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A Diagnostic Dilemma of a Subcutaneous Hibernoma: Case Report

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Manuscript Preparation E
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Patient: Male, 33-year-old
Final Diagnosis: Hibernoma
Symptoms: Mass in the thigh • increasing in size
Medication: —
Clinical Procedure: —
Specialty: Surgery

Objective: Rare disease

Background: Subcutaneous lipomatous lesions are commonly encountered in clinical practice. Hibernoma is a rare subtype of the benign lipomatous tumor, representing 1% of all types. It poses a challenge due to the difficulty of differentiating it from atypical lipomatous lesions and liposarcomas, which may lead to possible inappropriate diagnosis and management.

Case Report: We report a case of a 33-year-old male who presented with a right upper thigh swelling noticed some time prior to presentation that had started increasing in size prior to his presentation. The magnetic resonance imaging (MRI) was unable to rule out atypical lipomatous tumor and liposarcoma. An ultrasound-guided biopsy gave a diagnosis of hibernoma. The patient underwent a wide local excision, which confirmed the diagnosis of hibernoma. At the 3-year follow-up, there was no evidence of local recurrence.

Conclusions: Hibernoma has been reported in the literature to be discovered incidentally by radiological imaging done for other causes. However, hibernomas raise a diagnostic challenge because in most imaging modalities they are indistinguishable from other malignant tumors.

A wide local excision with negative margins is key to resolving the diagnostic dilemma that a hibernoma presents, as it will provide a definitive diagnosis differentiating it from other lipomatous lesions and prevent any future recurrence.

Caution is advised when dealing with lipomatous lesions, as they often overlap with malignancy. Furthermore, an MRI should be done for any subcutaneous lesion that is larger than 5 cm or shows recent growth. A biopsy can resolve the diagnostic dilemma with caution to the hypervascularity of such tumors.

MeSH Keywords: Lipoma • Liposarcoma • Neoplasms, Adipose Tissue

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/921447

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Background

Soft tissue tumors are some of the most common presentations encountered in clinical practice, particularly lipomatous lesions which represent 20% of all soft tissue tumors. These tumors can be both benign and malignant. Differentiating between them can be quite challenging in some cases [1,2].

Hibernoma is a rare subtype of the benign lipomatous tumor accounting for only 1% of all types of both benign and malignant lesions. It was first reported in the literature by Merkel in 1906 and then named by Gery in 1914 as "hibernoma" owing to its histological resemblance to the brown fat of hibernating animals. Since the first reported hibernoma in the literature, until the reported case by Greenbaum, et al. in 2016, there have been less than 250 reported cases. These tumors pose a challenge due to the difficulty of differentiating hibernomas from atypical lipomatous lesions and liposarcomas, even in terms of histopathology, leading difficulty in determining the accurate diagnosis and hence the appropriate preoperative management [3,4].

Case Report

We report a case of a 33-year-old male who had a known case of multiple sclerosis and was on medications for this condition, who presented to another facility complaining of a right upper thigh swelling first noticed around a year prior. It was stable in size until 1 month prior to his presentation when he noticed it becoming more visible than usual. There was no history of any other masses elsewhere in his body, lower limb weakness, or any constitutional symptoms; he also did not have a past surgical history. He underwent Tru-cut biopsy which revealed an atypical lipomatous tumor and the possibility of liposarcoma could not be ruled out. Since the possibility of malignancy could not be ruled out, he was referred to our surgical clinic for further workup and management.

Upon his presentation to our clinic, a physical examination of his right lower limb showed a 10×13 cm soft, non-tender, non-fluctuating mass with a smooth surface and defined edges on the anterior midline of his upper right thigh. There were no signs of local inflammation nor skin discoloration. Upon examination, the neurovascular bundle of whole right lower limb was intact.

An MRI of the right lower limb showed: a 10×13×14 cm oval mass with a bright signal in T1, and to a lesser extent T2, located between the pectineus, adductor brevis, and obturator externus muscles medially and between the sartorius, iliopsoas, and rectus femoris muscles laterally with the femoral neurovascular bundle running lateral to the lesion (Figure 1). These findings were suggestive of lipomatous lesion, such as atypical lipoma, and the possibility of liposarcoma could not be rule out.

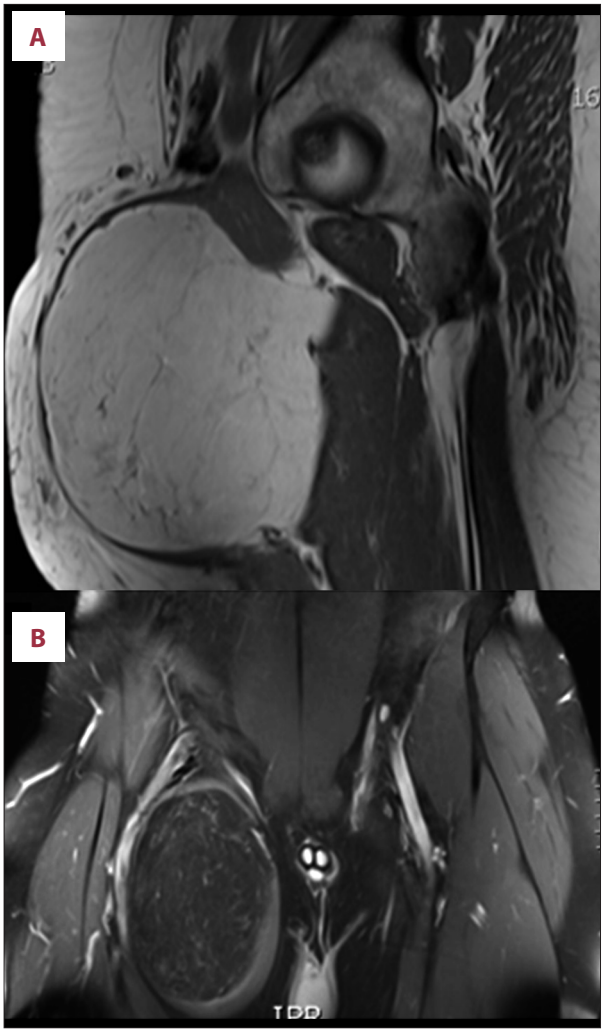

Figure 1. Magnetic resonance imaging (MRI) of the lesion. MRI of the pelvis and lower limb showing sagittal (**A**) and coronal (**B**) views of a lesion that is 14 cm long at its widest diameter.

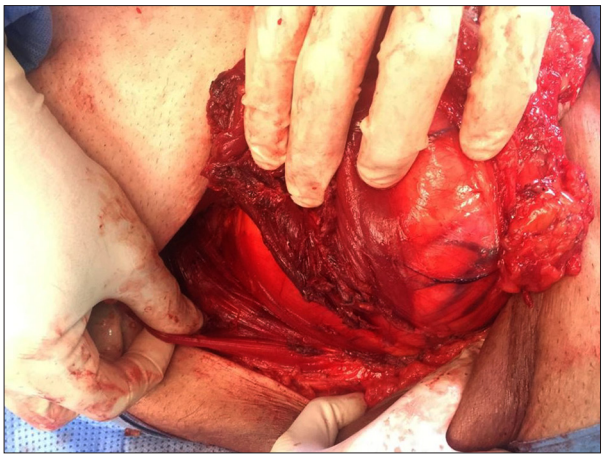

Figure 2. Intraoperative image of the mass.

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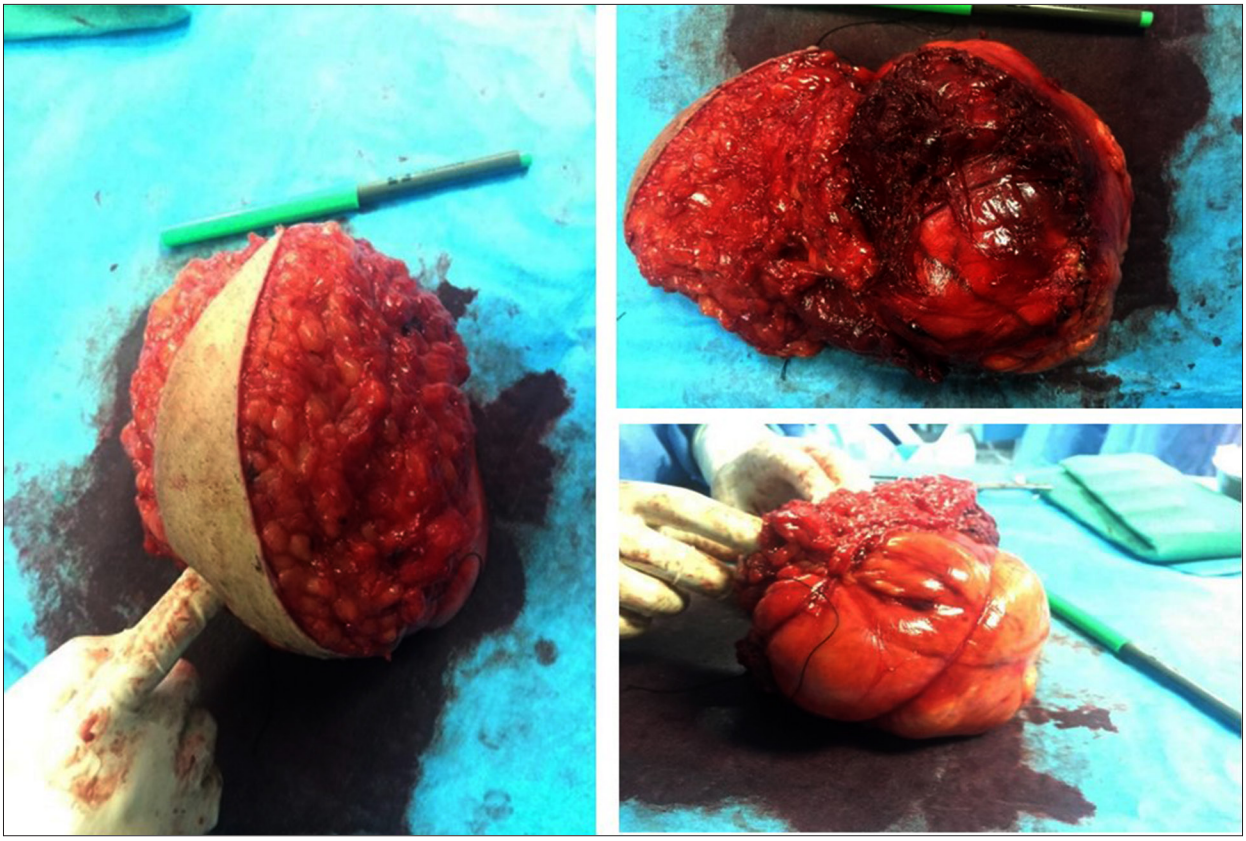

Figure 3. Macroscopic images of the mass. The mass is seen to have a yellow, greasy, and lobulated surface.

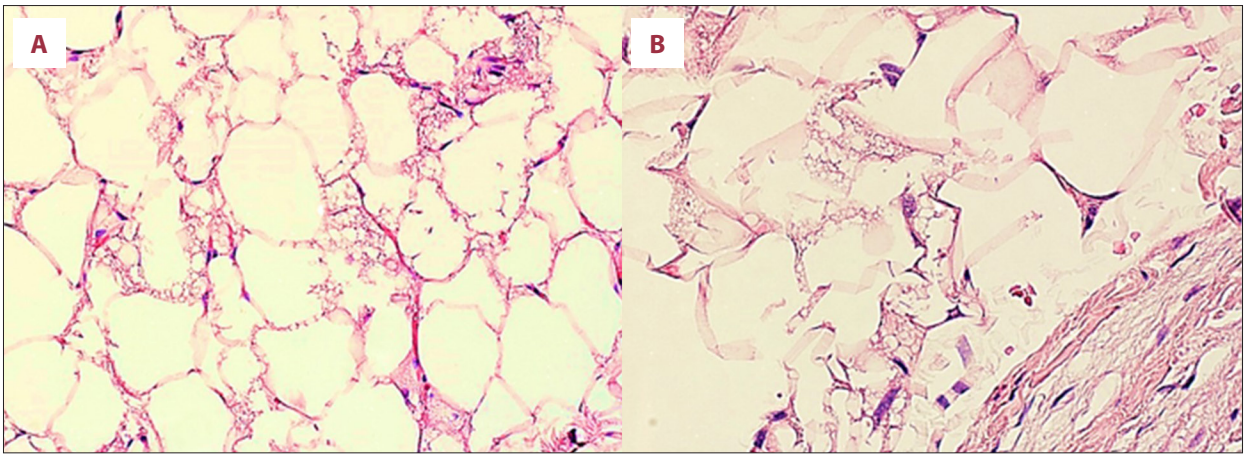

Figure 4. (**A, B**) Histopathology slides. Microscopic image showing organoid arrangement of uniform large cells resembling brown fat with pale coarsely granular to multivacuolated cytoplasm. Vacuoles are small with central nucleus and rare atypia.

Additionally, an ultrasound-guided biopsy was done with the histopathology analysis showing atypical lipomatous cells with hibernomatous features, leading to a preliminary diagnosis of begin hibernoma. We proceeded with a wide local surgical excision with a drain left in place on a negative pressure (Figures 2, 3).

The final histopathology showed a fatty lobulated tumor on cross-section composed of a mix of univacuolated and multivacuolated adipocytes alongside spindly fibroblastic cells with negative margins, thus confirming our diagnosis of a benign hibernoma (Figure 4).

The patient was discharged home on the third day after the operation with regular follow-up visits for the next 3 years.

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There has been no evidence of local recurrence or any other similar lesions.

Discussion

Lipomatous lesions can range from benign lipomas to poorly differentiated liposarcomas. Hibernomas are a rare subtype of the benign lipomatous lesions. They are usually clinically silent due to their benign origin and slow growth. They also usually remain asymptomatic unless there is a considerable growth that could compress any nearby structures. It is most commonly encountered in the thigh during adulthood [1]. But other locations have been reported in the literature such as neck, axilla, shoulder, thorax, breast, stomach, retroperitoneum, and bone [1,5–7].

Radiologically, hibernomas raise a diagnostic challenge because in most imaging modalities they are indistinguishable from other malignant tumors, such as well-differentiated liposarcoma. On computed tomography (CT) scans, hibernomas appear as well-circumscribed, hyperdense lesions in the subcutaneous tissue sometimes deeply embedded intramuscularly, as we encountered in our case; however, this represents less than 15% of all cases. Therefore, MRI should be a part of the investigations done for any soft tissue mass that is larger than 5 cm or located deep to the fascia [1,3,4,8,9].

A plain MRI of a hibernoma would be isointense or relatively hypointense to the surrounding subcutaneous fat, and relatively hyperintense in comparison to the surrounding muscles on both T1 and T2. While upon using contrast, a diffused heterogeneous enhancement will be noted secondary to the hypervascularity of hibernomas. However, there is no single radiological pathognomonic sign of hibernomas [10,11].

Due to its silent growth, it has been reported in the literature to be discovered incidentally by radiological imaging done for other causes [6]. Particularly on positron emission tomography (PET) scans done for other indications, owing to the fact that hibernomas are highly metabolically active because of their brown fat tissue content. Hibernomas show uptake of 8F-fluorodeoxyglucose (18F-FDG) similar to or higher than that of a liposarcoma, which makes obtaining an accurate preoperative diagnosis quite difficult. A hibernoma might even be considered a metastatic lesion in patients with a history of malignancy, or in some cases as a primary malignancy [9,12,13].

Obtaining an accurate diagnosis based on radiological modalities alone is very challenging, therefore, a biopsy, whether incisional or a fine needle aspiration (FNA), is usually done in order to confirm the diagnosis through histopathology, and to avoid an unpremeditated excision of lipomatous malignant lesions. Incisional biopsies can resolve the issue of resemblance with malignant lipomatous lesions; but hibernomas have a rich blood supply and can bleed heavily, which makes the FNA a safer option if a hibernoma is suspected. In any form of biopsy used, the biopsy tract must be marked to be included during the excision to ensure a complete oncological resection in case of malignancy [10,14]. On the other hand, hibernomas are well known for their variable histopathological characteristics, therefore obtaining a small specimen from FNA would make it difficult to determine if it is a hibernoma or any other lipomatous lesion [3,13].

The histology of typical hibernoma cells consists of a solid pattern of large multivacuolated brown fat cells, single small eccentric nuclei, and a large amount of granular cytoplasm. These brown fat cells have been divided into 4 histological subtypes that depend on the degree of cytoplasmic eosinophilia, as well as the presence of myxoid stroma, and spindle cell configuration. The different subcategories of hibernomas may vary in prevalence depending on multiple demographic factors [8,10,12,13].

Typical hibernomas accounts for around 82% of all cases. The myxoid variant is often confused with liposarcoma, and is the second most common subtype, accounting for 9% of all cases. It is so-called due to the presence of myxoid stroma separating the multivacuolated cells. The lipoma-like subtype is characterized by scattered hibernoma cells within univacuolated mature adipocytes, and it accounts for 7% of all cases. This variant is also commonly mistaken for liposarcoma. Finally, the least common subtype is the spindle-cell variant which accounts for 2% of all cases and is regarded as a combination of the features of hibernoma and spindle-cell lipoma [3,10,12,15]. All the aforementioned histologic subtypes have the same prognosis, and thus there is no clinical significance in their classification, therefore most surgical pathologists do not subtype hibernoma cases [8,13].

A wide local excision with negative margins can be the solution to the diagnostic dilemma that hibernomas present, as it will provide a definitive diagnosis differentiating the hibernoma from other lipomatous lesions and prevent any future recurrence [11,14].

A retrospective study reviewed all the cases of hibernomas in one center and found 19 cases over a period of 20 years. Only 7% of the cases were reported in radiology reports to be inconclusive, and other lesions such as atypical lipomatous tumor or well-differentiated liposarcoma could not be excluded. All such cases underwent surgical excision with no report of distant metastasis or recurrence, with only one exception being a case that recurred due to positive margins rather than a true recurrence [8]. Another study included 64 cases of hibernoma, of which 16 were followed for around 14 years.

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All cases, except for 4 cases, did not develop any recurrence or metastatic spread, and like the previously mentioned study, recurrrences were a result of positive resection margins rather than an actual recurrence [3].

Conclusions

Caution is advised when dealing with lipomatous lesions, as they often overlap with malignancy. Furthermore, due to the inconclusiveness of most imaging modalities, an MRI should be done for any subcutaneous lesion that is larger than 5 cm or shows recent growth. The dilemma regarding the diagnosis of a hibernoma can be resolved through performing a biopsy with the next step being – or in cases where biopsy is not a viable option – a wide local excision with negative margins, including the site of biopsy when performed, is recommended to prevent any future recurrence.

Conflict of interests

None.

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