agnostika i lechenie. Novyj vzglyad na problemu. Po materialam Proekta klinicheskih rekomendacij Rossijskoj associacii endokrinologov po diagnostike i lecheniyu pervichnogo giperal'dosteronizma [Primary hyperaldosteronism: diagnosis and treatment. A new look at the problem. According to the materials of the russian association of endocrinologists clinical guidelines for primary hyperaldosteronism diagnosis and treatment]. Consilium Medicum. 2017; 19(4): 75-85.

8. Osadchy O.E. Gipokaliemiya — klinicheskoe znachenie i rol' v mekhanizmah aritmogeneza serdca [Hypokalaemia: Clinical Signifi cance and Role in the Mechanisms Underlying Cardiac Arrhythmogenesis]. Kubanskij nauchnyj medicinskij vestnik [Kuban Scientific Medical Bulletin. 2019; 26(4): 94-106. DOI: 10.25207/1608-6228-2019-26-4-94-106 (In Russ.).]

9. Savin I.A., Goryachev A.S. Vodno-elektrolitnye narusheniya v nejroreanimacii [Water and electrolyte disorders in neurocritical car]. Izdanie vtoroe [Edition two, Moscow. 2016 (In Russ.).]

10. Sakovec T.G., Bogdanov E.I. Vtorichnye gipokaliemicheskie mioplegii [Secondary hypokalemic myoplegias]. Nevrologiya, nejropsihiatriya, psihosomatika [Neurology, Neuropsychiatry, Psychosomatics. 2019; 11(3): 52-56 (In Russ.).] DOI: 10.14412/2074-2711-2019-3-52-56.

11. Chihladze N.M. Arterial'naya gipotoniya u bol'nyh s vtorichnym giperal'dosteronizmom: sindromy Barttera i Gitel'mana [Arterial hypotension in patients with secondary hyperaldosteronism: Bartter and Gitelman syndromes]. Sistemnye gipertenzii [Systemic Hypertension. 2008; 3: 24-26 (In Russ.).]

12. Shurygina V.D. Diagnostika pervichnogo giperal'dosteronizma i algoritm vzaimodejstviya kardiologa i endokrinologa s pozicii kardiologa [Diagnosis of primary aldosteronism and the algorithm of interaction between a cardiologist and an endocrinologist from the position of a cardiologist]. Vestnik Sankt-Peterburgskogo universiteta. Medicina [Vestnik of Saint Petersburg University. Medicine. 2023; 18 (3): 214-232 (In Russ.).] DOI: 10.21638/spbu11.2023.301.

13. Blanchard A, Bockenhauer D, Bolignano D, et al. Gitelman syndrome: consensus and guidance from a Kidney Disease: Improving Global Outcomes (KDIGO) Controversies Conference Kidney international. 2017. 91(1). P.24–33. doi: 10.1016/j.kint.2016.09.046.

14. Ham Y., Mack H., Colville D., et al. Gitelman syndrome and ectopic calcification in the retina and joints. Clin Kidney J. 2021. 14(9). P.2023-2028. doi: 10.1093/ckj/sfab034.

15. Jiang R., Liu Q, Sun Y, et al. Finerenone as a Novel Treatment for Gitelman Syndrome: A Case Study of a 35-Year-Old Male with Adrenal Mass and Hypokalemia. Am J Case Rep. 2024. 25 (e944492). doi: 10.12659/AJCR.944492.

16. Louis Vincent, Frederick A. Moore, Rinaldo Bellomo, John J. Marini. Textbook of Critical Care. 8th Edition. 2022.

D.N. Kostromitsky, A.Yu. Dobrodeev, D.P. Koval,
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SURGICAL TREATMENT OF MESENCHYMAL HAMARTOMA OF THE LIVER: A CASE REPORT

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Mesenchymal hamartoma of the liver (MHL) is an uncommon benign tumor that mostly occurs in young children and is extremely rare in adults. Currently, only a few cases of MHL in adults have been reported. Due to the rarity of MHL and insufficient understanding of histogenesis of this tumor, this category of patients is most often followed-up by general surgeons and gastroenterologists. In our report, we present a case of a 45-year-old female patient with MHL. Before surgery, a differentiated diagnosis included cholangiocarcinoma and focal nodular hyperplasia. Attempts of morphological verification, including ultrasound-guided biopsies, were uninformative. The final diagnosis was established after laparoscopic anatomical liver resection with histological and immunohistochemical examination. This case highlights the importance of a comprehensive approach to the diagnosis of liver tumors in adults and the need to include MHL in the differential diagnosis in the case of atypical clinical and radiological presentation.

Keywords: mesenchymal liver hamartoma, benign liver tumor, laparoscopy, liver resection, pathomorphology

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Introduction. Mesenchymal hamartoma of the liver (MHL) is a benign tumor, which was first reported by H.A. Edmondson in the mid-20th century [3]. It generally occurs in young children and has been rarely observed in adults [7]. Currently, about 50 adult cases have been reported [4-6, 9-11]. Macroscopically, the tumor is a clearly defined mass varying in size from a few centimeters to 30 cm or more, and its structure may additionally contain a cystic component [9]. In 30% of cases, these tumors may have a vascular pedicle [10]. The microscopic image of MHL is characterized by lobular growth of myxomatous connective tissue containing scattered soft stellate mesenchymal cells. Branched bile ducts similar to ductal plate malformation may also often be present in the tumor [7].

Clinical symptoms of the disease are mild and depend on the location and size of the tumor. The most common complaints are general malaise, increased fatigue, and an increase in abdominal size [9]. It should be noted that approximately 75% of MHLs are located in the right lobe of the liver and most often manifest as a violation of the passage of the food bolus through the gastrointestinal tract,

weight loss, anorexia and respiratory failure. Most patients do not experience any symptoms that would prompt them to seek medical attention until the tumor has grown to a large size [10].

The etiology and pathogenesis of MHL is complex and poorly understood. To date, several theories of the origin of liver hamartoma have been described: 1) genetic, 2) vascular, 3) embryonic, and 4) toxic [1, 9, 10].

The main method of treating MHL is radical resection (R0). There is data on the development of angiosarcomas from MHL not only during follow-up for patients, but also over time after performing R1 resection [10]. Cases of liver transplantation have been also reported [1].

Case report. A 45-year-old female patient presented to the local clinic with complaints of general weakness and rapid fatigue. During the examination, moderate anemia of unknown origin was diagnosed. MRI of the abdominal cavity (12/23/2024) revealed 2 lesions in the left lobe of the liver (25×27 mm and 23×26 mm), closely adjacent to each other. Tumor markers, such as CEA, AFP, CA 19-9 were within normal limits. The patient was admitted then to the outpatient department of the Cancer Research Institute (Tomsk). The complexity of the diagnosis was caused by both the lack of convincing morphological findings for the tumor during two ultrasound-guided liver biopsies and the ambiguity of the CT image. The computed tomography scan of the abdomen with intravenous contrast (16.01.2025) demonstrated the presence of two merging hypodense lesions in the II+III segments of the left lobe of the liver, measuring 26×33 mm and 22×27 mm in diameter, located subcapsularly along the visceral surface. The lesions were clearly delineated, without signs of invasion into surrounding structures, with a homogeneous internal structure and no pronounced accumulation of contrast agent. Conclusion: tumor in the left lobe of the liver (cholangiocarcinoma, nodular hyperplasia of the liver) (Fig. 1).

The additional examination of the patient included chest X-ray, pelvic ultrasound, thyroid ultrasound, cervical and axillary lymph node ultrasound, breast ultrasound, video colonoscopy and esophagogastroduodenoscopy. The final clinical diagnosis was a tumor in II + III segments of the left lobe of the liver.

In January, 31, the patient underwent laparoscopic anatomical resection of segments II + III of the left lobe of the liver. The macroscopic examination of 9 × 11 cm specimen, including segments II + III of the left lobe of the liver, revealed a

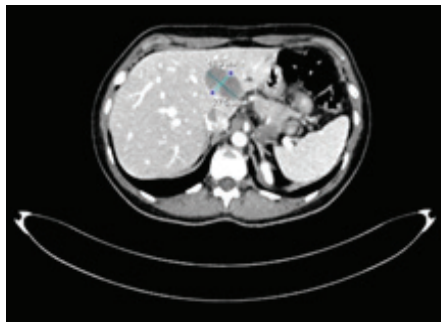


Fig. 1. CT scan of the abdominal cavity with intravenous contrast, axial section. Two merging hypodense lesions in the II+III segments of the left lobe of the liver, with total dimensions of 41×27 mm

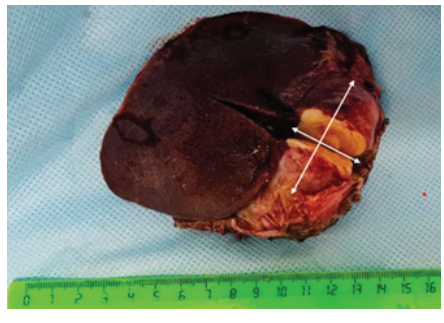


Fig. 2. Specimen macroscopy. A liver specimen measuring 9×11 cm with a subcapsular solid tumor measuring 3×5 cm. The tumor is clearly delimited from the surrounding parenchyma by a thick fibrous capsule, has a yellowish-amber color and a dense consistency

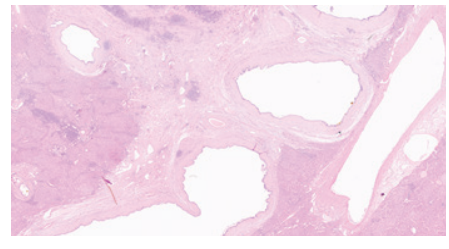
tumor of 3 × 5 cm in size. On the section, the tumor was amber in color, solid in structure with a pronounced capsule (Fig. 2).

The histological examination of the specimen revealed an encapsulated liver lesion with a thick fibrous capsule, heterogeneous in its morphological structure. The majority of cells in the lesion were spindle-shaped, with a normochromic nucleus, eosinophilic cytoplasm, and poorly distinguishable cytolemma. Mitotic figures were not determined. The cells formed bundle, vortex, perivascular structures lying in a moderate loose matrix with uneven diffuse-focal lymphoid infiltration. In places, the mesenchymal component alternated with liver parenchyma (Fig. 3) presented in the form of islets formed by typical hepatocytes. In some fragments, various-sized, cystically dilated, or branching bile ducts were identified. The bile ducts were surrounded by a moderately cellular fibrous stroma, in which numerous various-sized vessels and nerve fibers were seen (Fig. 4).

Considering the predominance of the mesenchymal component with a spindle cell structure in the tumor (Fig. 3), the differential diagnosis was mainly focused on excluding other tumors with similar morphology. It was necessary to exclude gastrointestinal stromal tumor (GIST), as well as inflammatory myofibroblastic tumor, which can demonstrate similar spindle cell and stromal-inflammatory histoarchitecture. In this regard, an extended immunohistochemical study with a panel of antibodies, including markers of mesenchymal, smooth muscle, neural and epithelial differentiation, was carried out.

The negative expression of S100 (polyclonal, Cell Marque), Cytokeratins (clone AE1/AE3, Leica), DOG-1 (clone (SP31, Cell Marque), CD34 (clone QBEnd 10, Leica), Calponin (clone CALP, Leica), SMA (clone asm-1, Leica) (Fig. 6), desmin (clone D33, Leica) and ALK (clone 5A4, Leica Biosystems) (Fig. 4) excluded the diagnosis of GIST and inflammatory

a



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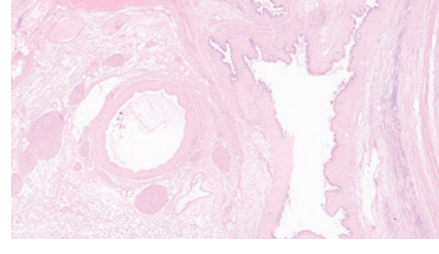

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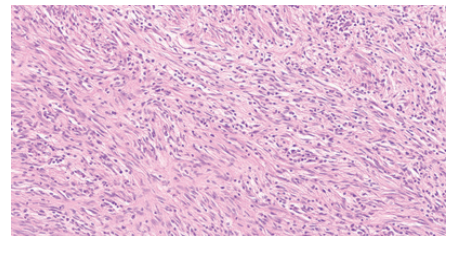

Fig. 3. Histological examination: a - fragment of a tumor with an area of preserved hepatic parenchyma, represented by typical hepatocytes, against the background of loose mesenchymal stroma, hematoxylin-eosin staining, UV. × 40; b - bile ducts surrounded by fibrous stroma. morphology resembles ductal plate malformation, hematoxylin-eosin staining, uv. × 100; c - the tumor component is represented by spindle-shaped cells with a fascicle and vortex arrangement in the connective tissue matrix with scattered lymphocytes, stained with hematoxylin-eosin, UV. × 200

myofibroblastic tumor. In the mesenchymal component, staining was observed only with the antibody to Vimentin (clone V9), and a low rate of proliferative activity was observed in the tumor cells (Ki-67 index less than 5%) (fig. 4). The results of immunohistochemical and histochemical analysis allowed us to verify the benign tumor and morphologically confirm the diagnosis of MHL.

Thus, based on a comprehensive examination and planned morphological study, the final diagnosis was: mesenchymal hamartoma of the left lobe of the liver in segments II + III. In January 2025, the patient underwent laparoscopic anatomical resection (R0) of segments II + III in the left lobe of the liver. The postoperative period was uneventful, and the patient was discharged from the hospital on the 6-th postoperative day. The patient is being followed-up and no recurrence has been noted.

Discussion. Mesenchymal hamartoma of the liver (MHL) is a benign tumor with a reported incidence of less than 1 case per million population per year [8]. Most of them are diagnosed during childhood and they are exceptional in adults [3, 7, 9]. Currently, approximately 50 cases of MHL in adults have been reported [4-6, 9-11]. As mentioned above, the diagnosis of MHL is difficult due to the lack of clinical symptoms specific to this disease, laboratory and radiological data, and the difficulty in differential diagnosis with cystadenoma, cholangiocarcinoma, nodular hyperplasia of the liver and embryonic sarcoma of the liver [2, 7, 8, 11]. The choice of strategy in the management of patients with MHL is of great importance. On the one hand, the possible malignant transformation of mesenhymal hamartoma is reported; on the other hand, there are risks of developing perioperative complications, including profuse bleeding, biliary fistulas and liver failure. All this emphasizes the rarity of this pathology, the relevance of diagnostic and treatment problems, and also clearly demonstrates the clinical significance and uniqueness of the case report we present.

The particular diagnostic difficulty in this case was associated with the absence of pathognomonic clinical and radiological signs. Thus, the patient presented non-specific complaints only of weakness and fatigue, and during the examination, anemia of unknown genesis was detected. This can be considered as a manifestation of paraneoplastic syndrome, especially in tumors with a pronounced inflammatory component. Differentiation of benign from malignant

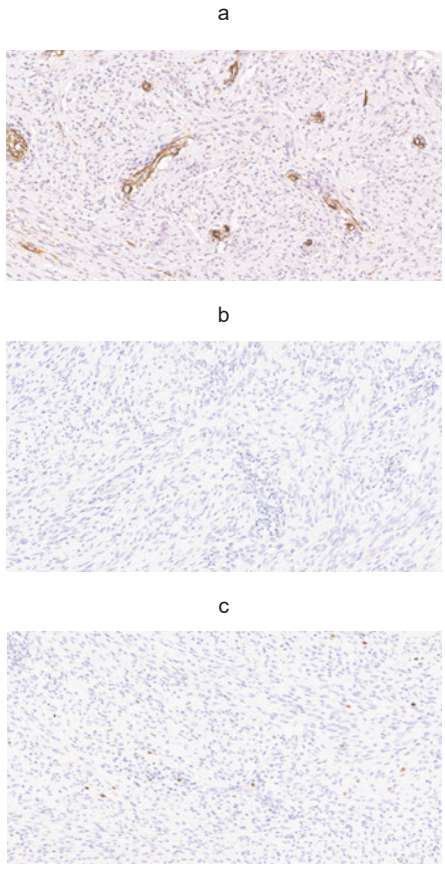

Fig. 4. Immunohistochemical study: a - negative expression of smooth muscle actin (SMA) in tumor cells with positive control in vascular smooth myocytes, reaction to SMA, uv. × 200; b - lack of expression of ALK (clone 5A4) in tumor elements, reaction to ALK, uv. × 200; c - weak proliferative activity of the tumor (Ki-67 index less than 5%), response to Ki-67, uv. × 200

tumors was not possible using CT, and repeated ultrasound-guided biopsies of the liver lesion were uninformative. Thus, the diagnosis was established only after surgery, which indicates the limitations of non-invasive diagnostic methods in cases of atypical course of rare benign liver tumors.

Macroscopically, the tumor was a solid amber-colored node with a dense capsule, which did not exclude the possibility of a malignant neoplasm. Microscopically, there was a spindle cell component located in a loose stroma, with areas of lymphoid infiltration and perivascular structures.

Differential diagnosis included GIST, inflammatory myofibroblastic tumor, and solitary fibrous tumor.

An extensive immunohistochemical study, including both epithelial and mesenchymal markers, was critical for establishing the diagnosis. The negative expression of DOG-1, CD34, SMA, calponin, desmin and ALK excluded the diagnosis of GIST and inflammatory myofibroblastic tumor. The diagnosis of MHL was supported by the presence of cystically dilated and branching bile ducts, surrounding fibrous structures and neurovascular bundles resembling a ductal plate malformation [7]. Low levels of proliferative activity (Ki-67 less than 5%) and the absence of cytological atypia also confirmed the benign nature of the lesion.

MHL may be a potentially premalignant lesion, with reported cases of malignant transformation to angiosarcoma in adults or to undifferentiated embryonal sarcoma in children [1, 9, 10]. These data justify an aggressive treatment strategy, with preference given to radical resection (R0). In some cases, liver transplantation may be considered for unresectable tumors [1].

Our case demonstrates the key challenges that clinicians and pathologists face when diagnosing rare liver tumors in adults. It highlights the need for a comprehensive multidisciplinary approach that includes imaging methods, clinical and laboratory data, comprehensive morphological and immunohistochemical studies, as well as knowledge of cancer-associated risks in benign neoplasms.

Conclusion. Mesenchymal hamartoma of the liver (MHL) is a benign lesion that is extremely rare in adults. This case report demonstrates the complexity of diagnosing MHL with atypical clinical and radiological manifestations, as well as the limitations of minimally invasive methods for diagnosis verification. A definitive diagnosis of MHL is possible only after comprehensive histopathological analysis of the resected specimen. Our case report confirms the need to include MHL in the differential diagnosis of liver tumors in adults and indicates the importance of a multidisciplinary approach that ensures accurate morphological verification and optimal treatment strategy.

The authors declare no conflict of interest.

References

1. Brownfield M.N., Adeyemi A., Tumolo N., et al. Infant With Beckwith-Wiedemann Requiring Transplant for Hepatic Mesenchymal Hamartoma. Pediatr Transplant. 2025 Feb;29(1):e70015. doi: 10.1111/petr.70015

2. Choi J.H., Thung S.N. Mesenchymal Tumors of the Liver: An Update Review. Biomedicines. 2025 Feb 15;13(2):479. doi: 10.3390/biomedicines13020479.

3. Edmondson H.A. Differential diagnosis of tumors and tumor-like lesions of the liver in infancy and childhood. AMA J Dis Child. 1956;91:168-86. doi: 10.1001/archpedi.1956.02060020170015.

4. Gou X.N., Xu W., Dong Z.H., Wang Z.B. [Clinicopathological analysis of adult hepatic mesenchymal hamartoma]. Zhonghua Gan Zang Bing Za Zhi. 2024 Jan 20;32(1):58-63. Chinese. doi: 10.3760/cma.j.cn501113-20231124-00230.

5. Idrees M., Chung K., Philipoff A., et al. Liver Transplant for Adult Recurrent Hepatic Mesenchymal Hamartoma and a Feasible Treatment Modality: A Case Report and Literature Review. Transplant Proc. 2022 Jul-Aug;54(6):1636-1639. doi: 10.1016/j.transproceed.2022.04.020.

6. Lee C., Yu J., Chang Y., Huang J. A Case Report of a Female Patient Who Presented a Hepatic Mesenchymal Hamartoma that Grew Since Childhood. Tungs’ Medical Journal. 2020;14:42-47. doi: 10.3966/207135922020061401008.

7. Martins-Filho S.N., Putra J. Hepatic mesenchymal hamartoma and undifferentiated embryonal sarcoma of the liver: a pathologic review. Hepat Oncol. 2020 Apr 7;7(2):HEP19. doi: 10.2217/hep-2020-0002.

8. Mocellin S. Hepatic Mesenchymal Hamartoma. In: Soft Tissue Tumors. 2021, Springer, Cham. doi: 10.1007/978-3-030-58710-9_124.

9. Mulrooney D.A., Carpenter B., Georgieff M., et al. Hepatic mesenchymal hamartoma in a neonate: a case report and review of the literature. J Pediatr Hematol Oncol. 2001 Jun-Jul;23(5):316-7. doi: 10.1097/00043426-200106000-00017.

10. Pinelli D., Guerci C., Cammarata F., et al. Huge mesenchymal hamartoma in a young adult: a case report. J Surg Case Rep. 2024 Apr 1;2024(4):rjae184. doi: 10.1093/jscr/rjae184.

11. Reza R., Nigam N., Kumar B. et al. Mesenchymal Hamartoma of the Liver: A Clinicopathological Analysis of Patients at a Tertiary Care Facility. Indian J Surg Oncol. 2025. doi:10.1007/s13193-024-02179-3.