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Full Length Research Paper

Osteoid osteoma of the radial shaft: An unusual location case report and literature review

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Osteoid osteoma has been found in nearly all bones and usually treated with radiofrequency ablation or laser photocoagulation. We reported a case of a radial shaft osteoid osteoma in a 37-year-old man and also reviewed the reported literature. The patient was presented with pain and swelling in his right forearm; conventional radiographic examination of forearm seemed to be normal; computerized axial tomography (CT or CAT) and magnetic resonance imaging (MRI) showed a typical image with a central 0.6 cm irregular lucent area surrounded by a zone of oval reactive bone sclerosis. The nidus was removed successfully by surgical excision and histological examination finally confirmed the diagnosis of osteoid osteoma.

Key words: Osteoid osteoma, radius, stress fracture.

INTRODUCTION

Osteoid osteoma is a benign bone tumor, which is most often found in the long bones, showing small in size and painful.It occurs relatively frequently and is typical of the second and third decades (Frassica et al., 1996).Osteoid osteoma is rarely seen before 5 and after 30 years of age, although patients as young as 3 years old have been reported (Illman et al., 1965; Rolf et al., 2006; Kaweblum et al., 1993). We present a case of an exceptional osteoid osteoma localized in the midshaft of radius with a 3 months history of right forearm pain due to recurrent wrist sprain that was initially identified as stress fracture by a local orthopedic surgeon until the tumor was finally diagnosed and a review of the literature with regard to diagnosis, differential diagnosis and treatment of the osteoid osteoma. The patient gave permission for this information to be submitted for publication.

MATERIALS AND METHODS

Case report

A 37-year-old man was referred to our department had pain in the right middle radius. He reported a 3-month history of pain, which had begun insidiously and increased over the previous 1 month and had recurrent right wrist sprain history. He was a People's Liberation Army (PLA) soldier and played basketball on a recreational level. The pain was elicited by contact in basketball field, which worsened at night. He attributed his complaints to overuse and consulted a local orthopedic surgeon in July, 2010. Physical examination showed there was a small swelling over the laterodorsal tegmental area of the right middle radius, initially diagnosed as stress fracture with no X-ray examination. There was no history of previous tumor and anti-inflammatory drugs provided pain relief.

The patient presented to our outpatient clinic with the relapse of bone pain in October, 2010. Forearm muscles atrophy was not obvious. Movements of wrist joint and elbow joint were mildly limited due to pain, which was aggravated by lesion squeezing. Laboratory studies including erythrocyte sedimentation rate (ESR), C-reactive protein (CRP) were unremarkable. Radiographic examination of lesion seemed to be normal (Figure 1A), thorough review of the radiograph (Lateral radiographs) showed a small radiodense lesion of the middle radius (Figure 1B). A computed tomography (CT) scan showed a typical image with a central 0.6 cm irregular lucent area surrounded by a zone of oval reactive bone sclerosis in bone cortex (Figure 2). The appearance was suggestive of an osteoid osteoma at the midshaft of the radius. Magnetic resonance imaging (MRI) showed a well-defined lesion with the same diameter (Figure 3). We had strong suspicion, due to characteristic MRI and CT findings, that it was an osteoid osteoma, no old stress fracture. There is anecdotal evidence that lesions closer to the cortical surface may have a higher propensity to resolve spontaneously (Motamedi et al., 2009). But for this patient,

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Figure 1. A, Antero-posterior radiographs of the right forearm showing no obvious abnormalities. **B,** Lateral radiographs of the right forearm, showing a region of thickened bone cortex at the middle radius (red arrow).

Figure 2. Axial view CT scan of the right forearm showing a clearly defined nidus with a hill-like protuberance.

with increasing pain and increased nidus, we seek definitive traditional treatment by surgical intervention.

A local curettage of the lesion with allogenous bone grafting was recommended. With surgical resection, it has been emphasized that lesion removal is essential for pain relief (Campanacci et al., 1999). However, in some cases where difficulty in localizing the lesion led to incomplete excision, symptoms have occasionally resolved (Sim et al., 1975). Under tourniquet ischemia, we used a dorsal approach, with palpation to determine the precise localization of lesion. After careful dissection, the lesion was found to be located on the surface of the right middle radius, sized approximately 0.8 cm, and the bone cortex had no violation. We

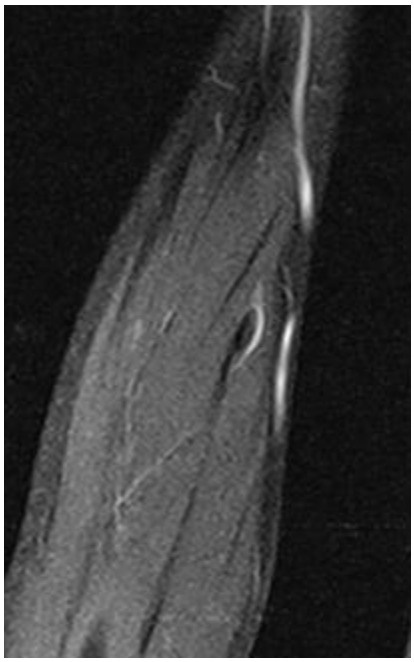

Figure 3. MRI showing a well-circumscribed oval lesion in the radius.

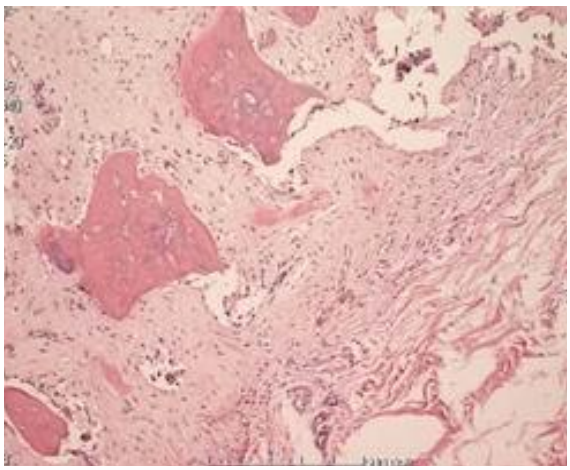

Figure 4. Histological section of the nidius showing irregular bone trabeculae and vascularized connective tissue (hematoxylin-eosin stain × 100).

opened a cortical window on the lesion that contained viscous bloody fluid. After thorough curettage using a oval curette, cancellous allografts were inserted into the bony defect. Immediate motion of adjacent joints was allowed, and the pain was relieved immediately after operation. Histological examination of this specimen demonstrated the nidus composed of irregular interconnecting trabeculae of woven bone in a cellular, strongly vascularized fibrous tissue background (Figure 4), thus confirming the diagnosis of osteoid osteoma combining with clinical symptoms and radiographic results. Skin sutures were removed 2 weeks postoperatively.

Figure 5. Postoperative radiographs taken 3 weeks following local curettage of the lesion with allogenous bone grafting.

The X-ray showed complete incorporation of the graft 3 weeks after surgery (Figure 5). At 2-year follow-up, the patient had no recurrence and had an excellent clinical outcome.

RESULTS AND DISCUSSION

Osteoid osteoma was first described by Jaffe as an entity in 1935 (Jaffe, 1935), which is a benign osteoblastic lesion characterized by a well-demarcated core usually less than 2.0 cm (reported more than 1200 cases) in diameter with surrounding reactive sclerotic bone (Kelly et al., 2002). With a peak incidence in the early decades of life, only 13% are older than 30 years of age, and there are approximately 3 times more commonly in males without any racial or ethnic predilection (Abnousi et al., 2008; Vallianatos et al., 2006).

Osteoid osteomas are traditionally classified into cortical, medullary or cancellous and subperiosteal or periosteal, which is most commonly located within the cortex (Aynaci et al., 2007). It can occur everywhere in the skeleton and the most common sites for this tumor are within the femur and tibia, with over 50% of all cases occurring in these locations (Ek and McCullough, 2010).

Although osteoid osteomas involving the midshaft of the radius is uncommon and it has ever been reported in the literature by Nakamichi and Tachibana (1996), the patient we reported had history of sprain of wrist joint, which was easy to lead to the misdiagnosis as stress fracture. To our knowledge, most of the osteoid osteomas are cortical, and cortical lesion shows more periosteal bone formation than medullary or subperiosteal osteoid osteomas, so it was normal for the patient in making the diagnosis delayed three months because of the atypical radiographic appearance. Despite the radiographic picture of osteoid osteomas may mimic stress fracture, and stress fracture also had the same pain and swelling at the injury site, which need taking non-steroidal anti-inflammatory medications for pain relief as well, CT or MRI can show the direct evidence to exclude it.

The differential diagnosis of osteoid osteomas includes tumors of bony origin, and fibro-osseous lesion should be taken into consideration. One of the challenging issues in the present case was differentiating the tumor from osteoblastoma. Some authors in their reports suggest that osteoblastoma and osteoid osteoma in gnathic regions are clinical and anatomic variants of the same osteoblastic-origin tumor and the pain symptom including type and intensity were similar when involving the sacrum in patients (Jones et al., 2006; Capanna et al., 1986). Despite the fact that osteoblastoma is called the giant osteoid osteoma because of its similarity in histopathologic features related to osteoid osteoma (Lucas et al., 1994), we believe that they are also distinguishable by clinical features and size.

We distinguished osteoid osteoma from osteoblastoma according to clinical features and radiographic findings. Osteoblastoma is less painful than osteoid osteoma and generally does not respond to salicylate treatment (Kransdorf et al., 1991). The osteoid osteoma nidus on MRI or CT usually measures less than 2 cm in diameter, and osteoblastoma usually has large size (Capanna et al., 1986). In addition, osteoblastoma presents with a more aggressive clinical course and cases of malignant transformation have been reported; (Canepa and Defabiani, 1965) described a radial osteoblastoma as an uncommon localization with tumor recurrence (Kransdorf et al., 1991; Canepa and Defabiani, 1965).

Differentiation of osteoid osteoma from osteosarcoma, especially low-grade types, may be difficult. In current case, osteosarcoma can be excluded because the tumor's behavior was consistent with a benign bone tumor, with respect to demarcated radiographic borders. In addition,the typical microscopic characteristics of osteogenic sarcoma includes cytological polymorphism, cellular atypia, or abnormal mitotic figures was lacking on this lesion.

Fibrous dysplasia is a benign, self-limiting disorder of the bone structure, the normal bone is replaced by cellular fibrous connective tissue, and it is usually painless (Neumann et al., 2003; MacDonald-Jankowski, 2009). The predominant initial radiographic appearance was that of a single eccentric patchy lesion with combined sclerotic/ground-glass features (Ippolito et al., 2003), neither of which were seen in this patient.

Osteoid osteoma is usually diagnosed by means of clinical examinations and radiological findings. Regarding the treatment method, surgical resection has been emphasized that it is essential for pain relief (Campanacci et al., 1999). Surgical options include en bloc resection and percutaneous minimally invasive methods that include resection, laser photocoagulation and radiofrequency ablation (Motamedi et al., 2009; Lee et al., 2006; Towbin et al., 1995; Gangi et al., 1998). The traditional surgical intervention is the direct excision or curettage, which can bring several disadvantages such as weakening the bone, using internal fixation, bone grafting or postoperative immobilization. The application of radiofrequency ablation was firstly described in the treatment of osteoid osteoma by Rosenthal et al. (1992). This method allows the precise delivery of heat under image guidance to the targeted tissue, therefore it is popular for surgeons because of difficulty in lesion localisation at surgery (Cantwell et al., 2004). Although invasive techniques seem to be very good which have also the disadvantage that it is more difficult to assure that all of the nidus is removed and lower the recurrence rate.In addition, radiofrequency ablation is still associated with some other complications. Using radiofrequency ablation have been damage to neighbouring neurovascular structures particularly if ablating cancellous bone (Jowett and Singh, 2010). Hoffmann et al. (2010) reported two major complications (broken drill, infection) and two minor complications (hematoma, prolonged pain) with this treatment; moreover, in 3 of 38 patients relapse occurred after 1, 14 and 32 months and biopsy was able to prove diagnosis in 14 of 29 (48%) cases (Hoffmann et al., 2010). Finstein et al. (2006) reported an unusual complication that a patient treated with radiofrequency ablation for tibial osteoid osteoma resulting in thermal necrosis of the skin (Finstein et al., 2006).

Although, radiofrequency ablation of osteoid osteoma is a highly effective, precise, minimally invasive and safe method of treating osteoid osteoma, we still performed an en block resection with allogenous bone grafting for treating this patient, which allowed a histological diagnosis to be made and there was no complications occurring; in early postoperative period the pain was relieved, no recurrence occurred after the resection of lesion on 2 year follow-up. However, radiofrequency ablation can play an important role in certain cases.

Conclusion

The shaft of radius is an uncommon location for osteoid osteoma and the tumor occurring in this part may not

always present with the typical features, which are frequently leading to missed or delayed diagnosis. A high index of suspicion is not only important, detailed imaging and examination are necessary to confirm the diagnosis, accurately localize the nidus and allow complete excision as well. Surgical resection remains the standard treatment in patients who tried conservative treatment have not obvious therapeutic effectiveness.

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