DOI: https://doi.org/10.2298/SARH200616036S
UDC: 616.36-006.326

CASE REPORT / ПРИКАЗ БОЛЕСНИКА

Liver angiomyolipoma

Filip Świątkowski[1,2], Dorota Zielińska[1], Wojciech Tański[3], Pawel Gajdzis[4], Mariusz Chabowski[1,2]

[1]Fourth Military Teaching Hospital, Department of Surgery, Wroclaw, Poland;
[2]Wroclaw Medical University, Faculty of Health Science, Department of Clinical Nursing, Division of Oncology and Palliative Care, Wroclaw, Poland;
[3]Fourth Military Teaching Hospital, Department of Internal Medicine, Wroclaw, Poland;
[4]Wroclaw Medical University, Faculty of Medicine, Department of Pathomorphology and Oncological Cytology, Wroclaw, Poland

SUMMARY

Introduction Benign tumors of the liver are rare. Liver angiomyolipoma is a rare benign mesenchymal tumor that usually occurs in adult female patients. There are four types of hepatic angiomyolipoma: (I) hybrid; (II) myoma type; (III) lipoma type; and (IV) hemangioma type.

Case outline We present a 44-year-old female without symptoms, admitted to the Department of Surgery due to the lesion of the third segment of the liver, measuring 35 × 30 × 15 mm. Tumor was totally excised, and the margins of resection were clean. The differential diagnosis based on radiological findings might be difficult. There are many liver disorders with fatty components, both benign and malignant, e.g., hepatic steatosis, adenoma, lipoma, hepatocellular carcinoma or liposarcoma.

Conclusion Its prognosis is good and the recommended treatment is surgical resection.

Keywords: angiomyolipoma; hepatic tumor; surgery

INTRODUCTION

Benign tumors of the liver are rare with the exception of cavernous hemangioma [1]. Liver angiomyolipoma is a rare benign mesenchymal tumor that usually presents in adult female patients; nonetheless, several cases have also been reported among males [2, 3]. Angiomyolipoma most frequently occur in the kidney, with the liver being the second most common site of involvement [3, 4].

CASE OUTLINE

A 44-year-old symptomless female was admitted to the Department of Surgery in order to have a lesion resection of the third segment of the liver. The patient suffered from posterior mitral valve prolapse with mild regurgitation (no medications) and underwent bilateral knee arthroscopy. Her grandmother has a liver tumor of unknown origin. Ultrasound abdominal examination revealed the liver lesion. Magnetic resonance imaging (MRI) showed the sharply contoured lesion of 30 mm (transverse) × 20 mm (anteroposterior) × 10 mm (craniocaudal), located in dorsal part of the third hepatic segment. The tumor manifested a slight T2 signal hyperintensity and fat-containing component, which quenched in the MRI p-phase. After administration of contrast agent, it was demonstrated that the lesion was heterogenous with enhancing linear partitions inside. In the hepatotropic phase the lesion was not enhanced. The MRI report was ambiguous, and the lesion might be: adenoma, lipoma, hepatocellular carcinoma or angiomyolipoma. The retroperitoneal lymph nodes were within normal limits. The biochemical tests, the tests for hepatic viruses and tumor markers were determined. No active liver disease nor impairment of the synthesizing function of the hepatocytes was revealed. The presence of anti-HBC total was confirmed in repeated tests. However, the other markers of hepatitis B infection were negative. The tumor markers were within normal limits: CA19-9 – 13.73 U/ml and CEA – 0.70 ng/ml. Due to inconclusive additional tests the patient was qualified for laparotomy with surgical resection of the lesion. Intraoperatively the soft tumor of the third hepatic segment measuring 35×30×15 mm was confirmed. Tumor was excised totally, and the margins of resection were clean (R0 resection). The intraoperative ultrasound did not show any other lesions within the liver (Figure 1 and 2). The postoperative course was uneventful. A drain from the subhepatic area was removed on the second postoperative day. The patient has been regularly followed up in the out-patient department.

Gross examination showed solid hepatic tumor. The tumor was gray-yellowish on cut surface. It did not have a capsule but it was well demarcated from liver parenchyma. Histologically, tumor was composed of large, epithelioid smooth muscle cells intermixed with few dispersed mature adipocytes. Smooth muscle cells had epithelioid morphology and abundant, clear to eosinophilic cytoplasm.

Received • Примљено: June 16, 2020
Revised • Ревизија: April 16, 2021
Accepted • Прихваћено: April 25, 2021
Online first: May 10, 2021

Correspondence to:
Mariusz CHABOWSKI
Fourth Military Teaching Hospital
Department of Surgery
5 Weigla street, 50-981 Wroclaw
Poland
mariusz.chabowski@gmail.com

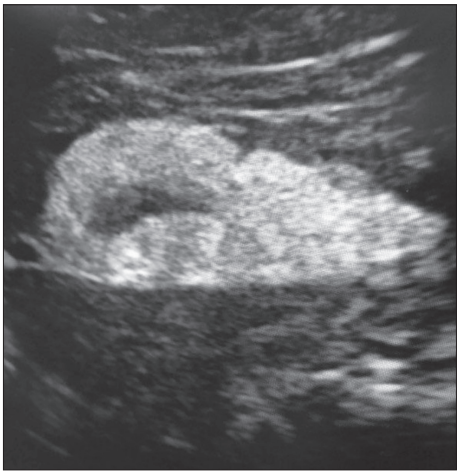

Figure 1. The intraoperative ultrasound view of liver angiomyolipoma

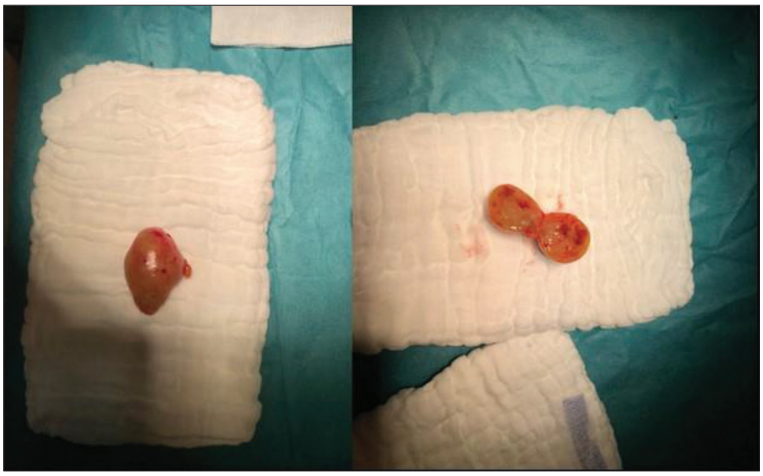

Figure 2. The intraoperative view of the lesion: the ocher tumor measuring 35×30×15 mm

Many blood vessels were also observed, some of them were thick walled. No necrosis, cellular atypia or mitotic figures were noticed. Smooth muscle cells presented characteristic co-expression of muscle markers (smooth muscle actin and focally Desmin) and melanocytic marker HMB45. Histological and immunohistochemical features were consistent with diagnosis of hepatic angiomyolipoma (Figures 3 and 4).

The study was approved by the Commission of Bioethics at Military Medical Chamber in Warsaw (no 173/20), and written informed consent in Polish was obtained from the patient for the publication of this paper.

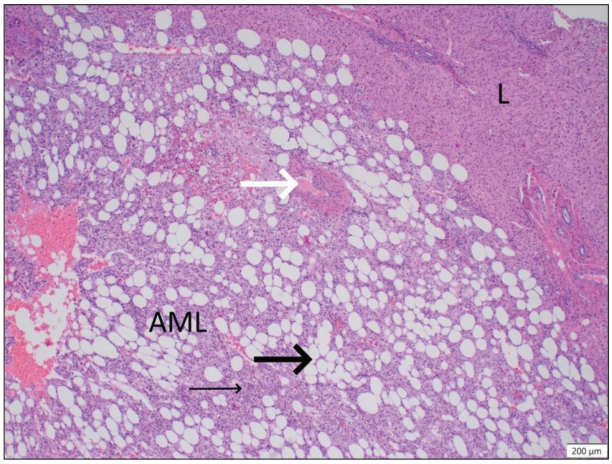

Figure 3. Hepatic angiomyolipoma; the tumor is well demarcated from liver parenchyma (L) and is composed of admixture of epithelioid smooth muscle cells (thin black arrow), mature adipocytes (thick black arrow) and blood vessels (white arrow) (hematoxylin and eosin stain, magnification 400×)

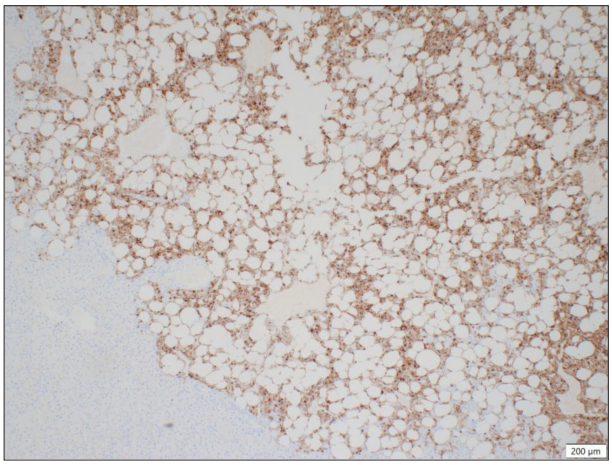

Figure 4. Positive cytoplasmic HMB45 staining of smooth muscle cells (magnification 400×)

DISCUSSION

Primary hepatic angiomyolipoma is a rare tumor. The first case was reported by Ishak [5] in 1976. Hepatic angiomyolipoma can be divided into ten groups based on the tissue components and type of dominant tissue:

(I) hybrid: typical and most common, contains similar proportions of each tissue components within the tumor;
(II) myomatous: smooth muscle cells are the dominant tissue type within the tumor;
(III) lipomatous: adipose tissue is the dominant tissue type;
(IV) angiomatous: vascular tissue is the dominant type;
(V) angiomyomatous;
(VI) myoangiomatous;
(VII) myolipomatous;
(VIII) lipomyomatous;
(IX) lipoangiomatous;
(X) angiolipomatous [6].

The origin of the liver angiomyolipoma is not clearly defined. Angiomyolipoma is associated with tuberous sclerosis in some cases [7]. The differential diagnosis based on radiological findings might be difficult. There are many liver disorders with fatty components, both benign and malignant, e.g., hepatic steatosis, adenoma, lipoma, hepatocellular carcinoma or liposarcoma. Those lesions might

be shown in ultrasound or computed tomography scan, but the MRI is the most sensitive tool [8]. Angiomyolipoma usually occurs as solitary tumor however, some multiple lesions have been described in literature [9]. In the presented case the diagnostics was extended to abdominal MRI with contrast, which allow to qualify whether the patient is for surgery. In MRI with contrast, angiomyolipoma tumors might be enhanced depending on the ratio of their fatty to vascular components. The complete lack of contrast enhancement may suggest liver lipoma [9].

Despite the fact that it is a benign neoplasm, malignant features such as infiltration of surrounding tissues, relapse after resection or distant metastases have sometimes been observed. The main predictor of malignancy is not so much the tumor size as its growth rate and the presence of atypical cells [10]. This rare, but non-negligible, potential for malignancy makes hepatic angiomyolipoma an even greater clinical problem. It is considered, that surgical management is suggested to patients with the following criteria: tumor size greater than 5 cm, with clinical symptoms, faster tumor growth, the tumor located at the first, fourth, fifth or eighth segment of the liver [11]. In case of a decision on conservative treatment, it is necessary to regularly monitor patients in order to observe the dynamics of the growth of the lesion and early detection of symptoms. However, sometimes large tumors may rupture and bleed. This causes the patient to develop acute abdominal symptoms and urgent surgery is required [12].

In conclusion, liver angiomyolipoma is a rare benign tumor, usually asymptomatic, and detected during routine radiological tests. The differential diagnosis with other hepatic tumors by means of radiological tests is very difficult and sometimes impossible. Its prognosis is good and the recommended treatment is surgical resection.

Conflict of interest: None declared.

REFERENCES

1. Lerut J, Iesari S. Vascular tumours of the liver: a particular story. Transl Gastroenterol Hepatol. 2018;3:62.
2. Damaskos C, Garmpis N, Garmpi A, Nonni A, Sakellariou S, Margonis GA, et al. Angiomyolipoma of the Liver: A Rare Benign Tumor Treated with a Laparoscopic Approach for the First Time. In Vivo. 2017;31(6):1169–73.
3. Zhou H, Guo M, Gong Y. Challenge of FNA diagnosis of angiomyolipoma: A study of 33 cases. Cancer Cytopathol. 2017;125(4):257–66.
4. Čolović R, Čolović N, Grubor N, Radak V, Micev M, Stojković M. Bilateral angiomyolipoma of the kidney in patient with tuberous sclerosis. Srp Arh Celok Lek 2005;133(9–10):433–7.
5. Ishak KG. Mesenchymal tumors of the liver. Hepatocellular carcinoma, Okuda K, Peters RL, eds. New York: John Wiley & Sons; 1976. p. 247.
6. Nonomura A, Enomoto Y, Takeda M, Takano M, Morita K, Kasai T. Angiomyolipoma of the liver: a reappraisal of morphological features and delineation of new characteristic histological features from the clinicopathological findings of 55 tumours in 47 patients. Histopathology. 2012;61(5):863–80.
7. Klompenhouwer AJ, Verver D, Janki S, Bramer WM, Doukas M, Dwarkasing RS, et al. Management of hepatic angiomyolipoma: A systematic review. Liver Int. 2017;37(9):1272–80.
8. Podgorska J, Anysz-Grodzicka A, Cieszanowski A. State-of-the-art MR Imaging of Fat-containing Focal Lesions of the Liver. Curr Med Imaging Rev. 2019;15(5):435–42.
9. Seow J, McGill M, Wang W, Smith P, Goodwin M. Imaging hepatic angiomyolipomas: key features and avoiding errors. Clin Radiol. 2020;75(2):88–99.
10. Machida M, Sugo H, Watanobe I. A huge hepatic angiomyolipoma with growth during 5 years of follow-up. J Surg Case Rep 2020;2020(9):rjaa353.
11. Yu F, Wang K, Yan ZL, Zhang XF, Li J, Dong H, et al. Clinical study of 169 patients with hepatic angiomyolipoma. Zhonghua Wai Ke Za Zhi. 2010;48(21):1621–4.
12. Mengesha RE, Esayas R. Bleeding with in a Huge Angiomyolypoma of Liver Presenting as an Acute Abdomen. HPB 2021;23(Suppl.1):S146.

Ангиомиолипом јетре

Филип Свјотковски[1,2], Дорота Зјелинска[1], Војћех Тањски[3], Павел Гајђис[4], Маријуш Чабовски[1,2]

[1]Четврта војнонаставна болница, Одељење хирургије, Вроцлав, Пољска;
[2]Медицински универзитет у Вроцлаву, Факултет здравствених наука, Одсек за онкологију и палијативну негу, Одељење за клиничку негу, Вроцлав, Пољска;
[3]Четврта војнонаставна болница, Одељење интерне медицине, Вроцлав, Пољска;
[4]Медицински универзитет у Вроцлаву, Факултет здравствених наука, Одељење за патологију, Вроцлав, Пољска

САЖЕТАК

Увод Бенигни тумори јетре су ретки. Ангиомиолипом јетре је редак бенигни мезенхимски тумор који се обично јавља код одраслих болесника. Постоје четири врсте ангиомиолипома јетре: (I) хибридни; (II) тип миома; (III) тип липома и (IV) тип хемангиома.

Приказ болесника Представљамо жену од 44 године, без симптома, која је примљена на одељење хирургије због лезије трећег сегмента јетре, димензија 35 × 30 × 15 милиметара. Тумор је у потпуности уклоњен, а маргине ресекције су биле чисте. Диференцијална дијагноза заснована на радиолошким налазима може бити тешка. Постоје многи поремећаји јетре са масним компонентама, и бенигни и малигни, нпр. хепатичка стеатоза, аденом, липома, хепатоцелуларни карцином или липосарком.

Закључак Прогноза је добра, а препоручени третман је хируршка ресекција.

Кључне речи: ангиомиолипома; тумор јетре; хирургија