Hibernomas of the upper extremity: a case report and literature review

Cara A. Cipriano • Robert R. L. Gray • John J. Fernandez

Published online: 17 December 2014
© American Association for Hand Surgery 2014

Abstract Hibernomas are rare, benign tumors of brown adipose tissue. While they have been found in a variety of locations, they are rare in the upper extremity and not previously described in the hand. Diagnosis is most often made histologically, and treatment consists of surgical resection. We present an unusual case of hibernoma in the hand treated with marginal excision, resulting in good function, cosmesis, and no recurrence.

Introduction

Hibernomas are benign tumors derived from brown adipose tissue. In contrast to lipomas, which are derived from white adipose, hibernomas are extremely rare, with fewer than 200 overall cases reported in the literature. These tumors most often occur in regions proximate to the axial skeleton where brown fat existed in the fetus and persisted into adult life; they have been described in various other locations as well [1, 3, 4, 6, 7]. To our knowledge, there have been no previously documented cases of hibernoma in upper extremity.

Lesions that are small, superficial, mobile, and otherwise consistent with a diagnosis of lipoma on physical exam may be safely excised without imaging or biopsy; however, if

intraoperative findings are not consistent with the presumed diagnosis, certain techniques and precautions must be followed. Our report describes an unusual case as well as recommendations for the appropriate management of unexpected findings in the excision of a hand mass.

Case Report

The patient is a 37-year-old man who presented with a mass on the dorsum of the right thumb webspace. The mass was painless but had gradually increased in size over several months. On physical examination, he had a palpable mass in the first webspace of the right hand measuring approximately 3 by 5 cm (Fig. 1). The mass was soft and mobile over bone, and it did not seem tethered to the surrounding soft tissues. The overlying skin was normal in appearance, and the mass was nontender to palpation. He had no neurologic complaints. There was no appreciable weakness or muscle atrophy, and range of motion was full and equal to the contralateral side.

Following a discussion of the risks and benefits involved, the patient elected to undergo excisional biopsy of the mass. A longitudinal incision of approximately 4 cm was made over the dorsum of the hand, centered over the thumb webspace. Dissection was carried down through the skin and subcutaneous tissues, taking care to preserve and protect the dorsoradial sensory nerve branches. This exposed the mass, which was noted to be tan in color, multilobulated, and fibrotic, unlike a typical lipoma. Due to this unexpected appearance, a small sample was sent to pathology for frozen section, which revealed no evidence of malignancy. A complete excision was then performed, taking care not to incise the mass, which was removed en bloc. The specimen was sent to pathology for permanent analysis and hemostasis obtained, after which a layered closure of the wound was performed.

C. A. Cipriano (✉)
Division of Musculoskeletal Oncology, Department of Orthopedics,
Washington University in St Louis, 425 South Euclid, Suite 5505, St
Louis, MO 63110, USA
e-mail: ciprianoc@wudosis.wustl.edu

R. R. L. Gray
Division of Hand Surgery, Department of Orthopaedics, North Shore
University Health System, Evanston, IL, USA

J. J. Fernandez
Division of Hand Surgery, Department of Orthopaedics, Rush
University Medical Center, Chicago, IL, USA

Fig. 1 Preoperative appearance of the patient's hand. The mass, clearly visible in the first web space, was soft, mobile, and nontender to palpation

Grossly, the excised mass was 5 by 3 cm in size (Fig. 2); histologic examination revealed benign adipocytes with peripheral nuclei but no signs of cellular atypia. Also noted were occasional areas of granular eosinophilic cytoplasm, consistent with the diagnosis of hibernoma (Fig. 3).

When the patient returned for evaluation 2 weeks postoperatively, he had no complaints. The incision was well healed with no signs of infection, and there were no changes in motor function, sensation, or range of motion in the hand. He had ability to make full composite fist and oppose to the small finger metacarpophalangeal head, with intact sensation in all digits and a Disabilities of the Arm, Shoulder, and Hand (DASH) score of 1.7. Due to the benign nature of the mass, there was no indication for radiotherapy or further treatment of any kind. At 2.5 years postoperatively, he has had no signs or symptoms or recurrence.

Discussion

Brown adipose tissue is present in the mammalian fetus and regresses with age in species that do not hibernate. Its dark color results from the higher concentration of mitochondria and increased vasculature that distinguish it from white adipose tissue. The primary metabolic function of this organ is nonshivering thermogenesis, which produces heat rather than ATP. In humans, brown fat develops during the 21st week of

Fig. 2 Gross appearance of the multilobulated mass at time of excision. The fibrotic texture and tan color are suggestive of a hibernoma rather than a typical lipoma

Fig. 3 Histopathologic analysis of the specimen at **a** $\times 10$ and **b** $\times 20$ magnification revealed benign adipocytes with peripheral nuclei and areas of granular eosinophilic cytoplasm, confirming the diagnosis of hibernoma. There was no evidence of cellular atypia or other features concerning for malignancy

fetal gestation in the neck, axilla, mediastinum, periaortic, and perirenal areas: interscapular area, posterior abdominal wall, in the suprailiac and peripancreatic adipose stores, and near autonomic ganglia. In adults, hibernomas have been most commonly found in locations where brown fat may persist, such as the interscapular region and subcutaneous tissues of the back, as well as the neck, axilla, thigh, and intrathoracic region [1, 6, 7]. They have also been reported in a variety of more unusual locations, including the scalp, intracranial and intraspinal spaces, abdominal wall, popliteal fossa, buttock, scrotum, and periureteral area [6]. There does not seem to be a significant gender predominance, and incidence is highest in the third decade, whereas lipomas are more common after the age of 30 [5].

Patients with superficial hibernomas most commonly present complaining of a painless, gradually enlarging mass, while deep lesions may be found incidentally. Symptoms are rare but may result from compression on an adjacent structure. Small, superficial hibernomas such as the one reported here will most likely be presumed to be lipomas, which are identical in presentation and much more common. Cross-sectional imaging is not indicated for lesions that are small, superficial, mobile, painless, and slowly or nonprogressive. In cases of tumors that are larger, deeper, or suspicious for malignancy, diagnostic imaging is indicated prior to embarking on surgical treatment. Hibernomas are readily apparent on CT and angiogram due to their hypervascularity; however, this quality may also confuse the diagnosis by suggesting malignancy and necessitate biopsy [5].

Grossly, the tumor may range from yellow to tan or red brown, according to the relative abundance of lipid content. It is well defined, soft, and mobile, composed of lobules separated by fibrovascular septa and capillaries. On the microscopic level, hibernomas consist of three principle cell types: cells with granular eosinophilic cytoplasm containing few, if any, lipid vacuoles; cells that are larger in size containing scarce granular eosinophilic cytoplasm and multiple lipid vacuoles; and large univacuolated adipocytes [2]. Location of the nuclei is eccentric in the adipocyte but varies in the other types, and there is generally no mitosis or cellular atypia.

Because of their low incidence and the imaging challenges described above, hibernomas are definitively diagnosed based on pathology. Histologically, hibernomas may be distinguished from rhabdomyosarcoma and granular cell tumors by the presence of lipid vacuoles. Round cell liposarcoma, however, may be composed of multivacuolar eosinophilic lipoblasts and therefore may be a challenging diagnosis to exclude.

Hibernomas are benign tumors, and recurrence has only been described in cases where complete resection was impossible due to the location of the tumor [3]. Excisional biopsy is

safe and appropriate for lesions that are small, superficial, and consistent with benign entities on physical examination. However, if any mass undergoing resection without a definitive diagnosis appears suspicious upon initial intraoperative encounter and is large enough to warrant incisional biopsy, tissue may be sent for frozen section before proceeding. In the case of malignant or uncertain pathology, meticulous hemostasis should be achieved and the patient should receive further evaluation and possibly treatment prior to definitive surgical management. For smaller, superficial lesions not closely approximating critical structures, an excisional biopsy can be performed, but it is critical that the mass be removed en bloc rather than in segments. In addition, excessive dissection should be avoided because, in the rare event of a malignant diagnosis, this could lead to further contamination of surrounding tissues and thereby complicate future management.

Acknowledgments The authors did not receive any financial support for this work and have no relevant disclosures to report. All work was performed at Rush University, in Chicago, IL.

Conflict of Interest Cara A. Cipriano declares that she has no conflict of interest.

Robert R. L. Gray declares that he has no conflict of interest.

John J. Fernandez declares that he has no conflict of interest.

Statement of Human and Animal Rights This article does not contain any studies with human or animal subjects.

Statement of Informed Consent Informed consent was not necessary for this report.

References

1. Ahn C, Harvey JC. Mediastinal hibernoma, a rare tumor. *Ann Thorac Surg.* 1990;50(5):828–30.
2. Gaffney EF, Hargreaves HK, Semple E, Vellios F. Hibernoma: distinctive light and electron microscopic features and relationship to brown adipose tissue. *Hum Pathol.* 1983;14(8):677–87.
3. Lele SM, Chudru S, Chalijub G, Adegboyega P, Haque, Abida J. Hibernoma: a report of 2 unusual cases with a review of the literature. *Arch Pathol Lab Med.* 2002;126(8):975–8.
4. Rigor VU, Goldstein SE, Jones J, Bernstein R, Gold MS, Weiner S. Hibernoma: a case report and discussion of a rare tumor. *Cancer Lett.* 1986;57(11):2207–11.
5. Rigor VU, Goldstone SE, Jones J, Bernstein R, Gold MS, Weiner S. Hibernoma: a case report and discussion of a rare tumor. *Cancer.* 1986;57(11):2207–11.
6. Stephen AH, Sanjay RP, John WB. Hibernoma of the pericardium presenting as pericardial effusion in a child. *Ped Pathol.* 1990;10(4): 575–80.
7. Udwardia ZF, Kumar N, Bhadura AS. Mediastinal hibernoma. *Eur J Cardiothorac Surg.* 1999;15(4):533–5.