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Intra-abdominal lymphangioma

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The historical background, aetiology, clinical features, radiographic findings and treatment of abdominal lymphangiomas are reviewed. The condition may give rise to the acute surgical abdomen.

Keywords: Retroperitoneal lymphangioma, acute surgical abdomen

Intra-abdominal cystic and cavernous lymphangiomas are rare benign tumours, usually found incidentally through lymphangiography, during elective abdominal surgery, in an acute surgical abdomen, or at post-mortem examination. The incidence of such tumours has been estimated to range from 1 per 27 400 to 1 per 100 000 hospital admissions[1–3].

Historical background

Braquehaye[4] credited Benievine, a Florentine anatomist, as being the first to note a mesenteric cyst during a post-mortem examination in 1507. Slocum[5] has stated that the earliest description of a chylous cyst, found at necropsy, was published by Rokitansky in 1842. Millard and Tillaux[6] in 1880 were the first to report on the successful treatment of a patient with a mesenteric cyst. According to Walker and Putnam[1], omental cysts were not described until Gairdner's report in 1852. This type, which is believed to be less common than mesenteric cysts, has been estimated to occur from three to ten times more frequently[1]. Pean in 1883, as quoted by Slocum[5], was the first surgeon to record the recovery of a patient who had a lymphangiomatous cyst treated by marsupialization. The initial American report of a chylous cyst of the mesentery was presented by Carson in 1890[7]. Some years later, Sarwey[8] successfully excised a unilocular chylous cystic lymphangioma located in the region of the head of the pancreas of a 9-year-old girl. Gaudier and Gorse[9] in 1913 surveyed the accumulated relevant literature, and included a 4-year-old patient of their own with a mass in the right lumbar region which was excised successfully. They claimed this to be the first description of a case of retroperitoneal cystic lymphangioma to appear in print. In 1941 Loeb[19] reviewed the literature on mesenteric cysts and estimated that reports on 550–600 patients had been published. Bearhs *et al.*[11], reporting from the Mayo Clinic, found only nine cases undergoing surgery between the years 1911 and 1947. Pack and Tabah[12], discussing over 870 patients with primary tumours of the retroperitoneum, including 120 of their own, found only five with cystic tumours. Harrow[13] estimated that of more than 600 patients with mesenteric cysts, 70 of whom had retroperitoneal cysts, only 14 fitted the criteria for lymphatic cystic tumours. Rauch[14] screened the literature and added two male patients aged 17 and 10 years with retroperitoneal lymphangiomas. Henzel *et al.*[15] gave an excellent overview of the published material on intra-abdominal lymphangiomas, while adding a 20-year-old patient treated by them.

Aetiology

Sabin[16] divided the development of the lymphatic system into two stages:

1. The primary stage which encompasses the development of isolated lymph sacs derived from veins that are united by the thoracic duct
2. The second stage, which involves peripheral growth of lymph vessels that sprout from the endothelial lining of the sacs. The lymph sacs are transformed into a plexus of lymphatic capillaries by bridging of the lumen with bands of connective tissue from which chains of lymph nodes are derived.

Numerous theories have been proposed concerning the origin of cystic lymphangiomas. Barnett and Branch[17] summarized these theories on the following aetiological basis:

1. Retention, which explains the lymphangioma on the basis of mechanical pressure
2. Disturbance of the endothelial secretory function of the vessels or of endothelial permeability
3. Inflammatory background
4. Embryonal origin.

The last theory, favoured by Barnett and Branch[17], explains the lymphangioma on the basis of a defect in embryonal organization of the lymphatic system. Some investigators express the opinion that retroperitoneal lymphangiomas arise from obstruction of existing lymphatic channels by an inflammatory fibrotic process or lymphatic hamartoma[18]. In general, the majority of authors subscribe to the last two theories.

Godart[19] believes that the aetiology is related to an abnormal development of the lymphatics in that there is failure of communication of a branch, or branches, with the central system, thus explaining why cystic lymphangiomas are found in the same position as fetal lymph sacs (cervical, mediastinal and retroperitoneal). He proposed that lymphatic spaces in the embryo fail to join the venous system, thus producing cysts. Elliot *et al.*[20] believe that lymphaticovenous shunts exist in the perinodal tissues as congenital deficiencies, their formation resulting in cysts.

Other suggested aetiologies include failure of the leaves on the mesentery to fuse or localized degeneration of lymph nodes[2]. Trauma has also been offered as a possible cause of lymphangioma[21,22].

Ewing[23] stated that retroperitoneal lymphangioma occurs in both children and adults as cystic tumours, originating along the spinal column and ramifying into the pelvis behind the kidney or colon, upward to the liver and spleen, as well as into the omentum. He included in this entity a variety of slowly growing congenital tumours of the skin and subcutaneous tissue, neck muscles, axilla, trunk, lip, tongue, eye and orbit.

Lymphangiomas that arise in the retroperitoneal space

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separate the leaves of the mesentery as they grow anteriorly and take up a mesenteric position, or they may push posteriorly in their development, to be recognized only as retroperitoneal tumours. They may encroach on the adjacent structures, including gastrointestinal organs, and so lead to partial obstruction.

Occasionally it is impossible to distinguish between retroperitoneal lymphangiomas and mesenteric cysts, as both these tumours may extend from the retroperitoneum into the mesentery[24] and, moreover, also have the same gross appearance. With these difficulties in mind, Takiff *et al.*[25] stressed the importance of differentiation between these two lesions in view of the fact that lymphangiomas often behave in an aggressive manner and invade the adjacent structures. They observed that lymphangiomas are principally a disorder of childhood and young adulthood, while mesenteric cysts may occur at all ages, although they are mainly found in the fourth decade of life.

Histologically, these two lesions differ. Cystic lymphangiomas display a single endothelial lining, foam cells, innumerable lymphatic channels between lobules of adipose tissue, fibrous and lymphoid tissue, blood vessels and smooth muscle[13,26]. The lining cells of the mesenteric cysts, on the other hand, are often cuboidal or columnar or, on rare occasions, may even be entirely absent.

While lymphangiomas are histologically benign, two patients with lymphangioendotheliomas have been reported[11]. Malignant lymphangiomas are known as lymphangiosarcomas[27]. A unique case of a retroperitoneal malignant tumour in a 44-year-old man has been described recently[28]. The tumour had infiltrated the duodenum and the head of the pancreas, and pancreatoduodenectomy was performed[28].

Classification

Lymphangiomas are classified primarily on a histological basis. The original classification established by Wegner in 1877[29], which is still considered to be the most reliable one[13,14,17,30–34], divides lymphangiomas into simple, cavernous or cystic tumours.

The simple form represents a new growth of small lymphatic channels lined with endothelium, usually situated superficially in the skin. This type consists of an ill-defined mass composed of dilated lymph vessels with rich cellular connective tissue stroma. It has a moderate number of channels through which it is connected to the adjacent lymphatic system.

The cavernous type comprises communicating lymph-filled locules separated by septa and found chiefly in macroglossia, macrocheilia and, more rarely, in other parts of the face and skin. This kind of lymphangioma is a spongy, compressible tumour with dilated lymph vessels and lymphoid stroma, having a moderate number of channels which connect it to the normal lymphatic system.

The cystic type may be uni- or multilocular and appears mainly in the neck, breast, axilla, lower sacral region, abdomen, thighs and inguinal region. When located in the mesentery, cystic lymphangiomas often contain chyle and are appropriately termed chylangiomas or 'chylous cysts'. The tumour is composed of one or many variously-sized cysts ranging from 1 mm to 5 cm in diameter, often communicating with each other[35], and lined with endothelium. The cyst may contain clear chylous, serous, bloody, purulent or chocolate-coloured fluid. Such cystic tumours have practically no connection with normal adjacent lymphatics. They may arise from cavernous forms in which one compartment enlarges, breaking the septa and compressing the other compartment[13]. Gerster[36] maintains that it is not always possible to draw a sharp line between cystic and cavernous lymphangiomas, since the former may contain areas that are cavernous in structure.

Henzel *et al.*[15] attempted to outline some features which will permit differentiation between the three types (*Table 1*). Two entries listed in *Table 1* warrant emphasis: (1) only cavernous and cystic lymphangiomas have been found in the retroperitoneum, and (2) only in the cavernous and cystic types does lymphangiomatous malignant degeneration occur.

Lymphangiomas most commonly manifest themselves in the neck (75 per cent), when they are called cystic hygromas. The rest are found mainly in the axillary region (20 per cent), while the remaining 5 per cent may appear at various sites in the body, among which are mediastinum[37,38], lungs[39], chest wall, arms, back, parotid, spleen[40–42], liver[43], uterus[44], rectum[45], inguinal region[46], and multiple other sites[33]. Skeletal involvement may be seen as part of a generalized lymphangiomatosis[43,47,48]. In the spleen[49], as in the stomach and omentum[50], the lymphangioma may be associated with intramural gastric pancreas. It may also be found in the oesophagus and stomach[51,52] and in the jejunum[53].

Less than 1 per cent of lymphangiomas are found in the retroperitoneum, those occurring in the mesentery and omentum being called mesenteric and omental cysts, respectively[21,22,33,34,47,54]. Most unilocular lymphangiomas are localized within the mesentery or the omentum, whereas

Table 1 *Differential features in the gross and microscopic characteristics of lymphangiomas (according to Henzel* et al.[15])

	Simple	Cavernous	Cystic
Anatomical location	Skin and subcutaneous tissue of face and neck	Cheek, tongue, buccal tissue, neck, retroperitoneal	Neck (hygroma), axillae, inguinal area, retroperitoneal
Compressibility (patent to adjacent normal lymph flow)	Yes	Yes	No
Unilocular/multilocular	Unilocular	Both	Both
Serous and chylous variants	No	Yes	Yes
Origin congenital	Yes	Yes	Yes
Encapsulated	No	No	No
Microscopic dilated lymphatic spaces	+	+ ±	+ + to + ± +
Endothelial lining within spaces	Yes	Irregularly occurring	Irregularly occurring
Inflammatory cell infiltration	Insignificant	+ +	+ + to ± + ±
Smooth muscle component	Very infrequent	Yes	Yes
Frequency of foam cell	No	±	+ ±
Benign growth	Slow, if at all	Yes	Yes
Potential for malignant transformation	Unknown	Rare but substantiated	Possible

−, Mild: + ±, moderate: − + +, substantial

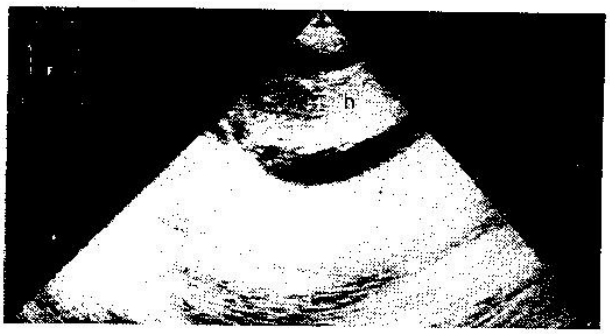

Figure 1 *Real-time ultrasound image showing a portion of the large cystic loculated mass situated in the left upper abdomen with an echogenic structure within the lesion representing the intracystic haemorrhage (h)*

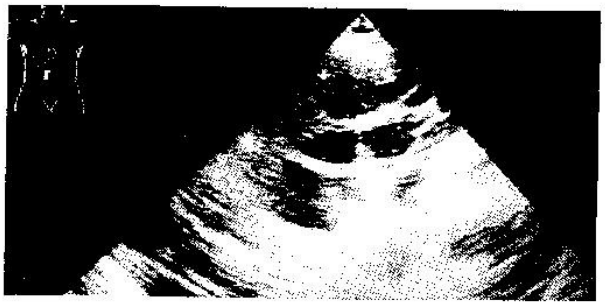

Figure 2 *Real-time ultrasound image showing smaller cystic areas situated in the epigastric region which were part of the large multiloculated mass*

the multiloculated variant may occupy the entire bowel, mesentery and much of the retroperitoneal space.

Clinical features

Retroperitoneal cystic lymphangiomas usually appear in early infancy with about 90 per cent being detected in the first 2 years of life[35]. Except for the series of Takiff *et al.*[25] no sex predilection has been observed. These authors describe 28 patients, eight of whom had cystic lymphangiomas and 20 of whom had mesenteric cysts. Of the eight cystic lymphangioma patients, six were symptomatic; four of the eight patients were found on examination to have ascites and large lesions (mean diameter 8·8 cm). Of the 20 patients with mesenteric cysts, six were male, and only five of the entire group of 20 were symptomatic. None of them had ascites, and the lesions were smaller than in the group with lymphangiomas (mean diameter 4·7 cm). There were no clinical features which served to differentiate retroperitoneal lymphangioma from other retroperitoneal masses.

The most common clinical manifestation of retroperitoneal cystic lymphangioma is that of a palpable, soft, cystic mass in the abdomen, which usually enlarges slowly. Some patients may complain of a 'dragging' sensation[15], while others may be asymptomatic, the mass being discovered only incidentally during examination for an unrelated complaint. Like most retroperitoneal masses, these, too, remain occult until pressure occurs, or the mass has become visible or palpable. Pressure symptoms depend largely on the location of the cystic mass, and in 40 per cent of cases pressure on the adjacent structures may cause partial intestinal obstruction[15], displacement of kidneys and ureters to the midline[31], ureteric obstruction[30,55] and displacement of the middle colic artery[56].

The cyst seldom causes acute clinical symptoms and, as the retroperitoneum is relatively inaccessible to physical examination, the diagnosis is often entirely accidental. When acute symptoms do occur, they are due either to pressure on the adjacent structures by the enlarging mass, or to complications, such as haemorrhage into the cyst, inflammation of the cyst wall, infection, perforation, torsion and rupture[17,57,58]. The patient may suffer from abdominal pains and tenderness with guarding and fever; leukocytosis may be present. These cases of acute clinical abdomen may mimic appendicitis, as occurred in 12 per cent of the patients reviewed by Gallifer *et al.*[59].

In our own practice we have treated a 25-year-old woman with a large upper mid-abdominal cystic multiloculated mass (*Figures 1* and *2*), and a 20-year-old woman with mesenteric cystic lymphangioma (*Figure 3*). Both patients presented with abdominal pain and fever (38°C) and complained of vomiting. On physical examination, psoas and Rovsing's signs were positive. They underwent laparotomy and excision of the cysts. *Figures 4* and *5* demonstrate the histological findings in these two patients.

Most of the cases reported in children also mention an acute surgical abdomen[37,54,60]. Coagulation disorders described in lymphangiomatosis[39] included features of consumptive coagulopathy[61].

A useful list of conditions to be considered in the differential diagnosis of lymphangioma has been compiled by Blumhagen *et al.*[62] (*Table 2*).

Radiographic findings

Previously, the diagnosis of lymphangioma was made during surgery or at autopsy. Currently, we have access to methods

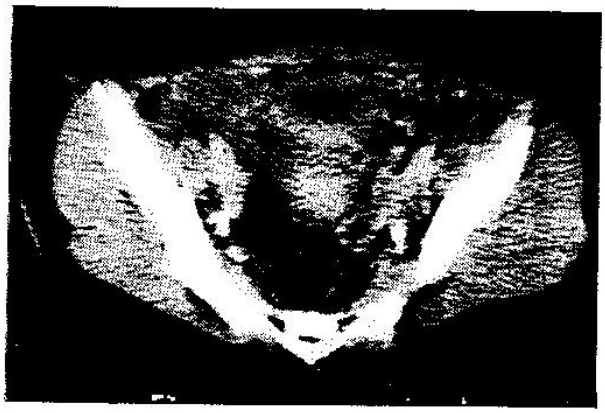

Figure 3 *Computed tomographic scan of the pelvis showing cysts (c) and ascites*

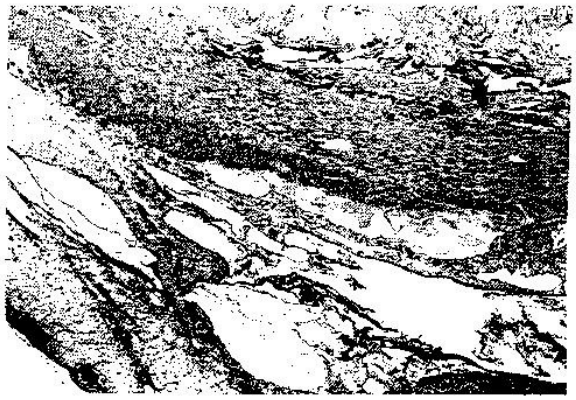

Figure 4 *Fragment of cystic wall covered with flattened endothelial cells. Extensive chronic inflammatory infiltrate with granulation tissue formation and some smooth muscle are seen*

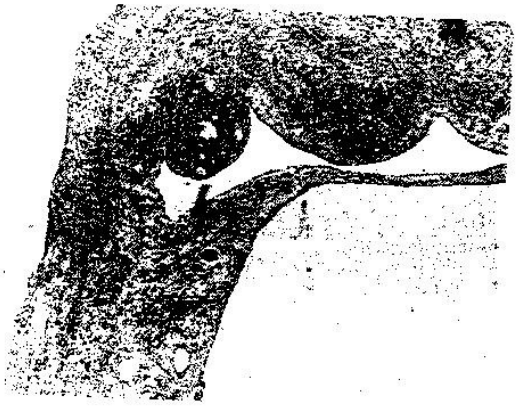

Figure 5 *Cystic lymphangiectasis beneath the serosal surface of the appendix*

of imaging that afford a preoperative diagnosis. Radiographs may be helpful in localization through displacement of surrounding viscera, and calcification has been described in a cavernous pancreatic lymphangioma[63]. Asch *et al.*[43] reported on a calcified cystic lesion in the spleen of a child with diffuse involvement of the liver.

Intravenous urography may be of value in revealing displacement of the kidney or ureters. A barium enema may demonstrate medial or lateral displacement of the ascending or descending colon without obstruction, while barium studies of the duodenum may assist in the diagnosis of a lymphangioma arising in the upper abdomen.

Other diagnostic imaging methods have proved of value in preoperative localization of these masses and in defining their internal architecture. Ultrasound contributes to the diagnosis of lymphangioma by showing a cystic mass with multiple thin septa. This mode of examination also shows the relationship of the mass to abdominal and retroperitoneal structures[62].

The computerized tomographical features of cystic lymphangiomas are uni- or multilocular massses of water density (*Figure 3*). The wall of the mass may be enhanced following intravenous administration of contrast material. The septa are of uniform thickness[32]. In one case, the computed tomograpical attenuation of 15 Hounsfield units within the cyst suggested the presence of fat within the lesion[34].

Although the efficacy of magnetic resonance imaging in displaying the cystic lesion is comparable to that of computed tomography, the signal intensities and T-1 and T-2 values, which differ between cysts, may reflect differences in the composition of cystic fluids[30].

With lymphangiography, only those lymphangiomas that communicate with the lymphatic system are directly visualized. A cystic lymphangioma of the mesentery (mesenteric cyst), which was shown to communicate with the thoracic duct, demonstrated dilatation of interconnecting lymphatic channels with passage of contrasting material into the mesenteric cystic spaces on lower extremity lymphangiography[64].

Angiography clarifies the manner in which the mass causes displacement of arterial branches, as well as stretching of an artery by the tumour, although the method does not show tumour vascularity.

Of the methods noted here in brief we advocate ultrasound and computed tomography as the most efficacious procedures for visualizing abdominal lymphangiomas.

Treatment

Complete excision is the treatment of choice and it is usually carried out with relative ease. Aspiration of the larger cysts lessens the difficulty with exposure. If the cyst has invaded an abdominal organ like the small bowel, spleen or the tail of the pancreas[25], total excision with resection of the involved organ is indicated. In the past, both external and internal marsupialization were performed. Although cystenterostomy has been suggested, the form of drainage preferred was internal peritoneal cavity marsupialization. Such surgical forms of treatment have become obsolete, and the modern approach advocates total excision of the mass in a patient with cystic retroperitoneal lymphangioma. With incomplete removal of the cyst, there is a possibility of recurrence at a later date, such as was observed by Raszkowski *et al.*[65] 7 years after the original surgical intervention.

Table 2 *Differential diagnosis of lymphangioma (according to Blumhagen et al.*[62]*)*

Huge lesions	
Ovarian:	Lymphoma (not truly cystic)
Teratoma	Kidney:
Cystadenoma	Polycystic kidney disease
Mucinous cystadenoma	Cystic dysplasia
Pancreatic pseudocyst	Clear cell sarcoma
Multilocular cystic nephroma	Spleen:
Lymphoma (not truly cystic)	Epidermoid cyst
Lesions of moderate size	Liver:
Haematoma	Mesenchymal hamartoma
Postoperative seroma	Cystic hepatoblastoma
Peritonitis	Haemangioendothelioma
Echinococcal cyst	Choledochal cyst
Enteric duplication	Carolis disease
	Embryonal sarcoma

Conclusions

The historical review demonstrates the relative rarity of cystic abdominal lymphangioma. The aetiology of the lesion is probably a primary malformation of the lymphatic system, essentially a hamartoma, which may either grow continuously, or cause blockage and subsequent enlargement of lymphatic channels.

Histologically, lymphangiomas can be distinguished into simple, cavernous and cystic types. The clinical features are typically those of a space-occupying lesion causing pressure on adjacent structures or of complications within the lesion itself. The patient often presents with an acute surgical abdomen. Ultrasound is the imaging modality of choice, owing to the cystic internal consistency. Other imaging modalities, especially computerized tomography, may be useful. The recommended treatment is complete excision.

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