

CASE REPORT/KAZUISTIKA

Osteoid Osteoma in S2: Percutaneous CT Guided Resection of a Double Nidus

Osteoid osteoma v S2: perkutánní CT navigovaná resekce dvojitého nidu

JIŘÍ SKÁLA-ROSENBAUM, JAKUB JEŽEK

Department of Orthopaedics and Traumatology, Third Faculty of Medicine of Charles University, and University Hospital Královské Vinohrady, Prague

Dedicated to the 70th anniversary of the birth of Prof. Martin Krbec, MD, CSc.

Corresponding author:

Jiří Skála-Rosenbaum, MD, PhD, Prof
Department of Orthopaedics and Traumatology
Third Faculty of Medicine of Charles University
and University Hospital Královské Vinohrady
Šrobárova 50
100 00 Prague 10, Czech Republic
jrosenbaum@volny.cz

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SUMMARY

The case report describes the option of percutaneous CT-guided ablation of osteoid osteoma with a double nidus to combine minimally invasive approach and CT(0-arm)-guided technique. Following X-ray, CT and MRI examinations, osteoid osteoma was diagnosed in the body of the second sacral vertebra (S2) in a 31-year-old female patient. CT reconstructions

detected a double nidus with a defective, sclerotic rim. MRI demonstrated reaction in the surrounding tissue and edema in the nerve root. In consideration of the anatomic location of the osteoid osteoma and vicinity of the irritated S2 root percutaneous CT-guided intralesional resection of a double nidus was performed.

There was immediate pain relief after the surgery, and the patient was discharged on the second postoperative day. The follow-up histological examination confirmed the diagnosis. During a five-year follow-up, the patient

remained symptom-free and without recurrence of the disease.

CT-guided minimally invasive resection of osteoid osteoma is an option in cases of benign tumor-like lesions located in the spine, as well as in cases of a double nidus. In comparison to radiofrequency ablation, percutaneous curettage enables obtaining valid histological specimens and does not bear the risk of thermal damage to neural tissue.

Key words: osteoid osteoma, sacrum, CT navigation, percutaneous resection, double nidus.

INTRODUCTION

Osteoid osteoma is a benign bone tumor occurring predominantly in children, adolescents and young adults up to the age of 30 years. The first pathologic description of this tumor is ascribed to Bergstrand (4). The term "osteoid osteoma" was first used by Jaffe in 1935 (9). A characteristic symptom of this tumor is nocturnal pain that is relieved with salicylic and non-steroid antirheumatic drugs, which act as antiagregans (the so called "aspirin test") (12). Thus, the symptoms are apparently related to a high local concentration of prostaglandins. Diagnosis is based on assessment of radiographic and CT findings with a typical central nidus of up to 2 cm and sclerotization of the surrounding bone. There have also been cases where two adjacent nidi have been described (1). Scintigraphic examination reveals accentuated activity at the nidus.

Osteoid osteoma is usually located eccentrically in long bones. According to location, cortical, subperiosteal and cancellous types of osteoid osteoma have been described. About 50% of cases are found in the femur and tibia. Only 7-10% of osteoid osteomas involve the spine, where a clear majority occurs in the thoracic and lumbar regions (6). In these regions, it typically occurs in the pedicle area, but location in the vertebral body, articular facet or arch is not uncommon either. Findings of osteoid osteoma in the sacrum is a rarity (16). Also, the incidence of a double nidus is rare (1). The combination of sacral location and a double nidus is therefore a very sparse finding.

The basis of treatment is elimination of the pain source, i.e. the nidus. This can be performed by conventional open en bloc resection, which may also be intralesional. Considering the anatomic conditions in spinal locations, the route of



Fig. 1. CT reconstruction of sacrum in a 31-year-old patient: a – transverse incision revealing a central clearing with surrounding sclerotic border and eroded rim, b – sagittal reconstruction, c – frontal reconstruction with a clearly defined double nidus.

choice is either CT-guided radiofrequency or thermal ablation. The disadvantages of these methods include the inability to obtain biopsies for histological examination but more particularly the arising heat producing cytotoxic effects on the adjacent tissues. Hence, in cases where the tumor is located in close contact with neural structures, all risks of damage to the involved structures have to be considered. A feasible option may be CT-guided intralesional curettage of the tumor.

CASE REPORT

A 31-year-old female patient underwent unsuccessful conservative therapy for pain in the lower lumbar region that lasted for the duration of over one year. The symptoms were characteristically nocturnal, but daytime pains at rest were not exceptional either. The patient felt relieved after non-steroid antirheumatic drugs, but her consumption amount already exceeded the recommended daily dosage. We completed CT and MRI examinations in our department. CT findings were characteristic for osteoid osteoma located in the body of S2 in close contact with the left-sided foramen with typical occurrence of a sclerotic rim around the nidus (Fig. 1). In frontal reconstruction, it became distinctly evident that the central clearing contained two neighboring nidi. The cortical rim was already partially eroded with edema of the adjacent root, which was visible on the MRI (Fig. 2). The neurological findings with pain in the region of the lumbosacral junction and the sacrum, and further on the medial side of gluteal line also corresponded with the findings. Subsequently, the applied aspirin test was positive.

Given the clear findings, we did not complete scintigraphy and continued by deciding on the type of surgical solution. The anatomic position excluded open resection, which would have certainly been associated with higher blood loss and increased postoperative pain. On the other hand, the close proximity of the already irritated root made radiofrequency

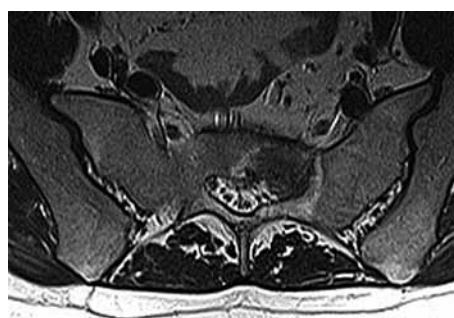


Fig. 2. MRI in the transverse plane with reaction in the adjacent tissue and S2 root edema on the left side.

ablation impossible for the risks of thermal damage to nervous tissue. Therefore, we proceeded to conduct a CT-guided intralesional resection of the double nidus.

We operated on the patient under full anesthesia in prone position. Through a mini incision, we inserted a screw to the right iliac wing, by means of which we calibrated the CT equipment interconnected with an intraoperative navigation system (O-arm® System & StealthStation® Navigation, Medtronic, Louisville, USA). From a left-sided, guided percutaneous access along the crest of the iliac wing, we created a 1 cm wide working “tunnel” all the way to the nidus (Fig. 3). With the use of calibrated tools, we performed curettage of both nidi and exochleation of sclerotic rims (Fig. 4). At the end of the surgery, we filled the cavity with Spongostan. We sent the obtained specimens for histological evaluation, which we had previously confirmed, re-established diagnosis of osteoid osteoma.

The surgery lasted 45 minutes. There was near to zero blood loss. The X-ray amplifier was not used at all. After the surgery, the patient felt immediate relief. She was discharged to home care on the second postoperative day. On the 6-month post-op follow-up, the patient was completely symptom-free; the nerve root irritation had even disappeared. Preoperatively, the VAS score was 9 and postoperatively it was 0. During a five-year follow-up, the patient remained symptom-free and without recurrence of the disease.



Fig. 3. Intraoperative image showing a calibration disc fixed to a screw in the right iliac wing, and navigation probe inserted from percutaneous approach left.

DISCUSSION

Spinal osteoid osteoma findings are not extremely rare, however, in regard to their location in close proximity of nerve structures, decision-making regarding the type of surgical approach is not easy. In the sacral region, these benign tumors are less common when compared to thoracic and lumbar areas of the spine (16). An osteoblastoma needs to

be considered in the differential diagnosis, as it has similar radiographic findings. In our patient, there was a highly presumptive diagnosis of osteoid osteoma for several reasons. Osteoblastoma occurs in younger patients up to the age of 20 years. The nidus is usually larger than 1.5 cm, a double nidus is uncommon and the aspirin test is positive only in 17% of cases (10). Also, the sclerotic rim is not commonly regular as in osteoid osteoma, but generally creates a lobular structure. On the other hand, osteoid osteoma within the sacral bone can be mistakenly considered as sacroiliitis (17).

Osteoid osteoma of the sacrum often leads to delayed diagnosis due to negative radiographic image findings (16). Only perifocal sclerosis may provide us with markers in cases of a potential primary tumor. The route of choice in diagnostics is therefore CT in combination with scintigraphy, thus enabling precise location of the nidus. Even MRI may be misleading. Nevertheless, in younger patients, it is beneficial in terms of differential diagnosis, particularly for purposes of eliminating primary malign tumors, such as the Ewing sarcoma. In terms of the CT and MRI findings, we decided not to perform scintigraphy, also with perspective on the scheduled CT-guided surgery.

Our case is rare not solely for its sacral location, but also for the presence of a double nidus. So far, the incidence of a multicentric nidus has been published in less than thirty case reports (1). The most common location of a multicentric nidus is in long bones, with the femur being represented the most (35%). Solely, Schajowicz and Lemos published one case of a multicentric nidus in the sacrum in 1970 (15). It is reported that the tumor

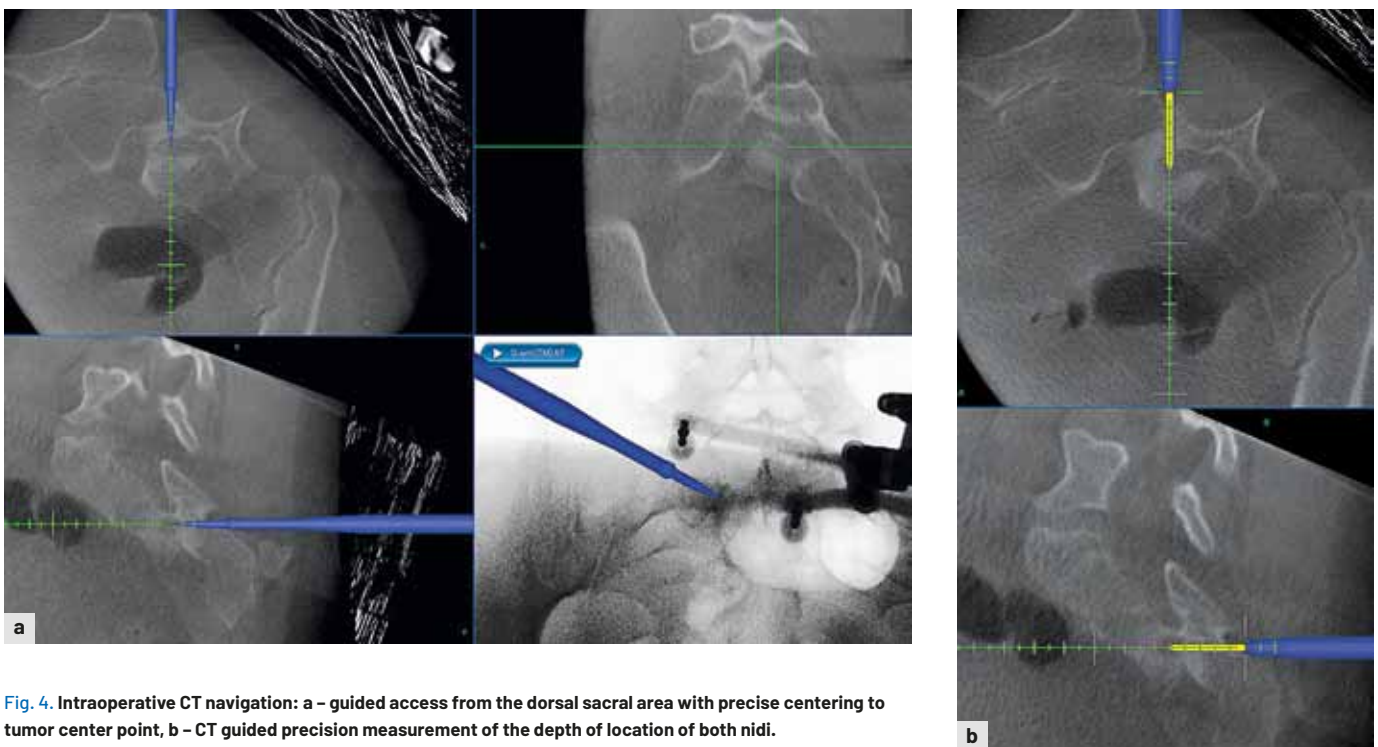


Fig. 4. Intraoperative CT navigation: a – guided access from the dorsal sacral area with precise centering to tumor center point, b – CT guided precision measurement of the depth of location of both nidi.

recurrence is rare and occurs in just under 10% of patients (5). A very probable cause of recurrence is inadequate excision of the nidus, particularly in case of double or multicentric nidus. Although the monitoring time of our patient has been too short to eliminate contingent recurrence, the double nidus diagnosis with guided curettage and complete, immediate relief represents a prerequisite for good future prognosis.

Surgical solution is indicated for a vast majority of patients. It may represent extensive en bloc resection, open intraleisional resection or percutaneous guided curettage. CT navigation offers multiple options for removal of the pain generator. Extensive resection yields the benefit of reliable tumor removal and valid histological specimens, but it is associated with a higher operative morbidity rate. Extensive resection in the spinal area may then become the cause of subsequent iatrogenic instability. In the case of S1 body location, transperitoneal or retroperitoneal anterior approach is recommended, which is, however, associated with non-negligible risks (3).

Percutaneous guided radiofrequency or thermal ablation of a nidus is a widely used treatment procedure for osteoid osteomas located in long bones and it may also be applied for spinal osteoid osteomas (2, 11, 14). Yet, it is necessary to realize that the heat produced during these techniques increases local temperature to 50–90°C for a period of 4–6 minutes, which is cytotoxic

both to the spinal cord and peripheral nerves (7). This applies particularly when the nidus is in direct contact with the spinal cord or a nerve root, and in case of defective sclerotic border, that normally protects the neural tissue (8). Furthermore, not even this technique can reduce the risk of tumor recurrence.

In consideration of the CT and MRI findings and the risk of thermal damage to the nerve root, we decided to perform percutaneous CT-guided curettage of the tumor. Intraoperative decision making left us with the options of either the transiliacal approach (13) or the angled dorsal percutaneous approach. The transiliacal approach may lead to pain in the area of sacroiliac joint and when performed on children, may result in mild pelvic asymmetry. In our case, however, favorable location of the tumor allowed us to create a percutaneous working “tunnel” along the crest of the iliac wing and therefore spare the sacroiliac joint. After the surgery, the patient felt immediate, complete pain relief in terms of local pain and S2 nerve root irritation. Moreover, as compared with thermal coagulation, we had a chance to obtain specimens for histological evaluation, re-confirmed our pre-established diagnosis.

Percutaneous CT-guided ablation of sacral osteoid osteoma yields safe removal of the nidus with feasibility of valid histological examination and without any risks of thermal damage to neural structures. ■

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