Sciatic Neuropathy From a Giant Hibernoma of the Thigh: A Case Report

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Hibernomas—rare, uniformly benign soft-tissue tumors of brown fat—were originally described in 1906 by Merkel.[1] These tumors are usually found in the scapular[2] and posterior cervical regions or (more rarely) in the folds of the buttocks or on the thigh.[3-5]

Sciatic neuropathy is an infrequently diagnosed focal mononeuropathy. Few case reports of lipomas compressing the sciatic nerve or its peripheral branches have appeared in the literature.[6-8] The present case report is to our knowledge the first on sciatic nerve palsy caused by a hibernoma.

Case Report

A woman in her early 30s was referred to our clinic with a painless left-side posterior thigh mass that had been slowly enlarging over 5 years. She complained of loss of sensation in the left lower leg and had left ankle weakness while walking during the month before she became our patient. She had no history of trauma and exhibited no systemic signs, such as weight loss, fatigue, fever, or night sweats.

Physical examination revealed a soft, nontender, left-side posteromedial thigh mass (~25×15 cm). No overlying warmth or erythema was noted. Neurovascular examination revealed marked weakness in dorsiflexion of the left ankle and toes in addition to ankle eversion. Sensory examination indicated hypoesthesia over the peroneal and tibial nerve distribution in the left anterolateral lower leg. The patient exhibited complete paralysis of the anterior tibialis, extensor hallucis longus, and extensor digitorum communis (deep peroneal nerve-innervated muscles); severe (2/5) motor loss in the peronei (superficial peroneal nerve-innervated muscles); moderate (3/5) weakness in the gastrocnemius/soleus, posterior tibialis, and toe flexors (tibial nerve-innervated muscles); and sensory abnormalities throughout the entire sciatic nerve distribution. The common peroneal, posterior tibial, superficial peroneal, and sural nerves were electrodiagnostically evaluated, and reduced amplitude in the peroneal and the tibialis posterior nerves was detected. Sensory nerve conduction of the superficial peroneal and sural nerves in the left leg was less than that in the right leg. These findings confirmed active motor and sensory sciatic mononeuropathy of the left leg.

Plain x-rays of the left leg showed a large soft-tissue mass without calcification. Magnetic resonance imaging (MRI) of the left leg revealed a well-circumscribed large soft-tissue mass that was 27 cm at its maximum dimension and that primarily involved the posterior and medial compartments of the left thigh. The mass was characterized by a heterogeneous increased signal intensity approaching that of subcutaneous fat on both T_1-weighted and T_2-weighted images. On T_1-weighted images, this increased intensity was intermediate (lower than fat, higher than muscle), but heterogeneous hyperintensity was most prominent in the T_2-weighted images with no cystic

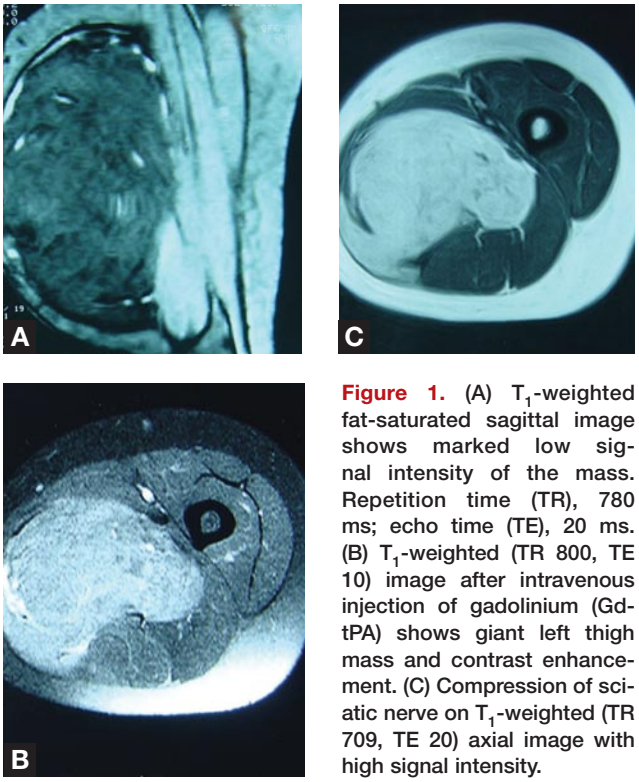

Figure 1. (A) T_1-weighted fat-saturated sagittal image shows marked low signal intensity of the mass. Repetition time (TR), 780 ms; echo time (TE), 20 ms. (B) T_1-weighted (TR 800, TE 10) image after intravenous injection of gadolinium (Gd-tPA) shows giant left thigh mass and contrast enhancement. (C) Compression of sciatic nerve on T_1-weighted (TR 709, TE 20) axial image with high signal intensity.

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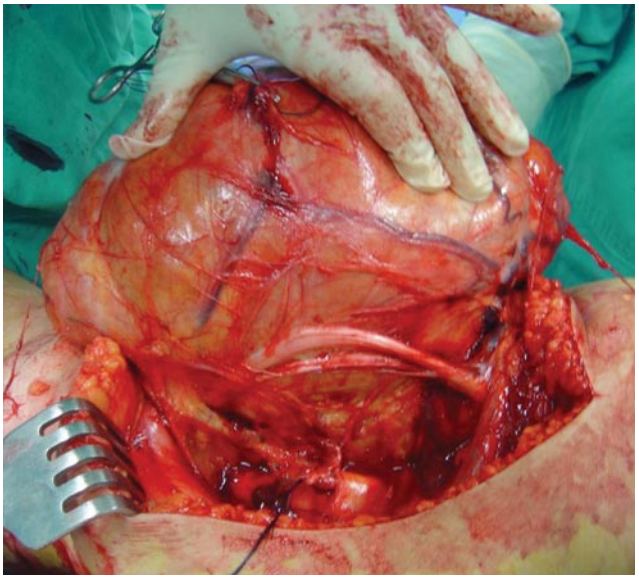

Figure 2. Intraoperative view of giant hibernoma that is compressing the sciatic nerve.

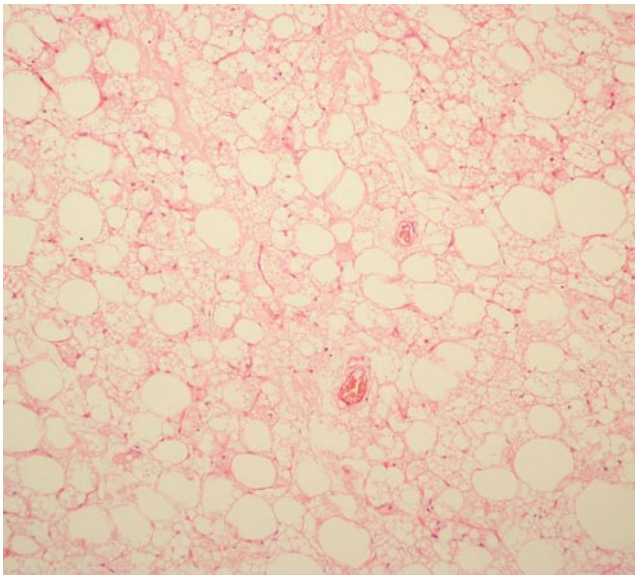

Figure 3. Histopathologic specimen of thigh mass with hematoxylin-eosin stain confirms diagnosis as hibernoma with large vacuolated fat cells with eosinophilic features.

components. The signal intensity of the mass was markedly suppressed on fat-saturated images (Figure 1A), and there was marked contrast enhancement after intravenous injection of gadolinium (Figure 1B). Although no necrosis or hemorrhage was observed, compression of the sciatic nerve was identified in the axial images (Figure 1C).

Surgery was performed after the patient had received a spinal anesthetic and had been placed in the prone position. A midline longitudinal surgical approach over the mass was used for exposure. The tumor extended from the gluteal region to the popliteal region. The sciatic nerve was stretched and compressed by a firm, yellowish tumor (Figure 2). The sciatic nerve dissection that was performed spared the sciatic nerve innervations to the surrounding musculature. The mass was excised, vacuum drains were placed, and the surgical wound was closed. After marginal excision, gross inspection of the mass revealed firm, well-circumscribed, partially encapsulated, lobulated, yellow-brown, adipose tissue 2500 g in weight and 29.0×19.0×12.0 cm in size. Microscopically, the tumor consisted of irregularly shaped multivacuolated fat cells with darkly stained small central nuclei. Some of the fat cells, which were not atypical or mitotic, exhibited pale staining and eosinophilic features typical of hibernoma cells (Figure 3). The final diagnosis was that of a benign brown-fat tumor (hibernoma).

The patient's hypoesthesia resolved 3 months after surgery, and the footdrop resolved completely 6 months after surgery. The patient was able to ambulate normally and returned to full activity.

Discussion

Hibernomas—rare, benign, soft-tissue tumors composed of brown fat[1]—have been reported in almost all age groups but develop most often during the third decade of life.[9] They usually present as firm, freely movable, painless, slow-growing masses.[10] All reported hibernomas have been benign and have not recurred after resection.[11]

Hibernomas, which can develop in a variety of anatomical regions, have been reported in the thigh[11] and mediastinum[12]; however, the most common sites are the pericapsular-interscapular,[13] neck,[14] and axillary, intrathoracic, and retroperitoneal regions.[15-17] About 14 hibernomas have been described as having developed in the gluteal region or on the lower extremities.[18] In such presentations, hibernomas are often found in the anteromedial or posterior compartments, and most develop within the proximal or middle third of the thigh. In our patient, the hibernoma was in the posterior proximal third of the thigh.

Adenomas of the sebaceous glands or *pseudolipomas*[19] were renamed *hibernomas* because their gross appearance mimics that of the brown fat of hibernating animals.[1,20] Although many investigators have analyzed the gross features of hibernomas,[21-23] histologic studies of the lesions are limited to a few cases or small series.[24] One of the most extensive pathologic studies of hibernomas included 170 histologic slides that were analyzed in detail.[25] In that study, morphologic review distinguished 4 histologic hibernoma variants with no clinical significance: typical, myxoid, spindle-cell, and lipoma-like. The typical hibernoma was classified into 3 subtypes: pale cell, mixed cell, and eosinophilic cell. The authors stated that the importance of this morphologic spectrum was to establish those variants as benign hibernomas. Our patient had a typical hibernoma characterized by irregularly shaped multivacuolated fat cells with darkly stained small, central nuclei. Pale staining and presence of eosinophilic hibernoma cells confirmed the diagnosis as a pale cell hibernoma.

Hibernomas develop in various sizes. Lewandowski and Weiner[5] reported a 40-year-old woman with a painless right-side medial thigh mass that had been enlarging

over 1 year. A marginal resection was performed, and an 840-g tumor was excised. The marginally excised tumor in our patient weighed 2500 g, probably because the patient delayed treatment. In a study by Alvine and colleagues,[11] a 42-year-old man presented with a large left-side thigh mass approximately 20 cm in length. Although that patient had a giant mass (13×10.8×21 cm), he exhibited no neurologic deficit. The resected hibernoma in our patient measured 29.0×19.0×12.0 cm and had grown large enough to compress the sciatic nerve, which produced neuropathy.

Aspects of Diagnosis

There is considerable overlap in presentation of benign and malignant soft-tissue tumors. For this reason, clinicians must take a systematic approach toward evaluation and biopsy in order to avoid treatment errors. After unplanned resections, incomplete removal of a malignancy may lead to a contaminated excision, and this can change the treatment course and may even require an otherwise unnecessary amputation.[26] Therefore, excisional biopsy (in which the entire lesion is resected without knowledge of the diagnosis) should be performed only when the surgeon, using clinical findings and imaging studies, is certain that the lesion is benign. Excisional biopsy may also be performed when a wide range can be obtained with no increased morbidity. When the nature of the soft-tissue mass cannot be determined (indeterminate lesions), a biopsy (fine-needle aspiration, core-needle, or incisional) is usually necessary to establish a histologic diagnosis.[27] However, incorrect or poorly performed biopsies may lead to contamination along the biopsy tract, and local recurrences, major diagnostic errors, or unnecessary amputations may result.[26]

For evaluation of the mass and final decision making, detailed history, physical examination, and imaging studies must be obtained to minimize diagnostic errors and biopsy complications. Although some soft-tissue tumors, including liposarcomas, must be considered in the differential diagnosis,[28] clinicians and radiologists are able to precisely identify some soft-tissue masses, such as lipomas, ganglions, hemangiomas, and popliteal cysts, with detailed history, physical examination, and MRI. Awareness of MRI appearances of hibernomas may allow for improved preoperative diagnosis. For this reason, MRI is the most helpful tool for differentiating hibernomas from other conditions. Anderson and colleagues[18] reported on the MRI features of hibernomas, which they described as hyperintense or isointense fatty lesions (on T_1-weighted images) that exhibit a heterogeneously increased signal intensity on T_2-weighted images.[18]

Our patient had a slowly growing soft-tissue mass, which was palpable on physical examination. Plain x-rays were normal and showed no signs of malignancy. MRI appearance was typical of a hibernoma (as previously described in the literature[3,18]): intermediate signal intensity (higher than muscles, lower than fat) on T_1-weighted images and heterogeneous, highly increased signal intensity on T_2-weighted images without any necrosis or hemorrhage should suggest this diagnosis. In addition, well-differentiated, low-grade liposarcomas may contain large areas of fat and have normal fatty attenuation on fat-suppressed images. In our patient's case, fat-suppressed images did not show normal fatty attenuation and lacked fatty signal characteristics, so low-grade liposarcomas were excluded.[28] The most common liposarcoma types, myxoid forms, have inhomogenous T_2 signal–intensity characteristics with cystic components.[29,30] On MRI, our patient had no characteristics of inhomogeneity with cystic components, excluding myxoid liposarcomas. High-grade liposarcomas can be also kept in mind in the differential diagnosis, but a diffusely homogenous appearance, evident on MRI in our patient's case, would be atypical for high-grade liposarcoma.[28] For all these reasons, the mass was accepted as benign (hibernoma), so no fine-needle aspiration or core-needle or incisional biopsy was planned before definitive surgery, and complete excision was performed. Indeed, for deep soft-tissue masses larger than 3 cm, there should be a definitive diagnosis with a proper biopsy. We performed an excisional biopsy because the tumor had the diagnostic appearance of a hibernoma, but, usually, when a lesion cannot be characterized by clinical and radiologic techniques, an incisional or needle biopsy should always be performed with a good technique before definitive surgery.

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Symptoms Are Rare

Hibernomas seldom cause symptoms, which usually result from the compression of adjacent structures, such as the sciatic nerve.[11] Although the literature includes accounts of sciatic neuropathy caused by lipoma of the thigh,[6] hibernoma of the thigh causing sciatic neuropathy has not to our knowledge been reported previously. Botwin and colleagues[6] described an infiltrating intermuscular lipoma of the thigh in a 52-year-old man with paresthesia throughout the right leg. Physical examination revealed normal motor function and hypoesthesia over the peroneal and tibial nerve distribution, which confirmed the diagnosis of neuropathy. After marginal excision, results of the gross pathologic examination revealed a mass of firm lobulated adipose tissue (1166 g in weight, 25×17×5 cm in size) compressing the sciatic nerve.

Our patient had both motor and sensory involvement, which included footdrop and sensory loss in the distribution of the sciatic nerve. During surgery, the sciatic nerve was found to be stretched and compressed by the giant tumor. The resultant symptoms probably caused the patient to seek medical care. Marginal resection of the mass was performed by careful dissection, and the branches of the sciatic nerve were spared.

To our knowledge, this is the first case report of a histopathologically proven hibernoma that caused sciatic mononeuropathy. Although there are many causes of sciatic mononeuropathy, hibernoma must be considered when the differential diagnosis is made.

Authors' Disclosure Statement

The authors report no actual or potential conflict of interest in relation to this article.

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