



Case report

A case report of hibernoma in the thigh comprising the femoral neuromuscular bundle

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ABSTRACT

Introduction: Hibernomas are rare, slow-growing, painless, benign tumors of soft tissue that develop from residual brown fat cell showing predilection for areas where brown fat is more common in fetuses and infants. Due to the rarity of the tumor its often overlooked or mistaken for other pathologies such as liposarcoma.

Case presentation: Using the CARE 2020 criteria we describe a 26-year-old man who presented with painful swelling of the thigh. A magnetic resonance imaging examination without contrast showed a mass in the medial thigh just lateral to the femoral artery. A biopsy specimen was obtained from his thigh for histopathological analysis, and the findings suggested a hibernoma. A wide resection was performed, and it showed that the femoral nerve was lateral to the mass, with the course of the nerve altered due to mass effect. The mass was sent for a histopathological examination, and the findings were consistent with a hibernoma. Two weeks post-operative, the patient reported a considerable decrease in pain intensity, and six months post-operative, he reported complete pain relief.

Discussion: Hibernomas are unusual tumors that are benign and usually painless, but the large size the tumor can reach and proximity to vital structures can explain the growing trends symptomatic nature of some hibernomas. A proper and step wise approach using clinical, radiological and histopathology is important to Diagnose and plan surgical management. Proper understanding of the proximity of adjacent structures and the high vascularity of the tumor.

Conclusion: Because of the rarity and under reporting of hibernomas they are often misdiagnosed as large lipomas or malignant soft tissue tumors, underscoring the importance of histopathology in ensuring accurate diagnosis and anticipating intraoperative. Careful dissection and ligation of the vasculature with the understanding of adjacent anatomy are key to safe tumor excision.

1. Introduction

Hibernomas are rare benign tumors of soft tissue that develop from residual brown fat cells. They are typically observed in adults aged 30–40 years, with equal gender distribution [1]. The tumors show a predilection for areas where brown fat is more common in fetuses and infants, such as the underarms, shoulders, mediastinum, and retroperitoneum [1], and they are uncommon in the cranium and popliteal fossa [2]. Other uncommon locations include the lower and upper extremities and the abdomen [1,2].

Hibernomas manifest as slow-growing, painless masses that can be confused with other tumor types, such as liposarcomas [3]. Among the

six histological subtypes identified [4], the lipoma-like variant is often misdiagnosed as an atypical lipoma or well-differentiated liposarcoma [1].

Treatment of hibernomas is usually excision [6,7] with complete excision usually being curative.

In our case we describe a case of hibernoma of the thigh presenting as a painful swelling compressing the femoral nerve in a tertiary care oncology center.

2. Case presentation

Using the SCARE 2020 criteria [15] we would like to report.

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A 26-year-old man medically free office worker and recreational athlete was referred to our center for a thigh mass of insidious onset. The patient noticed the swelling while playing sports six months prior to his presentation to our hospital. He noticed a significant difference in size between both thighs, with the diameter of his right thigh being much bigger. However, he experienced no pain, tenderness, or restriction in the range of motion of his right thigh. He did not initially seek medical advice. As time progressed, the patient started feeling pain in the anterior aspect of the right thigh associated with a considerable increase in the size of the thigh. Additionally, the pain was accompanied by an unintentional weight loss of five kilograms in four months; there was no history of night sweats nor fever. He presented for a consultation at his local hospital, where a magnetic resonance imaging (MRI) examination of the right thigh was performed prior to his referral to a tertiary center.

The patient presented with an aching type of pain severe enough to wake him up from sleep. The pain was localized with no radiation. It was neither aggravated nor relieved by movement or posture changes. Over-the-counter analgesics slightly improved the pain but not enough for a full night of sleep.

The swelling was in the anterior medial side of the right thigh. The mass was rubbery in consistency, ill-defined, non-pulsating, and tender to deep palpation, and it measured approximately 10×20 cm. The overlying skin was slightly warm, but it was not erythematous, and there were no wounds or ulcers overlying the mass. The mass was deep-seated, and it was difficult to assess its mobility. However, it did not appear to be fixed to surrounding tissue. A neuro-vascular exam of the limb was unremarkable with fully palpable pulses. A sensory and motor examination was normal, and hip and knee range of motion tests were painless with full power and maintained reflexes. The patient's thigh pain was exacerbated by a straight leg test.

An X-ray examination of the thigh showed soft tissue changes with no calcification. An MRI examination without contrast showed a mass in the medial thigh just lateral to the femoral artery measuring $5.5 \times 7.5 \times 13.0$ cm in anterior-posterior, transverse, and axial diameter (Figs. 1, 2, and 3). The mass appeared to be seated between the sartorius and rectus femoris, superficial to the vastus intermedius muscles, and it had a high T1 signal intensity that was suppressed with fat suppression techniques. Possible differential diagnoses included a hibernoma, a lipoma, or an atypical lipomatous tumor.

A percutaneous biopsy specimen was obtained from the patient's thigh for histopathological analysis, and the findings were suggestive of a hibernoma. A wide resection was offered.

The procedure was performed under general anesthesia by orthopedic oncology consultant and vascular surgery back up with cross-match of 2 units prepared. A mass of similar size as the original tumor was identified between the rectus femoris and sartorius muscle. Careful dissection was done around the mass, making sure not to rupture the capsule (Fig. 4). The mass was vascular in nature, with many perforating arteries arising from the femoral artery, which was identified just medial to the mass. Careful hemostasis and ligation of the perforators were done using both sutures and vascular clips. The femoral nerve was just lateral to the mass with the course of the nerve altered due to mass effect. After freeing the mass from blood vessels and all adhesions, the stalk of the mass was ligated, and the excision was completed. The femoral nerve was traced and freed from any adhesions, and careful hemostasis was done before closure. The mass was then sent for a histopathological examination, and the findings were consistent with a hibernoma.

The patient was assessed two weeks post-operative. He reported a considerable decrease in the intensity of his pain compared to before the procedure. Six months post-operative, the patient reported complete pain relief, and wound healing was unremarkable. There was no numbness or weakness on neuromuscular examination.

3. Discussion

Hibernomas were first described by Merkle in 1906 [5], and the tumor has been classically described as a slow-growing, mobile, painless mass that can reach up to $29.0 \times 19.0 \times 12.0$ cm in diameter [6] and 2500 g in weight [7]. However, hibernomas can become symptomatic due to mass effect and other anatomical variabilities, as reported in our case and several cases in the medical literature [6,8,9]. In our report, the patient presented with an aching pain that was severe enough to interrupt his sleep and was only slightly improved with the use of over-the-counter analgesics. 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.

Due to the rarity of the tumor, many clinicians and radiologists are unfamiliar with it, contributing to misdiagnosis and underreporting [1]. Although hibernomas are benign tumors, it is important for clinicians to correctly identify them pre-operatively due to their vascular nature and size. Notably, large tumors and those adjacent to major nerves and blood vessels may cause compression of adjacent organs [10], as was the case

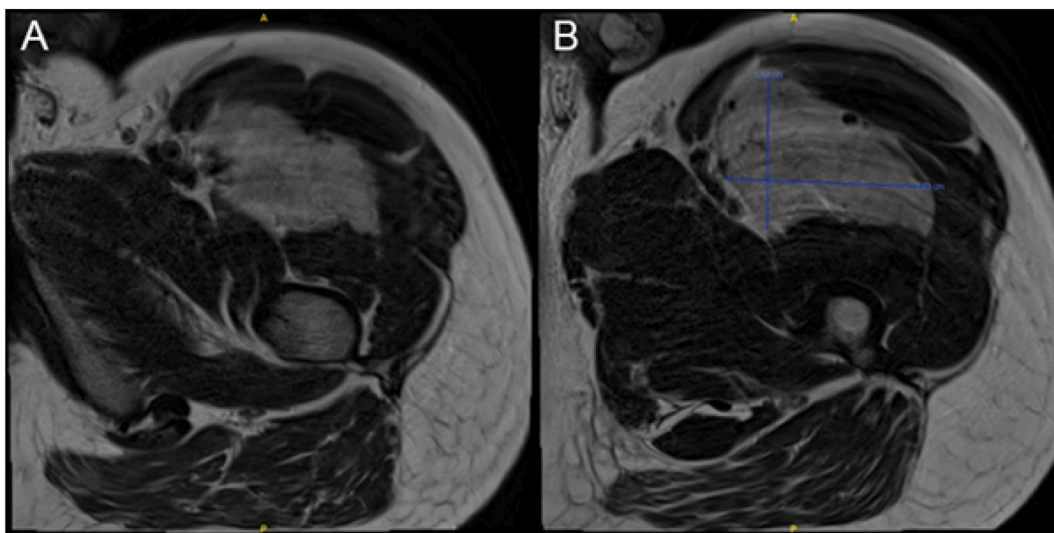


Fig. 1. Axial magnetic resonance imaging (MRI) T2-weighted image showing void flow representing the femoral artery with possible communication (A). Axial MRI T2-weighted image showing the mass measuring 5.64×7.73 cm (B).

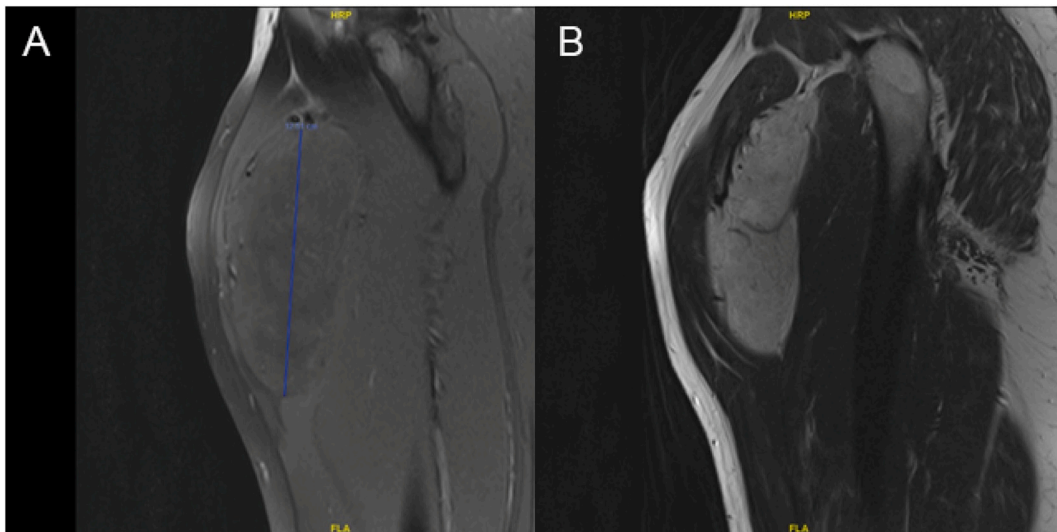


Fig. 2. Sagittal magnetic resonance imaging T1-weighted image showing the mass measuring 12.51 cm length (A). Although the mass is bright on a T2-weighted scan, it is not as bright as subcutaneous fat (B).

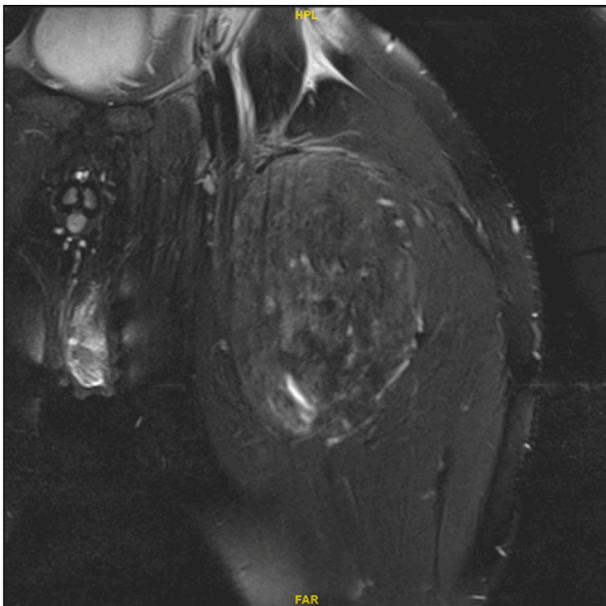


Fig. 3. Coronal T2-weighted image showing the mass.

in our patient whose tumor was compressing the femoral nerve. Additionally, the mass was vascular, with many perforating arteries arising from the femoral artery.

A histopathological examination is considered the gold standard for diagnosing hibernomas. In patients with deep soft tissue tumors larger than 3 cm in diameter, as was the case in our patient, a biopsy is recommended to reach a definitive diagnosis [6]. Nevertheless, a puncture biopsy may not be recommended because of the vascular nature of the tumor, which may increase the patient's risk of bleeding. Therefore, tumor excision is preferable.

Advanced imaging techniques can guide clinicians in making a pre-operative diagnosis. On ultrasonography, a hibernoma appears as a hyperechogenic mass, whereas angiography shows a highly vascularized tumor with occasional arteriovenous shunts. Technetium99 scintigraphy yields areas of increased uptake. Computed tomography shows the tumor density to be intermediate between muscle and fat, with possible heterogeneous enhancement after contrast enhancement and

the presence of intratumoral vessels [11]. On MRI, the radiographic study of choice, the mass shows high-signal intensity on T1-weighted images, and it is isointense or hypointense on T2-weighted images [12]; fat suppression can show the tumor vessels [13].

A careful understanding of tumor location and the relationship of the tumor with adjacent structures is important to guide the surgeon in careful dissection and hemostasis. In our patient's case, an aMRI was helpful to determine the relationship between the mass and adjacent structures, which can assist in planning the intervention. In general, hibernomas are not invasive, although there has been a report of incomplete excision due to the tumor being near a critical vascular structure [14]. The well-encapsulated tumor is non-adherent during excision, and complete resection is curative.

4. Conclusion

Hibernomas are unusual tumors whose onset and slow progression may contribute to underreporting and misdiagnosis. A proper diagnosis can be reached by using a systemic approach and incorporating many diagnostic modalities, such as imaging and biopsy. Due to the rarity of this tumor, it can be misdiagnosed as a large lipoma or a malignant soft tissue tumor, underscoring the importance of histopathology in ensuring accurate diagnosis. Some technical challenges must be considered during tumor excision, such as its size and vascularity. Therefore, careful dissection and ligation of the vasculature is key to safe tumor excision.

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Ethical approval

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Consent

Taken by the patient.

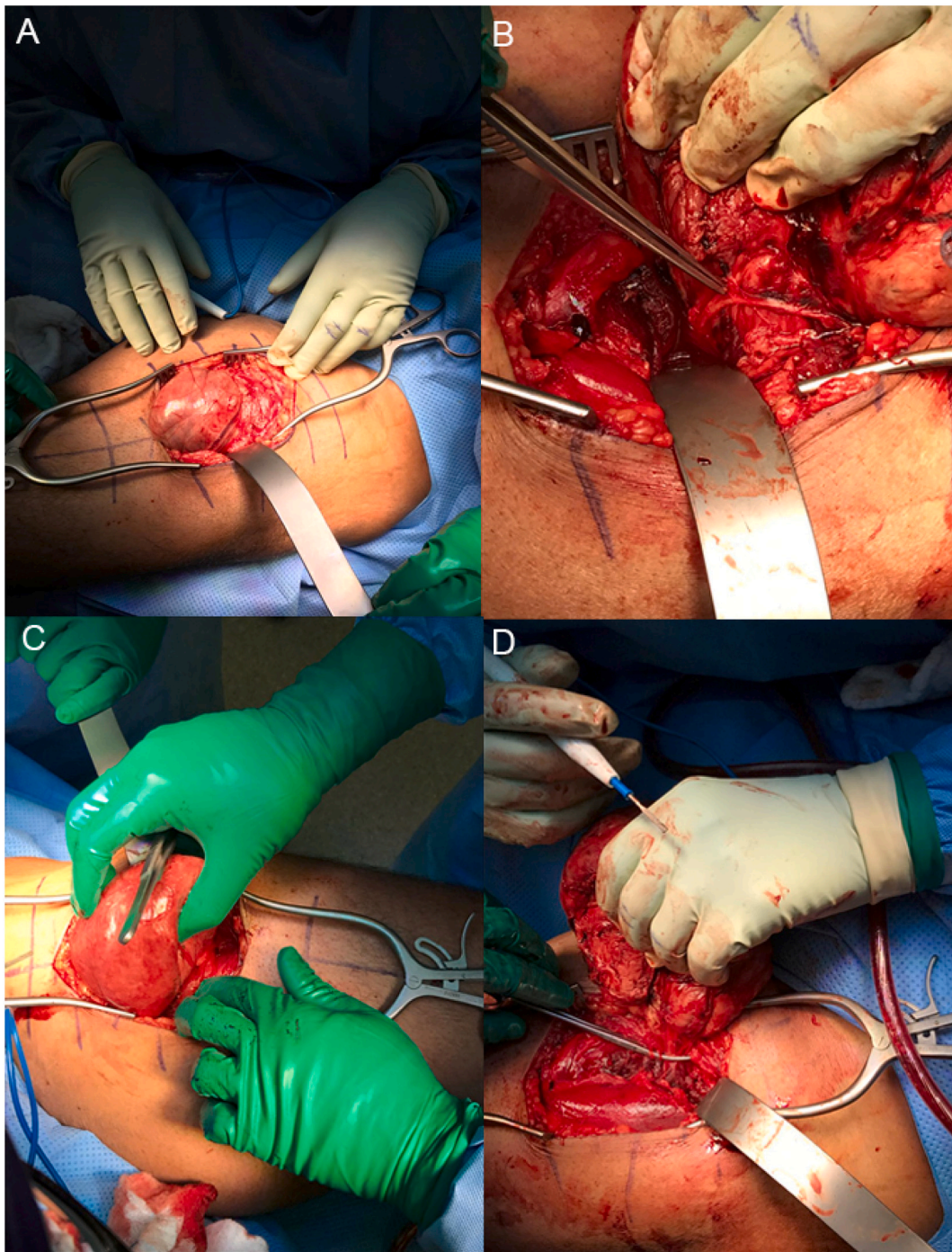


Fig. 4. Intraoperative image of the mass upon retraction of the sartorius and rectus femoris (A). Note the proximity between the femoral nerve and the mass (B). The femoral artery was palpated after it was identified medial to the mass (C). The deep perforators were ligated at the stalk of the mass (D).

Research registration

Non to declare.

Guarantor

Khalid Beidas

CRedit authorship contribution statement

Khalid F Beidas: primary author, reviewer.
Hasan Sawan: consultant operating and reviewer.

Fahad alwatban: radiologist that marked the images.

Khalid alhazazi: Data collection.

Tala F Beidas: literature Review.

Declaration of competing interest

Non-to declare.

References

- [1] M.A. Furlong, J.C. Fanburg-Smith, M. Miettinen, The morphologic spectrum of hibernoma: a clinicopathologic study of 170 cases, *Am. J. Surg. Pathol.* 25 (2001) 809–814.
- [2] M.D. Murphey, J.F. Carroll, D.J. Flemming, T.L. Pope, F.H. Gannon, M. J. Kransdorf, From the archives of the AFIP: benign musculoskeletal lipomatous lesions, *Radiographics* 24 (2004) 1433–1466.
- [3] S.D. Patil, A.R. Sheik, V. Tewari, D. Mutreja, Hibernoma: a missed diagnosis!., *Indian J. Pathol. Microbiol.* 62 (2019) 461.
- [4] C.D.M. Fletcher, *Diagnostic Histopathology of Tumors*, 3rd ed., Elsevier Health Sciences, Hong Kong, 2007.
- [5] H. Merkel, On a pseudolipoma of the breast, *Beitr Pathol Anat.* 39 (1906) 152–157.
- [6] S. Ersozlu, O. Sahin, A.F. Ozgur, T. Akkaya, Sciatic neuropathy from a giant hibernoma of the thigh: a case report, *Am. J. Orthop.* 37 (2008) E103–E106.
- [7] P.J. Lewandowski, S.D. Weiner, Hibernoma of the medial thigh. Case report and literature review, *Clin. Orthop. Relat. Res.* (1996) 198–201.
- [8] B. Salim, C. Belkacem, Hibernoma of the thigh: a report of four cases, *J. Orthop. Surg.* (Hong Kong) 22 (2014) 118–121. SAGE Publications Ltd STM.
- [9] D.C. DeRosa, R.B. Lim, K. Lin-Hurtubise, E.A. Johnson, Symptomatic hibernoma: a rare soft tissue tumor, *Hawaii J. Med. Public Health* 71 (2012) 342–345.
- [10] D. Daubner, S. Spieth, J. Pablik, K. Zöphel, T. Paulus, M. Laniado, Hibernoma—two patients with a rare lipoid soft-tissue tumour, *BMC Med. Imaging* 15 (2015) 4.
- [11] S.E. Anderson, C. Schwab, E. Stauffer, A. Banic, L.S. Steinbach, Hibernoma: imaging characteristics of a rare benign soft tissue tumor, *Skelet. Radiol.* 30 (2001) 590–595.
- [12] A. Datir, S.L.J. James, K. Ali, J. Lee, M. Ahmad, A. Saifuddin, MRI of soft-tissue masses: the relationship between lesion size, depth, and diagnosis, *Clin. Radiol.* 63 (2008) 373–378, discussion 379–380.
- [13] S. Atilla, S.S. Eilenberg, J.J. Brown, Hibernoma: MRI appearance of a rare tumor, *Magn. Reson. Imaging* 13 (1995) 335–337.
- [14] S.M. Lele, S. Chundru, G. Chaljub, P. Adegboyega, A.K. Haque, Hibernoma: a report of 2 unusual cases with a review of the literature, *Arch. Pathol. Lab. Med.* 126 (2002) 975–978.
- [15] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus Surgical CASE REport (SCARE) guidelines, *Int. J. Surg.* 84 (2020) 226–230.