Joint Diseases and Related Surgery

Jt Dis Relat Surg
2024;35(2):417-421

CASE REPORT

Cortical hypertrophy in intramuscular hemangioma mimicking bone tumor

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ABSTRACT

Although hemangiomas are the most common soft tissue tumors, intramuscular hemangiomas account for only 0.8% of all vascular tumors. These lesions are rarely located adjacent to the bone and cause changes in the adjacent bone. They are often mistakenly diagnosed as bone tumors. In this study, a case of a 19-year-old male patient with intramuscular hemangioma causing cortical thickening was reported.

Keywords: Bone tumor, cortical hypertrophy, hemangioma, intramuscular, periosteal reaction.

Vascular tumors are considered a subset of vascular anomalies and have historically been difficult to classify. These are lesions with proliferative ability ranging from local findings to distant metastases.[1]

Skeletal hemangiomas are rare and make up less than 1% of all hemangiomas.[2] About 90% of intramuscular hemangiomas occur in young people, particularly those under 30 years of age. The area where these lesions like to settle the most is the lower extremity, specifically on the thigh. The most common complaints of the patients at the time of admission are swelling or pain.[3]

Localization of intramuscular hemangioma in the periosteal region is extremely rare. When they develop adjacent to the bone, a periosteal reaction may occur. Deep localization of the hemangioma developing adjacent to the bone makes the diagnosis difficult.[4] In this study, a patient with intramuscular hemangioma referred to our clinic with a preliminary diagnosis of osteoid osteoma was reported.

CASE REPORT

A 19-year-old male patient was referred to our clinic with pain and weakness in the right thigh. The patient reported that he had occasional pain in the right thigh for the last six years and that his right thigh was weaker than the left thigh for about five years. There was no night pain or pain that did not increase at night; however, it increased with activity. The patient could not remember any trauma and did not have any systemic or metabolic disease. On inspection, it was observed that the diameter of the right thigh was significantly less than the left thigh (Figure 1a).

The hair growth on both thighs was similar. On clinical examination, the diameter of the right thigh was approximately 2 cm less than the diameter of the left thigh. There was no limitation in hip and knee joint movements. There was mild pain on palpation of the right thigh. There was cortical thickening in the distal posterolateral aspect of the right femur on direct radiographs (Figure 1b).

On magnetic resonance imaging (MRI), a cortical thickening of approximately 9.5 mm was observed at

Received: January 05, 2024
Accepted: March 19, 2024
Published online: April 26, 2024

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Doi: 10.52312/jdrs.2024.1604

Citation: Öztürk R, Bulut EK. Cortical hypertrophy in intramuscular hemangioma mimicking bone tumor. Jt Dis Relat Surg 2024;35(2):417-421. doi: 10.52312/jdrs.2024.1604.

the metadiaphyseal level in the distal posterolateral region of the right femur. In addition, in the soft tissue adjacent to the cortical thickening, a hypointense change on T1-weighted sequences, approximately 18×12 mm in size, and hyperintense signal increase on T2-weighted sequences were detected. The lesion contours could not be clearly distinguished. There was significant contrast enhancement after intravenous contrast agent injection (Figures 2a, b). Cortical thickening in the distal region was confirmed by computed tomography. A nidus was not found (Figure 2c).

True-cut biopsy was performed on the soft tissue mass. A lesion characterized by large-scale vascular spaces including erythrocytes, lined with flattened endothelium without atypia, was observed in the sections examined. The diagnosis was reported as hemangioma.

The patient did not accept surgical excision surgery, and sclerotherapy was applied (Figure 3). The pain regressed with sclerotherapy. However, there was no change in leg weakness. No changes were observed in the follow-up MRI, except for a

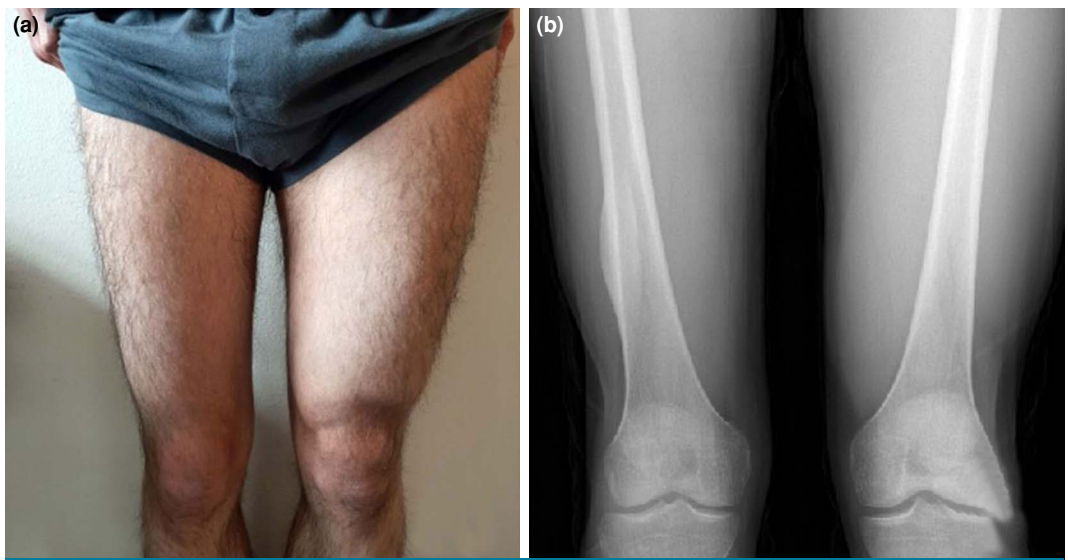

FIGURE 1. (a) On inspection, the right thigh appears weaker than the left. **(b)** Cortical thickening in the distal posterolateral right femur on direct radiography.

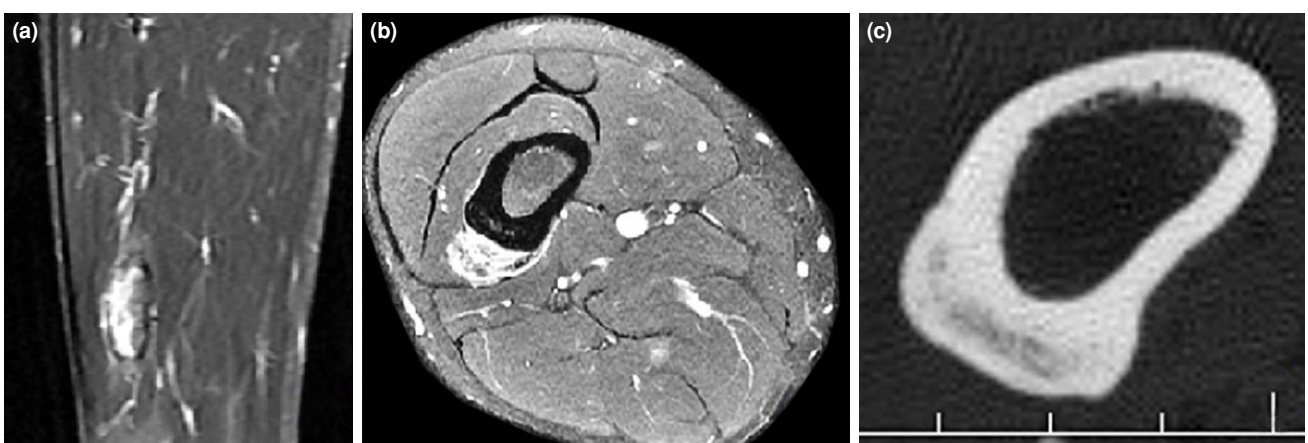

FIGURE 2. (a) A soft tissue mass with unclear borders adjacent to cortical thickening on coronal T2-weighted, fat-suppressed magnetic resonance imaging sequence and **(b)** axial T2-weighted sequence. **(c)** The nidus is not visible in the center of cortical thickening on computed tomography.

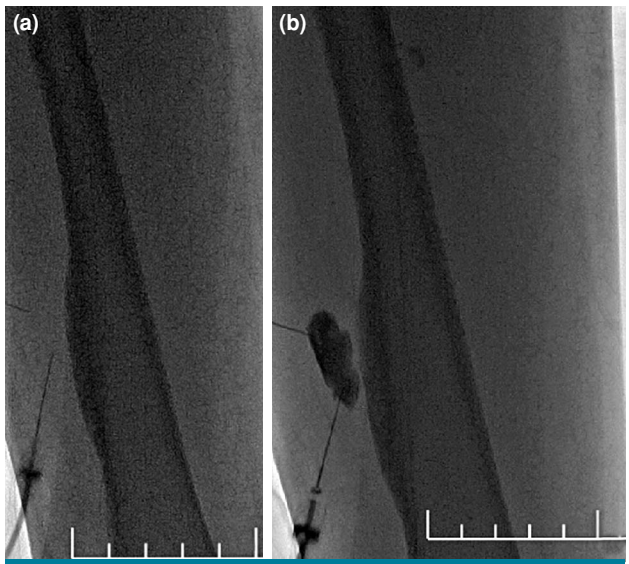

FIGURE 3. (a, b) An image of the percutaneous alcohol ablation therapy applied to the patient under general anesthesia.

slight decrease in size (Figures 4a, b). The patient has been under follow-up for 24 months.

DISCUSSION

Although hemangiomas are the most common soft tissue tumors, intramuscular hemangiomas constitute only 0.8% of all vascular tumors.[4,5] They may be asymptomatic or present with mass or pain. In the current case, the complaints were weakness and pain in the relevant lower extremity.

Magnetic resonance imaging is useful in the diagnosis of intramuscular hemangioma, and the images are diagnostic in about 90% of cases. On T1-weighted images, they typically appear hypointense or isointense relative to muscle. Intralesional fat areas may be observed. On T2-weighted sections, they have a heterogeneous and hyperintense appearance. There are tubular vessels aligned in parallel in the images. Signal gaps called phleboliths may also be observed. Conversely, in osteoid osteoma, there is intense edema around the lesion on MRI. It does not signal the sclerotic lesion. The diagnostic value of MRI in osteoid osteoma is low and may lead to misdiagnosis.[6,7] Direct radiographs should be performed in all tumors. Phleboliths can be observed in hemangiomas on direct radiographs.[5,6] In this case, the lesion was isointense compared to muscle on T1-weighted sequences and hyperintense on T2-weighted sequences. In all sequences, millimetric-sized hypointense areas (phleboliths) were present.

Most superficial cutaneous hemangiomas can often be diagnosed by history and physical examination. Diagnostic difficulty usually occurs in deep-seated lesions. In such cases, ultrasound is usually sufficient as an imaging method to confirm the diagnosis. This method stands out with its cheapness, speed, and

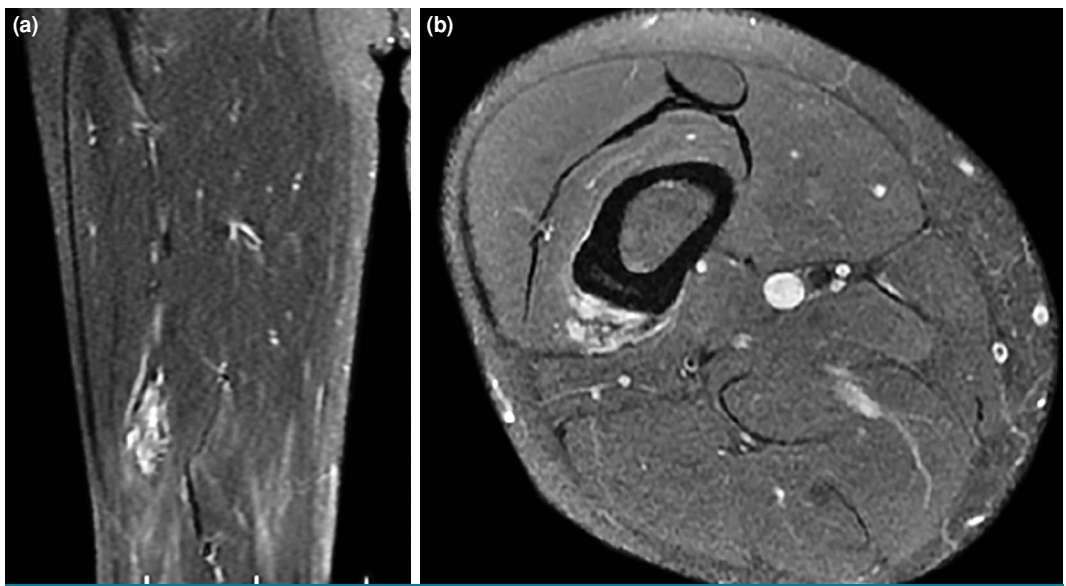

FIGURE 4. (a) Coronal T2-weighted, fat-suppressed sequence and **(b)** axial T2-weighted sequence on 24-month control magnetic resonance imaging showing a reduction in lesion size.

usefulness.[7,8] However, in this case, unfortunately, an ultrasound was not performed initially. The bone lesion observed on the direct radiograph, which was first performed due to thigh pain, made the physician think of a bone tumor. Therefore, MRI and computed tomography were taken for further evaluation. The diagnosis was confirmed by histopathological examination. Similarly, in most of the case reports in the literature, the lesions on plain radiographs were subsequently further evaluated with MRI. Afterward, a histopathological examination was performed since the clinical and radiology findings were not typical.[4,9-11] However, unlike these cases, bone biopsy or bone resection was not performed in our case. This case is a representation of the fact that benign vascular lesions causing bone findings can be managed without the need for bone intervention after a good preoperative analysis. In all of the case reports already mentioned, the histopathological evaluation results of the bone were reported as normal bone changes.

Localization of intramuscular hemangioma in the periosteal region is extremely rare. When an intramuscular hemangioma forms near a bone structure, it can cause cortical, medullary, and periosteal bone changes that are often misdiagnosed with plain radiography.[4] One of these misdiagnoses is osteoid osteoma. The best differential diagnosis of osteoid osteoma is made by computed tomography. In this case, osteoid osteoma was excluded since no nidus was detected in the thin-section tomography.

There are only rare case reports on this subject in the literature. In 1996, DeFilippo et al.[9] reported a hemangioma located adjacent to the proximal ulna. The lesion had caused a periosteal reaction and cortical expansion. The lesion was thought to stimulate a bone tumor. Biopsy samples were taken from the lesion, periosteum, and bone. Bone and periosteal biopsy results were reported as normal bone tissue and periosteal resection, respectively. In 2015, Shikhare et al.[10] reported an intramuscular hemangioma located adjacent to the tibia. An aggressive parosteal bone tumor was considered when imaging findings were examined. Osteoid osteoma, parosteal osteosarcoma, and Ewing's sarcoma were considered in the differential diagnosis. While the soft tissue tumor was completely excised, a part of the bone tissue was excised and sent for histopathological examination. No abnormality was found in the excised bone.

In 2017, Yeh et al.[4] reported a case of hemangioma confused with osteoid osteoma. Osteoid osteoma was considered in the preliminary diagnosis of cortical thickening in the tibial diaphysis. In the patient who was scheduled for surgery, the cortical thickening was excised. The soft tissue that was noticed adjacent to the lesion during surgery was also excised. No nidus was found in the histopathological examination, and the diagnosis of the excised soft tissue was reported as hemangioma.

Hemangioma located adjacent to the periosteum has been reported in the fibula, tibia, and ulna.[12] To our best knowledge, it has not been previously reported in the femur.[13] In the management of these lesions, only follow-up, corticosteroids, cryotherapy, sclerosing agent injection, arterial ligation, embolization, and surgical excision are among the options.[6,9] The main purpose of sclerotherapy in vascular lesions such as hemangioma is to cause thrombosis in the hemangioma by causing maximum damage to the endothelium of the vascular bundles.[14] In these lesions, ethanol is considered one of the most effective sclerosing agents with low recanalization rates.[14,15] In our case, sclerotherapy was applied using alcohol, and the patien''s pain symptoms regressed.

In conclusion, intramuscular hemangiomas located adjacent to the periosteum may cause hypertrophic periosteal reactions that mimic a periosteal or parosteal tumor. Contrast-enhanced MRI or ultrasound should be performed for the differential diagnosis of intramuscular hemangioma in cases with suspected osteoid osteoma. Osteoid osteoma should be suspected specifically when a nidus is not found on computed tomography and in patients with atypical symptoms.

Patient Consent for Publication: A written informed consent was obtained from patient.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Contributed significantly to data collection and assisted in manuscript preparation: E.K.B.; Wrote the manuscrpit and critically revised the manuscript for important intellectual content; All authors have read and approved the final version of the manuscript: R.Ö.

Conflict of Interest: The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding: The authors received no financial support for the research and/or authorship of this article.

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