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CASE REPORT

Interscapular hibernoma—Case report and literature review

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Abstract Hibernoma is a rare benign tumor that arises from the vestiges of fetal brown fat. We present a case of interscapular hibernoma. Computed tomography scan showed a well-circumscribed, hypodense mass with peripheral enhancement, and magnetic resonance imaging revealed intermediate high T1 and T2 signal intensities with incomplete fat suppression. Although it is rare, hibernoma should be included in the differential diagnosis of lipomatous soft-tissue tumors. This is a benign tumor with no malignant potential. Complete excision is the treatment choice.

Introduction

Hibernoma is a very rare benign tumor that arises from the vestigial remnants of fetal brown adipose cells and usually manifests as a slowly growing, painless, soft-tissue mass [1]. It mainly occurs in adults, slightly more in women, and is commonly seen in the subcutaneous regions of the back, especially the periscapular and interscapular regions, neck, axilla, shoulder, thorax, thighs, and retroperitoneum [2–5].

A hibernoma might be confused with a lipoma clinically and could not be completely distinguished from hypervascular lesions, such as liposarcoma, by radiographic images. Using a case report, we will discuss the diagnostic features in this case, followed by a literature review.

Case presentation

A 39-year-old man was found to have a mildly tender, slow-growing mass over his interscapular back region for more than 1 year. Palpation of the back region revealed an approximately 8-cm × 7-cm immobile soft mass. Computed tomography (CT) scan (Fig. 1) showed a 2-cm × 7-cm × 8-cm well-defined lobulated mass in the interscapular subcutaneous area, slightly toward the left and separated from the

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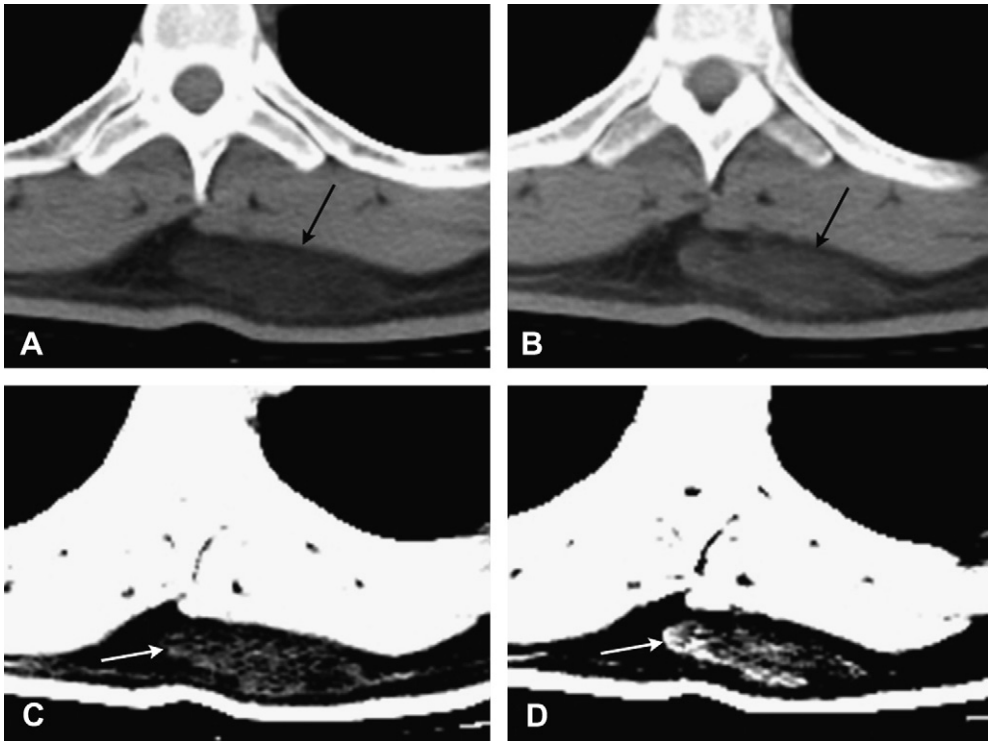

Figure 1. Axial chest CT scan with a focus on left side back before (A) and after (B) contrast administration. One well-defined mass (black arrows) located in the interscapular subcutaneous area with separation from the trapezius muscle. The mass showed an intermediate attenuation between fat and muscle attenuation and asymmetric marginal enhancement, which can be recognized more easily (white arrows) while comparing images before (C) and after (D) contrast administration by narrowing the window width of CT number. CT = computed tomography.

trapezius muscle. It presented with intermediate attenuation (−23 Hounsfield units) between fat and muscle attenuation and asymmetric marginal enhancement after contrast administration.

Magnetic resonance (MR) imaging (Fig. 2) showed a well-circumscribed septated mass with intermediate T1 and bright T2 signal intensity. On the T2 fat saturation and short TI inversion recovery images, the mass demonstrated uneven signal loss with an irregular peripheral high signal change. There was a clear fatty strap between the lesion and the underlying musculature. This patient refused to receive contrast medium administration; thus, no post-contrast-enhanced images were obtained. Because of the heterogenous enhancement of the lesion, a malignant tumor could not be excluded. An elective surgery was performed. The mass was completely excised.

Pathology

Gross examination of the tumor showed a tan to light brown color separated by fine fibrous trabeculae (Fig. 3) with lobulated, well-demarcated margins, and measured about 5 cm × 4 cm × 3 cm in size. On microscopic examination, this tumor was composed of a mixture of brown and white adipose cells (Fig. 4). The brown adipose cells were characterized by granular multivacuolated, eosinophilic, round to oval cells with centrally placed nuclei and prominent nucleoli and multiple small lipid droplets. The white adipose cells varied in size and shapes and were characterized by clear cytoplasm with peripherally located nuclei. Gradual transition between two types of cells was noted. Atypia of nuclei or mitotic activity was not seen. There were small vessels in these tissues. The picture favored the following diagnoses: hibernoma, metastatic granular renal cell carcinoma, granular cell tumor, and alveolar soft-part tumor. For differential diagnosis, S-100 protein staining (Fig. 5A) was positive, which ruled out metastatic granular renal cell carcinoma. Because periodic acid-Schiff staining (Fig. 5B) was weakly positive and had no organoid pattern of the tumor, granular cell tumor and alveolar soft-part tumor were excluded. The diagnosis of hibernoma was made.

Discussion

Hibernoma is a benign tumor of brown fat that is also known as fetal lipoma, lipoma of embryonic fat, and lipoma of immature adipose tissue [1]. Brown fat and white fat are two types of adipose tissues found in mammals. Brown fat is especially abundant in newborns and in hibernating mammals and is thought to play a role in thermoregulation. In contrast to white adipocytes, which contain a single lipid droplet, brown adipocytes contain numerous smaller droplets and a much higher number of mitochondria that contain iron and make the tissue brown in color [6]. Because of a greater need for oxygen than that for most tissues, brown fat also contains more capillaries than white fat. In neonates, brown fat makes up about 5% of the body

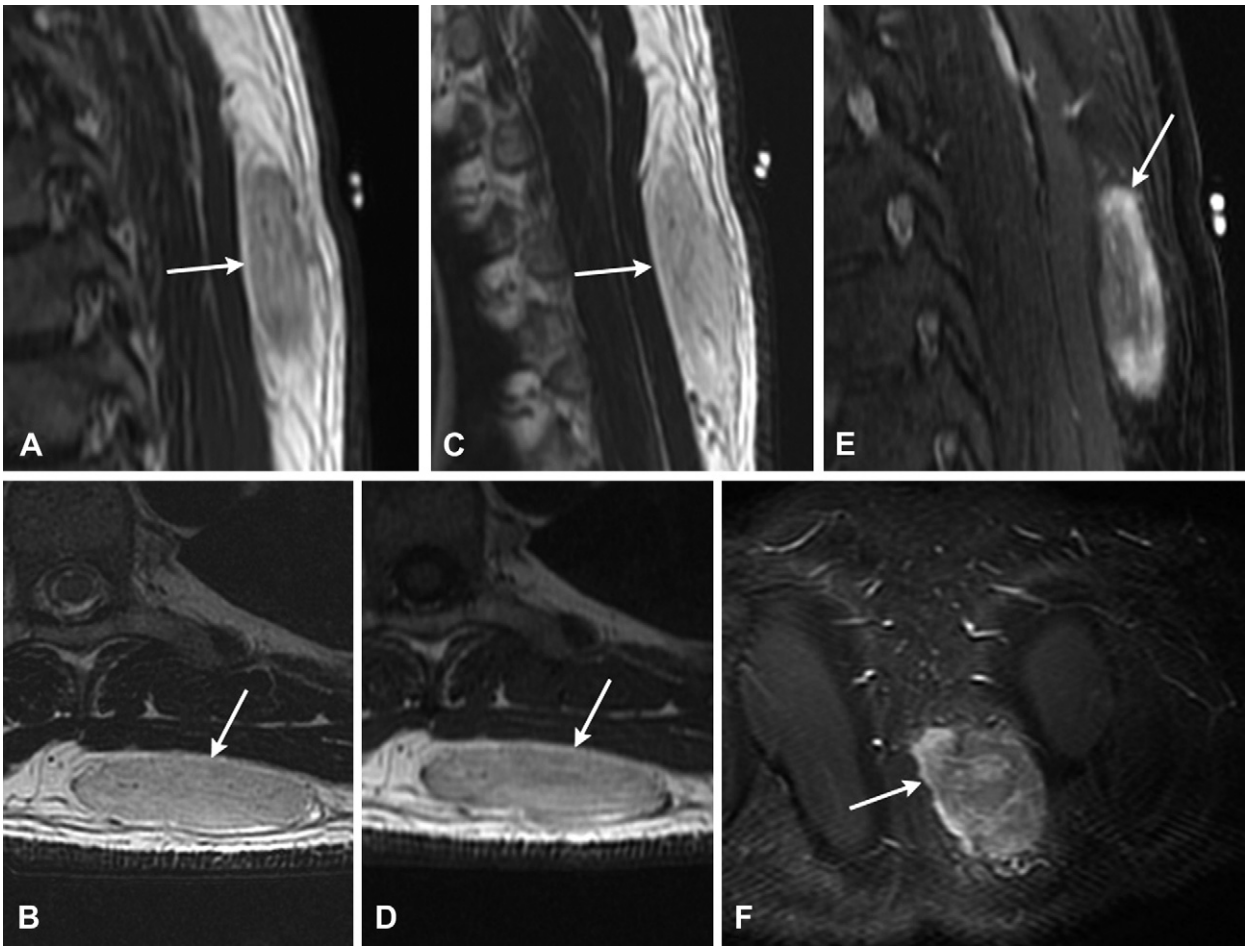

Figure 2. On magnetic resonance imaging, there was one well-circumscribed septate mass (arrows) with intermediate signal intensity on the sagittal (A) and axial (B) T2-weighted images and the sagittal (C) and axial (D) T1-weighted images. On T2 fat saturation (E) and short TI inversion recovery (F) images, the mass demonstrated uneven signal loss with an irregular peripheral high signal change. There was a clear fatty strap between the lesion and the underlying musculature.

Figure 3. The tumor, 5 cm × 4 cm × 3 cm in size, showed tan to light brown fat separated by fine fibrous trabeculae.

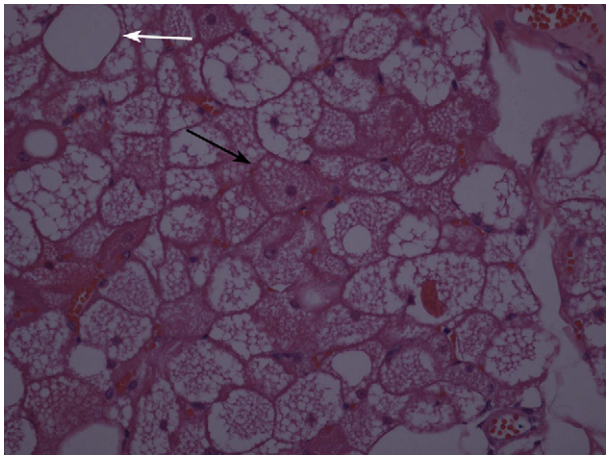

Figure 4. Mixture of brown and white adipose cells in hematoxylin and eosin stain, 20×. The brown adipose cells (black arrows) were granular, multivacuolated, eosinophilic, round to oval cells with centrally placed nuclei and prominent nucleoli and multiple small lipid droplets. The white adipose cells (white arrows) were clear cytoplasm with peripherally located nuclei.

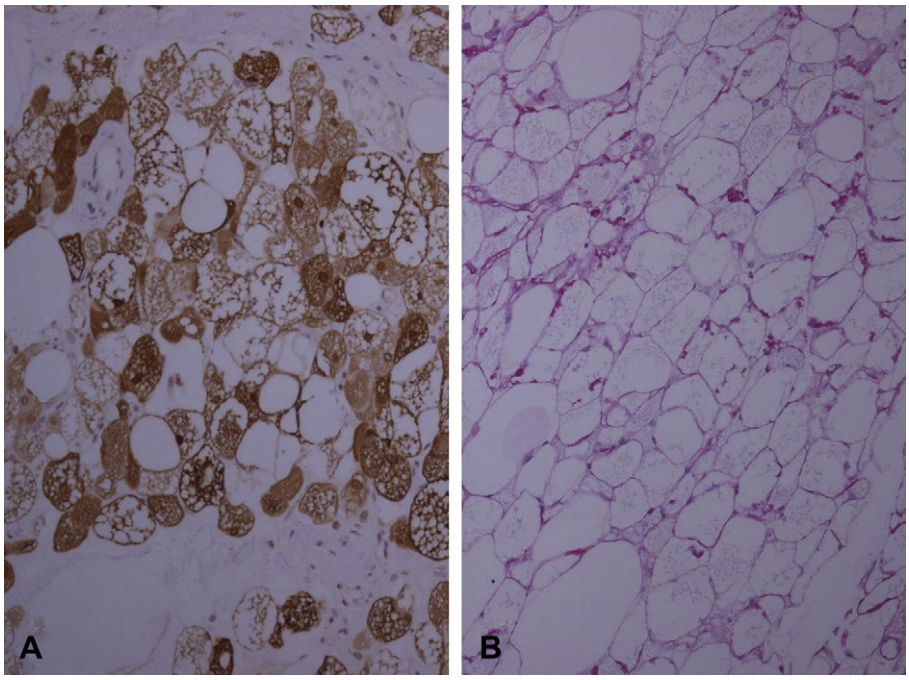

Figure 5. S-100 protein stain, 10×, (A) was positive, which ruled out metastatic granular renal cell carcinoma. Because periodic acid-Schiff stain, 10×; (B) was weakly positive and had no organoid pattern of the tumor, granular cell tumor and alveolar soft part tumor were excluded.

mass and is located on the back, along the upper half of the spine and toward the shoulders. The amount of brown fat decreases during childhood. In adults, it accounts for only 1% of adipose tissue [7].

The presence of brown fat in humans was first described by Hatai [8] in 1902, as an "interscapular gland" in embryos. Merkel [9] first described a tumor composed of brown fat tissue in 1906. In 1914, Gery [10] proposed the name "hibernoma" because of its resemblance to the brown fat that is prominent in hibernating mammals. Hibernomas usually arise from areas in which vestiges of brown fetal fat persist beyond fetal life. They include the periscapular/interscapular region, neck, axilla, shoulder, and retroperitoneum [2]. Recently, the thigh has been described as a very common location [3].

Hibernoma usually presents as a firm, movable, slow-growing, and asymptomatic soft-tissue mass when located in the subcutaneous tissue. Physical examination typically results in a mobile and pliable mass, often suggestive of a lipoma. Because of hypervascularity, sometimes, the mass area may feel warm. The symptoms caused by hibernoma are the result a mass effect on adjacent structures. In a recent report of 170 cases [3], 61% of the patients had hibernomas in the third and fourth decades of life. It was slightly more in males (58% of the cases) than in females, although a slight female predominance has been described in the literature [4].

The tumors are well circumscribed, partially encapsulated, and lobulated. The cut surface varies from yellow to brown and is occasionally mucoid with rare areas of hemorrhage [3]. The diameter usually ranges from 5 cm to 10 cm, but it may reach up to 20 cm [4]. Microscopically, the presence of hibernoma cells, multivacuolated fat cells with small, central nuclei, was common to all tumors. Four histological variants have been identified based on the tinctorial quality of hibernoma cells, the nature of the stroma, and the presence of a spindle cell component: typical (82% of cases), myxoid (8%), lipoma-like (7%), and spindle cell (2%) [3]. The color of hibernoma results from a combination of hypervascularity with abundant mitochondria.

Conventional radiography often demonstrates a radiolucent mass without adjacent osseous involvement. Sonography may show a well-circumscribed hyperechoic mass with increased vascularity. As it is derived from brown fatty tissue and depends on the histological variants, hibernoma may have similar but not identical imaging characteristics to fatty tissue on CT scan and MR imaging. The lesion demonstrates as a well-demarcated, intermuscular, intramuscular, subcutaneous, or retroperitoneal mass with low density on enhanced CT scan and intermediate high signal on T1- and T2-weighted images, imaging characteristics between subcutaneous fat and adjacent musculature [4]. Because of the nature and amount of lipids [11], there may be incomplete fat suppression on fat suppression MR pulse sequences, as shown in our case. The vascular structure in hibernoma has received attention in recent reports. In addition to heterogenous enhancement after contrast administration, large feeding surface vessels have been found on Doppler ultrasound [12], and increased T1 signal intensity and large intratumor vessels indicating a diagnosis of hibernoma have been suggested in some reports [12—14]. By using recent advances in MR time-resolved contrast imaging, these large vessels clearly demonstrated arteriovenous shunting [14]. The differential diagnoses of a high T1 signal mass include angiolipoma, lipoma, hemangioma, liposarcoma, alveolar soft-part sarcoma, and clear cell sarcoma; however, none of them contains large intratumoral vessels typically seen in hibernomas [13].

Increased vascularity of a lesion could raise the suspicion of malignancy. Such large intratumoral vessels, however, could be an important feature for differentiation [4]. Unfortunately, if such a characteristic (i.e. large intratumor vessels) is absent, as in our case, it is still difficult to distinguish hibernomas from other hypervascular malignant or benign soft-tissue tumors. In this kind of a circumstance, pathological examination is often necessary for diagnosis.

During scintigraphic examination, increased radiotracer uptake identified on technetium 99m (Tc-99m) tetrofosmin and fluorine 18 fluorodeoxyglucose positron emission tomography can differentiate a benign lipoma from other tumor. Intense uptake within hibernoma and higher standardized uptake values (SUVs) than those of liposarcoma were reported [15,16]. However, in a recent report [17], all hibernomas have SUVs that overlapped with those reported for liposarcoma. It may not be possible to differentiate hibernoma from liposarcoma on the basis of SUV alone.

Treatment is complete surgical resection. Local recurrence does not occur with complete excision. No reports of metastases or malignant transformation have been identified in the reviewed literature.

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