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Cavernous medullary hemangioma of the distal left femur: a case report

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Bone hemangiomas occur in only one percent of primary bone neoplasms, and their diagnosis is difficult. The location of these benign neoplasms in long bones is even more rare. Objective. To describe the features of the diagnosis and surgical treatment of a woman with cavernous medullary hemangioma of the distal femur. Methods. The main diagnostic methods were computed tomography, radiography, and histopathological examination of the surgical specimens. Treatment was surgical removal of the neoplasm followed by rehabilitation with dosed loading of the limb. Results. A 45-year-old woman presented to the clinic with persistent aching pain in the left knee joint that did not stop after taking analgesics. Radiologically and using magnetic resonance imaging, an area of destruction and a neoplasm of irregular shape with clear uneven contours were found in the distal epimetaphysis of the left femur. Over 2 years of observation, the volumetric neoplasm did not increase on the radiographs, but the pain syndrome did not disappear and intensified during physical exertion. The results of the trephine biopsy did not allow to determine the exact diagnosis. Surgical treatment was carried outby means of parietal resection of the pathological focus of the distal part of the left femur and combined replacement of the defect with bone allograft with cement and fixation with a plate and screws. Morphological changes detected in the surgical specimens during histopathological examination corresponded to the diagnosis of cavernous hemangioma of the bone. At six months postoperatively, the patient demonstrated near-complete painless weight-bearing on the operated limb with minimal use of a cane, and the knee joint's range of motion was fully restored. Conclusions. Cavernous hemangioma of the femur is difficult to diagnose using trephine biopsy alone; accurate diagnosis is typically possible only through analysis of the surgical specimen. Surgical treatment enabled painless weight-bearing on the left limb by the sixth month of follow-up.

Кісткові гемангіоми зустрічаються лише в одному відсотку первинних новоутворень кісток, а їхня діагностика є складною. Розташування цих доброякісних новоутворень у довгих кістках є ще більш рідкісним. Мета. Описати особливості діагностики та хірургічного лікування жінки з кавернозною медулярною гемангіомою дистального відділу стегнової кістки. Методи. Основними діагностичними методами були комп'ютерна томографія, рентгенографія, патогістологічне дослідження операційного матеріалу. Лікуванням було хірургічне видалення новоутворення з подальшою реабілітацією з дозованим навантаженням кінцівки. Результати. Жінка 45 років звернулася до клініки з постійним ниючим болем у лівому колінному суглобі, який не припинявся після прийому анальгетиків. Рентгенологічно та за допомогою магнітно-резонансної томографії виявили в дистальному епіметафізі лівої стегнової кістки ділянку деструкції та новоутворення неправильної форми з чіткими нерівними контурами. За 2 роки спостереження на рентгенограмах об'ємне новоутворення не збільшувалося, але больовий синдром не зник і посилювався під час фізичних навантажень. Результати трепан біопсії не дали змогу визначити точний діагноз. Провели хірургічне лікування шляхом пристіночної резекції патологічного вогнища дистального відділу лівої стегнової кістки та комбінованого заміщення дефекту алоімплантатом із цементом і фіксацією пластиною та гвинтами. Морфологічні зміни виявлені в операційному матеріалі під час патогістологічного аналізу відповідали діагнозу кавернозна гемангіома кістки. Через 6 міс. після хірургічного втручання пацієнтка майже повністю навантажувала оперовану кінцівку без болю з частковим використанням тростини, обсяг рухів колінного суглоба відновився. Висновки. Кавернозну гемангіому в стегновій кістці складно виявити за допомогою трепан біопсії, лише аналіз операційного матеріалу дає змогу точно встановити діагноз. Хірургічне лікування допомогло відновити навантаження на ліву кінцівку без больового синдрому на 6-й місяць спостереження. Ключові слова. Новоутворення кісток, внутрішньокісткова гемангіома, рентгенографія, комп'ютерна томографія, магнітно-резонансна томографія, гістологія.

Keywords. Bone neoplasm, intraosseous hemangioma, radiography, computed tomography, magnetic resonance imaging, histology

Introduction

Vascular tumors of long bones are rare and difficult to diagnose. They are classified as benign (hemangioma), intermediate-locally aggressive (epithelioid hemangioma), intermediate with rare metastasis (pseudomyogenic hemangioendothelioma), and malignant (epithelioid hemangioendothelioma and angiosarcoma) [1]. Hemangioma is a proliferation of blood vessels lined by a single layer of flattened endothelial cells. Among primary bone neoplasms, they occur in less than 1 % of cases [2]. In the skeleton, hemangioma is most often found in the vertebral bodies or skull, while it is not common in other areas. The difficulty of diagnosing cavernous hemangioma is due to its extremely diverse presentation. Namely, from asymptomatic lesions and incidental findings on radiographs to severe pain syndrome [3]. Differential diagnosis of hemangioma of long bones includes giant cell tumor, aneurysmal bone cyst, plasmacytoma, etc. In the vast majority of patients with hemangiomas, surgical removal of the lesion is effective with a favorable prognosis, and there is usually no need for specific treatment.

Purpose: to describe the features of diagnosis and surgical treatment of a female patient with cavernous medullary hemangioma of the distal femur.

Material and Methods

In the described clinical case of a 45-year-old woman with cavernous medullary hemangioma in the distal part of the left femur, the following diagnostic methods were used: palpation of the knee joint, ultrasound diagnostics of the vessels of the lower extremities, complete blood count, biochemical blood test, magnetic resonance imaging (MRI), computed tomography (CT), radiography, pathohistological examination of biopsy and surgical material. Surgical removal of the neoplasm was performed for treatment. In the postoperative period, active rehabilitation with dosed loading of the operated limb was prescribed.

Results

The presented clinical case describes the results of the diagnosis and treatment of a patient at the State Institution Professor M. I. Sytenko Institute of Spine and Joint Pathology of the National Academy of Medical Sciences of Ukraine (Kharkiv) with a benign vascular tumor of the femur — cavernous medullary hemangioma. The patient signed an informed consent for the publication of her clinical case report.

In 2022, the 45-year-old woman was first hospitalized in the Department of Emergency Traumatology and Reconstructive Surgery with the Department of Bone Oncology presenting with constant aching pain in the area of the medial surface of the left knee joint (6–7 points on the VAS), which did not stop after taking analgesics and anti-inflammatory drugs. According to the patient, the pain first appeared in 2020. The severity of the pain syndrome gradually increased, so the patient had an appointment at our institution for further examination.

During a local examination of the left knee joint, edema of the medial surface was detected. The surrounding tissues had no signs of inflammation. During palpation of the knee joint, the patient felt pain in the projection of the medial condyle of the left femur. During flexion or extension of the joint, the pain increased, and the flexion angle was limited to 85°. No neurocirculatory disorders were found.

Further, a more detailed examination was performed using instrumental methods such as Dopplerography, ultrasound diagnostics of the vessels of the lower extremities, electrocardiography, all of which did not show any abnormalities. As a result of general and biochemical blood tests, an increase in the erythrocyte sedimentation rate to 20 mm/h, an increase in cholesterol levels (7.7 mmol/l) and B-lipoproteins (83 units) was determined. The blood coagulation system indicators were normal. After a general urine test, a moderate amount of amorphous phosphates was found.

The patient underwent spiral tomography and was diagnosed with osteochondrosis of the thoracic and lumbar spine.

After an X-ray examination of the left femur, an irregular oval-shaped area of destruction with indistinct contours and a heterogeneous structure was detected in the projection of its distal epimetaphysis, located on the anteromedial surface of the internal condyle of the bone (Fig. 1 a, b), which at this level was slightly thickened due to swelling, the cortical layer was thinned with signs of destruction on the anterior surface. The para-articular tissues were inhomogeneous and enlarged.

According to MRI in May 2022, an irregularly shaped neoplasm with clear uneven contours, measuring 39×43×37 mm, was identified in the distal metaepiphysis of the left femur, along the medial edge, which was hyperintense on T1, T2, in STIR, PD modes (Fig. 1 c–d).

All images revealed hypointense inclusions inside the neoplasm and its growth into the metaepiphyseal plate with extension into the diaphysis, which caused swelling of the bone (Fig. 1 c–d). The neoplasm caused moderate edema of the surrounding bone marrow and fatty tissue. A periosteal reaction of the cortical layer was recorded along the medial edge of the femur.

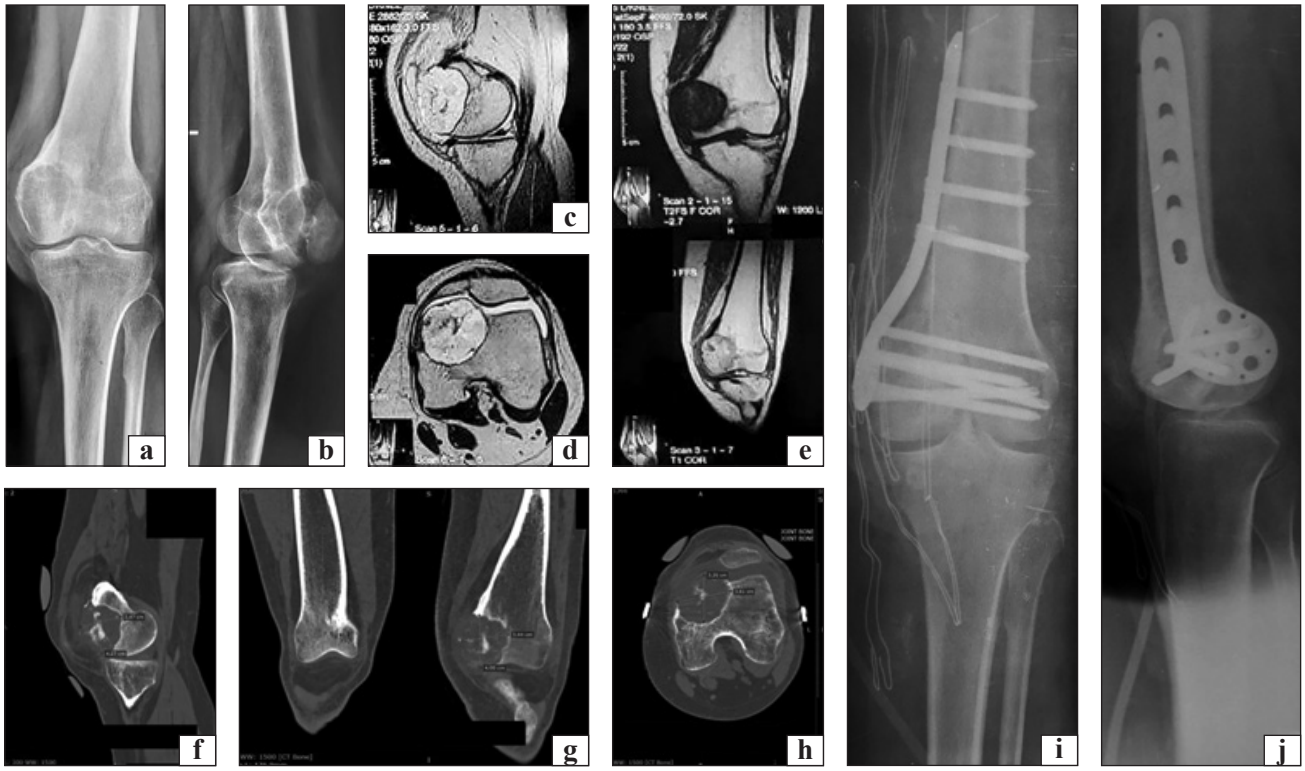

Fig. 1. Visualization of the left knee joint of a 45-year-old woman before (a–h) and after (i, j) surgical treatment of cavernous medullary hemangioma of the femur. Hemangioma in the distal femur on radiographic images (a — direct, b — lateral projections) and MRI scans in 2022 (c — lateral, d — axial, e — coronal projections); CT scans in 2024 (f — lateral, g — axial, h — coronal projections). Radiographic images after surgical removal of the hemangioma (i — direct, j — lateral projections) with replacement of the defect with a bone allograft and fixation with a plate and screws

The relationship of the bones in the knee and patellofemoral joints was preserved. The intercondylar tubercles of the tibia were sharpened, and the presence of minor marginal osteophytes of the condyles of the femur and tibia was determined.

The thickness of the articular cartilage of the condyles of these bones was preserved and with the corresponding MR signal characteristics. The articular cartilage of the patella was thinned, with defects with a depth of more than 50 % of its thickness and a size of 8×18 mm. Bone marrow edema was detected in the subchondral bone of the patella. Excessive content of homogeneous fluid was noted in the suprapatellar bursa and the cavity of the knee joint; the synovial membrane was not thickened, and its folds were not changed. Fat bodies were unremarkable. However, the fatty tissue around the joint was swollen.

During the observation of the patient's condition, control radiographs of the left knee joint were performed every 2, then 3 months. during 2022–2024, an increase in the volume of the neoplasm was not recorded. The formation of sclerosis of the contours of the neoplasm, densification of the bone structure, thickening of the newly formed bone trabeculae with the formation of a cellular structure and areas of calcification of uneven shape were observed. According to the CT scan in January 2024, in the anterior part of the medial epimetaphysis of the femur, a relatively delineated area of destruction with conditional dimensions of 41×29×49 mm remained, separated from the surrounding bone by a sclerotic rim (Fig. 1, a–h). At this level, swelling of the bone and thinning of the locking plate were detected, which was not clearly visible in some areas. Calcification of the matrix of the pathological area and a local calcification zone on the contact surface of the kneecap measuring 4×3×9 mm were noted. An enostomy measuring 3×6×6 mm was found in the lateral condyle of the left femur.

After examination, the preliminary diagnosis was determined as a focus of pathological reorganization of the distal part of the left femur, which is accompanied by impaired function of the left lower limb with persistent pain syndrome.

Given the presentation and history of the disease, the patient underwent a trepan biopsy, the results of which did not allow determining an accurate diagnosis. Namely, macroscopically these were fragments similar to bone and cartilage tissue. During microscopic examination, cartilage tissue and individual

bone trabeculae with destructive dystrophic changes in them were found.

Given the persistent pain syndrome, which intensified during physical exertion and interfered with the usual state of life, the patient's young age and active lifestyle, visible deformation of the knee joint due to edema and neoplasm, high risk of pathological fracture of the femur, she was offered surgical treatment. The intervention was performed by parietal resection of the pathological focus of the distal part of the left femur with preservation of subchondral and cartilage tissues and combined replacement of the defect with an alloimplant with fixation with a plate and screws.

After obtaining consent, the patient underwent surgical intervention. A linear incision of approximately 30 cm in length was made on the anterior surface of the left knee joint in the projection of the medial condyle. The soft tissues were separated in layers. Hemostasis was performed during surgical access. The medial condyle of the left femur was isolated and revised. There was soft tissue swelling around the tumor. A 4×4 cm zone of destruction of the cortical layer of the bone was detected with preservation of the periosteum and replacement of the spongy bone tissue of the medial condyle with a soft tissue component. A parietal resection of the tumor was performed with preservation of the articular surface and subchondral layer of the medial condyle. Careful debridement and pulse lavage were performed. The defect was replaced with a combined bone allograft in combination with Calcemex bone cement (Fig. 1, i, j). The fragments were fixed using a plate and screws, then the correct placement of the fragments was checked radiographically. The wound was washed with antiseptics, hemostasis was performed, and sutured in layers. There were no complications intraoperatively and in the early postoperative period. The removed surgical material was sent for pathological histological examination.

Macroscopic assessment of the surgical material showed a fragment of the femur with an oval-shaped neoplasm measuring 5.5×5×3.5 cm, of bone density, gray, red and brown in color. Cavities with bone trabeculae containing bloody fluid were identified on the section.

During microscopic examination, a chaotic arrangement of bone trabeculae adjacent to the defect zone was recorded, which had destructive changes (Fig. 2, a). Namely, uneven staining of the bone matrix of the trabeculae and microcracks were noted, and in some places dead osteocytes in the lacunae were found. Numerous capillary-type vessels surrounded by loose connective tissue were found in the intertrabecular spaces (Fig. 2, b, c). The vessels were arranged in a disordered manner, sometimes their groupings were determined as lobules, which were separated from each other by wide layers of fibrous tissue, there were cavernous structures, which consist of irregularly shaped and variously sized vascular cavities (Fig. 2, d). The elongated endothelial cells lining the vascular cavities were enlarged in size, had a hyperchromic nucleus and eosinophilic cytoplasm (Fig. 2, d), in which the presence of hemosiderin foci was noted in some places. Thus, the detected morphological changes corresponded to the diagnosis of cavernous hemangioma of the bone. In the early postoperative period, the patient had a significant decrease in pain syndrome. Three months after surgery, its complete regression was established. The patient was recommended to undergo active rehabilitation with dosed load on the limb. In 6 months the patient presented for follow-up with almost full weight-bearing on the operated limb and range of motion in the knee joint, with only partial support on a cane and no presenting complaints at the time of examination.

Discussion

Hemangioma of the long bones is a rare clinical case. In the population, this diagnosis is more common among women aged 40–50 years (60 %) [3, 4]. In our case, the patient was 45 years old.

The lower extremities are the predominant location of skeletal hemangioma in the appendicular skeleton. A. Rigopoulou and A. Saifuddin (Greece) [4] described 15 cases of histologically confirmed intraosseous hemangioma of the appendicular skeleton, in 9 of them the neoplasm was found in the lower extremities, mostly in the long bones (tibial — 4, femur — 3, fibula — 1). In a publication by authors from the USA, which described 5 episodes of skeletal hemangioma of the extremities, in 2 cases it was found in the lower extremities in the fibula [5]. In another series of cases (n = 24) in China, in most patients (n = 20) hemangioma was also diagnosed in the bones of the lower extremities (femur — 9, tibia — 7, fibula — 4) [2].

Depending on the location in the bone, hemangiomas are more often medullary (66 %), and less often periosteal (33 %) and intracortical (12 %) [5]. In our clinical case, a medullary hemangioma was observed in the metaphysis.

However, most hemangiomas of long bones are found in the diaphysis (80 %) or metadiaphysis, and the metaphysis occurs in only 10 % of such episodes [5]. A clinical case similar to ours was described by researchers from India, where a 38-year-old woman was also diagnosed with cavernous medullary hemangioma of the proximal metaphysis of the tibia [3].

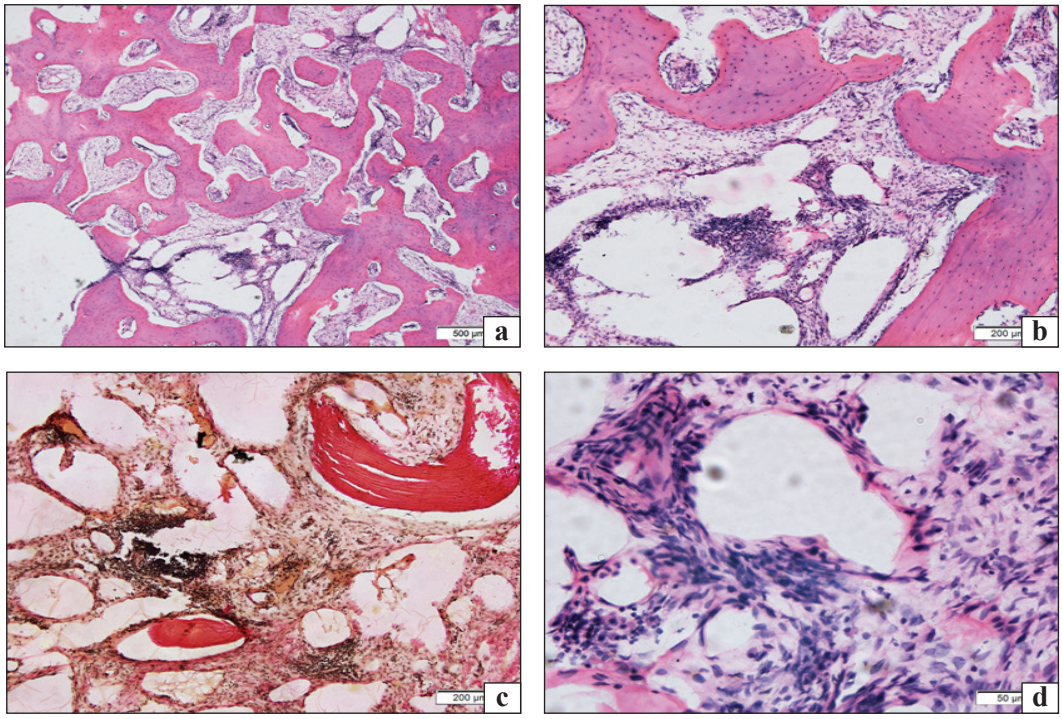

Fig. 2. Morphological features of cavernous medullary hemangioma of the femur in a 45-year-old woman. Bone trabeculae are arranged chaotically (a). Irregular sinusoidal capillaries (b–c), lined by a single layer of typical endothelium and surrounded by collagen fibers (d). Hemosiderin foci (b). Staining: hematoxylin and eosin (a, b, d); van Gieson (c). Magnification: a) 40; b-c) 100; d) 400

Common features were pain and swelling of soft tissues. According to other authors, these are characteristic symptoms specifically for hemangiomas of long bones, while hemangiomas of the vertebral bodies or skull are more often asymptomatic [4–6].

Radiological and clinical manifestations of hemangiomas in long bones are not characteristic and are nonspecific, which makes differential diagnosis difficult. Thus, according to the analysis of a series of 5 cases of this pathology, the preoperative diagnosis of venous medullary hemangioma was considered in only one. At the same time, in this series of cases, two were with cavernous medullary hemangioma of the long bones of the extremities, as described by us [5]. In a study of 36 cases of intraosseous hemangiomas outside the skull and spine, a common feature on CT scans was the germination of the neoplasm through the cortex [7]. The authors believe that this feature distinguishes bone hemangiomas with this localization from those in the skull or spine [7]. That is, in their opinion, this location is more aggressive, probably because of this they are often confused with malignant neoplasms. Similar features were found in our case in the form of periosteal reaction of the cortical layer and germination of the hemangioma into the metaepiphyseal plate with spread into the diaphysis of the femur.

The complexity of the diagnosis is confirmed by another case similar to ours, in which a patient with intraosseous hemangiomas of the distal femur and diaphysis of the left tibia was suspected of *cystic angiomatosis* and multiple myeloma until an open biopsy was performed [8].

Histologically, the most common type in the long bones of the extremities is cavernous hemangioma [2]. The morphological changes that we found were consistent with this diagnosis. Pathological and histological examination for its determination is not difficult. However, artifacts that arise from the collection of material during trephine biopsy, histological processing, and the preparation of histological sections can complicate the interpretation of microscopically detected changes in tissues. The reason for the appearance of such artifacts is the combination of tissues of different densities in the fragment: delicate thin-walled vessels and denser bone trabeculae.

Because of the above, in our opinion, a comprehensive approach is important for the diagnosis of cavernous hemangioma in severe clinical cases. Namely, the comparison of clinical indicators, imaging results (radiography, CT or MRI) and preoperative trepan biopsy data. At the same time, the final diagnosis of "cavernous hemangioma" can be confirmed only by the results of pathological histological examination of the surgical material.

Conclusions

Cavernous medullary hemangioma of long bones, especially with an unusual location in the distal femur, is a rare vascular tumor. Its differential diagnosis is difficult due to nonspecific clinical and radiological

manifestations, but surgical treatment has a favorable prognosis.

In the described clinical case, the 45-year-old woman underwent the necessary clinical and radiological examination, but this diagnosis was confirmed only after surgical intervention to remove the tumor due to the difficulty of obtaining the necessary material during trepan biopsy.

Surgical treatment by parietal resection of the tumor, replacement of the defect with a bone alloimplant with fixation with plates and screws was effective and helped to almost completely restore the load on the operated limb of the patient without pain syndrome after 6 months.

Conflict of interest. The authors declare the absence of a conflict of interest.

Prospects for further research. Development of a set of methods to improve the diagnosis of cavernous hemangioma localized in the long bones.

Information on funding. None.

Authors' contribution. Shevchenko I. V. — participation in the diagnosis of the disease, performed surgical intervention with a description of the technique; Gubsky S. S. — participation in the diagnosis of the disease, performed treatment in the pre- and postoperative period and described it; Zlatnik R. V. — participation in the diagnosis of the disease, performed radiological examinations with a description of the results; Danyshchuk Z. M., Ivanova N. V. — participated in the diagnosis of the disease, performed histopathological studies and description; Maltseva V. Ye. — drafted the discussion and final version of the manuscript, created figures. All authors approved the final manuscript.

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