A Rare Tumor: Hibernoma in The Thigh: Case Report

Abdullah Alhazmi¹, Othman Saeed Alghamdi², Almufarrh Dhafer Saad², Abdulaziz Ahmed N. Alanazi³

¹Department Orthopedic Surgery, King Abdulaziz Medical City, Jeddah, Saudi Arabia
²Department Orthopedic Surgery, East Jeddah Hospital, Jeddah, Saudi Arabia
³Department Orthopedic Surgery, King Faisal Specialist and Research Center Jeddah, Jeddah, Saudi Arabia
*Corresponding author: Abdulaziz Ahmed N. Alanazi, Mobile: +966509988659, E-Mail: a.alanzee7@gmail.com

ABSTRACT

Background: Hibernomas are soft tissue tumors that form from lingering brown fat cells and tend to form in locations where brown fat is more prevalent in fetuses and babies. They are uncommon, grow slowly, cause no discomfort, and are benign. Because of its rarity, this tumor type is commonly misdiagnosed as liposarcoma or dismissed altogether.

Objective: The objective of this case study was to achieve accurate diagnosis for the rare tumor hibernoma in the thigh. **Case report:** 33-Years-old female not known to have any chronic medical illness, gastric sleeve before 4 years, cholecystectomy 1 year back, and her condition started 5 year back when she noticed left knee lump. Initially she sought medical advice and was told it is lipoma. She was advised for weight loss. No history of trauma or falling. After she lost weight, it become more prominent with knee pain for the last year.

Conclusion: As a result of their rarity and underreporting, hibernomas are frequently misinterpreted as either big lipomas or malignant soft tissue tumors, which necessitate the need of histology in achieving an accurate diagnosis and anticipating intraoperative outcomes. Tumors can only be removed safely through meticulous dissection and ligation of the vasculature, which requires knowledge of the surrounding anatomy.

Keywords: Rare tumor, Hibernoma, CT, X-ray, Orthopedic surgery.

INTRODUCTION

Hibernomas are uncommon soft tissue tumours that originate in the body's remaining brown fat cells and are considered benign ⁽¹⁾. They are rare in the skull and popliteal fossa, and are seen most often in individuals between the ages of 30 and 40 years ⁽²⁾. In addition to the lower and upper extremities, the abdomen is a rare site ^(1,2). Hibernomas present as slow-growing, painless masses that are often mistaken for other tumour forms like liposarcomas ⁽³⁾. With six distinct histological types recognized ⁽⁴⁾, atypical lipomas and well-differentiated liposarcomas are common misdiagnosis of the lipomalike variety ⁽¹⁾.

In most cases, surgical removal of a hibernoma is the treatment of choice. Generally speaking, a total surgical removal is curative ^(5, 6).

CASE REPORT

Adult oncology clinic as consultation and history of patient:

33-Years-old female not known to have any chronic medical illness, gastric sleeve before 4 years and cholecystectomy 1 year back. Her condition started 5 years back when she noticed left knee lump initially sought medical advice and was told it is lipoma that recommend to lose weight. No history of trauma or falling, after she lost weight, it become more prominent with knee pain for the last year.

She asked for medical advice in Altaif (Alamin hospital). Initially ultrasound showed suspicious lesion invading muscles recommended for resection but patient refused for resection or biopsy, before 3 months she sought another medical advice.

MRI was done 9 cm X 3 cm and was recommended for surgery.

Social history:

-She was single living in orphanage in Altaif (Alamin hospital). Currently, she is living with her friend in Jeddah, currently unemployed and currently taking multivitamins since her gastric sleeve.

Discussed with most responsible physician (MRP): On 20 October (2020), patient was informed to come to be assessed.

Follow up clinic with oncology orthopedic: Finding:

Patient was 33 years old female and not having any medical illness. She came to the clinic complaining of left thigh swelling and pain for 5 years not increasing, not associated with fever, trauma, weight loss or other masses, MRI done outside without contrast.

Assessment plan:

Patient had left thigh middle to distal anteromedial mass 10 CM X 8 CM mobile with skin and subcutaneous not adherent to deep soft tissue with no color change. It was mildly painful and there was no hotness or signs of inflammation. MRI from outside showed a mass originating from the fatty tissue not invading the muscular structure and separated by the muscular septa.

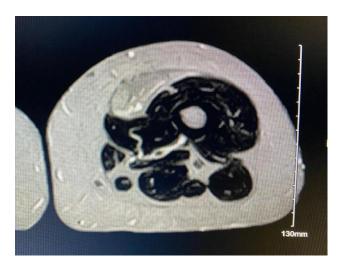
Diagnosis: US-guided biopsy, lipoma vs liposarcoma (myxofibrosarcoma).

CT guided Biopsy:

Received: 11/06/2022 Accepted: 18/08/2022 **Clinical history and indication:** The patient was referred to interventional radiology suite for percutaneous left thigh lesion biopsy.

Procedure: The patient was prepped and draped in a standard sterile fashion. Lidocaine 1% was used as a local anesthetic. A skin nick was made. Under ultrasound guidance, the left thigh lesion was targeted with 15g introducer needle followed by 16g coaxial advancing of biopsy needle (MaxCor- Bard). Seven passes were performed. The needle tract was embolized with gel foam sponge slurry. Post biopsy ultrasonography showed no abnormal finding or active bleeding.

Conclusion: Percutaneous left thigh lesion biopsy under ultrasound guidance.



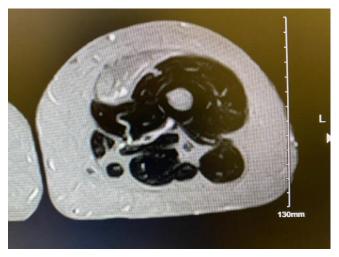


Figure (1): Axial view of magnetic resonance T1 imaging findings of a well-defined, heterogeneous mass, measuring 9 cm \times 5 cm, clearly hypo-intense to subcutaneous fat on T1-weighted spin-echo images, with prominent thin low signal bands throughout the tumour.

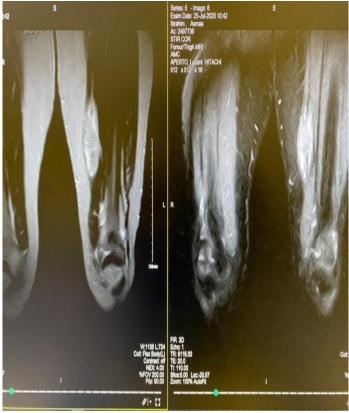


Figure (2): Coronal view of magnetic resonance imaging findings of a well-defined, heterogeneous mass, measuring 9 cm \times 5 cm, clearly hypo-intense to subcutaneous fat on T1-weighted spin-echo images, with prominent thin low signal bands throughout the tumour. The lesions failed to fully suppress on STIR or fat-saturated T2-weighted images. No bone or chest wall invasion was demonstrated.

Orthopedic follow up-clinic:

Finding: 33 years old female known left thigh mass for the past five years she complained of mild pain with range of motion and no trauma. MRI showed heterogeneous mass, pathologically reported as hibernoma.

We discussed with her the pathologic result and explained surgical vs non-surgical options. She was keen for resection and we explained to her the possible complications including infection, neurovascular injury and recurrence. We booked her for next week. She was smoker and we advised her to quit to avoid complications.

Pre-Operative Diagnosis: Left thigh swelling for 5 years, admitted for CT guided biopsy.

Surgical technique:

Pre-operative diagnosis: Neoplasm of uncertain or

unknown behavior, unspecified.

Operative name: Excision of soft tissue tumor.

Post- operative diagnosis: Hibernoma.

Anesthesia Type: Spine **Operative date:** 10/11/2020

Surgeon: AL HAZMI, ABDULLAH

Surgical Report:

Operation Performed:

-Left anterior thigh Hibernoma resection

-Estimated Blood Loss: 50 Drains: 0

Complications:

Specimen(s) Removed: tagged and sent for

histopathology.

Procedure Details:

Under Spinal anesthesia in supine position the leg was prepped and draped in sterile fashion, a skin incision was done over the lesion with a small ellipse around the previous biopsy tract. Dissection done down to the fascia which was carefully incised over the mass. The lesion was carefully dissected and resected completely in one piece. Thorough hemostasis ensured and irrigation done. The fascia was closed with Vircryl size 0. The wound was closed in layers and injected with 10 cc of 0.25 Marcaine for post-operative pain relief covered with a sterile dressing.

Patient Condition/Disposition: stable

Orthopedic oncology post-operative clinic follow up:

- After 2 weeks post-operative left thigh hibernoma resection histopathology reported as hibernoma wound dry and clean.
- After 4 weeks post-operative patient contacted through mobile, doing fine no new complain, wound healed.
- After 6 months post-operative patient came for follow up, no new active issue, full range of motion full weight bearing.

Declaration of patient consent:

An approval of the study was obtained from Done in King Abdul-Aziz Medical City Hospital in Jeddah (Saudi Arabia) Academic and Ethical Committee. The patient and her relative were informed that the case would be published as case report and this was accepted. This work has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for studies involving humans.

DISCUSSION

Hibernomas typically affect younger persons, with a mean age upon diagnosis of 38 years. ⁽⁷⁾. While, most

often found in the thigh, trunk, upper extremities, and head/neck, hibernoma can also develop in the thoracic and abdominal cavities. The sizes of these slowgrowing tumours can vary from 1 centimeter to 24 centimeters, with 10 centimeters being the average. Histologically, most hibernomas look like regular brown adipose tissue, making them easier to spot. However, a few of them are atypically big or lack the multivacuolated cells that characterize hibernoma. Oftentimes, these "lipoma-like" variations can be mistaken for liposarcomas that have undergone adequate differentiation (also known as atypical tumors when they occur lipomatous in the extremities)(8).

Brown adipose tissue hibernomas occur seldom. The thigh, the shoulder, and the back are the most common sites where they manifest themselves. These lipomatous tumours are completely safe. In 1906, Merkle was the first to describe hibernomas (9). The traditional descriptions of the tumour state that it is a slow-growing, movable, painless mass that can develop to a maximum of 29.0 x19.0 x12.0 cm in size (5) with a total of 2500 grams (6). However, hibernomas can become symptomatic due to mass effect and other anatomical variabilities, as reported in our case and several cases in the medical literatures (5, 10, 11). The most frequently reported site of hibernomas is the back, specifically the inter-scapular area. Some investigators have reported the thigh to be also frequently involved, accounting for about 30% of all cases (1). Other common sites include the neck, axillae, and intra-thoracic area

In our case report, magnetic resonance imaging findings showed a well-defined, heterogeneous mass, measuring 9 cm × 5 cm, clearly hypo-intense to subcutaneous fat on T1-weighted spin-echo images, with prominent thin low signal bands throughout the tumour. The lesions failed to fully suppress on STIR or fat-saturated T2-weighted images. No bone or chest wall invasion was demonstrated. After 2 weeks post-operative left thigh hibernoma resection histopathology reported as hibernoma. Wound was dry and clean, and after 6 months post-operative patient came for follow up, no new active issue, full range of motion full weight bearing.

The rarity of the tumor means that many doctors and radiologists have never seen one before, which can lead to incorrect diagnosis and underreporting ⁽¹⁾. Despite their benign status, hibernomas are large, vascular tumors that require careful pre-operative diagnosis. Compression of neighboring organs can be a symptom of tumor growth, especially when the tumor is big or located near a major nerve or blood stream⁽¹²⁾, similar to how our patient's tumor was putting pressure on her femoral nerve. The bulk was vascular as well, with several perforating arteries branching from the femoral artery.

The definitive method for identifying hibernomas is a histological analysis. A biopsy is recommended to achieve a clear diagnosis in patients with deep soft tissue tumor greater than 3 cm in diameter, as was the situation in our patient ⁽⁵⁾.

Diagnostic imaging advancements helped doctors zero in on potential issues before surgery. A hibernoma shows up as a hyperechogenic mass on ultrasound, whereas angiography reveals a highly vascularized tumor with irregular arteriovenous shunts. Increased uptake can be seen on a technetium99 scintigraphy image. Tumor density on CT scans is between that of muscle and fat, with possibly heterogeneous augmentation following contrast enhancement and the presence of intratumoral arteries ⁽¹³⁾. The mass is very signal-intense on T1-weighted images and isointense or hypointense on T2-weighted pictures on magnetic resonance imaging (MRI), the preferred radiographic investigation ⁽¹⁴⁾ by reducing body fat, tumor blood vessels can become visible ⁽¹⁵⁾.

The surgeon's precision in tumor dissection and hemostasis is greatly aided by a thorough comprehension of tumor location and the interaction between the tumor and neighboring structures. The MRI scan of our patient was essential in elucidating the link between the mass and neighboring structures, which in turn aided in the development of an intervention strategy. Even though hibernomas are often non-invasive, there has been a case of partial excision due to the tumor's proximity to an important arterial structure (16). Well-encapsulated tumor do not cause any problems when being removed surgically, and total resection can be curative.

CONCLUSION

As a result of their rarity and underreporting, hibernomas are frequently misinterpreted as either big lipomas or malignant soft tissue tumors, which is highlighting the need of histology in achieving an accurate diagnosis and anticipating intra-operative outcomes. Tumors can only be removed safely through meticulous dissection and ligation of the vasculature, which requires knowledge of the surrounding anatomy.

Financial support and sponsorship: Nil. Conflict of interest: Nil.

REFERENCES

- 1. Furlong M, Fanburg-Smith J, Miettinen M (2001): The morphologic spectrum of hibernoma: a clinicopathologic study of 170 cases. Am J Surg Pathol., 25: 809–814.
- 2. Murphey M, Carroll J, Flemming D *et al.* (2004): From the archives of the AFIP: benign musculoskeletal lipomatous lesions. Radiographics, 24: 1433–1466.
- 3. Patil S, Sheik A, Tewari V *et al.* (2019): Hibernoma: a missed diagnosis. Indian J Pathol Microbiol., 62: 461-65.
- 4. Fletcher C (2007): 3rd ed. Elsevier Health Sciences; Hong Kong. Diagnostic Histopathology of Tumors. Churchill Livingstone Elsevier, Philadelphia, Pp: 136-149. https://www.elsevier.com/books/diagnostic-histopathology-of-tumors-2-volume-set/fletcher/978-0-323-42860-6
- 5. Ersozlu S, Sahin O, Ozgur A *et al.* (2008): Sciatic neuropathy from a giant hibernoma of the thigh: a case report. Am J Orthop., 37: 103–106.
- **6. Lewandowski P, Weiner S (1996):** Hibernoma of the medial thigh. Case report and literature review. Clin Orthop Relat Res., 330: 198–201.
- 7. Cohade C, Osman M, Pannu H *et al.* (2003): Uptake in Supraclavicular Area Fat (USA-FAT) Description on 18F-FDG PET/CT. J Nuc Med., 44 (2): 170-176
- **8. Wimmer M, Conrad E, Eary J (2008):** Hibernoma in the Thigh Mimicking Soft Tissue Sarcoma on FDG-PET. Radiology Case Reports, 3 (3): 1-4.
- **9. Merkel H** (**1906**): On a pseudolipoma of the breast. Beitr Pathol Anat., 39: 152–157.
- **10. Salim B, Belkacem C (2014):** Hibernoma of the thigh: a report of four cases. J Orthop Surg., 22: 118–121.
- **11. DeRosa D, Lim R, Lin-Hurtubise K** *et al.* **(2012):** Symptomatic hibernoma: a rare soft tissue tumor. Hawaii J Med Public Health, 71: 342–345.
- **12. Daubner D, Spieth S, Pablik J** *et al.* **(2015):** Hibernoma–two patients with a rare lipoid soft-tissue tumour. BMC Med Imaging, 15: 4-7.
- **13.** Anderson S, Schwab C, Stauffer E *et al.* (2001): Hibernoma: imaging characteristics of a rare benign soft tissue tumor. Skelet Radiol., 30: 590–595.
- **14. Datir A, James S, Ali K** *et al.* **(2008):** MRI of soft-tissue masses: the relationship between lesion size, depth, and diagnosis. Clin Radiol., 63: 373–378.
- **15. Atilla S, Eilenberg S, Brown J (1995):** Hibernoma: MRI appearance of a rare tumor. Magn Reson Imaging, 13: 335–337.
- **16.** Lele S, Chundru S, Chaljub G *et al.* (2002): Hibernoma: a report of 2 unusual cases with a review of the literature. Arch Pathol Lab Med., 126: 975–978.