

Occipital Condyle Osteoid Osteoma with Severe Occipital Pain that Disappeared after Surgical Resection

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Osteoid osteoma is a benign bone tumor characterized by local pain that typically increases at night. The tumor commonly occurs in the long bones of the lower extremities, and in rare instances in cranial bones. Here we report the case of a 25-year-old man diagnosed with an osteoid osteoma of the right occipital condyle. The patient suffered from severe occipital pain in the 3 years leading up to surgery, and the pain disappeared after surgical resection of the tumor. Due caution must be taken to avoid vertebral artery injury in the surgical approach in this region. An intraoperative navigation guidance system and preoperative analysis using three-dimensional reconstructed computed tomography (CT) images improved the accuracy and safety of the resection. The typical pain in osteoid osteoma is presumed to be associated with prostaglandin E2 secretion. Plasma prostaglandin E2 of this patient was elevated preoperatively and normalized after the operation. This is the first report describing an elevation of plasma prostaglandin E2 before surgical resection followed by a normalization of serum prostaglandin E2 after surgical resection.

Keywords: occipital bone, osteoid osteoma, surgical resection, prostaglandin E2

Introduction

Osteoid osteoma is a benign bone-forming tumor first described by Jaffe in 1935.¹⁾ The tumor occurs predominantly during the first three decades of life (90% of patients are younger than 25 years old). The typical symptom is local pain that increases at night.²⁾ In some cases the pain may abate after 5 years to 6 years of conservative medical treatment.^{2,3)} When conservative treatment fails, the most common treatment option is total removal of the nidus, the round nodule composing the core of the osteoid osteoma. Histologically, the osteoid osteoma consists of the nidus and surrounding reactive sclerosis. The lesion commonly occurs in the long bones of the lower extremities, and only rarely in the cranial bones.^{2,4)} We report a rare case of osteoid osteoma of the occipital condyle with severe occipital pain that disappeared after surgical resection. The typical pain in osteoid

osteoma is presumed to be associated with prostaglandin E2 secretion. While some articles have reported elevation of prostaglandin E2 locally,^{5,6)} none have reported elevated plasma concentration of prostaglandin E2 in peripheral blood. The present report is the first to describe a change of the plasma prostaglandin E2 concentration in patients with osteoid osteoma.

Case Presentation

I. History and examination

A 25-year-old man visited the Department of Orthopedic Surgery in our hospital with a history of severe occipital pain that worsened at night. The patient had received nonsteroidal anti-inflammatory drugs (NSAIDs) and nerve block in other institutions for pain over the previous 2 years, but the pain gradually worsened and interfered with his sleep. Computed tomography (CT) scans revealed a 10-mm radiolucency in the left occipital condyle (Fig. 1a, b). Bone scintigraphy showed localized uptake of Technetium 99m (^{99m}Tc) in the same lesion (Fig. 1e). The lesion was hypointense in T₁ and T₂ weighted images in magnetic resonance image (MRI) (Fig. 1c, d). After another year of conservative treatment at our hospital without improvement, the patient decided to undergo surgical resection. The preoperative concentration of plasma prostaglandin E2 was 12 pg/mL, somewhat above the normal range (< 8.4 pg/mL).

II. Operation

The patient was placed in the prone position. A curved linear skin incision was made from the right asterion to behind of the tip of mastoid process, and then curved medially inside the hairline. The vertebral artery and C1 lateral mass were identified in the suboccipital triangle. The C0/C1 facet was exposed by dissecting the obliquus capitis superior muscle and rectus capitis posterior major muscle from the occipital bone. The medial half of the occipital condyle was drilled with the help of CT navigation. The lesion could not be clearly distinguished from the surrounding bone tissue (Fig. 2).

III. Pathological findings

Microscopic examination revealed trabeculae of woven bone and surrounding fibrovascular stroma (Fig. 3a). Those findings were consistent with a diagnosis of osteoid osteoma, but no osteoblasts were detectable in the small specimen

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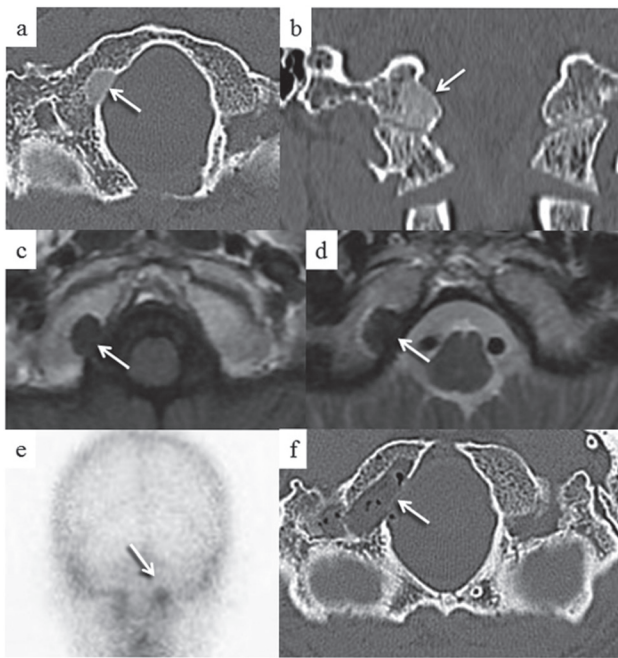


Fig. 1 Radiological findings. The radiolucent nidus and surrounding sclerosis in axial (a) and reconstructed coronal (b) computed tomography (CT) scans (arrow). Increased uptake of ^{99m}Tc in the lesion (arrowhead), posterior view (c). The nidus was depicted as a hypointense lesion in both T₁ (d) and T₂ (e) weighted images (arrow). Postoperative CT scans confirmed complete resection of the nidus and a small remnant portion of partial sclerosis (arrow) (f).

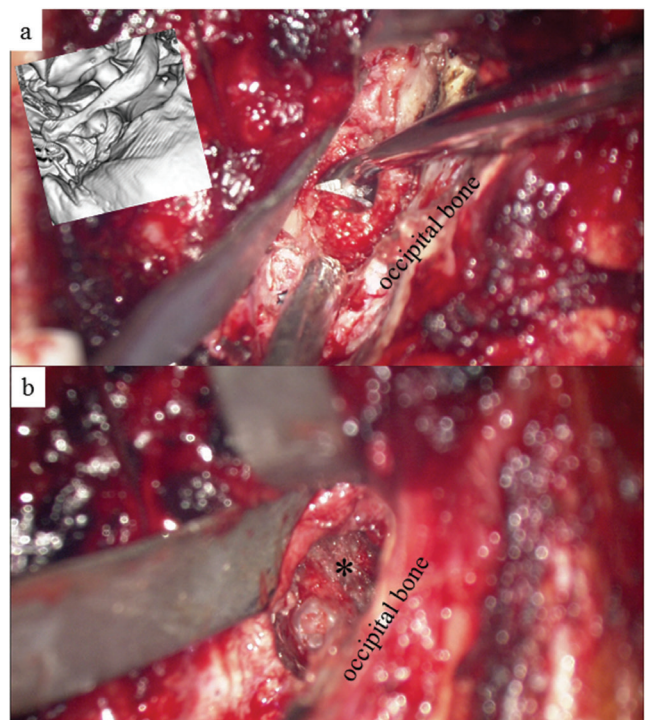


Fig. 2 Intraoperative photographs. Curretting the nidus (a). The nidus was bony hard and yellowish white in color with a tinge of red (asterisk). The lesion was difficult to distinguish from the surrounding bone tissue (b).

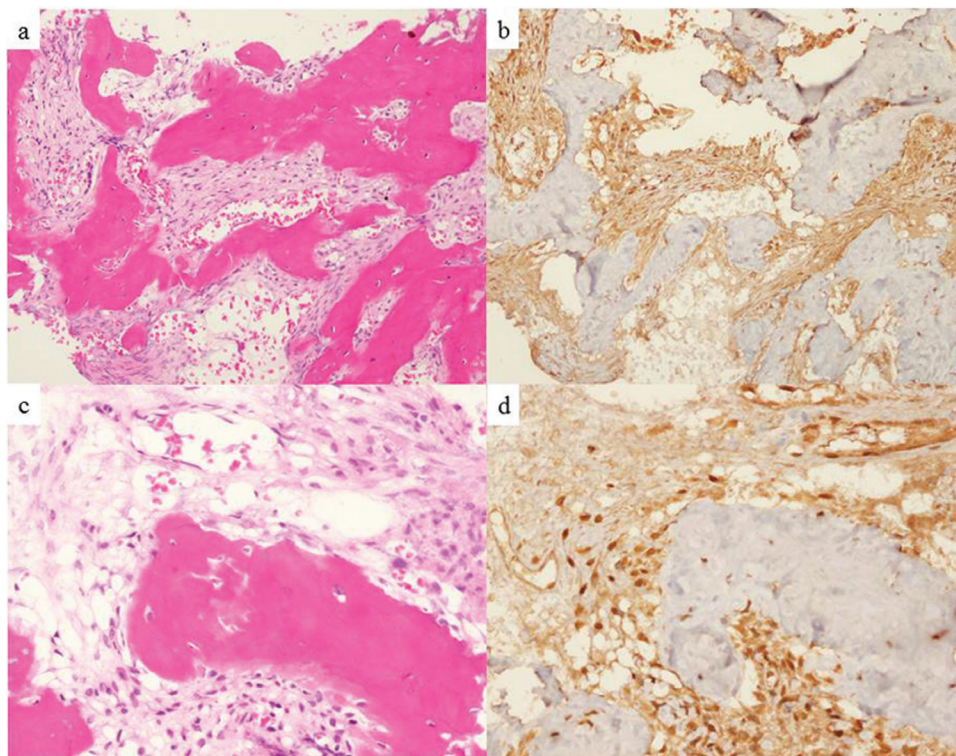


Fig. 3 Pathological findings. Hematoxylin and eosin stain showed the irregular trabeculae of woven bone and surrounding fibrovascular stroma (a) and (c). Stromal cells were positive for anti-prostaglandin E₂ antibody in immunohistochemical staining (b) and (d). Original magnification $\times 20$ (a) and (c), $\times 40$ (b) and (d).

resected. Immunohistochemical staining with anti-prostaglandin E2 antibody ab2318 (abcam) showed the production of prostaglandin E2 in the cellular cytoplasm of the stromal cells (Fig. 3b).

IV. Postoperative course

The patient was discharged 9 days after the operation without any neurological deficit and has been free from pain in the 12 months since. A postoperative CT scan confirmed that the lesion was almost completely resected, though a small anterior area of reactive sclerotic tissue remained (Fig. 1f). The postoperative concentration of serum prostaglandin E2 fell to 6.8 pg/mL, a level within the normal range (< 8.4 pg/mL).

Discussion

While osteoid osteoma is fairly common, accounting for about 10–12% of all benign bone tumors, its appearance in the bones of the cranium is fairly rare. Osteoid osteomas of the tibia and femur account for 50% of all cases. They also affect the spine in 10% of cases, usually in the thoracic or lumbar area.^{1,2,4} Only rarely do the tumors appear in the higher cervical spine or occipital condyle. To our knowledge, only two earlier cases involving the occipital condyle have been reported.⁷

Bruneau et al. reported seven cases of osteoid osteoma or osteoblastoma in the C0-C2 region, and two of the seven lesions were located in the occipital condyle. Due caution must be taken to avoid vertebral artery injury in the surgical approach in this region. One of Bruneau's cases underwent an emergent operation for hemorrhage caused by vertebral artery dilacerations.² Another case involving the occipital condyle required a second operation for recurrent symptoms caused by incomplete resection.⁷ The nidus resection in our case was completed without injury to the vertebral artery, and the patient has been free from symptoms in the 12 months since. An intraoperative navigation guidance system and preoperative analysis using three-dimensional reconstructed CT images improved the accuracy and safety of the resection.

Clinical presentation can help the diagnosis of osteoid osteoma. Worsening local pain at night is a hallmark symptom,^{2,4} and NSAIDs are typically effective in relieving pain from the tumor.⁸ Patients may be compelled to choose surgical resection, however, as medication alone sometimes fails to bring relief.^{8,9}

Regarding differential diagnosis, osteoid osteoma manifests as a round or oval radiolucency (the nidus) with surrounding sclerosis^{2,4} in radiological findings. The localized high uptake of ^{99m}Tc in bone scintigraphy is thought to reflect the increased osteoblastic activity in the lesion.^{10,11} The value of MRI for the diagnosis of osteoid osteoma has remained controversial.^{12,13} MRI can be helpful for depicting bone marrow and peripheral soft tissue edema.^{13,14} Pathologically, osteoid osteoma is impossible to differentiate from osteoblastoma. Most authors have differentiated osteoid osteoma from osteoblastoma based on the size of the tumor. Lesions with maximum diameters of less than 2 cm and larger than 2 cm are classified as osteoid osteomas and osteoblastomas,

respectively.^{2,4} The lesion in our case had a maximum diameter of 1 cm, so we diagnosed it as an osteoid osteoma. Our case lacked one of the typical histological features of osteoid osteoma, namely, osteoblasts surrounding the trabeculae of woven bone. This was unimportant for the diagnosis of osteoid osteoma in our case, however, as the lesion specimen was small and the quantity of osteoblasts varies according to local mineralization.²

The goal of surgical treatment is complete excision of the nidus. Removal of the reactive bone sclerosis is inessential.² Symptoms may recur if the resection is incomplete,^{5,15} but the surrounding sclerosis can be partially left in place if en bloc resection is anatomically difficult. Postoperative CT scans of our case revealed a small portion of surrounding sclerosis left in place. The nidus was completely removed, however, and our patient has now been symptom free for 12 months.

The typical pain in osteoid osteoma is presumed to be associated with prostaglandin E2 secretion. Prostaglandins are known to mediate inflammation, to induce tissue edema via a powerful vasodilating effect, and to mediate pain and hyperalgesia by lowering the nociceptive threshold. Immunohistochemical studies have identified nerve fibers in the fibrous zone of the nidus of osteoid osteoma.¹⁵ Prostaglandins may elicit pain in osteoid osteoma via two mechanisms: the increased pressure from enhanced blood flow may stimulate nerve endings, or prostaglandins may directly stimulate nerve endings by lowering the nociceptive threshold. An immunohistochemical study on osteoid osteoma by Mungo et al. revealed elevated expression of cyclooxygenase (COX), the rate-limiting enzyme in prostaglandin biosynthesis.¹⁶ Hence, NSAIDs probably relieve the pain of osteoid osteoma by inhibiting COX bioactivity.

Some articles have reported elevation of the local density of prostaglandin E2 in the nidus.^{5,6} Yet as far as we can ascertain from a literature search, elevated plasma prostaglandin E2 concentration has never been reported in osteoid osteoma patients. Hence, the present report is the first to describe a change from an elevated concentration of plasma prostaglandin E2 preoperatively to a normalized concentration postoperatively. The concentration of plasma prostaglandin E2 changes by only a small amount, because prostaglandin E2 secreted locally is soon inactivated and diluted. In observation by Sunouchi in patients with colon cancer, the level of plasma prostaglandin E2 was about 8.8 times lower in peripheral blood than in blood from tumor drainage veins.¹⁷ The concentration of plasma prostaglandin E2 was 1.23 times higher in patients with hypopharyngeal and laryngeal squamous cell carcinoma than in non-cancer patients, and was also found to decrease after surgical removal of those tumors and to be predictive of tumor recurrence.¹⁸ Though never reported in osteoid osteoma, the plasma concentration of prostaglandin E2 may facilitate tumor diagnosis and help predict the effects of non-invasive therapy with NSAIDs, assess the indication of surgical resection when NSAIDs are ineffective, and indicate whether the surgical resection is complete. Further data on plasma prostaglandin E2 concentrations in osteoid osteoma cases is needed.

Conclusion

We reported a rare case of occipital condyle osteoid osteoma. The clinical presentation and radiological findings were helpful for the diagnosis, and the symptom of pain disappeared after the nidus was surgically resected. This is the first report describing an elevation of plasma prostaglandin E2 before surgical resection followed by a normalization of plasma prostaglandin E2 and an abatement of pain after resection. The clinical role of plasma prostaglandin E2 in osteoid osteoma will be more fully known when data on more cases accumulates.

Conflicts of Interest Disclosure

None declared. All authors who are members of The Japan Neurosurgical Society (JNS) have registered online Self-reported COI Disclosure Statement Forms through the website for JNS members.

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