The Journal of Laryngology and Otology
October 1985. Vol. **99.** pp. 1055–1058

Cervical hibernoma
Review of the literature and a new case

by

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Introduction

Hibernoma is the well-recognized designation for a rare, benign, soft-tissue tumour composed of cells remarkably similar to those of brown adipose tissue (Seemayer *et al.*, 1975; Lawson and Biller, 1976).

Mercel in 1906 first described a tumour composed of brown adipose tissue, and the term 'hibernoma' was proposed in 1914 by Gery because of its similarity to hibernal fat (Mesera and Batsakis, 1967).

A review of the world literature contains about 80 cases of hibernomas, of which only six occurred in the cervical area (Simon, 1950; Cox, 1954; Apatenko and Poroshin, 1962; Vetner, 1966; Mesera and Batsakis, 1967; Lawson and Biller, 1976). This paper reports a case, drawing attention to the salient features of this neoplasm, which presented as a swelling in the neck.

Case History

A 54-year-old, previously healthy man, was admitted with a slowly growing tumour in the left side of the neck during a period of three months, with no general symptoms or pain. The tumour was initially located close to the angle of the mandible but gradually descended. Physical examination revealed a 6×5×4 cm. soft, not well-delimited, freely movable, non-tender tumour in the lower left region of the neck just above the clavicle. Pre-operative blood tests were normal.

Surgery showed a pale brown tumour, well-delimited with a well-defined capsule, placed beneath the sternocleidomastoid muscle posterior to the carotid sheath. The tumour was completely removed. The post-operative course was uneventful, and the patient was free from recurrence at follow-up five years later.

Macroscopically, the tumour was covered by a thin, wispy capsule. The cut surface was bright yellow-brown and lobulated. Microscopy showed a well-delimited, thinly encapsulated tumour consisting of round-to-polygonal cells varied in size, with a single central nucleus containing a large, round nucleolus. The foamy cytoplasm was granular to multivacuolated, eosinophilic between the vacuoles (Fig. 1). There were small groups of normal-looking fat cells, staining strongly with Sudan stain. The tumour was divided into many lobules by fibrous septa containing thin-walled vessels; in some places there were also bundles of skeletal muscle fibres. The histologic picture was characteristic of a hibernoma.

Discussion

Brown adipose tissue, first discovered in 1670 by Velsch who thought it was related to the thymus (Rasmussen, 1923), is prominent in hibernating animals as hibernal fat or the hibernating gland. This specialized adipose tissue has been described in more than 50 animal species including such non-hibernating forms as the mouse, rat, monkey and man (Rasmussen, 1923; Mesera and Batsakis, 1967; McLane and Meyer, 1978). It is mainly present in the superior mediastinum with extension along the intermuscular septa into the neck, axillae and interscapular region, but has also been found in the retroperitoneum and inguinal area (Lawson and Biller, 1976; McLane and Meyer, 1978).

Brown adipose tissue is brown-tan in colour and quite vascular. The cells (lipocytes) are polygonal, 25 micra in size, multivacuolated with granular cytoplasm and a central ovoid nucleus (Rasmussen, 1923; Mesera and Batsakis, 1967). White adipose tissue is avascular, uniformly distributed, white-yellow in colour, and lardaceous in appearance. These fat cells (lipocytes) are round, 50 micra in

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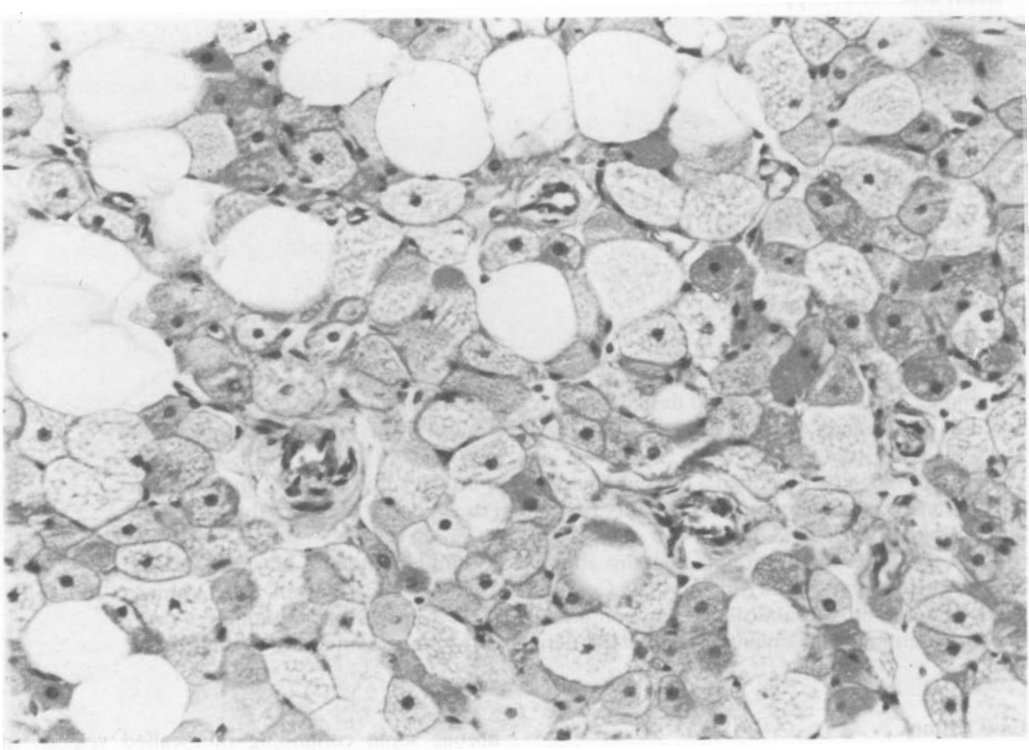


FIG. 1

The tumour is composed of round or polygonal multi-vacuolated cells, also called 'mulberry' cells. A few unvacuolated adipose tissue cells are also seen. There are several scattered capillaries surrounded by the tumour cells. (H and E ×250).

size, and univacuolated with a peripheral, flattened nucleus (Rasmussen, 1923).

The histologic, histochemical, biochemical, and ultrastructural differences between white and brown adipose tissues have promoted the suggestion that they are histogenetically different tissues (Rasmussen, 1923; Napolitano and Fawcett, 1958; Dardick, 1978). The subject has nevertheless been debated for almost a century without reaching a resolution (Dardick, 1978; Enterline *et al.*, 1979). Some authors claim that brown adipose tissue represents a stage in the development of white adipose tissue (Sidman, 1956; Simon, 1965), while others favour the view that there are two different types of fat cells with unique developmental characteristics and different metabolic functions (Aherne and Hull, 1966; Hull, 1966; Barnard, 1969).

Ultrastructural studies suggest that both white and brown adipose tissues arise from endothelial ingrowths into the mesenchyme (Wasserman, 1965; Dardick, 1978). At some stage in tissue differentiation adipocytes of both brown and white adipose tissues have multiple cytoplasmic lipid vacuoles (Napolitano, 1965; Wasserman, 1965), but though adipocytes of brown adipose tissue appear to be capable of becoming univacuolated (Simon, 1965), the question remains whether these cells are similar to the mature adipocytes of white adipose tissue (Dardick, 1978).

Normally the amount of identifiable brown adipose tissue decreases markedly after birth (Novy and Wilson, 1956; Fentiman *et al.*, 1975), although small quantities persist in all age groups as small islands of brown adipocytes in the white adipose tissue. From these residua hibernomas may develop (Fentiman *et al.*, 1975; Lawson and Biller, 1976).

The great majority of hibernomas were, according to McLane and Meyer's (1978) review of 67 cases, situated in the subcutaneous tissues of the back, especially in the interscapular area, the axilla, intrathoracic area, neck, and perirenal area. The thigh, buttock, popliteal fossa, lumbosacral area, abdominal wall, and anterior chest wall were

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uncommon sites of location and difficult to understand from the anatomical distribution of brown adipose tissue in man (Seemayer *et al.*, 1975; Enterline *et al.*, 1979). The reports of intracranial, intraspinal, and intramyocardial hibernomas were even more unusual from the viewpoint of location (Vagn-Hansen and Osgaard, 1972; Shuangshoti and Menakanit, 1974; Kindblom and Svensson, 1977).

In those cases for which the patient's sex is specified, hibernomas appear to be slightly more common in females (Fentiman *et al.*, 1975; McLane and Meyer, 1978), and the majority of patients present with a hibernoma in the third and fourth decades.

The youngest patient reported with a hibernoma was six weeks old (Cox, 1954), the oldest 84 years of age (Kindblom and Svensson, 1977) and the duration of symptoms ranged from a few weeks to more than 20 years (Novy and Wilson, 1956), generally averaging several years. All seven cervical hibernomas (Table I) arose in males and, with the exception of the case presented by Cox (1954), in the posterior triangle of the neck. This hibernoma was present at birth in the anterior and lateral aspect of the neck.

Clinically, the hibernomas generally present as a firm, freely movable, non-tender mass. They are usually ovoid in shape and generally asymptomatic, but may occasionally produce symptoms by pressure, displacement, or both, of vital regional structures, such as tracheal compression (Cox, 1954) and cough occurring with intrathoracic tumours (Lawson and Biller, 1976; Mesera and Batsakis, 1967). The tumour is typically slowly growing and may suddenly show rapid enlargement (Lawson and Biller, 1976). Two cases of intermittently expanding hibernomas have been reported (Vetner, 1966; Tizes and Tizes, 1974), and the present case showed an unexplainable descending behaviour. Because of their slow growth they may attain considerable size before treatment is instituted. Novy and Wilson (1956) reported an average size of 5 to 10 cm. after a growth time of six years. Reports of malignant types (Symmers, 1944; Apatenko and Poroshin, 1962, 1963; Enterline *et al.*, 1979) have been noted by some authors (Mesera and Batsakis, 1967; Lawson and Biller, 1976).

The tumours are likely to be diagnosed preoperatively as lipoma, fibroma, or neurofibroma, but the usually prominent vascularity of the hibernoma may be detected in the overlying skin (McLane and Meyer, 1978). Thus, angiography of the affected area may be helpful in the pre-operative diagnosis, particularly if the mass is not easily amenable to palpation (McLane and Meyer, 1978). Angiography would also be valuable in identifying the feeding vessels prior to surgery, as the extensive vascularity of these lesions may cause troublesome bleeding.

Although (Symmers, 1944; Apatenko and Poroshin, 1962, 1963; Enterline *et al.*, 1979), histologically malignant hibernomas may exist, but most hibernomas were clinically benign. Surgical resection is curative for hibernomas, and no recurrence after complete removal has been described (Mesera and Batsakis, 1967; Enterline *et al.*, 1979).

Summary

Hibernoma is the designation for a rare, benign, soft-tissue tumour composed of cells similar to those of brown adipose tissue. Review of the world literature reveals about 80 cases, but only six hibernomas in the cervical region have previously been reported.

A case of cervical hibernoma showing descending behaviour is presented, and the histological, histogenetical, and clinical features of hibernomas are discussed.

It is stated that the tumours are adequately treated by local excision, as the neoplasms are histologically and clinically benign, but the extensive vascularity may cause troublesome bleeding in some instances.

TABLE I

SEVEN CERVICAL HIBERNOMAS

Authors	Sex	Age	Duration of symptoms	Size of tumour in centimetre
Simon, 1950	M	21	Twenty years	8×3×3
Cox, 1954	M	0	Six weeks	5×3×2
Apatenko and Poroshin, 1962	M	44	Twenty-two months	6×4×3
Vetner, 1966	M	30	Two months	7×4×4
Mesera and Batsakis, 1967	M	36	Nineteen months	4×3×2
Lawson and Biller, 1976	M	20	One month	8×5×3
Kristensen, 1984	M	54	Three months	6×5×4

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