Unusual “Dumbbell”-Shaped Hibernoma

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Summary: Hibernoma is considered as a rare soft tissue benign tumor originating from brown fat. Few cases have been described so far in the literature; hibernoma was first described by Merkel in 1906, but it was named by Gery in 1914. We present the case of 33-year-old woman who presented with a palpable lump over superior aspect of her thigh associated with pain. Magnetic resonance imaging examination reported a “dumbbell”-shaped soft tissue tumor involving the right side of the pelvis and adductor compartment passing through the obturator foramen into the adductor compartment infiltrating the hip joint. Clinical and imaging features initially suggested liposarcoma, but ultrasound-guided biopsy and final surgical resection yielded the diagnosis of hibernoma. This rare soft tissue tumor can imitate malignancy, but magnetic resonance imaging and possibly guided biopsy are essential for preoperative planning as surgical resection remains the best management pathway. (Plast Reconstr Surg Glob Open 2019;7:e2142; doi: 10.1097/GOX.0000000000002142; Published online 24 April 2019.)

Hibernoma is considered a rare soft tissue benign tumor originating from brown fat. Few cases have been described so far in the literature. This was first described by Merkel in 1906, but it was named “hibernoma” by Gery in 1914 who noted that the morphologic features of this tumor was similar to the brown fat present in hibernating animals.1,2 There are 4 subtypes of hibernomas: classic, lipoma-like, myxoid, and spindle cell (CD34+).3 They are rare as they represent 2% of lipomas and occur predominantly in adults aged 20–50 years. The most frequent anatomic areas that hibernomas can occur are the scapular area, the neck, the axilla, the thigh, the brain, and the retroperitoneal area.4 The aim of this case report is to highlight the importance of preoperative imaging and possibly histopathological diagnosis to achieve satisfactory surgical resection.

CASE REPORT

Case Presentation

A 33-year-old woman with no significant medical history presented in our department with a noticeable slowly growing lump in her superior right thigh region associated with progressively deteriorating discomfort that forced her to be bedridden. Magnetic resonance imaging (MRI) scan has revealed a “dumbbell”-shaped soft tissue tumor, with the upper limit of the mass being in the right pelvis and lower part passing through the obturator fossa into the adductor compartment in close relation to the adductors, the gracilis, and potentially the gluteal muscles (Fig. 1). Laterally, it was encroaching the right hip joint. The patient was breastfeeding, and therefore, we performed pre-operatively ultrasound-guided biopsy and not computed tomography (CT)-guided biopsy, which was considered a contraindication. The core biopsy has shown no atypical spindle, and fluorescence in situ hybridization analysis for MDM2 gene amplification has been performed and showed no evidence of MDM2 copy number change. The appearances in the biopsy material were in keeping with a hibernoma.

After multidisciplinary team discussion, surgical resection of the mass was planned. A midline subumbilical “S”-shaped incision has been performed extending around the mons pubis and reaching the upper right thigh medially. A retroperitoneal dissection to identify the mass has been performed, and multiple branches arising from the iliac vessels have been ligated carefully. The obturator nerve and the inguinal ligament have been preserved. To access the mass into the femoral space, longitudinal split of the pectineus and adductor longus muscles has been performed followed by the blunt dissection of the tumor capsule down to the acetabulum. The tumor was bisected above the obturator foramen to allow the removal of pelvic and thigh portions separately. One drain has been inserted, and closure in layers of the wound has been performed. Patient had excellent recovery without immediate postoperative complications.

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The preoperative diagnosis has been confirmed with the surgical resection of the mass. Two specimens have been sent for histological examination due to the local-ization and the “dumbbell” shape of the mass, and both specimens presented similar histological features. The histological examination showed 2 firm fatty tissue masses partially covered by a thin capsule measuring 110 mm × 80 mm × 50 mm and 65 mm × 55 mm × 45 mm with a homogenous yellow cut surface. Sheets of adipocytes with the abundant microvacuolated cytoplasm and small, inconspicuous nuclei have been shown, with no atypical spindle cells present, and all features were similar to the preoperative core biopsy and were consistent with the hibernoma.

DISCUSSION

Hibernomas originate from the persistent fetal brown fat tissue, which can be noticed in humans during gestation and specifically after the 21st week of gestation. This brown fat is gradually replaced with white fat as maturation occurs; however, it does persist in humans in specific areas of the body, particularly the neck, axilla, mediastinum, and periaortic and perineal area.

Clinically, a majority of patients with hibernoma are usually asymptomatic and present with slow growing mass as incidental finding. In our case, the patient presented with slow growing tumor compressing the adjacent structures in the pelvis and upper thigh and causing debilitating pain.

Preoperative radiologic imaging with CT and MRI scans is essential. Usually CT scan shows variably homogeneous mass whose density is between the subcutaneous fat and the skeletal muscle. On MRI scan, hibernomas show slightly higher signal intensity on T1- and T2- weighed image with some loss of signal on fat suppression images and remain more vascularized with noticeable presence of large septa. In our case, an initial ultrasound demonstrated the adductor muscular group to be displaced but not infiltrated by the echogenic fatty character tissue posteri-orly, and it was reported as benign fatty tumor in keeping with hibernoma and not sarcoma. The MRI scan reported a large fat signal dumbbell-shaped mass partly located in the pelvis and partly located in the proximal thigh. The waist of the dumbbell shape was passing through the obturator foramen. The intrapelvic part was medial to the iliopsoas muscle and displaced the intrapelvic contents medially. Distally the mass was passing through the obturator foramen invading the adductor compartment.

Preoperative imaging and histopathological diagnosis are important to implement a well-planned approach in difficult anatomic areas such as the pelvic region. Core biopsy and fine-needle aspiration confer a hemorrhage risk due to their vascularity, but on the other hand, it is important to recognize hibernomas preoperatively with histopathology and imaging investigations to achieve a complete surgical resection and a decrease in chances of recurrence.

In our case, preoperative core biopsies have been performed that showed cores of adipose tissue invested by narrow fibrous strands with many of adipocytes presenting abundant microvacuolated cytoplasm. There were no genuine lipoblast-type cells identified, and adipocytic nuclear atypia was not seen. Fluorescence in situ hybridization analysis for MDM2 gene amplification has been performed and showed no evidence of MDM2 copy number change. The overall findings did not support a diagnosis of liposarcoma. The final histological examination showed fatty tissue containing adipocytes with abundant microvacuolated cytoplasm and small, inconspicuous nuclei with no atypical spindle cells present, and all features were consistent with a hibernoma.

Generally, hibernomas are most commonly asymptomatic as they do not infiltrate surrounding structures, but if they grow to large sizes, like in our case, they can become symptomatic. Ideally they should be treated surgically, and it is well known that incomplete resection can lead to local recurrence of the tumour. The overall prognosis is excellent, and there is no known reported case with malignant transformation.

In our case, the “dumbbell” shape of the mass passing through the obturator canal did not allow us to resect the mass in 1 specimen. The pelvic and the thigh portions of the mass have been separated over the obturator canal after a subumbilical incision, which was extending down to the medial aspect of the thigh. The mass was encapsulated and adhered to the surrounding muscles with some level of the intramuscular growth. It is very common for hibernomas to occur predominantly in subcutaneous location but sometimes they can be found adherent to deep soft tissues in combination with intramuscular growth.

This report demonstrates an atypical localization and shape of this rare benign tumor. We believe that preoperative diagnosis is essential as many times it is a clinical challenge for surgeons, pathologists, and radiologists. The most effective treatment of choice whenever hibernomas become symptomatic is complete excision of the tumor.
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