CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 34 (2017) 106–109

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Intramuscular hemangioma causing periosteal reaction and cortical hypertrophy misdiagnosed as osteoid osteoma☆

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ARTICLE INFO

Article history:
Received 6 December 2016
Received in revised form 12 March 2017
Accepted 13 March 2017
Available online 16 March 2017

Keywords:
Intramuscular hemangioma
Periosteal reaction
Osteoid osteoma

ABSTRACT

INTRODUCTION: Intramuscular hemangioma in the periosteal region is rare. Although comprising less than 1% of all hemangiomas, they represent the most common type of intramuscular tumors. When located adjacent to bone, a periosteal reaction can occur. The deep localization of the hemangioma poses the diagnosis difficult. Only 8% to 19% of cases were diagnosed before surgery according to the literature review.

PRESENTATION OF CASE: We present a case of forty-one-year-old female diagnosed with intramuscular hemangioma, mimicking osteoid osteoma, adjacent to the periosteal region of tibia diaphysis treated by surgical excision.

DISCUSSION: When intramuscular hemangioma occurs nearby a bone structure, it can cause cortical, medullary and periosteal bone changes that are frequently misdiagnosed by plain radiography. Due to their infrequency, deep location, and atypical presentation, these lesions are seldom diagnosed at presentation. The hemangioma of the periosteal region can be locally destructive due to compression exerted on neighboring structures. It does not regress spontaneously, and surgical excision is frequently needed.

CONCLUSION: Intramuscular hemangioma of periosteal region occurs most commonly adjacent to long bones of the lower limb. They can cause hypertrophic periosteal reactions mimicking a periosteal or parosteal tumor. Although osteoid osteoma was considered in the differential diagnosis, MRI with enhancement should be performed to exclude intramuscular hemangioma. This may avoid unnecessary aggressive en-bloc tumor excisions resulting in bone weakness and prolonged rehabilitation.

This case report has been written in line with the SCARE criteria (Agha et al., 2016 [1]).

1. Introduction

Intramuscular hemangiomas are benign neoplastic proliferations of blood vessels that arise from skeletal muscle. Although comprising less than 1% of all hemangiomas, they represent the most common type of intramuscular tumors [2,3] with a preoperative diagnostic rate of only 8%–19% [4]. When intramuscular hemangioma occurs nearby a bone structure, it can cause cortical, medullary and periosteal bone changes that are frequently misdiagnosed by plain radiography. Due to their infrequency, deep location, and atypical presentation, these lesions are seldom diagnosed at presentation. The hemangioma of the periosteal region can be locally destructive due to compression exerted on neighboring structures. It does not regress spontaneously, and surgical excision is frequently needed [5]. By recognizing that skeletal muscle is the most common site of deeply located hemangiomas, should orthopedic surgeons avoid misdiagnosis of this pathology and evolve a better understanding of its clinical and radiographic presentation. We reported a case of intramuscular hemangioma diagnosed by histology and localized adjacent to the bone in the lower limb with periosteal reaction mimicking an osteoid osteoma.

2. Presentation of case

A 41-year-old female presented with a 6-year history of calf pain in her right lower leg. Initial conservative therapy at a local medical doctor that consisted of aspirin or no steroid anti-inflammatory drugs failed to alleviate the patient's symptoms. The pain pro-

☆ No proprietary interest by any of the authors was given to any product or manufacturer mentioned.

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http://dx.doi.org/10.1016/j.ijscr.2017.03.013

gressed to the point of limiting her normal daily activities in the last three months. Although typical night pain was not observed, minimal pressure on the affected area resulted in severe pain that awakens her from sleep. There was no history of prior trauma or infection. The remaining medical history was unremarkable. On physical examination, there was a significant amount of pain elicited with palpation on mid-calf. No local palpable mass, warmth or bruit was noted. There was no obvious skin change overlying the painful area. The range of movements of knee and ankle were full and free. All laboratory studies were normal. Plain radiographs showed the irregular sclerotic periosteal reaction of the posterior mid-diaphyseal cortex of the right tibia. Although the mass lesion was not well visualized on images in the anteroposterior plane, it was well pictured on the lateral projection. There was no associated osseous destruction or fracture, nor were there calcifications adjacent to the mass. Soft tissue abnormalities were not identified within the tibia, and the visualized knee and ankle joints appeared normal (Fig. 1). Axial computed tomography (CT) scans with contrast revealed a cortically based sclerotic lesion that emanated from the posterior aspect of the tibial diaphysis (2.01 cm × 4.79 cm × 0.67 cm in the sagittal, coronal, and axial planes, respectively). There was no identifiable cortical breakthrough (Fig. 2).

Although the clinical symptoms and the radiological assessment were not typical, osteoid osteoma was still a concern, and other pathological osteo skeletal processes such as periostitis, eosinophilic granuloma, hemangioma of bone, non-ossifying fibroma, Paget's disease and avascular necrosis of the bone also should be considered. An open biopsy was performed. The operative strategy was pre-planned based on the preoperative imaging findings, dictating the position of the patient on the operating table. The palpable tender point over the posterior aspect of the right lower leg was marked using dermographic pencil. The lesion was localized and needle trajectory planned using a fluoroscopic guiding. The patient was set in prone position and under spinal anesthesia. A tourniquet was applied to the affected extremity prepped aseptically in the usual way. An incision was made longitudinally over middle calf according to the landmark prepared previously. The skin and soft tissue were dissected deep to periosteal area. The periosteum was dissected with elevator and cortical margins just underneath the posterior border of the shinbone. A large friable soft, lobulated mass, measuring 10 cm × 4 cm × 2 cm in length, wide and thickness respectively, was located on the surface of the tibial bone, interpolated within the soleus muscle fibers, and eroding the tibial cortex without evidence of intramedullary involvement (Fig. 3A). The thickened cortex was excised en-bloc with the adjacent soft tumorous mass (Fig. 3B and C). The resection was made with a little saw and osteotome.

Histology revealed poorly circumscribed dilated vascular channels in a loose fibrous stroma interspersed between striated muscle bundles (Fig. 4A). The presence of cells that stained positively for factor VIII confirmed the diagnosis of intramuscular cavernous hemangioma (Fig. 4B). The cortical bone was normal histologically, and a reactive sclerosis was identified. No nidus was found.

The patient was involved in light exercises and partial weight bearing walk for three weeks and then allowed to increase activity level as tolerated. Physical therapy was focused on muscle strengthening, proprioception, and range of motion exercises. Within one month, the patient reported much pain relief and discontinued all pain medications. Full weight bearing walk with no external support was permitted at the sixth-month follow-up. The symptoms resolved after the surgery and at eight months' follow-up, she was pain-free without evidence of tumor recurrence.

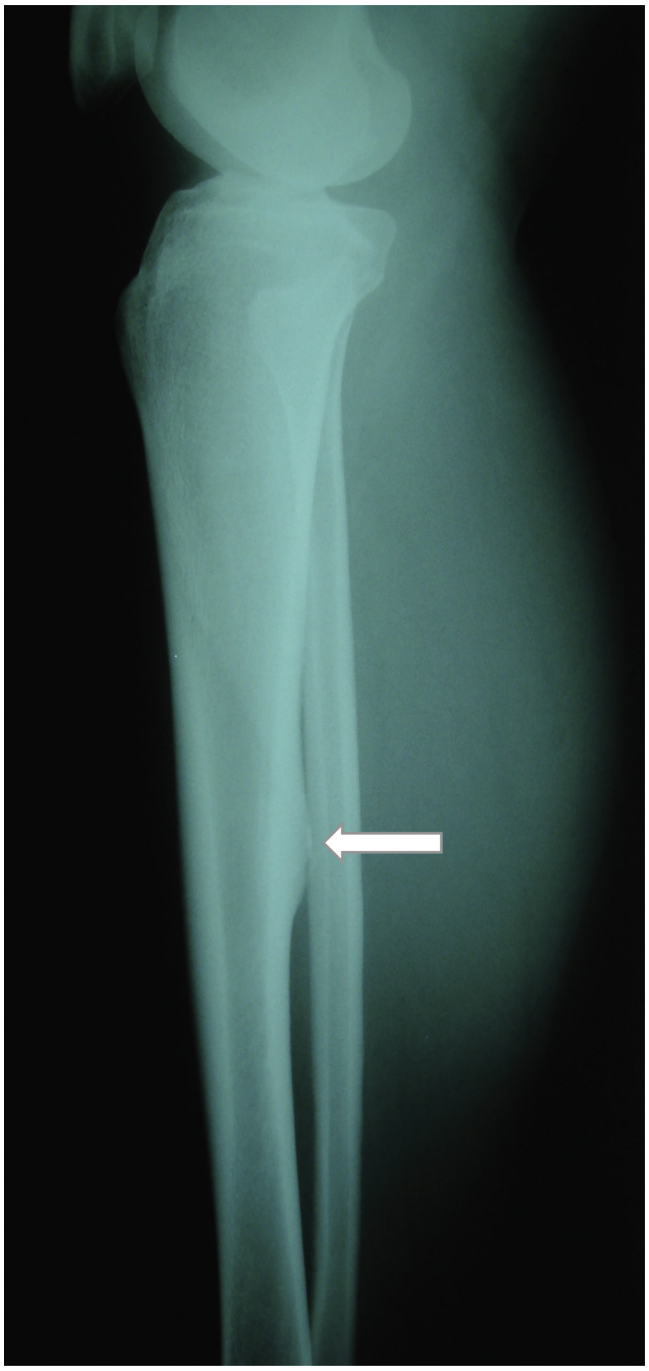

Fig 1. Plain radiograph showing periosteal reaction with cortical hypertrophy of the posterior mid-diaphyseal segment of the right tibia (white arrow).

3. Discussion

Intramuscular hemangioma in the periosteal region is rare [3,6,7] and account for 0.8% of all benign vascular tumors [8]. The diagnosis is usually difficult due to the deep localization of the hemangioma. Approximately 45% of described cases were located in the lower extremities, occurring predominantly in the 3rd decades of life in 90% of cases [4,9,10]. The highest prevalence was noted in young women [9]. The typical presentation is a painful mass, usually not associated with cutaneous changes [2,11]. Because of its location in the deep tissue, painful tendon contracture and functional impairment of the extremity can be complications of this slow growing tumor [4].