Case Report

The lipoma-like hibernoma: A case report of a rare entity

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ABSTRACT

Hibernoma is a rare tumor developing from fat cells. It is a slowly evolving benign tumor that is rarely pain-inducing. The most frequently encountered histological form is the typical hibernoma. The main differential diagnosis is liposarcoma. Here we present a case of a lipoma-like tumor of the arm: a rare variant of hibernoma. A 45-year-old man presents with a swelling of the left arm evolving for one year. Physical examination revealed a mobile, firm, and well-defined mass of the lateral left upper arm measuring 5 cm in length with no cutaneous lesions overlying. MRI and ultrasound confirmed the presence of a highly vascularized mass suggestive of a liposarcoma. A biopsy of the mass was performed concluding to a lipoma with no evident signs of malignancy. The patient underwent a surgical resection of the mass. Histopathological examination showed a well-differentiated adipose proliferation arranged in diffuse patterns of mature adipocytes. Large hibernoma-like foci were also noted. The diagnosis of a lipoma-like hibernoma was confirmed. Hibernoma represents an uncommon benign tumor. It usually occurs in areas where the brown fat persists, including the thighs, shoulders, back, and neck in decreasing frequency. Commonly, this tumor occurs between the second and third decades of life. Clinically, it presents as a slow growing, painless mass. It may occasionally be painful due to compression of the surrounding structures. MRI shows T1w and T2w hyperintensity, with contrast enhancement after gadolinium injection. On histopathological examination, the structure is distinguished by an association of mature cells, round cells with central nuclei and eosinophilic cytoplasm, and multivacuolated cells. Surgical excision is the optimal treatment. The differential diagnosis concerns lipomas and well-differentiated liposarcomas. Lipoma-like hibernoma is an uncommon benign tumor which might imitate a liposarcoma clinically and radiologically. Histopathological examination is necessary to establish the diagnosis.

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Introduction

Hibernoma is an uncommon benign tumor developing from the residual brown fat cells [1]. It usually affects middle-aged adults with a predilection for subcutaneous tissue of the thigh, upper trunk, and neck. It can have several histological forms, including the lipoma-like variant which remains very rare [2]. The main differential diagnosis of this variant is well-differentiated liposarcoma. Here, we report a case of a lipoma-like hibernoma of the left arm. This case report has been reported in line with the SCARE Criteria [3].

Case presentation

A 45-year-old Tunisian man presented with an isolated swelling of the left arm, evolving for 1 year and increasing progressively in pain and size without other signs of infection, fever, or weight loss. The patient had neither medical/surgical history nor allergies. There was no mention of recent trauma or injuries. Physical examination revealed a 5 cm mass of the lateral side of the left upper arm. The mass was firm, well-defined, painful, and movable over the deeper tissues (Fig. 1). No overlying cutaneous lesions or ulceration were noticed. Ultrasound of the left arm showed a fatty tissue tumor with clear margins to the surrounding muscle. MRI confirmed the presence of a highly vascularized mass of the left arm, measuring 5 cm in length, with clear margins to the surrounding muscle. Signals in T1w and T2w were hyperintense with contrast enhancement upon injection of gadolinium, suggestive of a well-differentiated liposarcoma (Figs. 2, 3 and 4).

After a multidisciplinary reunion, a primary incisional biopsy was performed to rule out malignancy. It concluded to a lipoma with no signs of malignancy. The patient underwent then a marginal resection of the tumor. The intervention was performed by a senior orthopedic surgeon in an orthopedic surgery department of a university hospital in Tunisia. Macroscopically, the mass measured $9 \times 6 \times 2.5$ cm. It had a fatty appearance, yellowish color, was encapsulated with soft consistency (Fig. 5). Histopathological examination revealed a well-differentiated fat cell proliferation arranged in diffuse patterns of mature adipocytes.

These adipocytes had multivacuolated cytoplasm with a central or eccentric nonatypical, recessed nucleus. This aspect supports the diagnosis of lipoma-like hibernoma. The resection margins were free from tumoral proliferation. After 3 years of follow-up, the patient did not present any sign of local recurrence nor sensory-motor disorders.

Fig. 1 – Mass of the lateral side of the left upper arm.

Fig. 2 – Axial T1 fat-suppressed post-gadolinium MRI shows hypointense signal of the mass measuring 5.8 centimeter.

Fig. 3 – Axial T1 MRI shows hyper intense mass measuring 5.8 centimeter.
Discussion

First described by Merkel in 1906 as a pseudo lipoma, the term hibernoma was introduced in 1914 by Gery when he noticed the similarities between the glands of hibernating animals and this tumor. It represents about 1.6% of all benign tumors [1]. In adults, brown fat represents only less than 1% of the body mass and is distributed mainly in the inter scapular region and mediastinum. Hibernoma usually occurs in areas where this type of fat persists, including the thighs (30%), shoulders, back, neck, thorax, arms, and abdomen, in decreasing frequency. However, unusual locations have been described such as the retroperitoneum, the pericardium, the spermatic cord, the extremities, and the scalp [4–8]. Commonly, this tumor occurs between the second and third decades of life with a slight female predominance [7–10].

There are no known risk factors for hibernoma. Typically, the patient presents with a slow growing, movable, firm, painless mass. It may occasionally be painful due to compression of the surrounding structures. Some authors have reported a loss of weight due to the thermogenicity of the malignant tissue, responsible for heat production in hibernating animals [6,10]. Ultrasound, the first line test, usually shows a homogeneous tumor, highly vascularized on Doppler with a fatty density on CT scan. MRI shows T1w and T2w hyperintensity, with contrast enhancement after gadolinium injection [11,12].

Cytogenetically, hibernoma is characterized by chromosome 11q13 abnormalities that probably correspond to a tumor suppressor gene. This chromosomal region is located in the same area as the multiple endocrine neoplasia type1 (MEN1) gene, but the association “MEN-hibernoma” could not yet be proved [2,4,9,13]. Macroscopically, hibernoma appears as a 5-10 cm sized, encapsulated, firm, brownish-yellow tumor that rarely infiltrates surrounding tissues. On histopathological examination, the structure is distinguished by an association of mature cells, round cells with central nuclei and eosinophilic cytoplasm, and multivacuolated cells. The tumor proliferation is architecturally patterned by richly vascularized connective tissue. According to stromal characteristics, 4 different histological subtypes of hibernoma can be distinguished: the most common variant is typical hibernoma, representing 82% of the cases. The lipoma-like (7%), myxoid (9%), and spindle-cell (2%) variants are more uncommon [1,7]. Despite the disputed degenerative potential, surgical excision remains the optimal treatment [2,5,8,14]. The resection margins must be wide to avoid any risk of local recurrence, which was achieved with our patient.

The differential diagnosis concerns lipomas and well-differentiated liposarcomas and only the histological analysis of the resected piece will allow the diagnosis to be made [15].

Conclusion

Hibernoma is a rare tumor of the brown fat tissue with a good prognosis. It should be considered in front of any fatty mass with a slow progression and a large blood supply on imaging
but has to be included in differential diagnosis to prevent false treatment.

Provenance and peer review

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Author contribution

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

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