Case Report

Ossified hemangioma of the popliteal fossa: imaging findings with pathologic correlation

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Abstract: Ossified hemangioma is a rare benign hemangioma, composed of mature bones and numerous dilated vascular channels lined by benign endothelial cells. Here, we report a case in a 30-year-old man presenting a painless mass in the right popliteal fossa. On imaging studies, x-ray revealed a soft tissue mass, occupying the right popliteal fossa with massive and spotty calcification. Magnetic resonance imaging (MRI) showed a mass of intermediate signal intensity on T1WI and high signal intensity on T2WI. Total resection of the lesion was performed. The diagnosis of ossified hemangioma was made based on morphological examination. Clinicians and radiologists should recognize the features of such lesion.

Keywords: Hemangioma, ossification, popliteal fossa, X-ray, magnetic resonance imaging

Introduction

Hemangiomas are common benign vascular tumor, which usually appear in infancy, presenting as a slowly growing mass [1]. Even though calcification in hemangioma is relatively common, extensive ossifications within the hemangioma is rare, with only sporadic cases reported. Herein, we report a rare case of ossified hemangioma in the right popliteal fossa of a 30-year-old man. The final diagnosis was established by pathological examination after an open excision of the lesion. To the best of our knowledge, such a presentation of the lesion in the right popliteal fossa has not been reported before. In the present study, the pathological results and x-ray and magnetic resonance imaging (MRI) findings of ossified hemangioma are described.

Case report

A 30-year-old man was admitted to the Eighth Affiliated Hospital, Sun Yat-sen University, Shenzhen, China with complaints of a history of three-year of popliteal fossa mass and occasional distending pain. He had no medical history of trauma or infection. On admission, there

was a bump occupying his right popliteal fossa with an ill-defined boundary. The movement of the right knee was slightly limited when compared to normal knee function.

The lateral x-ray showed a soft tissue mass, occupying the right popliteal fossa with massive and spotty calcification (Figure 1). Nonenhancement MRI of the right knee showed a juxta-articular mass with intramuscular component. T1-weighted images revealed an isointense lesion, but inhomogeneous signal intensity within it (Figure 2A). T2-weighted images demonstrated a high signal intensity lesion (Figure 2B).

Intraoperatively, a juxta-articular bluish lobulated mass was found to infiltrate the gastrocnemius muscle in the right popliteal fossa. On gross examination, the mass was ill-defined, measuring 11×3×2.8 cm in size. The cut surface demonstrated a vascular lesion with irregularly shaped bone.

On microscopic examination of the HE sections, the lesion was mainly consisted of thick, mature bones and numerous dilated vascular channels lined by benign endothelial cells (**Figure 3A**). In



Figure 1. X-ray lateral imaging of the right popliteal fossa. A soft tissue mass occupying the right popliteal fossa with massive and spotty calcification.

some area, the vessels infiltrated between skeletal muscle fibers (Figure 3B).

To date, one year after operation, the patient is well without evidence of recurrence.

Discussion

Hemangiomas are benign tumors of the blood vessels and often present with slowly enlarging soft tissue masses. The lesion predominantly affects infant and child. Although the etiology of hemangiomas remains unclear, some authors believe that they are attributed to congenital lesion. Calcification is a common feature of hemangioma. However, ossification within the hemangioma is a rare radiologic and histological phenomenon, which is considered to be the result of dystrophic calcification within organizing thrombi [2]. On the basis of the endothelial walls and vascular lumen, hemangiomas are classified into the capillary, cavernous and mixed types. The most common type of ossified hemangioma is cavernous hemangioma [3]. To the best of our knowledge, only a few cases of ossified hemangioma have been reported in the English literature. Most of them are found in head and neck regions [4-7]. Ossified hemangioma originated from popliteal fossa has not been reported before.

Histologically, ossified hemangioma contains thick, mature lamellar bones and numerous dilated vessels lined by bland endothelial cells. The presence of adipose tissue is common. The striking feature is the presence of cavernous vascular channels in between the ossified component. Sometimes, the mature bone may be misinterpreted as an evidence of osteoma. Pathologists should be aware of this rare feature to avoid erroneous diagnose. Similar to the previous reports, the lesion of our case was composed of extensive mature bone and large vessels lined by flattened epithelium of cavernous type. These dilated and sinusoidal vessels were separated by connective septa, located within the bone. These findings confirmed the diagnosis of ossified cavernous hemangioma.

Although ossified hemangioma has distinct clinicopathological features, they are not familiar to most radiologists. On x-ray, ossified hemangioma within muscle has been reported to have a unique feature, classically described as "Swiss cheese" appearance [8]. This appearance reflects the architecture of the ossified component interspersed with cavernous veins. This typical appearance was also present in our case. Due to the presence of irregularly ossified lesions in the soft tissues on radiologic examinations, myositis ossificans, ossifying fibromyxoid tumor, early extraskeletal osteosarcoma and synovial sarcoma should be considered in the differential diagnosis. Myositis ossificans is characterized by zonal phenomenon presenting peripheral ossification in the late stage. Ossifying fibromyxoid tumor of soft parts has faint ossification in the periphery of the lesion. Extraskeletal osteosarcoma can appear as a progressively enlarging lesion with central ossification. Tippled amorphous calcification is an important radiological imaging feature of synovial sarcoma. Therefore, Swiss-cheese-like appearance is suggestive of a hemangioma with ossification rather than other lesions. When ossified hemangioma is suspected, adequate preoperative planning can be made to minimize blood loss.

MRI of the present case showed isointense signal on T1WI and high signal intensity on T2WI. In addition, the lesion contained heterogeneous signal intensity, corresponding to ossification. The finding was consistent with previous report on the intramuscular ossified haemangiomas [9, 10]. It has been reported that

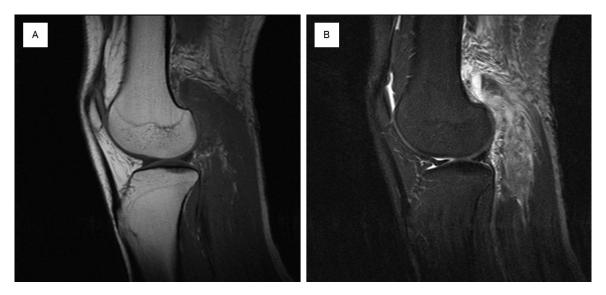


Figure 2. MRI scans of the mass located in the popliteal fossa. A. T1-weighted MR images revealed an isointense lesion mixed with heterogeneous signal intensity. B. T2-weighted images demonstrated a high signal intensity lesion.

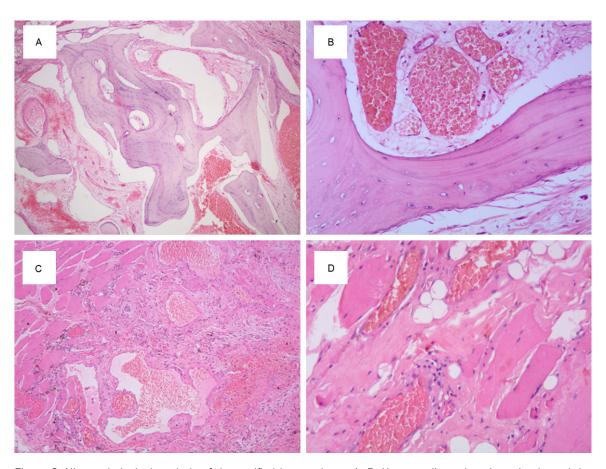


Figure 3. Histopathological analysis of the ossified hemangioma. A, B. Hematoxylin and eosin stain showed the lesion was mainly consisted of thick, mature bones and numerous dilated vascular channels lined by benign endothelial cells (magnification, ×20 and ×100). C, D. In some area, the vessels infiltrated between skeletal muscle fibers (magnification, ×40 and ×100).

the lesion exhibited serpentine or lattice like density on MRI with contrast enhancement

[11]. Unfortunately, enhanced MR scan was not performed in our case. In our view, MRI is the

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investigation of choice which may facilitate the correct preoperative diagnosis of ossified haemangiomas.

Ossified hemangiomas involving muscles have a high local recurrence rate because of their infiltrating growth pattern. Thus, complete excision is the best choice of treatment. Consequently, the role of preoperative imaging is to determine scope of the lesion and guide the clinical treatment.

In conclusion, we here present a rare case of ossified hemangioma of popliteal fossa in a young man. This unusual presentation can cause misdiagnosis. Ossified hemangioma is a benign lesion associated with a good prognosis. Therefore, understanding the imaging, pathological and clinical features of ossified hemangioma is important in the diagnosis and differentiation.

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Disclosure of conflict of interest

None.

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References

[1] Ly JQ, Sanders TG, Mulloy JP, Soares GM, Beall DP, Parsons TW and Slabaugh MA. Osseous change adjacent to soft-tissue hemangiomas of the extremities: correlation with lesion size and proximity to bone. AJR Am J Roentgenol 2003; 180: 1695-1700.

- [2] Nagira K, Yamamoto T, Marui T, Akisue T, Yoshiya S and Kurosaka M. Ossified intramuscular hemangioma: multimodality imaging findings. Clin Imaging 2001; 25: 368-372.
- [3] Wang YC, Jeng CM, Wu DY, Chang CY and Resnick D. Giant ossified cavernous hemangioma of an extremity associated with an equinovarus deformity. Skeletal Radiol 1998; 27: 522-524.
- [4] Curtin HD, Jensen JE, Barnes L Jr and May M. "Ossifying" hemangiomas of the temporal bone: evaluation with CT. Radiology 1987; 164: 831-835.
- [5] Shimoji T, Murakami N, Shimizu A, Sato K and Ishii S. Cavernous hemangioma with bone formation in a child: case report. Neurosurgery 1984; 14: 346-349.
- [6] Freeman JL, Shemen LJ, Alberti PW, Holgate R, Pritzker KP and Noyek AM. Ossifying capillary hemangioma of the maxillary and ethmoid sinuses - a case report. J Otolaryngol 1981; 10: 481-492.
- [7] Naim R, Steinhoff I, Hormann K and Maurer JT. Ossifying haemangioma of the frontal sinus. ORL J Otorhinolaryngol Relat Spec 2004; 66: 98-100.
- [8] Engelstad BL, Gilula LA and Kyriakos M. Ossified skeletal muscle hemangioma: radiologic and pathologic features. Skeletal Radiol 1980; 5: 35-40.
- [9] Jin W, Kim GY, Lee JH, Yang DM, Kim HC, Park JS and Ryu KN. Intramuscular hemangioma with ossification: emphasis on sonographic findings. J Ultrasound Med 2008; 27: 281-285.
- [10] Brinda MA, Manjunath S, Balasubrahmaniya KS, Manjunath RD and Nanjaiah B. Intramuscular ossified haemangioma: a rare case report. J Clin Diagn Res 2015; 9: PD19-21.
- [11] Greenspan A, McGahan JP, Vogelsang P and Szabo RM. Imaging strategies in the evaluation of soft-tissue hemangiomas of the extremities: correlation of the findings of plain radiography, angiography, CT, MRI, and ultrasonography in 12 histologically proven cases. Skeletal Radiol 1992; 21: 11-18.