

CASE REPORT

Nagoya J. Med. Sci. 75. 125 ~ 129, 2013

A SURFACE-BASED HEMANGIOMA OF THE RADIUS WHICH POSED RADIOLOGICAL DIAGNOSTIC DIFFICULTIES

TAKAAKI SHINOHARA, MASAHIRO TATEBE, MICHIRO YAMAMOTO,
SHIGERU KURIMOTO and HITOSHI HIRATA

Department of Hand Surgery, Graduate School of Medicine, Nagoya University, Nagoya, Japan

ABSTRACT

Surface-based bone hemangiomas are uncommon, mostly occurring in long bones of the lower extremities. Radiographic surface-based bone hemangioma images may present various features that provide a diagnostic challenge. We report a case of surface-based hemangioma of the radius that posed radiological diagnostic difficulties. The tumor size was small and the cortical reaction was subtle. MRI findings were nonspecific and angiography was useful in the assessment of painful but less obvious surface-based bone hemangioma. The patient's pain improved following surgery and no tumor recurrence was observed at 3 years after surgery.

Key Words: angiography; diagnostic imaging; hemangioma; radius; recurrence

INTRODUCTION

Surface-based bone hemangiomas are relatively rare benign soft tissue tumors, mostly occurring in lower extremity long bones^{1,2}. There have been a few case reports of upper extremity surface-based hemangioma of the ulna or humerus, but a radius lesion has not been reported. Many of the lesions show localized cortical thickening and some cases show cortical erosion on plain radiographs¹. These hemangiomas can provide a diagnostic challenge radiographically, as the cortical reaction may mimic many different lesions³. Magnetic resonance imaging (MRI) findings are nonspecific with low signal intensity on T1-weighted images and high signal intensity on T2-weighted images. We report a case of recurrent surface-based hemangioma of the radius that posed radiological diagnostic difficulties.

CASE REPORT

A 22-year-old woman presented with left radial wrist pain that began 1 month earlier. She had a history of a resected hemangioma of the wrist twice at the age of 9 years and 13 years

Received: November 21, 2012; accepted: December 17, 2012

Corresponding author: Takaaki Shinohara

Department of Hand Surgery, Graduate School of Medicine, Nagoya University,
65 Tsurumai-cho, Showa-ku, Nagoya 466-8550, Japan

Tel: +81-52-744-2957, Fax: +81-52-744-2964, E-mail: hand_surgery1966@yahoo.co.jp

at another hospital. There was a surgical scar on the radial part of the flexor carpi radialis tendon, but there were no other abnormal physical findings. Radiographic examination of the distal radius metaphysis demonstrated slight cortical bone thinning on the radial side (Fig. 1). MRI showed ambiguous lesions with low signal intensity on T1-weighted images and high signal intensity on T2-weighted images (Fig. 2). Angiography revealed a pooling of contrast medium along the area of cortical thinning (Fig. 3). Pain increased and interfered with daily life 2 years after the first visit. Therefore, surgical treatment was performed at the patient's request 2 years and 4 months after the first visit. A second MRI examination was performed 1 month before surgery; however, the findings were same as for the first MRI. A $15 \times 7 \times 3$ mm blood-tinged multilocular cystic mass was identified at the radial side of the distal radius metaphysis upon surgical exploration. The tumor was contained in the periosteum and was removed en-block with the thickened periosteum (Fig. 4). Histologic examination confirmed a diagnosis of surface-based bone hemangioma (Fig. 5). The patient had no pain and no signs of tumor recurrence on MRI and magnetic resonance angiography at 3 years and 3 months after surgery.

DISCUSSION

Surface-based bone hemangiomas are uncommon; there are approximately 30 previously reported cases, mostly occurring in long bones of the lower extremities^{1,2}. Bone hemangiomas are benign, malformed vascular lesions that account for less than 10% of all hemangiomas. Bone hemangiomas usually occur in the medullary cavity of the vertebral body and skull, accounting for 75%, and in the scapula, ribs, clavicle, and pelvic bones, accounting for an additional 15–20% of all cases⁴. Since most of these lesions are asymptomatic, they are usually discovered



Fig. 1 Anteroposterior radiographs of the left wrist, showing slight cortical bone thinning (arrows) of the radial side of the distal radius metaphysis.

A SURFACE-BASED HEMANGIOMA OF THE RADIUS



Fig. 2 Coronal T2-weighted magnetic resonance image of the left wrist, showing ambiguous lesions (arrows) with high signal intensity in the area of cortical thinning.



Fig. 3 Angiography of the left wrist, showing the pooling of contrast media (arrows) along the area of cortical thinning.



Fig. 4 15 × 7 × 3 mm blood-tinged multilocular cystic tumor contained in the periosteum.

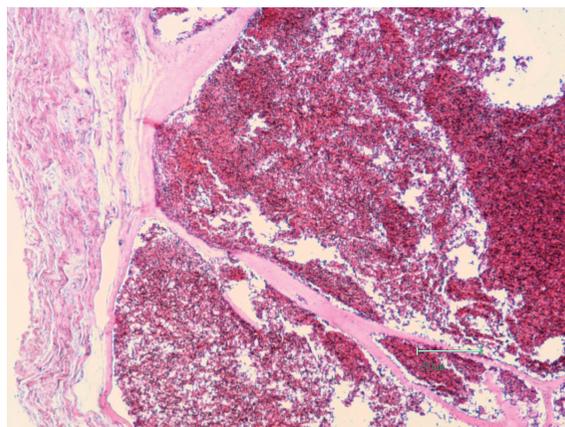


Fig. 5 Photomicrograph of the lesion, showing a cavernous hemangioma with dilated, thin-walled vascular spaces that contain blood cells in the vicinity of the periosteum (hematoxylin-eosin stain; magnification × 40).

incidentally on imaging examination. In contrast, bone hemangiomas of the extremities, though much less frequent than the axial bones, are symptomatic in over 90% of cases^{2,5,6}. Kaleem conducted a meticulous review of English-language medical literature and found 39 cases of extremity bone hemangioma with detailed clinical data. Forty percent of the patients had pain alone, 37% had pain associated with local swelling and or a mass, 7% had swelling alone, and 7% presented because of pathologic fractures². Although previous papers emphasized the extreme rarity of surface-based bone hemangiomas, these tumors may be more common than previously reported^{3,7,8}. Kaleem reported that the ratio of medullary lesions to surface-based lesions is three to one². The largest single series of surface-based lesions was reported by Devaney (11 cases). Among 22 cases (11 previously reported cases listed by Kaleem plus 11 cases reported by Devaney), the tibia (8 cases) was the most frequent site of involvement, followed by the fibula (7

A SURFACE-BASED HEMANGIOMA OF THE RADIUS

cases), ulna (3 cases), femur (2 cases) and humerus (2 cases)^{1,2)}. To the best of our knowledge, there is no previously published case report of surface-based hemangioma of the radius.

Medullary lesions usually show circumscribed, lucent lesions with a soap bubble appearance. However the diagnosis of surface-based lesions is often difficult, especially when the tumor size is small. On plain x-rays, surface-based lesions usually show localized cortical thickening, while some cases show cortical erosion¹⁾. The periosteal reaction around the surface-based lesion, if present, offers an excellent diagnostic clue. In the absence of the periosteal reaction, the cortical reaction can easily be missed, especially when the finding is subtle, as seen in the presented case. Additionally, cortical thinning is not typically observed on radiography of surface-based bone hemangioma. MRI findings are nonspecific, with low signal intensity on T1-weighted images and high signal intensity on T2-weighted images, and the surface-based lesion always lacks bone marrow changes. Therefore, it can be overlooked when the lesion is extremely small. In the presented case, the patient had a history of a resected hemangioma twice at the age of 9 years and 13 years. MRI images taken on two independent occasions did not clearly depict a tumorous lesion in the area of cortical thinning. Therefore, we could not confirm tumor recurrence until we carried out angiography. A blood-tinged multilocular cystic mass was confirmed at the contrast medium pooling area and the histological diagnosis was surface-based bone hemangioma. Although contrast medium pooling along the thinned cortex was not obvious, angiography findings were consistent with both macroscopic and microscopic findings.

Reports in the literature indicate that there is a high rate of recurrence after incomplete resection and emphasize the importance of en-block excision to prevent recurrence^{1,3)}. The patient underwent twice surgical resection of the hemangioma at the radial side of the wrist and surface-based bone hemangioma was located in the deep layer of a previous surgical wound this time. Therefore, this case was considered recurrence after incomplete resection. We strongly believe that meticulous localization of the lesion with angiography is essential in the assessment of painful but less obvious surface-based hemangioma of bones.

REFERENCES

- 1) Devaney K, Vinh TN, Sweet DE. Surface-based hemangiomas of bone: a review of 11 cases. *Clin Orthop Relat Res*, 1994; 300: 233–240.
- 2) Kaleem Z, Kyriakos M, Totty WG. Solitary skeletal hemangioma of the extremities. *Skeletal Radiol*, 2000; 29: 502–513.
- 3) Rougraff BT, Deters ML, Ivancevich S. Surface-based hemangioma of bone: three case studies and a review of the literature. *Skeletal Radiol*, 1998; 27: 182–187.
- 4) Unni KK. Dahlin's bone tumors: general aspect and data on 11,087 cases, 5th ed. 1996, Lippincott-Raven, Philadelphia.
- 5) Jaffe HL. Tumors and Tumorous conditions of the bones and joints. 1958, Lea & Febiger, Philadelphia.
- 6) Wilner D. Radiology of bone tumors and allied disorders. 1982, WB Saunders, Philadelphia.
- 7) Kenan S, Bonar S, Jones C, Lewis MM. Subperiosteal hemangioma: a case report and review of the literature. *Clin Orthop Relat Res*. 1988; 232: 279–283.
- 8) Sugiura I. Tibial periosteal hemangioma. *Clin Orthop Relat Res*. 1975; 106: 242–244.