

Case Report

A rare anterior sacral osteochondroma presenting as sciatica in an adult: a case report and review of the literature

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Abstract

BACKGROUND CONTEXT: Osteochondroma is the most common primary benign bone tumor and is usually located in the metaphyses of long bones and rarely in the spine or anterior sacrum. To the best of our knowledge, en bloc excision of a solitary osteochondroma of the anterior sacrum in an adult patient has not been previously reported in a peer-reviewed journal.

PURPOSE: The purposes of this study were to document the first report of an osteochondroma of the anterior sacrum along with the clinical course and operative management and review the literature on solitary osteochondroma of the sacrum.

STUDY DESIGN/SETTING: The study setting is an academic institution. This is a case report and review of the literature.

PATIENT SAMPLE: The patient is an adult female.

OUTCOME MEASURES: The outcome measure is the visual analog score for pain.

METHODS: A 54-year-old woman presented with 9/10 disabling low back and radicular pain in the left lower extremity. Radiologic studies showed a pedunculated mass occurring from the anterior sacrum thought to be causing nerve root compression. The patient received en bloc excision of the mass through an abdominal retroperitoneal approach. We also conducted a literature review of solitary sacral osteochondroma in peer-reviewed journals.

RESULTS: Histologic studies confirmed the mass to be an osteochondroma without pathologic signs of malignant transformation. The patient complained of dysesthesia in the left leg after surgery, which progressively improved completely over 8 months after the operation. At the 2-year follow-up, there was no evidence of local recurrence and she was pain free. A literature review revealed one previous case of en block resection of a solitary osteochondroma, but it involved the posterior sacrum.

CONCLUSIONS: Solitary osteochondroma can rarely present in the sacrum as low back pain and sciatica. In general, when osteochondroma causes pain in an adult, we should think that some structure is impinged or that it could have initiated a malignant transformation, so en bloc excision should be used to remove the tumor and histologic studies performed to assess for malignant transformation. Battered nerve root syndrome may take up to 8 months to resolve. © 2010 Elsevier Inc. All rights reserved.

Keywords: Osteochondroma; Spine; Sacrum; Tumor; Sciatica

Introduction

Solitary osteochondroma (osteocartilaginous exostosis or exostosis) was first described by Reid in 1843 [1], and it accounts for 10% of all primary osseous tumors [2]. Osteochondromas increase in size via endochondral ossification from an abnormal cartilaginous epiphyseal growth plate tissue and, therefore, corresponds to the sites of most rapid bone proliferation such as in the distal femur and

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proximal humerus [3]. Because of its association with the growth phase of bone, osteochondromas are considered to be the product of puberty and typically do not progress into adulthood [4].

Solitary osteochondroma of the spine beyond skeletal maturity has been uncommonly reported in the literature [5,6] but not on the anterior sacral ala. In all age groups, only 3% of osteochondromas occur in the spine, and less than 0.5% of those occur in the sacrum [7,8]. In this case report, we present a solitary osteochondroma in an adult located on the anterior surface of the sacrum with associated low back pain and radiculopathy. The patient's presenting clinical symptoms, operative procedure, and postoperative outcome are described in detail.

Case report

We obtained written informed consent from the patient regarding surgery and publication of the case and all additional components. A 54-year-old obese woman presented to our clinic after a series of unsuccessful nonoperative management for 9/10 low back pain and sciatica of 8 months' duration. She did not have bowel or bladder symptoms. The patient's family history screening was negative for bone tumors. The patient's examination was notable for lower back tenderness diffusely in her paraspinal muscles, and she complained of low back pain with flexion more than 60°. She had absent reflexes in both ankles. Her strengths in the lower extremities were decreased (4/5) throughout and slightly worse on her left side, and this was thought to be because of her complaints of pain with resistive motor testing. Overall, her examination was not indicative of a focal neurologic deficit attributed to her spine. Magnetic resonance imaging of her lumbar spine revealed no significant stenosis or compressive lesion to explain her weakness and sciatica. A computerized tomography (CT) scan was done to evaluate for any bone foraminal encroachment. The CT scan revealed a large pedunculated mass emanating from the left anterior surface of her sacral ala with potential impingement on the left lumbosacral trunk and on the S1 nerve root (Fig. 1). Based on the CT scan appearance of the cortex and spongiosa appearing to be contiguous with the bone beneath, the patient was given a diagnosis of an osteochondroma. Although the cartilage cap was thought to be less than 1 cm, the axial location of the mass in the pelvic girdle, along with pain, raised the suspicion for malignant transformation of the osteochondroma. The patient was informed and consented to open excision and biopsy of the tumor.

Based on the location of the tumor, the decision was made to perform the surgery through a retroperitoneal abdominal approach using an anterolateral oblique incision on the left side of the abdomen. Ureteral stents were placed preoperatively to lessen the risk of ureteral injury in this obese patient during the exposure and operation. A vascular



Fig. 1. Preoperative computerized tomography scan of an osteochondroma demonstrated as a left-sided pedunculated mass on the anterior surface of the sacrum.

surgeon performed the exposure. The osteochondroma could be palpated and visualized, tenting the soft tissues as we approached the sacrum. A soft-tissue capsule was separated and bluntly elevated off the cap with Cobb elevators to expose down to the stalk. We were able to expose the stalk without visualizing any direct neurovascular compression. We would have preferred to identify and dissect the neurovascular structures that were being compressed during our exposure, so we could directly protect the nerve but felt it would be safer to remove the tumor first to gain better exposure in this obese patient and then if needed we would expand our dissection to see the neurovascular structures. Once the exposure of the osteochondroma was made, we placed malleable retractors around the osteochondroma along its length and the stalk of the pedunculated tumor was amputated using a curved osteotome, and the residual pieces were sequentially removed using Kerrison rongeurs to lessen the risk of injuring the surrounding neurovascular structures (Fig. 2). Hemostasis was achieved with Gelfoam and bone wax over the bleeding bony surfaces. After copious irrigation, the wound was closed in layers. The specimen was sent to pathology for frozen and permanent analysis, and no pathologic cells were found.

Postoperatively, the patient's symptoms improved with the strength of 5/5 in both legs and substantial pain relief. She had dysesthesia in the S1 distribution, which was managed with gabapentin 400–600 mg three times a day. Approximately 8 months after the surgery, the patient had complete relief of symptoms, and no further medications were prescribed.

Follow-up CT scan showed removal of the entire cap and part of the stalk (Fig. 2). No further imaging was recommended because this was a solitary pedunculated lesion with no pathologic signs of tumor.



Fig. 2. Postoperative computerized tomography scan after “marginal” en bloc excision of the osteochondroma through the stalk with complete removal of the cap.

Discussion

Osteochondromas are benign bone tumors that rarely originate in the spine. They are cartilaginous tumors capped by bony projections from an osseous surface and may assume either pedunculated or sessile morphology. The cortex and spongiosa of osteochondromas are contiguous with the bone beneath, requiring CT scan for better visualization of cortical and medullary continuity between the tumor and the normal bone [9]. Osteochondromas are more accurately described as hamartomas that grow from an aberrant epiphyseal tissue commonly from patients with skeletal immaturity. Eventually, the growth of osteochondromas may stop or slow down once the growth plate fuses and rarely progress beyond 30 years of age. Hence, the average age of patients presenting with osteochondromas is between 20 and 30 years, most commonly affecting the long bones around the knee such as the distal femur and the proximal and distal tibia [3,10].

Approximately 3% of osteochondromas occur in the spine, and the cervical vertebrae are the most common site of tumor growth [8]. Similar to other benign tumors, osteochondromas have predilection for young male patients, typically younger than 20 years of age with male-to-female ratio of 3:1 [8]. Recently, Samartzis and Marco [11] reported a case study of an 11-year-old boy with a sacral osteochondroma treated with posterior en bloc excision.

Clinical presentation of osteochondromas can vary widely based on size and location. A substantial number of these tumors may remain asymptomatic, but there have been reports of dangerous consequences of nerve compression including sudden death and quadriplegia [12,13]. Most spinal or sacral osteochondromas would be expected to cause symptoms as a direct result of spinal canal or foraminal stenosis. Overall, manifestations of neurological symptoms are considered uncommon. However, based on the study of Albrecht et al. [8], as many

as 30% of spinal osteochondromas may eventually lead to cord compression. Sacral osteochondromas are rarely diagnosed with less than five reported cases in the literature to date, and most patients present with back pain and/or lower extremity pain.

Choosing the appropriate surgical approach is dictated by the location of the tumor and can be best visualized on CT scans. Preoperative planning with axial imaging allows measurement of the cartilaginous cap. A cap greater than 1 cm is concerning for malignancy. The CT scan is also recommended to clarify the site of attachment of osteochondroma [14] where the flaring of the bone cortex into the osteochondroma cortex is a pathognomonic feature [9]. In our case, based on the location and extent of the tumor, anterior approach was used through an oblique abdominal retroperitoneal incision. Because of the low risk of malignancy, “marginal” en bloc excision constitutes definitive treatment for osteochondromas where complete removal of the cartilaginous cap is still necessary to decrease the likelihood of local contamination and local recurrence [11,15,16]. Although it is important to resect as much tumor as possible, the risk of injuring nearby pelvic organs and neurovascular structures must be carefully weighed against potentially more damaging surgical intervention. Sacral osteochondromas near nerve roots are especially challenging because these neural functions are crucial to independent daily activities including the control of anal/urethral sphincter, parasympathetic stimuli to distal colon, and the function of certain muscle groups in lower extremities.

The risk of osteochondroma recurrence in the spine or the sacrum is not well known because of the rare occurrence. Based on literature review, Gille et al. [9] estimated 4% risk of recurrence in spine, slightly higher than the estimated 2% recurrence in long bones [17]. Multiple osteocartilaginous exostoses (also known as osteochondromatosis, hereditary multiple exostosis, Bessel-Hagel syndrome), which often result from an autosomal dominant inheritance, have much higher risk of malignant transformation at 10% to 15% as opposed to 1% in isolated solitary osteochondromas [17]. Uncommon diagnosis of osteochondromas in the spine and pelvis also results in relatively higher frequency of secondary chondrosarcoma, which could suggest higher risk of malignant transformation in axial skeleton [4]. After the surgical excision of the spinal or sacral osteochondroma, it is important to evaluate the patients for tumor recurrence and the thickness of the cartilaginous cap, which may all indicate risk factors for malignancy [4,17,18].

In conclusion, this report documents a rare solitary osteochondroma on the anterior sacrum presenting as low back and sciatic pain in an adult. When osteochondroma causes pain in an adult, we should expect that some structure is impinged or that it could have initiated a malignant transformation, so en bloc excision should be used to remove the tumor and histologic studies performed to

assess for malignant transformation. In this case, a battered nerve root syndrome occurred, which took 8 months to resolve.

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