Recurrent Lipoma-Like Hibernoma

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Abstract

This is the fifth report of the rare lipoma-like variant of hibernoma, the 19th case reported, and the first documented as recurring after excision. Our patient was a 56-year-old man whose painful lipoma-like hibernoma (LLH) in the pelvis/buttock was initially treated with marginal excision 15 years earlier. Nine years after treatment, the LLH recurred locally. The recurrence was treated with partial excision and embolization, which alleviated symptoms. The disease was stable 26 months after repeat excision and 202 months after initial treatment.

LLH is 1 of the 4 histologic variants of hibernomas, which are rare benign lipomatous tumors distinguished from other lipomas by their brown-fat component. Only minimal information is available regarding the clinicopathologic characteristics of the individual variants. Reviewing the literature, we found that LLH predominantly develops in the fifth and sixth decades, and mean age at diagnosis is 41 years (age range, 2-66 years). LLH has a slight male predilection of 10:9. It most commonly develops in the thigh, with other occurrences reported in the hip, trunk, knee, and calf. The magnetic resonance imaging signal of LLH mirrors fat in all sequences. Presence of internal septations and enhancement with contrast are variable. Histologically, LLH is defined as a hibernoma composed predominantly of univacuolated white-fat cells and only scattered granular or pale hibernoma cells. The literature provides only a few treatment details regarding this variant.

Lipoma-like hibernoma (LLH) is 1 of 4 histologic variants of hibernomas, which are rare benign lipomatous tumors distinguished from other lipomas by their brown-fat component.[1,2] Up until now, only 18 LLHs have been reported, but none in the orthopedic or major oncologic literature,[1-4] and none had ever recurred after surgical treatment.

Hibernomas were first described by Merkel[5] in 1906. In 1914, Gery named them for their morphologic similarities to the brown fat in hibernating animals.[6-8] Since then, only a few case reports and series on these uncommon and unusual tumors have been published. The largest series, 170 cases reported by Furlong and colleagues[1] in 2001, described for the first time 4 distinct variants of hibernomas: typical, myxoid, lipoma-like, and spindle-cell. In their study, LLH was the second rarest variant, accounting for just 7% of all hibernomas. These variants were added to the *World Health Organization Classification of Tumours* in 2002,[9] but still little information is available on their clinicopathologic characteristics.

This is only the fifth report of LLH in the scientific literature, and the 19th case reported.[1-4] To add to the small body of data on hibernomas and LLHs, we describe a case that recurred in the pelvis/buttock of a 56-year-old man. This was the first and only known LLH to recur after excision. We also examine the literature on hibernomas and compare their clinical, radiographic, and histologic patterns with those of the lipoma-like variant.

Our patient provided informed written consent for print and electronic publication of this case report. Each author certifies that his institution approved or waived approval for the human protocol for this investigation and that all investigations were conducted in conformity with ethical principles of research.

Case Report

A 56-year-old man returned to our orthopedic oncology service for worsening of his painful, recurrent left pelvis/buttock mass. Fifteen years earlier, he had had a similarly painful left pelvis/buttock hibernoma treated with marginal excision. He remained disease-free for 9 years. Then the pain returned, and imaging confirmed recurrence. The pain was now severe and worse with activity. He denied fevers, chills, night sweats, and weight loss. His medical history was otherwise unremarkable.

Physical examination revealed mild tenderness in the buttock. No mass was palpable. Associated swelling, erythema, warmth, and lymphadenopathy were not present. Strength, range of motion, neurologic examination, and vascular examination were unremarkable.

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Am J Orthop. 2010;39(6):E57-E60.

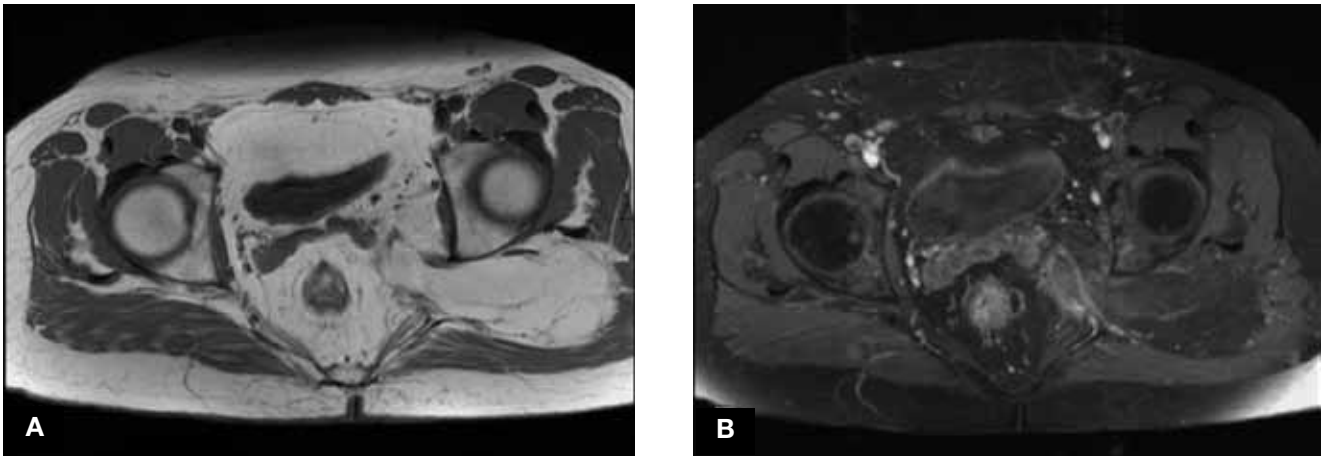

Figure 1. (A) T_1-weighted axial magnetic resonance imaging (MRI) shows fat-intense mass in left buttock extending into pelvis through sciatic notch. (B) T_1-weighted fat-suppressed axial MRI shows partial enhancement after contrast of medial/deep component of mass.

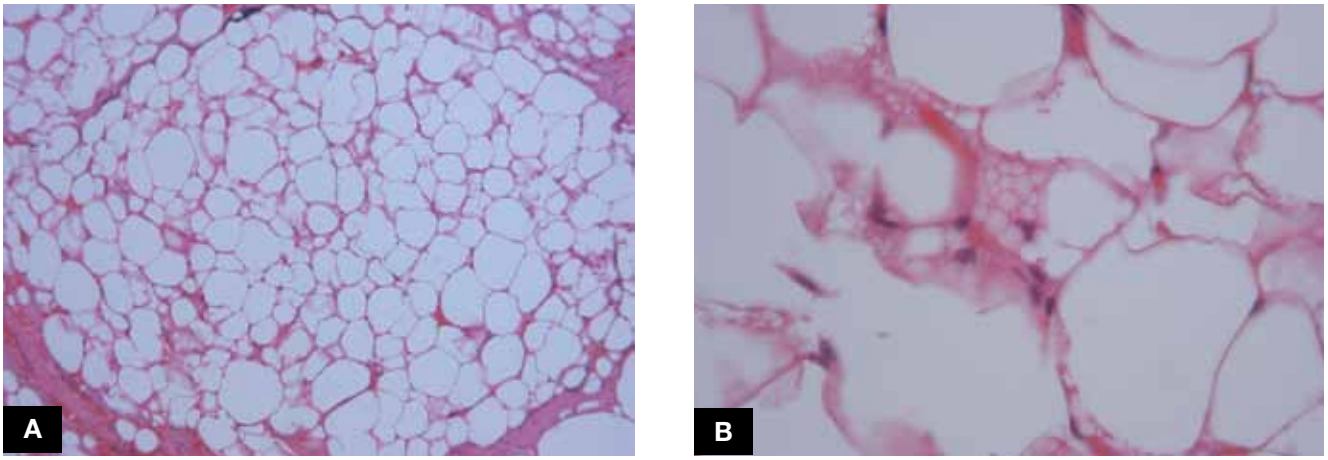

Figure 2. (A) At low power, predominantly univacuolated adipocytes (white fat) are visible with only scattered multivacuolated adipocytes (brown fat). (B) At high power, a cluster of multivacuolated adipocytes (brown fat) is visible in the center.

Magnetic resonance imaging (MRI) showed a 13×12×6-cm mass near the gluteus musculature (Figures 1A, 1B). The signal intensity of the mass followed fat in all sequences, and septations were present (Figures 1A, 1B). Portions of the mass were enhanced with contrast (Figure 1B).

A repeat marginal excision was planned, but only partial excision was possible, as the mass tightly adhered to the sciatic nerve and extended deep into the pelvis. Grossly, the tumor was described as yellow-tan fatty tissue. Microscopically, it consisted primarily of univacuolated adipocytes (Figures 2A, 2B). Five percent of the adipocytes were multivacuolated with centrally located round nuclei consistent with brown-fat cells (Figures 2A, 2B). Fibrous septa and abundant vasculature were also present. These findings were diagnostic for LLH.

There were no perioperative complications. Local embolization was performed 3 months later to minimize risk for recurrence from residual tumor. The patient, last seen at postoperative month 26, was doing well and had stable but minimally symptomatic residual disease.

Discussion

Hibernomas are rare benign lipomatous tumors, accounting for 1.1% of all adipocytic tumors in the Armed Forces Institute of Pathology files.[9] LLH, the second rarest of the 4 histologic variants of hibernoma, was reported only 18 times[1-4] before the present case. Clinical characteristics, treatments, and outcomes for all known cases of LLH are listed in the Table.

All hibernomas are derived from brown fat.[1,2,5] Human fetuses have deposits of brown fat, but the vast majority of this fat turns into white fat as fetus and newborn grow.[10,11] Adults retain minimal amounts of brown fat (~1% of total fat), which typically persists in the neck, axilla, mediastinum, periaortic area, and perirenal area.[10-12]

Hibernomas classically were thought to arise from regions of residual brown fat, with most reports describing their development in the upper back, shoulders,

Table. Clinical Characteristics, Treatments, and Outcomes for All Known Lipoma-Like Hibernomas

Authors	Pts (n)	Age (y) at Diagnosis	Sex	Tumor Location	Tumor Size (cm) at Diagnosis	Presenting Symptoms	Initial Treatment	Recurrence	Follow-Up (mo)
Furlong et al[1]	12	36.5 median	7M, 5F	Thigh, hip, trunk	—	—	—	—	—
Reis-Filho et al[4]	1	17	F	Thigh	9.5	Painless mass	Marginal excision	None	4
Sakabe et al[3]	1	53	F	Thigh	—	Painless mass	—	—	—
Ritchie et al[2]	4	66	F	Calf	15	—	—	—	—
		52	M	Thigh	27	—	—	—	—
		54	F	Knee	4	—	—	—	—
		47	M	Thigh	6	—	—	—	—
Moretti et al (present case)	1	41	M	Pelvis/buttock	—	Painful mass	Marginal excision	Local (109 mo)	202

neck, mediastinum, and retroperitoneum.[6,7,13,14] One large series recently suggested they are most common in the thigh, an area not known in adults to contain brown fat.[1] In comparison, the lipoma-like variant of hibernoma is also most often seen in the thigh, with additional occurrences reported in the hip, trunk, knee, and calf.[1-4] Noncategorized hibernomas have been reported in nearly all age groups but are most commonly seen in the fourth and fifth decades.[1] We similarly found that the lipoma-like variant predominately occurs in the fifth and sixth decades, with a mean age of 41 years at diagnosis (age range, 2-66 years).[1-4] This is 1 of only 2 variants known to affect pediatric patients. Noncategorized hibernomas display a slight male predilection of 4:3, and the lipoma-like variant shows a similar slight male predilection of 10:9.[1-4,12,15-19]

Details on the presenting symptoms for the previous 18 LLHs are limited to 2 case reports, describing painless masses.[3,4] Noncategorized hibernomas similarly tend to present as asymptomatic, slow-growing, painless soft-tissue masses.[1,6,7,16] Some are found only incidentally, during radiologic examination.[17,20] As was the case with our patient, however, hibernomas can also on rare occasion cause pain or paresthesia from the mass effect.[13,21,22] Interestingly, as hibernomas are typically hypervascular, the skin overlying superficial hibernomas may be warm.[23] Such was not the case in our patient.

Histologically, hibernomas are characterized by multivacuolated brown-fat cells with small central nuclei.[1,13,24] The common lipoma, by comparison, is a tumor of white-fat cells with a single large lipid vacuole and a peripheral nucleus. Another distinguishing characteristic of hibernomas is their abundant vascularity, a feature not seen in the typical benign lipomas composed of white-fat tissue.[13,20,21,24] In 2001, Furlong and colleagues[1] identified 4 distinct hibernoma variants on the basis of the tinctorial quality of the their cells, the nature of the stroma, and the presence of a spindle-cell component. LLH was the second rarest variant, accounting for only 12 (7%) of their 170 cases.[1] In our patient's case, this variant was composed predominantly of univacuolated white-fat cells and only scattered granular or pale hibernoma cells.[1] This histologic appearance can be confused with that of atypical lipomas or well-differentiated liposarcomas when the multivacuolated brown-fat cells are mistaken for lipoblasts.[4] Such was initially the case in 23% of the LLHs reported by Furlong and colleagues.[1]

There are few imaging reports on histologically proven lipoma-like variants.[2,3] Ritchie and colleagues[2] reported on 4 LLHs with MRI findings nearly identical to ours: a mass appearing slightly hypointense to isointense to fat on T_1 sequences and isointense to slightly hyperintense to fat on T_2 sequences. Half of their cases also had thin linear structures, similar to the septations in our case. MRI appearance after contrast is unclear, though, as 1 reported case enhanced, whereas ours did not. For most hibernomas, a range of MRI appearances has been reported.[6,15,21,25,26] The most common description is a mass hypointense to fat on T_1 sequences and isointense to fat on T_2 sequences.[6,15,21,25,26] After contrast, noncategorized hibernomas tend to display at least scattered slight enhancement.[15,25,26]

As hibernomas are benign tumors, marginal excision is generally considered curative.[1] No metastases or deaths have been reported from these lesions. There is a handful of cases of hibernomas with nuclear atypia, but a malignant course has never been documented.[27,28] Although there were no recurrences among the 170 cases reported by Furlong and colleagues,[1] there are 5 other reports of hibernomas that recurred after surgical resection.[13,15,27] Two of these recurrent cases were typical variant hibernomas, and the other 3 were not categorized. Therefore, our case is the only known LLH, and one of the few known hibernomas, to recur after treatment. However, patient numbers are too small to suggest whether any single variant is more likely than another to recur.

In summary, LLH is a rare hibernoma variant that historically has arisen in the thigh of middle-aged adults. Our case, the 19th to be reported, recurred in the pelvis/buttock of a 56-year-old man. Physical examination and imaging can be suggestive of hibernoma, but on imaging internal septae can also mimic liposarcoma. Histologically, LLH is distinguished from the other variants by its relative paucity of multivacuolated brown-fat cells. Treatment of hibernomas by marginal excision is

generally considered curative, with there being only a few reports of recurrence and none of metastasis. Our patient, with the rare lipoma-like variant, was similarly treated with marginal excision, but he had a recurrence almost 9 years later. Then partial excision and embolization were required to control the disease and symptoms. The disease was stable 26 months after repeat excision and 202 months after initial treatment. Although uncommon, these tumors should be considered in the differential diagnosis of all lipomatous lesions.

Authors' Disclosure Statement

Dr. Moretti received funding in the form of a research fellowship from Stryker Orthopaedics (Mahwah, NJ), and Dr. Lackman is a consultant for Stryker Orthopaedics. Dr. de la Cruz reports no actual or potential conflict of interest in relation to this article.

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