Abdominal cystic lymphangioma. A case report

Piotr Szymański, Marcin Zeman, Agnieszka Czarniecka

Introduction. Cystic lymphangiomas are benign tumors that are most prevalent in children in the head and neck region. Abdominal location of these tumors in adults is rare. Radical surgery is the treatment of choice.

Case presentation. We present a diagnostic and therapeutic approach to cystic lymphangioma. A 20-year-old male patient was diagnosed with an extensive multicystic lesion involving the hepatic hilum, hepatoduodenal ligament, aorta and inferior vena cava. Histological examination confirmed the diagnosis of cystic lymphangioma. Non-radical resection was performed as it was impossible to dissect the lesion from large vessels. The patient underwent a consecutive follow-up.

Results. An increase in residual lesions was not observed during a 7-year follow-up. No clinical manifestations were reported.

Conclusions. Non-radical resection can be considered in the case of cystic lymphangioma if total tumor resection is associated with a high risk of serious intra- and postoperative complications.

Key words: abdominal cystic lymphangioma, non-radical surgery
fragments of adipose and connective tissues with multiple spaces that might correspond to lymphatic vessels. The result was inconclusive.

Cytological examination of the fluid showed the presence of lymphocytes, eosinophils, macrophages containing hemosiderin, endothelial cells and protein masses. Biochemical analysis of the fluid showed the following composition: triglycerides 109 mg/dl, total protein 37 g/l and amylase activity 21 U/l. Bacteriological examination of the fluid showed negative culture results. Conversion to laparotomy was performed due to the difficulties in the accurate assessment of the tumor. Most of the tumor was removed. However, due to the fact that it was impossible to dissect the mass from hepatoduodenal ligament structures and large vessels, this part of the tumor was left. Postoperative histological examination revealed multiple cystically dilated vessels (most likely lymphatic) and an irregular distribution of smooth muscle bundles. After the immunohistochemical examination, which confirmed the presence of the CD34 antigen, the final histological diagnosis revealed cystic lymphangioma. The postoperative course was uncomplicated and the patient was discharged in good general condition on day 5 after the procedure. During a 7-year follow-up, several abdominal imaging examinations were performed, which showed a stable residual lesion (Figs. 2, 3).

Discussion

Differential diagnosis of retroperitoneal cystic tumors includes cystic lymphangioma, mucinous cystadenoma, cystic teratoma, cystic mesothelioma, Mullerian cyst, epidermoid cyst, tailgut cyst, bronchogenic cyst, cystic lesions in solid neoplasms, pseudomyxoma retroperitonei and perianal mucinous carcinoma [8]. Computed tomography and magnetic resonance imaging are the most useful tools in the assessment. A fine-needle aspiration biopsy can be of value [9].

When abdominally lymphangioma is diagnosed, the treatment of choice is total resection by laparotomy or laparoscopic-assisted approach [1, 7]. However, it is not always possible. The question arises whether in cases requiring extensive organ resection, tumor should always be resected in total. Non-radical resection with no local recurrence during several years of follow-up was reported [10]. In such cases it seems reasonable to perform non-radical resection with a follow-up (as in the case of our patient) or with additional sclerotherapy of residual lesions [11]. Many substances
(e.g. bleomycin, doxycycline, polidocanol, zein solution, OK-432 or fibrin glue) are successfully used in sclerotherapy of lymphangiomas which can be additionally given during surgery or percutaneous drainage. These substances can be also applied as primary treatment alone [4, 12, 13]. Single cases of successful treatment of lymphangiomas using oral drugs, such as propranolol [14] and sirolimus [15], were reported. However, the effectiveness of these drugs has not been fully confirmed yet due to the small number of cases and short follow-ups.

**Conclusions**

Our case and the reports of other authors show that total tumor resection which could expose patients to extensive multi-organ resection and complications should not be performed in each case. Non-radical resection with a consecutive follow-up is a reasonable approach in such cases.

**Acknowledgments**

We wish to thank Arkadiusz Badziński DHSc for the assistance in the translation of the manuscript.

**Conflict of interest:** none declared

**Piotr Szymański**

The Maria Sklodowska-Curie Institute – Oncology Center Branch in Gliwice

The Oncologic and Reconstructive Surgery Clinic

ul. Wybrzeże AK 15

44-101 Gliwice, Poland

e-mail: szymanskpiotr@mp.pl

Received: 11 Feb 2019

Approved for printing: 8 Mar 2019

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