Intrathoracic hibernoma

Sharon Y. Ong, MD;* Donna E. Maziak, MD CM;* Farid M. Shamji, MD;* Frederick R. Matzinger, MD;† D. Garth Perkins, MD‡

A benign tumour composed of multiloculated fat cells morphologically similar to the hibernating gland tissues found in animals was first recognized 95 years ago. It was subsequently termed "hibernoma." In 1966, 38 cases had been recorded, 4 of which were intrathoracic.[1] Thirty years later, approximately 100 cases had been reported, of which only 7 were intrathoracic. A recent review of the English literature reveals 13 intrathoracic cases to date. We report a 14th case here

Case report

A 40-year-old woman was referred to the Division of Thoracic Surgery for assessment of an abnormal chest radiograph obtained during a routine clinical examination. She had noticed a mild nonproductive cough for 5 months with no associated symptoms. Her medical history was unremarkable, without any chest illness. Findings on physical examination were normal.

The chest radiograph showed a mass in the right lower thorax. It was sharply demarcated, noncavitary and noncalcified. Computed tomography of the chest and upper abdomen showed a 5-cm, smooth, rounded pleural mass arising from the anterolateral and basal surface of the right side of the pleura. The mass showed homogeneous low density relative to the chest wall musculature and heart (Fig. 1). With contrast enhancement, it showed a heterogeneous increase in density. Otherwise the CT scan was normal. A core biopsy of the lesion showed epithelial-like cells, with uniform, centrally placed small nuclei and abundant eosinophilic cytoplasm. This was interpreted as possible ectopic adrenocortical tissue. With no confirmed diagnosis, a decision was made to resect the lesion.

The chest was entered through a right posterolateral thoracotomy, at the sixth intercostal space. A firm, well-encapsulated tumour, 5 cm in dimension, was visualized arising from the parietal pleura of the anterior chest wall. The base of the tumour was firmly attached to the posterior aspects of the fifth and sixth ribs, and there was no invasion to the diaphragm or the lung. Because biopsies of the tumour on frozen section were inconclusive, the tumour was resected en bloc with the fifth and sixth ribs. Resection margins were free of tumour. The postoperative recovery was uncomplicated and the patient was well with no sign of tumour recurrence at 31-months' follow-up.

The surgical specimen consisted of a

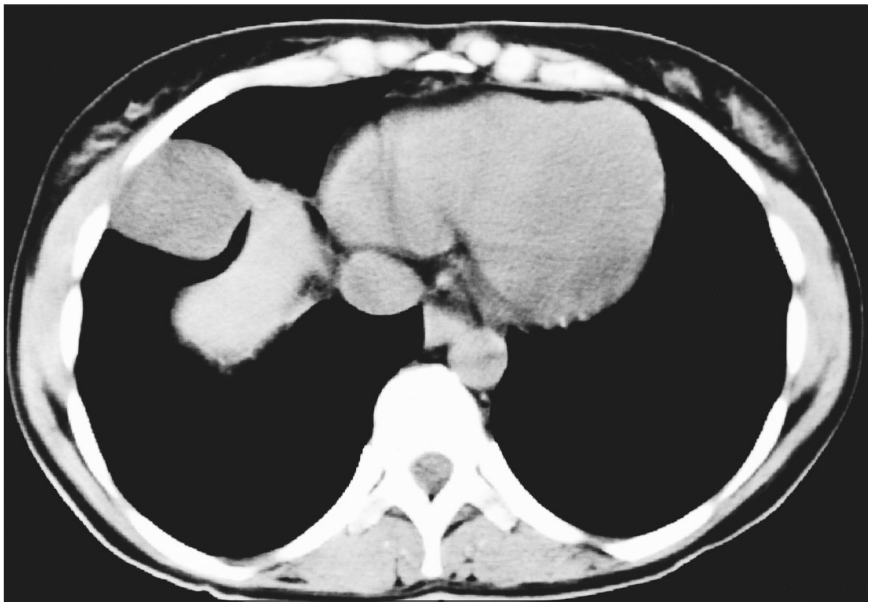

FIG. 1. Unenhanced computed tomography scan shows a 5-cm smooth mass arising from the anterolateral chest wall near the diaphragm with low attenuation similar to subcutaneous fat. Mean CT pixel values were +5.

*From the *Division of Thoracic Surgery, †Department of Radiology and ‡Department of Pathology, The Ottawa Hospital, Civic Campus, Ottawa, Ont.*

Accepted for publication Dec. 5, 2000.

Correspondence to: *Dr. Donna E. Maziak, The Ottawa Hospital — Civic Campus, C.P.C. Rm. 162, 1053 Carling Ave., Ottawa ON K1Y 4E9; fax 613 761-4452, dmaziak@ottawahospital.on.ca*

FIG. 2. Cross-section of the gross surgical specimen showing a smooth surface and finely lobulated appearance characteristic of hibernomas.

well-circumscribed, tan coloured mass, measuring 4.5 × 4.0 × 3.5 cm, covered by parietal pleura (Fig. 2). Microscopic examination revealed cells with abundant granular eosinophilic cytoplasm interspersed with cells containing lipid vacuoles resulting in a foamy appearance. Salient features on electron microscopy included numerous mitochondria and lipid vacuoles. This provided the confirmatory evidence of hibernoma.

Discussion

Hibernoma is a rare soft-tissue tumour arising in fetal brown fat. Merkel first described it in 1906.[2,3] The term "hibernoma" was coined by Gery in 1914, because of its similarity to brown fat cells found in the glands of hibernating animals.[3,4] Brown fat was first described by Velch in 1670.[2,3] He noted a gland-like structure in the mediastinum of a woodchuck and thought it was associated with the thymus. Barkow later separated and distinguished this gland from thymus, calling it a "hibernating gland."[4] In 1902, Shaw studied the fat of human fetuses and demonstrated that certain areas such as the axilla consisted largely of brown fat in contrast to the yellow subcutaneous fat.[2,3,5] A few years later, Bonnot confirmed the existence of brown fat and showed that it persisted in humans from embryonic, neonatal to adult stages and was found in approximately the same location as its proposed homologue in animals.[2,3] In adults, brown fat is usually seen in scattered foci as persisting vestigial remnants along the esophagus, trachea, posterior neck, interscapular area and around the great vessels of the mediastinum.[1,2] Hibernomas are commonly found in these same areas, particularly in the interscapular area.

Grossly, hibernomas are encapsulated and lobulated, of light brown colour. Histologically, the cells contain multiple, small fatty droplets with a centrally placed nucleus. This appearance differs from mature adipose cells, which contain a solitary large globule and peripherally situated signet ring nucleus.[6]

Imaging features of a hibernoma reflect the circumscribed, encapsulated nature of the mass and the hypervascularity of brown fat. The tumour displaces rather than invades adjacent structures. On unenhanced CT, a hibernoma shows homogeneous low density owing to its predominantly lipid composition. With intravenous contrast enhancement, it acquires a heterogeneous characteristic. On ultrasonography, hibernoma was described as a homogeneous, hypoechoic lesion with striking hypervascularity demonstrated by colour Doppler.[2] The characteristic appearance of hibernoma on magnetic resonance imaging is near-fat intensity with hyperintensity on both T_1- and T_2-weighted images and marked enhancement with gadolinium.[7] Because of the highly vascular nature of hibernoma, needle biopsy carries a risk of hemorrhage.

The origin of hibernoma and its exact relation to adult adipose tissue has never been clarified. It has been claimed that hibernoma is only a remnant of a phase in the development of yellow adipose tissue and is not identical to the hibernating gland of animals. Others, like Brines who studied the tumour in 1949, regard it as a special tissue arising from embryonic fatty tissue, which becomes differentiated from ordinary fat and may be related to a special organ comparable to the hibernating gland in animals.[4,6]

All 13 reported intrathoracic hibernomas have been benign, solitary lesions, well encapsulated, firm and with little infiltrative capacity.[1–6,8] Most were found incidentally on routine chest radiography, presenting at various ages. They occurred equally in men and women (average age at presentation 37 yr). In the subpleural tumours, the clinical differential diagnoses have included mesothelioma, neurofibroma or pleural fibrous tumour. Definitive diagnoses were based on surgical resection. All intrathoracic hibernomas were surgically excised with no reported recurrences. There was no endocrine involvement or other physiological consequences.

Although these tumours are considered benign and no malignant tumours have been reported, they have a tendency to grow slowly to a large size and may impinge on neighbouring structures to cause symptoms. Therefore complete surgical resection is necessary for diagnosis and treatment of hibernoma. No adjuvant therapy is required.

References

1. Morgan AD, Jepson EM, Billimoria JD. Intrathoracic hibernoma. *Thorax* 1966;21: 186-92.

2. Santambrogio L, Cioffi U, Simone MD, Nosotti M, Pavoni G, Caputo V, et al. Cervicomediastinal hibernoma. *Ann Thorac Surg* 1997;64:1160-2.

3. Ahn C, Harvey JC. Mediastinal hibernoma, a rare tumor. *Ann Thorac Surg* 1990; 50:828-30.

4. Merikallio E, Halttunen P. Intrathoracic hibernoma. *Ann Chir Gynaecol Fenn* 1966; 55:330-2.

5. Udwadia Z, Kumar N, Bhaduri A. Mediastinal hibernoma. *Eur J Cardiothorac Surg* 1999;15:533-5.

6. Peabody JW, Ziskind J, Buehner HA, Anderson AE. Intrathoracic hibernoma. *N Engl J Med* 1953;249:329-32.

7. Chitoku S, Kawai S, Watabe Y, Nishitani M, Fujimoto K, Otsuka H, et al. Intradural spinal hibernoma: case report. *Surg Neurol* 1998;49:509-13.

8. Rogeaux Y, Leduc M, Faillon JM, Maelfait J, Grenier JL. [Hibernoma or brown fat tumors. Review of the literature apropos of an intrathoracic localization.] *LARC Med* 1984;4:602-6.