was relieved transiently by NSAIDs. External immobilization with a soft neck collar could not control her symptoms. Neither medical history nor family history was contributory.

Physical examination revealed a temperature of 36.7°C and limitation of rotation and extension with neck pain, but otherwise the results of the neurologic examination were normal. There was no spinal tenderness.

Her laboratory data fell within the reference ranges. A radiograph showed a radiolucent ring at C2; however, the image was not very clear because shadows of the mandible and teeth overlapped C2. CT scans clearly showed an oval nidus and marked sclerosis around this lesion at the right pedicle of C2 (Fig. 1). Furthermore, expansion of the medial and inferior cortical bone of the C2 pedicle was seen. The size of this lesion was 11 mm × 12 mm × 10 mm. These findings suggested an osteoid osteoma of the right C2 pedicle. As nonoperative treatment was ineffective, we initially planned CT-guided radiofrequency ablation. However, this technique was regarded as too risky by interventional radiologists because this lesion had a close anatomic relationship to the spinal cord, right C2 nerve root, and vertebral artery and thermal damage to these vital structures was possible. Therefore, we planned surgery using a CT-based navigation system (StealthStation®; Medtronic, Minneapolis, MN).

One day before surgery, a CT scan of C2 was obtained. The scan data were transferred to the computer workstation and reformatted into coronal, axial, sagittal, and three-dimensional (3D) views of C2.

The patient underwent surgery under general anesthesia in a prone position on a Mayfield frame and four-point frame. C2 was exposed bilaterally to the lateral edge of the facet joints to obtain accurate registration. After the reference frame was attached to the spinous process of C2, registration was performed and accuracy of the images was verified using a pointer. Using the navigation system, location of the nidus was checked, and the entry point and projection of a hole toward this lesion were planned (Fig. 2). Using a 3-mm high-speed diamond burr, a hole was created. During this procedure, we confirmed the accuracy of the projection using a pointer on the navigation system several times. During curettage of the nidus, a pointer was used to confirm the location of the curette to avoid damage to the spinal cord, root, and vertebral artery. The walls were not burred out because applying this technique to walls showing expansion of the medial and inferior cortical bone was considered risky. After we confirmed adequacy of the curettaged area using a pointer on the navigation system, the rectus capital posterior major muscles and semispinalis cervicis muscles were reattached to the spinous process of C2 and the wound was closed in layers (Fig. 3). The resected tissue was sent for histopathologic examination and confirmed the diagnosis of osteoid osteoma (Fig. 4). After surgery, a cervical collar was applied for 2 weeks and walking was initiated the following day.

Twenty-eight months after surgery, her pain was relieved with no limitation of cervical movement and there has been no evidence of recurrence to date. Moreover, dynamic radiographs showed there was no segmental instability (Fig. 5).

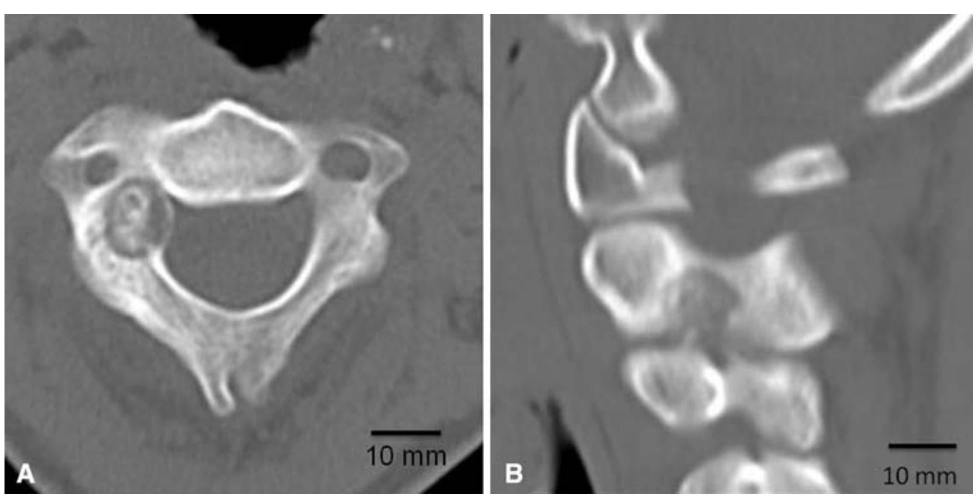

Fig. 1A–B Preoperative (**A**) axial and (**B**) sagittal reconstruction CT images clearly show an oval nidus and marked sclerosis around the lesion at the right pedicle of C2. Expansion of the (**A**) medial and (**B**) inferior cortical bone of the C2 pedicle was seen.

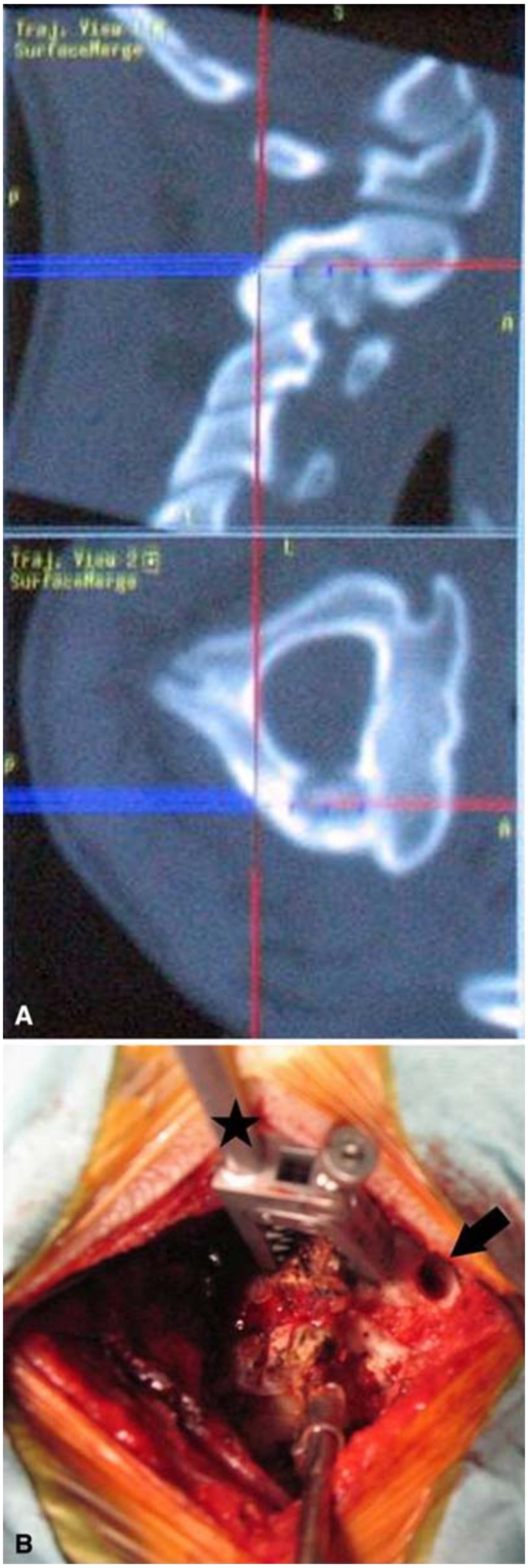

Fig. 2A–B (**A**) Two intraoperative images on the navigation system show the entry point and direction of the hole drilled toward the osteoid osteoma. (**B**) An intraoperative photograph of the operative field shows the hole (black arrow) drilled toward the osteoid osteoma at the right lamina. The reference frame (black star) is attached to the spinous process of C2.

Fig. 3 A postoperative CT scan shows excision of the osteoid osteoma and the hole drilled toward this lesion.

Discussion

Osteoid osteoma and osteoblastoma are benign bone tumors with similar histologic features showing osteoblastic bone formation [10]. These two lesions simply are separated on the basis of size. A lesion with a nidus of 15 mm or less in diameter is diagnosed as an osteoid osteoma, whereas an osteoblastoma typically is larger than 15 mm [19]. Osteoblastomas, which have more aggressive characteristics and sometimes metastasize, must be treated more aggressively [3]. Therefore, it is important to distinguish between these two tumors. The lesion in our patient was diagnosed as an osteoid osteoma because its was 11 mm × 12 mm × 10 mm.

Patients with an osteoid osteoma can be treated successfully nonoperatively, and spontaneous healing of an osteoid osteoma may occur after 3 to 4 years of medical management with NSAIDs [5, 10, 22]. Neumann and Dorn [22] reported the efficacy of celecoxib for treating an osteoid osteoma. A patient with an osteoid osteoma of the odontoid process of C2 was given 200 mg celecoxib orally once daily for 2 years and became free of symptoms, with disappearance of the sclerosed nidus on CT [22]. However, some patients cannot tolerate such long-term nonoperative treatment. If nonoperative treatment fails, the subsequent alternative would be invasive treatment.

Radiofrequency ablation currently is recommended if the site of the osteoid osteoma permits [23]. However, applying this technique to the spine remains controversial. Numerous authors [1, 11, 15, 17] have reported the efficacy and safety of radiofrequency ablation for osteoid osteoma of the spine, but some radiologists believe radiofrequency

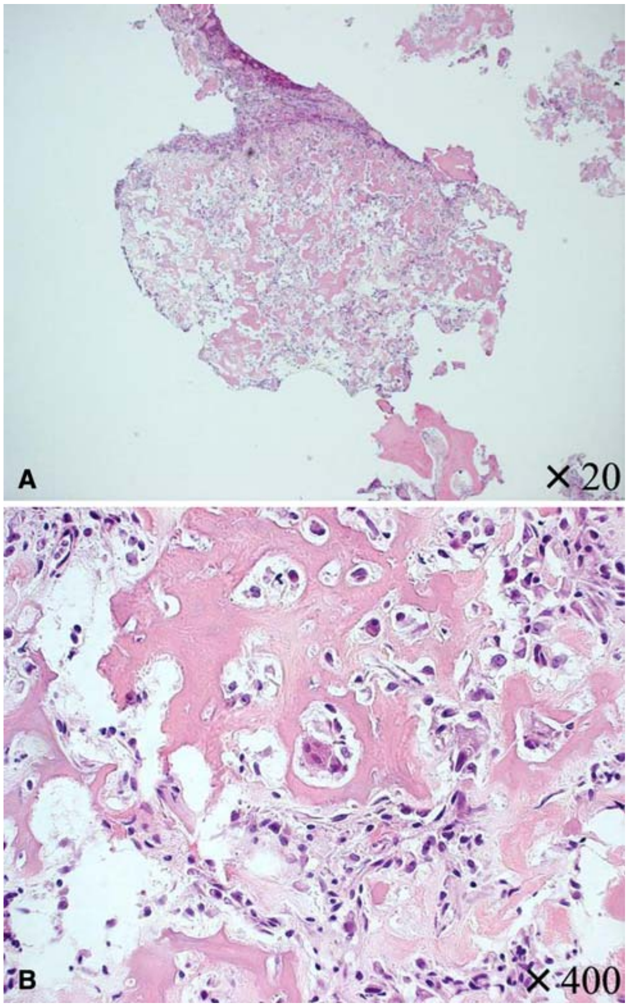

Fig. 4A–B Photomicrographs at (**A**) ×20 magnification and (**B**) ×400 magnification show osteoid trabeculae separated by fibrovascular tissue and lined by osteoblasts (Stain, hematoxylin and eosin).

ablation of a spinal osteoid osteoma carries a risk of neural damage [27] because thin cortical bone is not a reliable heat insulator and the electrode should be at least 10 mm away from the neural system. In our patient, the CT scans showed expansion of the medial and inferior cortical bone of the C2 pedicle, which also could be a risk factor for damage to the spinal cord and nerve root. For these reasons, we selected surgical treatment.

Surgery for an osteoid osteoma of the cervical spine, especially the pedicle, also carries a risk of damage to the spinal cord, nerve root, and/or vertebral artery. Surgery-related complications have been reported, including a vertebral artery injury [29], delayed vertebral artery rupture [4], and spontaneous fusion with deformity [24]. Moreover, an osteoid osteoma of the spine sometimes recurs because of incomplete resection [4, 24]. To prevent damage to vital structures and to achieve adequate resection, Rajasekaran et al. [25] reported using a navigation system for surgery of spinal osteoid osteomas. They used a 3D fluoroscopy navigation system, which has some advantages over the traditional CT-based navigation system we used [13]. First, anatomic registration is not necessary for a 3D fluoroscopy navigation system. Second, multiple adjacent levels automatically are registered simultaneously. Third, the images for navigation are taken after reduction or optimal alignment is obtained during surgery. We had only a traditional CT-based navigation system; therefore, the entire posterior compartment of the vertebra would have been exposed for accurate registration. Even using a traditional CT-based navigation system, we were able to perform curettage of the nidus through a small hole. When there is extensive loss of posterior elements, additional arthrodesis would be necessitated to prevent progressive kyphosis [7]. In the upper cervical spine, arthrodesis between C1 and C2 would cause limitations of rotation and activities of daily living.

Our technique was not minimally invasive for the muscles; however, curettage of the nidus through a small hole allowed minimal exposure of the C2 vertebra. Without using a navigation system, the hole accessing the lesion would have been larger and it would have been difficult to confirm whether the area of curettage was adequate. Moreover, even if the surgeon used an image intensifier, there would be a higher risk of damaging the spinal cord, nerve root, and/or vertebral artery compared with that using a navigation system. Therefore, we believe even a CT-based navigation system offers advantages over open surgery. Although computer-navigated surgery can accurately localize the nidus, there still may be errors. Geerling et al. [9] measured point accuracy and found Iso-C-based navigation and CT-based navigation had mean deviations of 0.5 mm and 1.0 mm, respectively. Holly and Foley [12] reported 5% of pedicle screws were placed outside the cortical pedicle margins in their cadaver study using Iso-C-based navigation. Therefore, careful registration is necessary. We consider surgery for cervical spinal osteoid osteoma using a navigation system safe and efficient, especially for surgery in the pedicle, which has a close anatomic relationship to the spinal cord, nerve root, and vertebral artery.

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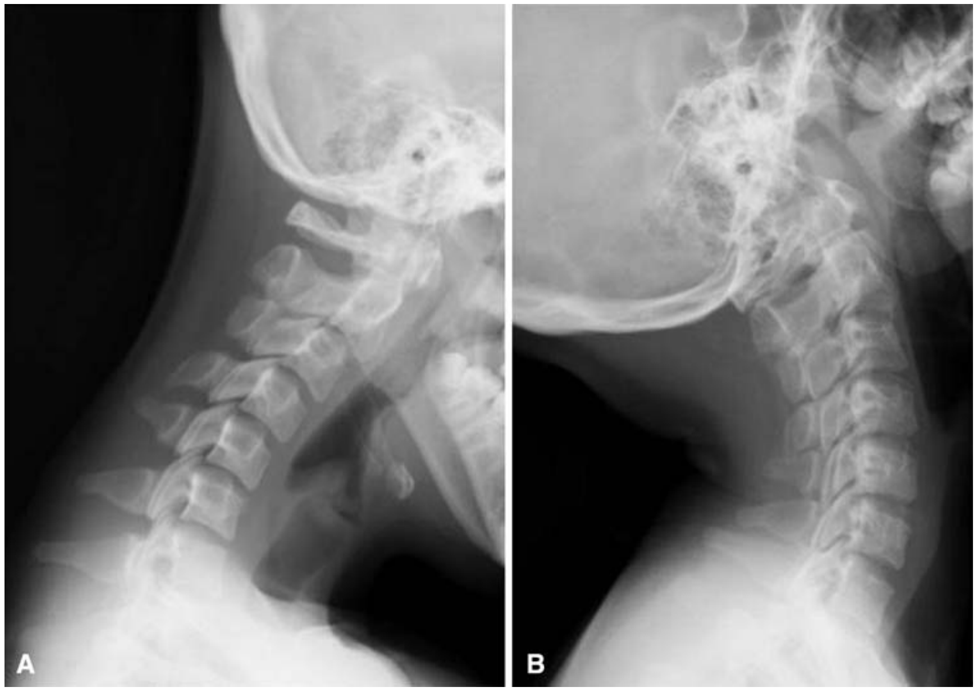

Fig. 5A–B Radiographs obtained at the final followup (28 months after surgery) of the patient's neck in (**A**) flexion and (**B**) extension show no segmental instability.

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