Eur Spine J
DOI 10.1007/s00586-017-5124-5

GRAND ROUNDS

Paraspinal hibernoma: Grand Round presentation of a rare benign adipocytic tumor

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Received: 26 August 2016 / Revised: 20 April 2017 / Accepted: 30 April 2017

Abstract

Introduction We report an uncommon case of paraspinal hibernoma with a T12–L1 foraminal extension and discuss the potential differential diagnoses of paraspinal adipocytic tumors.
Materials and methods A 32-year-old woman consulted our department with a right subscapular and paraspinal mass. There was no associated neurological deficit. The MRI revealed a paraspinal adipocytic tumor with a T12–L1 right foraminal extension. Percutaneous biopsy suggested a diagnosis of hibernoma.
Results Hibernoma is a rare and benign adipocytic tumor arising from embryologic remnants of brown fat. Specific MRI findings are discussed to differentiate hibernoma from other soft-tissue tumors. A planned marginal resection was undertaken with the final histopathology confirming the diagnosis of hibernoma.
Conclusion Based on the Grand Round case and relevant literature, we discuss a rare case of paraspinal hibernoma with a foraminal component and no recurrence at 3-year follow-up.

Keywords Brown fat · Hibernoma · Atypical lipomatous tumors · Lipoma · Liposarcoma

Case presentation

A 32-year-old woman with no prior medical history presented with a right subscapular and paraspinal mass in January 2013. The mass had been slowly growing over the past few years. The primary motive to consult was anxiety as the mass had become unsightly.

There was no associated skin lesion. A deep, subcutaneous, homogeneous and smooth mass was palpated. There was no associated neurological deficit.

Diagnostic imaging section

An initial MRI performed in 2012 (Figs. 1, 2) showed a homogeneous lesion of 165 mm in the long axis. This paraspinal, intramuscular mass extending from the T12–L1 right intervertebral foramen had the following

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Published online: 07 June 2017

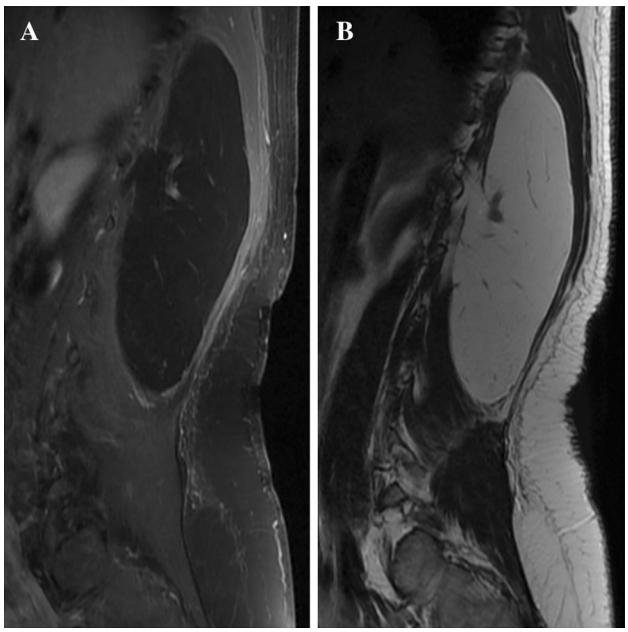

Fig. 1 **a** Parasagittal view with fat suppression showing residual hyperintensity within the tumor. **b** T1-weighted parasagittal view on the lesion long axis

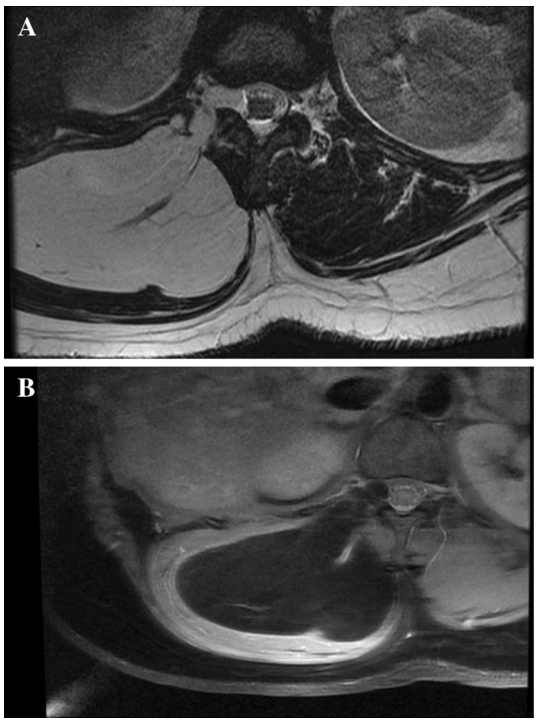

Fig. 2 **a** T1-weighted axial view showing a hyperintense paraspinal lesion with a right T12–L1 foramen. **b** Enhanced periphery after gadolinium injection

measurements: 165 mm height × 94 mm depth × 52 mm width. The lesion had a fatty signal and was well circumscribed. It contained vascular structures and regular fibrous septae in its inferior portion. An intraforaminal extension to the duramater on the right side was noted with no mass effect or deviation of the dural sac. There was no associated morphological or signal anomaly on the spinal cord. A follow-up MRI showed the stability of the lesion 10 months later.

A percutaneous biopsy with ultrasound guidance was performed under local anesthesia: histological analysis reported the presence of both white and brown adipocytes, suggesting the diagnosis of hibernoma. This evaluation was useful for diagnostic orientation and to rule out malignancy.

Procedure

The patient was positioned in the prone position on padded bolsters under general anesthesia. A paraspinal approach on the long axis of the tumor was performed. Following blunt dissection, a brown (due to its high vascularity and cytochrome content) subfascial mass was exposed with meticulous hemostasis using bipolar cautery. Its vascular pedicle was ligated and a marginal resection was performed. Histological analysis was carried out on the resected specimen (Fig. 3a). Layered closure was performed with closed suction drainage. Postoperative recovery was uneventful. The drain was removed at day 2. The patient was discharged from the hospital at day 3.

Histological analysis

Upon microscopic inspection, the final pathological analysis revealed the presence of a well-differentiated mature adipose tissue consisting of white and brown adipocytes (Fig. 3b), in favor of a hibernoma.

Outcome, follow-up

The patient was examined at a 3-year follow-up: she did not report any functional complaint and did not present any sign of local recurrence. No additional documentation was required.

Description about the condition (condition, epidemiology, diagnosis, pathology, and differential diagnosis)

Brown fat is found in newborns in superficial regions (posterior neck and interscapular region) or in deep regions (peri-carotid, retrosternal, pericardiac, perirenal, and intervertebral) and gradually decreases through adulthood.

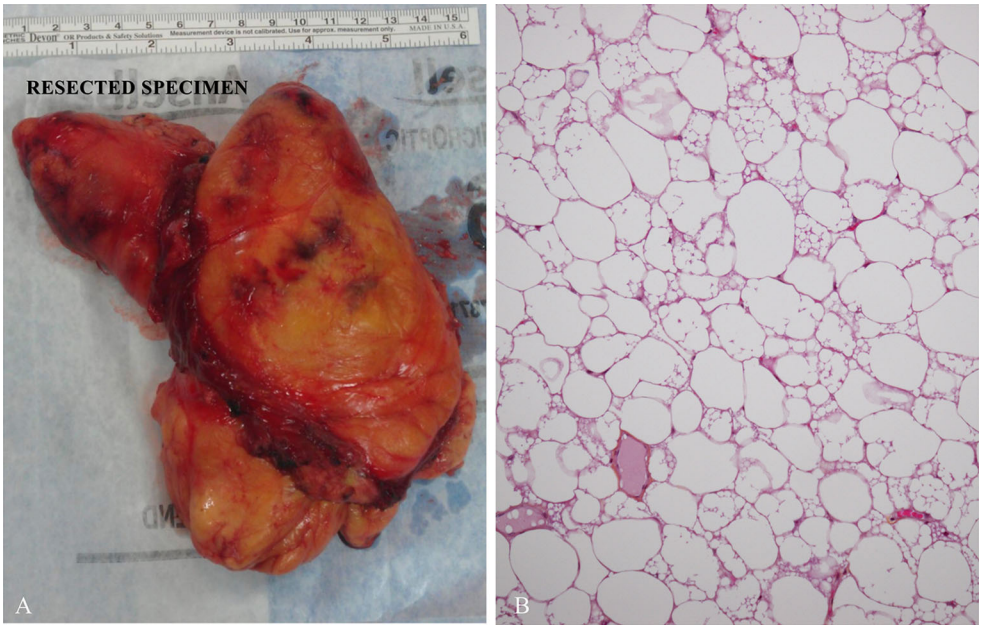

Fig. 3 **a** Resected specimen displaying grossly both *yellow* and *brown* adipose tissues. **b** Adipocytes with multivacuolated granular cytoplasm admixed with the conventional adipose tissue (HES, ×100)

Table 1 Comparative descriptions between lipoma, hibernoma, and atypical lipomatous tumors

	Lipoma	Hibernoma	Atypical lipomatous tumors
Topography	Subcutaneous fat: back and limbs (superficial lipoma). Muscle: thigh, shoulder, arm (deep lipoma)	Subcutaneous or intramuscular. Thigh, shoulder, back, neck	Muscle and fascia. Retroperitoneum (WDLPS: well-differentiated liposarcoma) or thigh
Size	<5 cm superficial lipoma often >5 cm deep lipoma	Variable often >5 cm	Usually >5 cm
Septations	Thin	Thin or thick and vascularized	Thick and vascularized
MRI	Homogeneous, fat signal identical to subcutaneous fat	Heterogeneous, fat signal but intermediate intensity between fat and muscle on T1-weighted images	Heterogeneous, with fat and hypo intense nodules on T1 and T2-weighted images
Enhancement	Absent	Variable, most often heterogeneous	Present and heterogeneous

It plays a role in nonshivering thermoregulation in newborns (unlike muscular thermogenesis) and represents 4% of their total body weight [1–3].

Hibernoma is a rare and benign adipocytic tumor arising from embryologic remnants of brown fat. It was first described by Merkel et al. in 1906 [4], and later, Gery et al. showed similarities with tissues from hibernating animals in 1914 [5]. Hibernoma occurs more frequently in patients between 20 and 40 [6]. The largest hibernoma cohort included 170 cases [6]. There was no recurrence, metastasis, or death at an average follow-up of 7.7 years. Kim et al. [7] reported the case of a small intra-thoracic hibernoma with an extension to paraspinal muscles. Most cases of hibernoma are intermediate forms between lipoma and hibernoma. They usually manifest as superficial, soft, well-defined, painless, and slow-growing masses that, however, may suddenly increase in size. It may also present with local heat due to its hypervascularity. In general, they are well-circumscribed, encapsulated tumors, though infiltration of the surrounding tissue may occur. To this date, several studies reported the imaging characteristics of hibernoma in the literature [8–11]. MRI is the imaging study of choice for hibernoma [12]. The MRI diagnosis of hibernoma can be difficult due to inconsistent results on MRI sequences, even following injection of gadolinium. Lesions with the following characteristics should raise suspicion: a well-defined lesion with a fatty content and a homogeneous, heterogeneous, or septated mass, slightly hypointense to subcutaneous fat on T1-weighted MR images but hyperintense to normal muscle (Figs. 1, 2). Hibernoma usually demonstrates inhomogeneous enhancement after intravenous contrast administration (Figs. 1, 2), but a diffuse enhancement and

even absence of enhancement were also described. On T2-weighted MR images, the mass is nearly isointense to subcutaneous fat. Fat-suppression techniques fail to completely suppress tumoral fat because of the different nature and amount of lipids compared to white adipose tissue.

Differential diagnoses include lipoma and atypical lipomatous tumors. The latter may dedifferentiate, leading to the development of dedifferentiated liposarcoma, the most feared differential diagnosis. However, the dedifferentiation risk is highest in the retroperitoneum (15%) and lower in deep extremity lesions (5%) or other locations, including the back or spine (only 6% prevalence) [13]. This aggressive high-grade tumor with metastatic potential (15% rate) presents very different MRI features. Indeed, focal, nodular non-lipomatous regions should be identified as they differ from their surrounding tissue: their MR aspect is nonspecific presenting a low-to-intermediate signal intensity on T1-weighted MR images and intermediate-to-high signal intensity on T2-weighted MR images [14]. Dedifferentiated liposarcoma should be suspected for deep soft-tissue masses with recent increase in size in patients in their 6th decade [13].

Such diagnostic uncertainties make hibernoma, along with lipoma and atypical lipomatous tumors, a differential diagnosis of choice for adipose tumors. Comparative descriptions are summarized in Table 1.

Hibernoma is a rare differential diagnosis of adipose soft-tissue tumors. As in cases of lipoma or atypical lipomatous tumors, MRI combined with histological analysis of preoperative biopsy samples plays a fundamental role in the diagnosis and further surgical management.

Compliance with ethical standards

Conflict of interest The authors declare that they have no competing interest.

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