

A rare case of misdiagnosis: sacrococcygeal osteoid osteoma that was treated surgically as a pilonidal sinus in a pediatric patient

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Summary

Background. We report a misdiagnosed pediatric case of osteoid osteoma in sacrococcygeal region. Previously it was said by general surgery that the source of pain was the pilonidal sinus and was therefore operated. A CT scan was taken and showed that the patient had sacrococcygeal osteoid osteoma which nidus was very close the right S4 spinal root. The lesion was therefore marked on CT scans and resected by performing an open surgery.

Methods. This report reviewed the patient's medical records, imaging studies, treatment, and related literature.

Results. Our patient, who had a continuous pain for 2 years, applied to our orthopedic clinic due to aches and pain at sacrococcygeal region. The characteristic clinical symptom was nocturnal pain especially night that decreases or resolves completely with salicylates. Previously it was said by general surgery that the source of pain was the pilonidal sinus and was therefore operated. Our patient who had no decrease pain after surgery was advised to use salicylate which resulted in dramatic responses. A CT scan was taken and showed that our patient had sacrococcygeal osteoid osteoma which nidus was very close the right S4 spinal root. The lesion was therefore marked on CT scans and resected by performing a mini-open surgery.

Conclusion. Osteoid osteoma is a benign tumor of bone, especially in children and young adults. Often seen in the lower bones of the lower extremity, the sacral vertebra is very rare and causes severe localized pain at night. When such a chart is encountered in children and young adults, osteoid osteoma should be considered carefully in the differential diagnosis.

KEY WORDS: osteoid osteoma; misdiagnosis.

Introduction

Osteoid osteoma is a benign tumor of the bone that was first described by H.L. Jaffe in 1935 (1), usually seen in children and young adults. Osteoid osteomas with a frequency of approximately 10% seen in benign bone tumors are more common in males than females (2). The size is characterized by nidus that contains calcification in variable amounts not exceeding 1.5-2 cm. Especially at night, it is a clinical chart which is manifested by pain that is exacerbated by salicylate and nonsteroidal anti-inflammatory agents. It is usually seen in long bones and may occur in any skeletal structure, even though it is more localized to the lower extremities. The spine is an area where osteoid osteomas are rarely localized. The vertebrate site is mostly in the lumbar vertebrae and at least in the sacrum (3). We present a rare case of osteoid osteoma in sacrum.

Case presentation

A 7-year-old female patient presented to our clinic with pain in the sacrococcygeal region. Our patient, who had pain for 2 years, had been operated due to the diagnosis of pilonidal sinus at another center. But postoperative pain still did not pass. The pain was especially present at night. She suffered insomnia due to the pain mostly occurring during night hours, and started having psychological problems and treatment for it. Osteoid osteoma was considered for our patient and salicylate 100 mg was initiated and a dramatic response was obtained. On this, our patient had sacral CT scan and right S4-level posteriorly located osteoid osteoma was detected (Figure 1). CT-guided Kirschner wire marking was done under general anesthesia (Figure 2). Afterwards, the nidus in our patient who was taken to the operating room was removed with open surgery (Figure 3). Histologically, hematoxylin and eosin staining demonstrated differentiated osteoblasts lining the osteoid, and interconnected trabeculae of woven bone, which was compatible with osteoid osteoma (Figure 4). Our patient's pain just passed after surgery, and was discharged 1 day later. Our patient did not have any complaints. Insomnia and the psychiatric treatment were terminated.

Discussion

Osteoid osteoma (OO) with a frequency of 2-3% seen among all bone tumors can be found in any skeletal structure (4). It is frequently localized to the long bones of the lower extremity. Spinal OO is rare (10%) (5), and present more often in the posterior spinal elements. The region is most com-

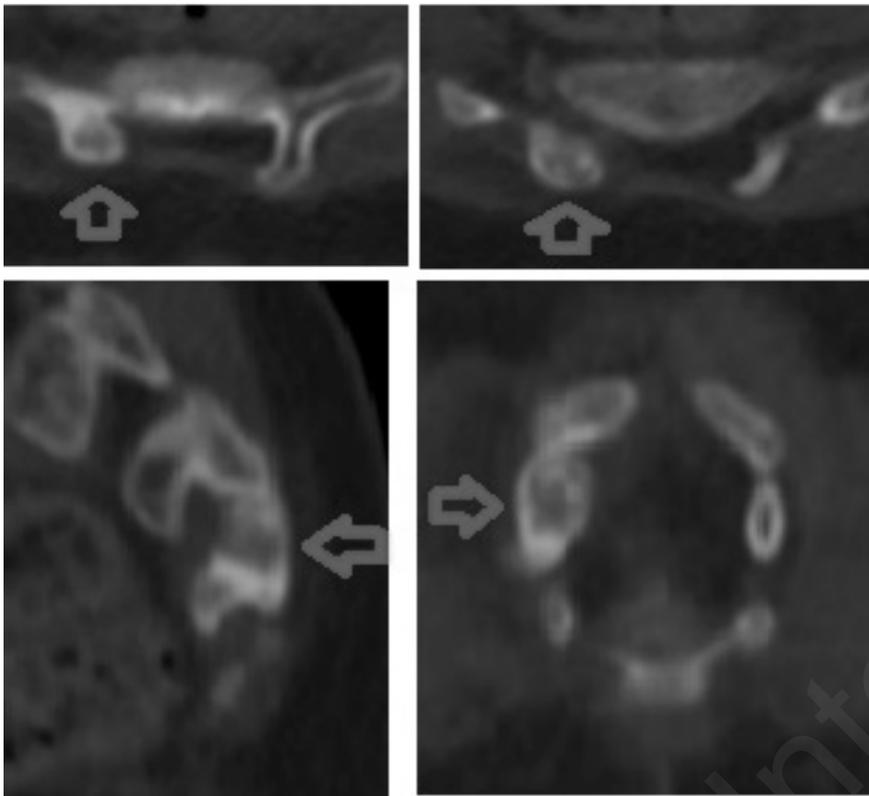


Figure 1 - Sacral CT scan of right S4-level posteriorly located osteoid osteoma (axial, sagittal and coronal views).

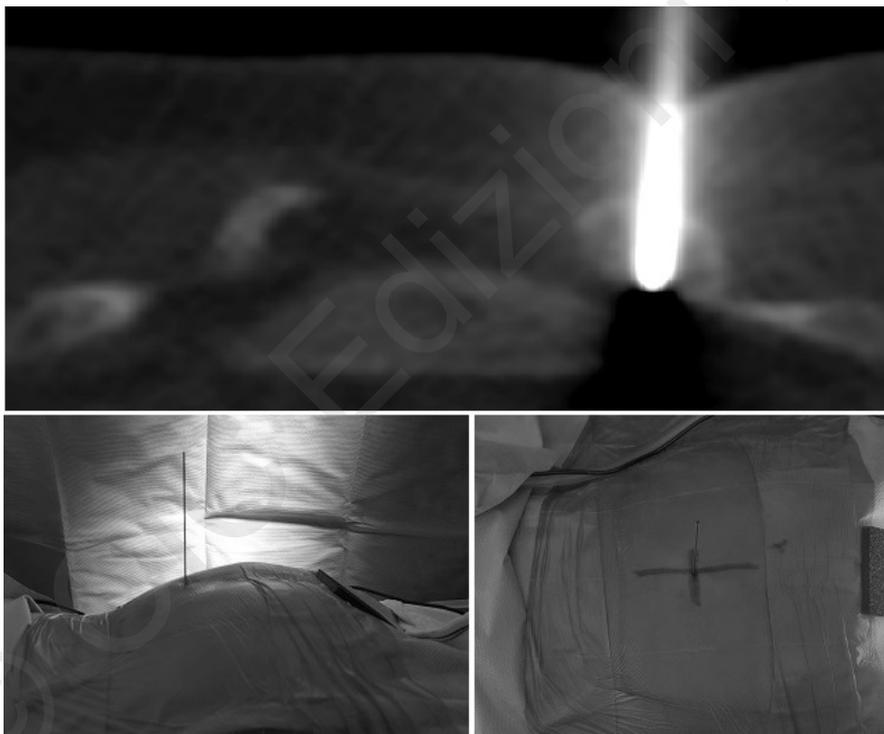


Figure 2 - CT-guided kirschner wire marking.

only seen in the lumbar vertebrae (59%) followed by the cervical vertebrae (27%) in the thoracic vertebrae (12%) and at least in the sacrum (2%) (3). Only 1 of the 19 cases reported in the literature on OO in sacrum was posteriorly located (6). In our case, the lesion is posteriorly located. The fact

that the events reported in sacrum are mostly anterior region, unlike other vertebrae's, suggests that it is due to its unique shape (7). In clinical reflection, pain is an important factor in the differential diagnosis of OO. Makley and Dunn (8) reported that the pain caused by OO was caused by abun-

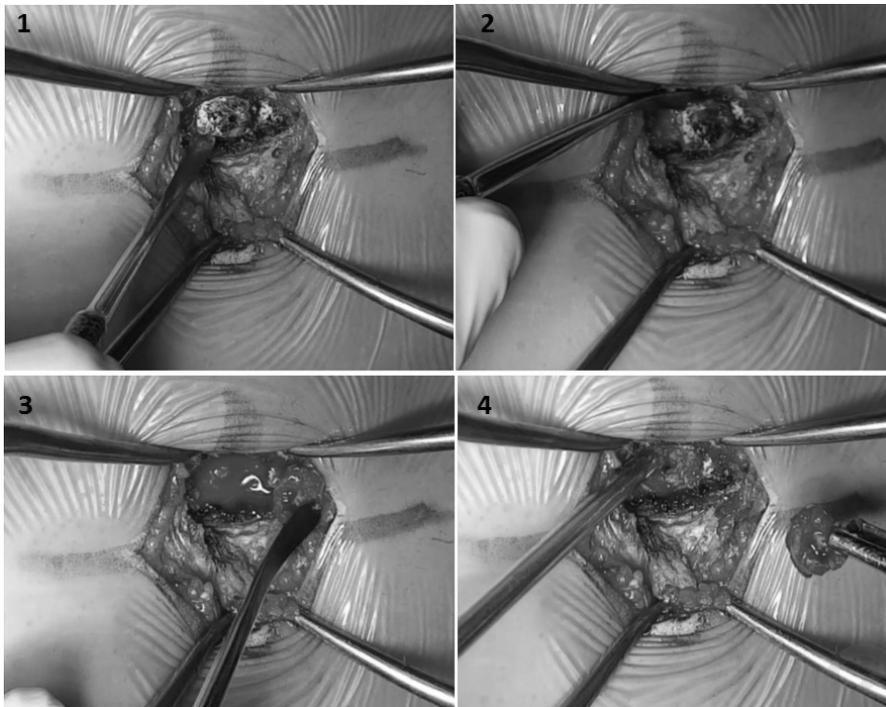


Figure 3 - Perioperative views.

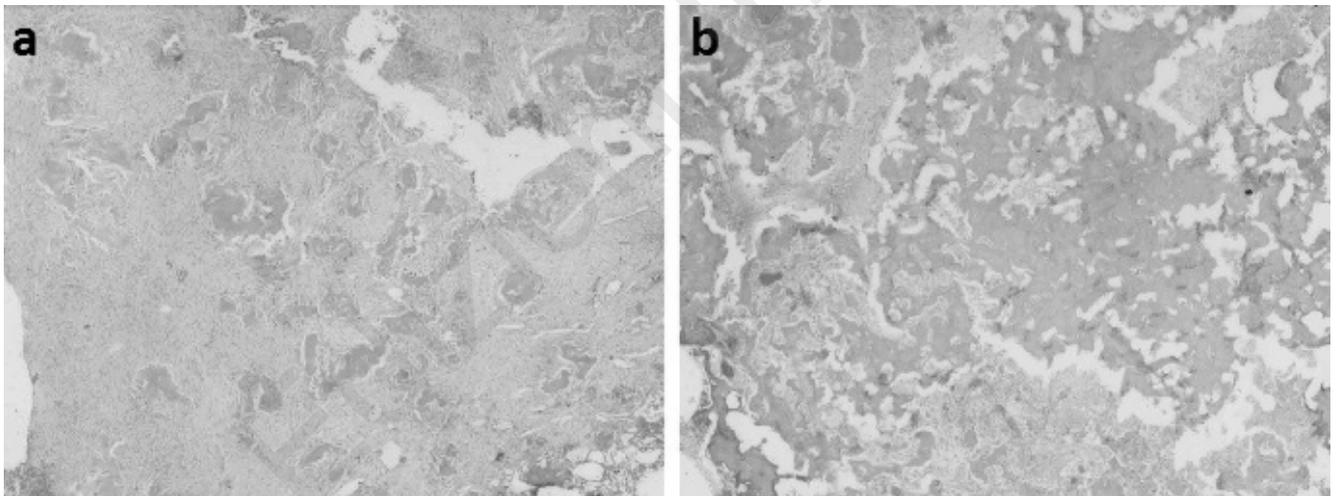


Figure 4 - a) Anastomosing, irregular trabeculae rimmed by single layer of osteoblasts in loose, fibrovascular stroma (HEX40); b) Sclerotic nidus of woven bone (HEX40).

dance of nodular nerve fibers and excessive prostaglandin synthesis. In a study involving a large number of cases, it was found that 95% of the cases of indole was aggravated at night and that prostaglandins caused by pain were increased (9). Our patient had localized pain that became very severe at night. This caused our patient not to sleep at night and to suffer deformities such as redness, burning, swelling in the eyes from insomnia. The insomnia state was making the patient restless and depressed by affecting the psychological state of our patient.

The operative treatment of the OO is the complete resection of the nidus. The CT guided resection was first reported by Voto et al. (10) who reported that this technique is useful for localization and removal of the nidus, and nowadays CT gui-

dance is applied with treatment options such as trephine percutaneous excision (11) and CT guided percutaneous radiofrequency ablation (RFA) (12). It has been reported that 16% recurrence is caused by inadequate ablation of RFA (6). Another deficiency in RFA is the lack of adequate tissue retrieval for histological and pathological diagnosis (13). The use of RFA in lesions close to the nerve structure is limited in its use because it can cause the nearby nerve tissue to become warmed and thus damaged (13, 14). 15 of the 19 OO cases reported in the sacrum were treated with open resection. 4 cases were treated with resection in CT guideline (6). RFA was not considered in our case because it was very close to right S4 spinal root. It is appropriate to have resection treatment under the guidance of CT which gives good results.

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