Case Report:

Hibernoma - an unusual presentation

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ABSTRACT

Hibernomas are rare benign tumours of brown adipose tissue. They are usually encountered in young males usually in their third decade of life. A 50-year-old lady presented with a swelling over the right forearm region that was soft, non-tender and was gradually increasing in size over the last one year. The patient underwent surgical excision of the mass which was confirmed to be hibernoma (mixed variant). This case is being reported for documenting the occurrence of hibernoma in a rare location (forearm) and in an older women aged 50 years.

Key words: *Hibernoma, Forearm, Benign tumour*

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INTRODUCTION

Hibernomas are rare benign slow-growing, painless neoplasms composed of brown adipose tissue admixed with variable proportion of white adipose tissue without any tendency for recurrence after complete surgical excision. Hibernoma are mostly documented in case reports and small series.[1-4] In Armed forces Institute of Pathology/ American Registry of Pathology Press (AFIP) series, hibernoma comprised 1.6% of benign lipomatous tumours.[5] They are described mainly in young adults, mostly in the third decade of life, with slight male predominance.[6] The reported age range is from 2 to 72 years.[1,4] Compared to lipoma, which is one of the most common soft-tissue tumours originating from white adipose tissue, hibernoma is listed among the rarest of the adipocytic neoplasms.[3] Brown adipose tissue is generally present in the foetus and is gradually replaced by white adipose tissue with advancing age. In the foetus, brown adipose tissue is noted in various sites such as the interscapular area, posterior abdominal wall, suprailiac and peripancreatic adipose tissue and near autonomic ganglia whereas in adults neck, axilla, mediastinum, periaortic and perirenal zones are the areas where brown fat generally persists.[3,4] So hibernomas are preferentially seen in these sites.

CASE REPORT

A 50-year-old lady presented with a swelling over the right forearm that was non-tender, soft, and gradually increasing in size over the last one year. With the clinical diagnosis of lipoma the mass was excised completely and the excised specimen was subjected for histopathological examination. On gross pathological examination, the specimen was irregular with nodular external surface measuring $8 \times 7 \times 2$ cm. Cut-surface was greasy with yellowish areas (Figure 1). Microscopically the lesion was well encapsulated and revealed predominantly foetal looking adipocytes with vacuolated cytoplasm with little intervening fibrous element, a few congested capillaries mild lymphomononuclear infiltrate without the presence of mitotic figures and necrosis (Figure 2). Since the present case showed an admixture of pale and eosinophilic stained multivacuolated cells, it was diagnosed as hibernoma (mixed variant).

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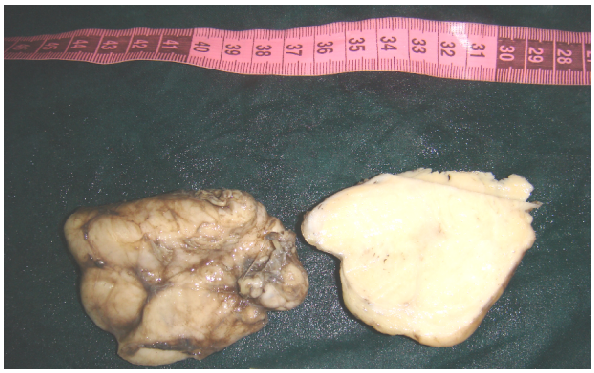

Figure 1: Cut-section of the excised specimen showing lobular, greasy yellowish areas

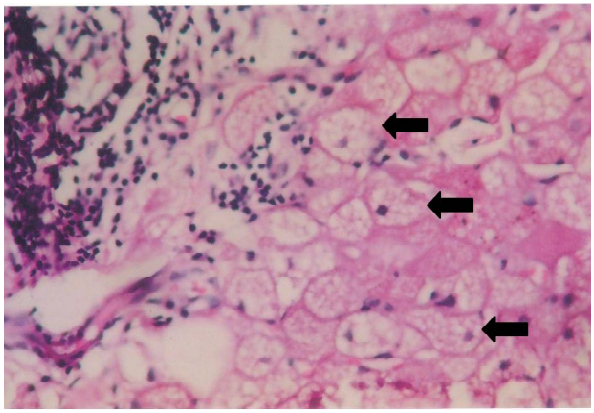

Figure 2: Photomicrograph showing foetal looking adipocytes with vacuolated cytoplasm (arrows) admixed with lymphomononuclear infiltrate (Haematoxylin and eosin, × 400)

DISCUSSION

Merkl first described this unusual tumour in 1906; Gery in 1914 termed it as hibernoma because of its resemblance to the brown adipose tissue of the hibernating glands of animals which helps in thermoregulation.[7] Apart from several mammalian animals, it may also be present in non-hibernating animals, such as mice, rats, monkeys and humans.[4] Clinically, hibernomas typically present as progressive, painless swellings without localized tenderness as was also seen with the present report.[1,3] Symptoms are usually because of mass effect resulting from pressure and displacement.[4] In our case no such effects were observed.

Brown adipose tissue is brown-tan in color and vascular, microscopically comprising of polygonal, multivacuolated cells with granular cytoplasm and ovoid nucleus. The brown colour of hibernoma is said to be due to its vascularity and mitochondria rich eosinophilic granular cytoplasm and is an important source of non-shivering thermogenesis.[4,5] The size of hibernoma is variable ranging from 1 to 24 cm with an average dimension of 9.3 cm. It is usually yellow to brown in color, lobular and well demarcated with soft, greasy cut surface.[1] Histo-pathologically six variants of hibernoma have been described. They are eosinophilic, pale cell, mixed, spindle cell, myxoid and lipoma like.[5] The present case was a mixed variant of hibernoma with both pale and eosinophilic stained cells. Immunohistochemically they stain variably for S-100 protein.[1,4] Increased expression of p53 protein has earlier been reported.[8]

The aetiology of hibernoma is unknown, although many lesions arise at the sites where brown fat is normally found in hibernating animals and human foetuses or newborns.[5] According to one theory proposed, in order to explain the origin of hibernoma, the tumour grows starting from some islands of brown adipose tissue that may persist in the white fat tissue; on the contrary, tumoural brown fat cells may develop from white adipose tissue.[4]

In a large published series[1] the most common locations for hibernoma included the thigh, shoulder, back, neck, chest, arm, and abdominal cavity/retroperitoneum. Though hibernomas are described in the upper extremity in literature, the usual site is upper arm whereas our case presented with a forearm swelling. We found only one such case in literature.[6] Ultrastructural features of hibernoma include investment of each tumour cell by basal lamina, an inverse relationship between lipid droplet size and the number of mitochondria per unit of cytoplasm, pleomorphic mitochondria with dense matrices or large round mitochondria with transverse lamellar cristae, undulating plasmalemmal invaginations, micropinocytotic vesicles, periodic short plasmalemmal densities, and a conspicuous lack of cytoplasmic membrane systems.[9] Though cytogenetic analyses of hibernomas

have consistently revealed rearrangements of chromosome bands 11q13-21,[9] these findings have also been reported in lipomas and liposarcomas.[10]

The differential diagnoses of hibernoma may include lipoblastoma and atypical lipomas.[1,11] Lipoblastomas are seen in children , shows presence of lipoblasts and 18q11-13 abnormalities.[11] The atypical lipomatous tumours are mostly superficial, have atypical nuclei and are positive for murine double minute2 (MDM2), Cyclin dependant kinase 4 (CDK4) and p16. Myxoid liposarcomas can be considered as differential diagnosis of hibernoma with myxoid stroma. However hibernomas lack the typical chicken wire vascular pattern of myxoid liposarcomas and frequently exhibit 11q13 rearrangement.[1]

Hibernomas are essentially benign tumours where complete surgical excision results in good prognosis. Since the vascular supply in hibernomas is more prominent compared to lipomas, it should be treated with care to avoid postoperative bleeding or haematoma formation. Follow-up data in a large series did not reveal any local recurrences or evidence of aggressive behaviour.[1,4]

This case report documents another unusual presentation with respect to age and location of a rare benign tumour.

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