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Case Report

Pathology Section

Intramuscular Hibernoma: A Rare Tumour in Buttock

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ABSTRACT

Hibernomas are benign tumours of brown fat that does not recur after complete excision. These tumours are found most often in adults and most commonly in thigh. Four morphologic variants of hibernoma are identified: typical, myxoid, spindle cell, and lipoma-like. The most common histologic type is typical variant. In this report, we present the clinical, morphological features and discuss the differential diagnosis of a typical variant of intramuscular hibernoma.

Keywords: Brown fat cells, Lipoblasts

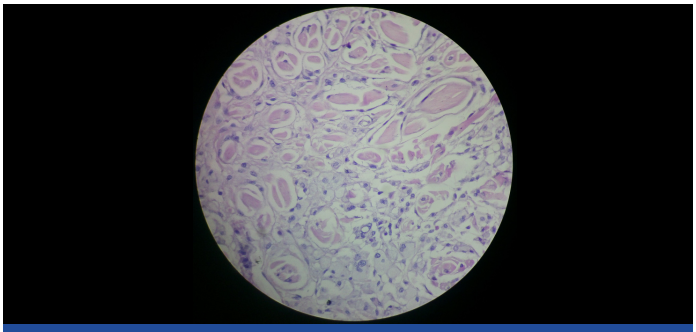
CASE REPORT

A 35-year-old otherwise healthy man presented to the surgical OPD on month of December 2014 with complaint of slow growing painless mass in right buttock since eight years. Clinical examination revealed a mobile tumour, soft to firm in consistency and non tender. No warmth, erythema and lymphadenopathy found. A provisional diagnosis of fibrolipoma was done and complete excision of the tumour was done.

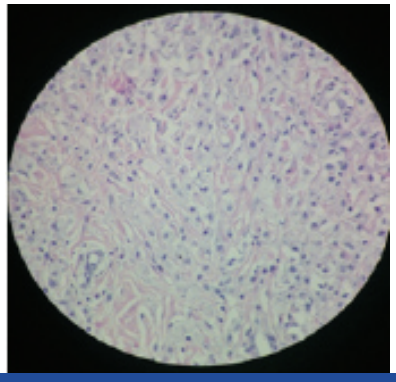
Grossly the resected specimen consisted of a skin covered mass measuring (3.5x3x2.5) cm. On cut section tumour was well circumscribed, homogeneous, grey tan and soft to firm in consistency [Table/Fig-1]. No areas of haemorrhage and necrosis seen.

Histopathological examination revealed an unencapsulated tumour composed of bland looking tumour cells with variable morphology irregularly infiltrating skeletal muscle bundles [Table/Fig-2]. The tumour cells showed numerous small cytoplasmic vacuoles and pale to intensely eosinophilic granular cytoplasm. The characteristic hibernoma cells were predominantly seen, which are large multivacuolated fat cells with indented small central vesicular nuclei, single round central nucleolus and evenly dispersed chromatin [Table/Fig-3]. Minimal focal nuclear pleomorphism was seen. But mitotic figures were not conspicuous. Abundant vascularity was also noted. Finally a histologic diagnosis of typical variant of intramuscular hibernoma was made.

[Table/Fig-1]: Gross photograph showing skin covered mass with homogeneous gray tan cut surface



[Table/Fig-2]: Tumour cells irregularly infiltrating skeletal muscle (H&EX40X)



[Table/Fig-3]: Bland tumour cells with multivacuolated, pale to granular cytoplasm and small central Nuclei (H&E X40X)

DISCUSSION

Hibernoma is a rare benign tumour composed of multivacuolated brown fat cells admixed with white adipose tissue [1]. It most commonly occurs in thigh followed by trunk, chest, head & neck, upper extremity and a few cases have been reported in the buttock [2]. Beals et al., and Mavrogenis et al., reported 19 & 17 cases of hibernoma during a period of 20 & 23 years respectively [3,4]. Furlong et al., found eight intramuscular tumours out of 170 hibernomas studied [5]. Recent WHO classification classifies hibernoma as benign adipocytic tumour and intramuscular tumours constitute 20%. A case of intramuscular hibernoma in buttock region is described.

Hibernoma represents an unusual benign lesion occurring predominantly in subcutaneous location, but sometimes in intramuscular or in deep soft tissues and the tumour cells show differentiation towards brown fat [1]. The entity was first reported in 1906 by merkel [6]. However, Gery was the 1st to coin the term hibernoma, in 1914 because of its resemblance to the brown fat in hibernating animals [7]. It accounts for 1.6% of benign lipomatous tumours and approximately 1.1% of all adipocytic tumours. It is more common in the third and fourth decades of life, only 5% occur in children and teenagers and 7% occur in patients aged >60 years. Clinically it grows slowly and usually presents with painless swelling. Symptoms related to the compression of adjacent structures rarely develop [1]. This case presented in fourth decade with a history of painless, slow growing mass for eight years.

Grossly hibernomas are usually well defined, soft, greasy to rubbery, and lobulated. The cut surface varies from yellow to red-brown

depending on the amount of intracellular lipid and is occasionally mucoid with rare areas of haemorrhage. Hibernomas ranging from 1 to 24 cm size have been reported. But recently described hibernomas are usually smaller with a greesy, soft and spongy cut surface [1,4]. Light microscopy typically shows the "hibernoma cells": large multivacuolated fat cells with central nuclei admixed with other cells with pale or granular cytoplasm. Cytoplasmic vacuoles stain for neutral fat. Some tumour cells resemble mature adipocytes and some resemble lipoblasts. Abundant vascularity is characteristic and atypia is rare. Rare nuclear atypia, infiltrative pattern & intramuscular location are not necessarily criteria for malignancy [8,9].

All hibernomas are composed partly or principally of multivacuolated hibernoma cells. Four histologic variants are recognized; 1) Typical variant (82%): Composed of a mixture of eosinophilic cells, hibernoma cells and pale cells (white fat cells); 2) Myxoid variant (9%): Composed of multivacuolated cells with focal eosinophilic cytoplasm separated by a myxoid stroma; 3) Lipoma-like variant (7%): Composed of scattered hibernoma cells among univacuolated mature adipocytes; 4) Spindle cell variant (2%): Composed of the typical hibernoma cells, as well as adipocytes, spindle cells, mast cells, and collagen bundles [4,7]. For all subtypes, the vascular supply is considerably more prominent in hibernoma than in lipomas. Histologically this case was diagnosed as typical variant of intramuscular hibernoma because it was infiltrating skeletal muscle bundles and was composed of a mixture of hibernoma cells, eosinophilic cells and pale cells.

Differential diagnosis of this particular variant are lipoblastoma, atypical lipomatous tumour, well differentiated liposarcoma and hybrid tumours [1,4,5]. Lipoblastoma is distinguished from hibernoma by absence of hibernoma cells and presence of a spectrum of maturation ranging from primitive mesenchymal cells to mature adipocytes to multivacuolated or signet ring lipoblasts. Atypical lipomatous tumour & liposarcoma have aggressive cytologic appearance with presence of atypical pleomorphic nuclei in lipoblasts. Hybrid tumours are hibernomas mixed with ordinary lipoma or spindle cell lipoma and such type of combination is not seen in this case.

Although Immunohistochemistry is not necessary to render a diagnosis, these lesions usually stain strongly for S-100 protein. CD34-positive spindle cells are present only in the spindle cell variant. Newer marker includes UCP1 [1]. Pathogenesis of hibernoma is unknown. Certain cytogenetic abnormality like rearrangements of 11q13-21 and deletion of MEN1 gene has been described. Recent evidence suggests loss of MEN1 and AIP tumour suppressor genes, is likely to be pathogenetically associated with hibernoma development [1,8,9]. Ultrastructural features are similar to brown fat and hibernoma cells contain multiple lipid droplets and numerous large mitochondria [1].

All hibernoma variants have good prognosis after complete excision. Metastases or malignant transformation have not been reported. Despite benign, some authors have experienced recurrent hibernomas and cases of significant bleeding. They have postulated that recurrence is related to the resection type and/or the pathologic variant and atypical histologic findings correlate with excessive bleeding during excision [10].

CONCLUSION

Hibernomas are derived from brown fat, in which the clinical and imaging presentation can mimic other neoplastic conditions. Histologically it should be distinguished from other adipocytic tumours including lipoblastomas, atypical lipomatous tumour and liposarcomas.

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