Rare disease

Hibernoma of the thigh: a lipoma-like variant rare tumour mimicking soft tissue sarcoma

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Summary

A 43-year-old Caucasian woman appeared with painless progressively enlarging tumour of the anterior part of the left thigh of approximately 6 months' duration. A differential imaging diagnosis of liposarcoma or large lipoma was made. An excisional biopsy was performed with complete removal of a large lobulated and encapsulated grossly ovoid mass. Frozen section diagnosis was unidentified lipomatous tumour with no evidence of malignancy. The final pathological diagnosis was lipoma-like hibernoma. The patient's postoperative course was uneventful and discharged on the seventh postoperative day with full function of the involved leg. Hibernoma is benign tumour and it is not known to recur after complete local excision which is the treatment of choice.

BACKGROUND

Hibernomas are rare benign lipomatous tumours originating from residual brown fat cells. The term hibernoma was coined by Gery in 1914 to describe a specific type of fatty tumour which bore a strong resemblance to the hibernating glands of certain mammals.^{1–3} They usually affect adults, with a slight male predominance and a peak of incidence between the third and fourth decades of life and usually grow in the vestiges, where brown fat is found in fetuses and infants such as, neck, shoulder, axilla, mediastinum, upper thorax and retroperitoneum.^{4–7} Other uncommon locations include the abdomen, thigh, buttock, popliteal fossa and intracranial sites.⁶ The hibernoma is classified into six histological subtypes.⁸ The lipoma like variant easily can be misdiagnosed as atypical lipoma or well-differentiated liposarcoma.⁴

CASE PRESENTATION

A 43-year-old Caucasian woman who was examined every 6 months in follow-up breast clinics for operated invasive ductal carcinoma stage IIA (T₂, N₀, M₀), appeared with a painless progressively enlarging mass of the anterior-lateral part of the left thigh of approximately 6 months' duration. She denied any injury to the affected thigh, and had no pain or numbness in lower extremity. Physical examination revealed that the presence of an oval, diffuse swelling of the anterior-lateral part of the left thigh. The size of the mass was approximately 16×9 cm and its consistency was that of a firm soft tissue mass which was unmovable and non-tender. There was no discolouration of the skin, no increased temperature, no bruit and the mass did not transilluminate or pulsate. The range of the motion of all joints of both extremities was normal and there was no motor, sensory, reflex or circulatory deficit in either leg. No regional lymphadenopathy was present. Family history was unremarkable.

INVESTIGATIONS

An MRI was performed and revealed a 16×9 cm lobulated and encapsulated lipomatous tumour of the anterior compartment of the left thigh (figure 1).

DIFFERENTIAL DIAGNOSIS

Various differential considerations based on MRI can be suggested when a complex lipomatous mass is encountered, including benign entities like lipoma, angiolipoma and haemangioma as well as malignant tumours like liposarcoma. Lipoma present as homogenous fatty mass with a few scattered internal septa and no signs of enhancement.⁹ Angiolipoma and haemangioma can be distinguished in terms of different morphology of internal vasculature.^{10–12} Some studies stress the significance of large branching intratumoral vessels with early contrast enhancement and arterio-venous shunting in the differential diagnosis of hibernomas.^{6 11 12} However, these features are not always present, although fine enhancing strands may be seen.⁹ Well-differentiated liposarcoma is characterised by the presence of irregularly thick (>2 mm) and/or nodular septa, foci of high T2 and prominent areas of enhancement.^{9 13} Moreover, the fatty component of the well-differentiated liposarcoma appears isointense to subcutaneous fat, on T1-weighted spin-echo; distinguished them from hibernomas.⁹ It is reported that hibernomas similarly to malignancy has a high FDG (fluoro-2-deoxy-D-glucose) standard uptake value, for this reason it is important to consider hibernoma in the differential diagnosis of soft tissue lipomatous neoplasms, because they are clinically benign.¹⁴

TREATMENT

The patient underwent surgery with clinical diagnostic hypotheses of well-differentiated liposarcoma or lipoma. Complete excision of a large lobulated and encapsulated grossly ovoid mass measuring 16×9×4.5 cm was

Figure 1 MRI of lobulated and encapsulated lipomatous mass of the anterior compartment of the left thigh.

performed. Histopathological report confirmed lipoma-like hibernoma (figure 2).

OUTCOME AND FOLLOW-UP

The patient is in follow-up and doing well, and her follow-up examinations for the breast cancer are normal so far.

DISCUSSION

Brown adipose tissue in hibernating animals was first described by Welch in 1670.¹⁵ However, brown fat is also found in more than 50 non-hibernating animal species, as well as in man.³ It is abundant in newborn humans and gradually decreases in quantity during life compromising less than 0.1% of total body weight by the age of

Figure 2 H&E×200. Multivacuolated brown fat cells with pale eosinophilic cytoplasm.

70 years.^{3 6 12} Unlike white adipose, the function of brown adipose is thermogenesis.⁴

Hibernomas are benign tumours that develop in any location where brown fat remains.^{9 11} They usually measure from 5 to 10 cm in diameter, but may reach up to 20 cm.^{6 11} Despite the initial belief that these neoplasms would be restricted to the areas of residual brown fat⁵ such as neck, axilla, back and mediastinum, the thigh was the most frequently affected body site in the largest series published so far.⁴

The first case report of this neoplasm appeared in the world literature in 1906.¹⁶ The term hibernoma was introduced by Gery Louis in 1914 to describe a specific type of fatty tumour which bore a strong resemblance to the hibernating glands of certain mammals.¹⁻³ Less than 200 cases have been reported in the literature.^{4 17} In 2001, Furlong MA *et al*⁴ reported the prototypic histological features of hibernomas, defining the classic pattern, as well as the three most common histological variants: myxoid, lipoma-like and spindle cell variants. According to the 2002 WHO classification there are six histological subtypes of hibernomas.¹⁸ According to Furlong MA *et al*⁴, the lipoma-like variant is the most troublesome for the surgical pathologist due to the presence of occasional multivacuolated adipocytic cells mimicking lipoblasts. In their series, the referral pathologist entertained a diagnosis of atypical lipoma or well-differentiated liposarcoma in 23% of the cases.⁴ Only 12 cases of the lipoma-like lipoma were included in this series, accounting for 7% of the cases.⁴

Until today, in the English literature has not been reported a malignant form of hibernoma, except for the case published as an abstract that involved a sarcoma with hibernoma-like features.¹⁹

Incomplete excision results in local recurrence of the tumour; therefore complete resection is considered as the treatment of choice for these neoplasms.^{4 5 8}

Learning point

- We would like to stress that a correct diagnosis of Lipoma-like hibernoma must be achieved in order to avoid an overtreatment, because while atypical lipomatous tumours frequently recur and might dedifferentiate to dedifferentiated liposarcoma,^{5 8} hibernomas are benign tumours that do not recur with complete local excision.^{4 5 8}

Competing interests None.

Patient consent Obtained.

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