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CASE REPORT

Liver transplantation for a giant mesenchymal hamartoma of the liver in an adult: Case report and review of the literature

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Abstract

Mesenchymal hamartomas of the liver (MHLs) in adults are rare and potentially premalignant lesions, which present as solid/cystic neoplasms. We report a rare case of orthotopic liver transplantation in a patient with a giant MHL. In 2013, a 34-year-old female sought medical advice after a 2-year history of progressive abdominal distention and respiratory distress. Physical examination revealed an extensive mass in the abdomen. Computed tomography (CT) of her abdomen revealed multiple liver cysts, with the diameter of largest cyst being 16 cm × 14 cm. The liver hilar structures were not clearly displayed. The adjacent organs were compressed and displaced. Initial laboratory tests, including biochemical investigations and coagulation profile, were unremarkable. Tumor markers, including levels of AFP, CEA and CA19-9, were within the normal ranges. The patient underwent orthotopic liver transplantation in November 2013, the liver being procured from a 40-year-old man after cardiac death following traumatic brain injury. Warm ischemic time was 7.5 min and cold ischemic time was 3 h. The recipient underwent classical orthotopic liver transplantation. The recipient operative procedure took 8.5 h, the anhepatic phase lasting for 1 h without the use of venovenous bypass. The immunosuppressive regimen included

intraoperative induction with basiliximab and high-dose methylprednisolone, and postoperative maintenance with tacrolimus, mycophenolate mofetil, and prednisone. The recipient's diseased liver weighed 21 kg (dry weight) and measured 41 cm × 32 cm × 31 cm. Histopathological examination confirmed the diagnosis of an MHL. The patient did not experience any acute rejection episode or other complication. All the laboratory tests returned to normal within one month after surgery. Three months after transplantation, the immunosuppressive therapy was reduced to tacrolimus monotherapy, and the T-tube was removed after cholangiography showed no abnormalities. Twelve months after transplantation, the patient remains well and is fulfilling all normal activities. Adult giant MHL is extremely rare. Symptoms, physical signs, laboratory results, and radiographic imaging are nonspecific and inconclusive. Surgical excision of the lesion is imperative to make a definite diagnosis and as a cure. Liver transplantation should be considered as an option in the treatment of a non-resectable MHL.

Key words: Liver; Mesenchymal hamartoma; Adult; Organ donor; After cardiac death; Transplantation

Core tip: Mesenchymal hamartoma of the liver is a rare disease in adults. Only 45 patients with this condition have been reported worldwide. This report presents a rare case of adult giant mesenchymal hamartoma of the liver that could not be treated by partial hepatectomy. Orthotopic liver transplantation relieved compression of other organs and avoided the risk of malignant change. Liver transplantation should be considered as an option in the treatment of non-resectable benign hepatic tumors.

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INTRODUCTION

Mesenchymal hamartoma of the liver (MHL) was first described by Edmondson in 1956[1]. It is a rare mesenchymal tumor affecting almost exclusively infants and children in the first two years of life, with a slight male predilection. Its occurrence in children older than five years is rare (about 5% of cases) and is extremely rare in adults[2-4]. MHL is a potentially premalignant lesion that presents as a solid/cystic neoplasm. The patient's symptoms are typically nonspecific, though abdominal pain is the most common. Laboratory results are noncontributory and radiographic imaging is variable and inconclusive. Needle biopsy is rarely diagnostic and surgical excision of symptomatic or enlarging lesions is recommended to exclude the possibility of malignancy and to establish a diagnosis[5].

CASE REPORT

A 34-year-old, previously healthy, woman presented in 2011 with abdominal fullness and loss of appetite. She took no medications, had no history of liver disease, and denied alcohol and drug use, including the use of anabolic steroids. She presented to our hospital with increasing abdominal girth, abdominal pain, and vomiting. Physical examination revealed a grossly distended abdomen without evidence of ascites, a firm and massively enlarged liver extending below the umbilicus, and tenderness in the upper quadrant. Contrast enhanced computed tomography (CT) of the abdomen revealed near replacement of the liver with diffuse cystic masses of low density (Figure 1). Initial laboratory test results were unremarkable. Hematological, biochemical investigations and the coagulation profile were within normal limits. Tumor markers, including levels of α-fetoprotein, and carcinoembryonic antigen, carbohydrate antigen 19-9, were within the normal ranges. Serology for hepatitis B virus, hepatitis C virus and human immunodeficiency virus was negative. The extensive hepatic involvement precluded resection, and so she was evaluated and placed on the waiting list for liver transplantation.

The patient underwent orthotopic liver transplantation in November 2013. Our techniques of organ procurement and preservation have been previously described[6,7]. The liver graft was procured from a 40-year-old male donor after cardiac death. The liver graft was preserved in 4℃ UW solution. The warm ischemia time was 7.5 min and cold ischemia time was 3 h.

The native diseased liver filled about 80% of the abdominal cavity and displaced the normal vascular anatomy. The excised diseased native liver weighed 20 kg (dry weight) and measured 41 cm × 32 cm × 31 cm (Figure 2). The recipient operation was conducted according to the classical orthotopic liver transplantation procedure[8]. The whole transplant procedure took 8.5 h and the total blood volume loss was 5500 mL. A blood reinfusion system replaced 3000 mL, and an additional 10 units of packed RBC and 1000ml of plasma were infused. The anhepatic phase lasted for 1 h without the use of venovenous bypass. After release of the vascular clamps, Doppler ultrasound demonstrated the liver graft to be well perfused (Figure 3). The patient was extubated on the second day after surgery.

The immunosuppressive regimen included intra-operative induction with basiliximab and high-dose methylprednisolone, and postoperative maintenance with tacrolimus, mycophenolate mofetil and prednisone. No acute rejection episode was documented. The

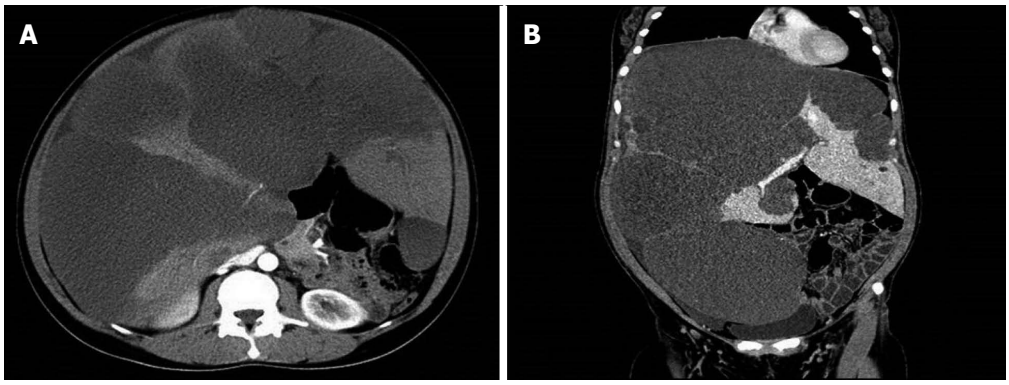

Figure 1 Contrast enhanced computed tomography of the abdomen revealed the near replacement of the liver with diffuse cystic masses of low density. A: Enhanced computed tomography scan shows the near replacement of the liver with diffuse cystic masses, leaving only small amounts of liver parenchyma. The portal vein and inferior vena cava are obviously compressed; B: The massively enlarged liver essentially occupies the entire abdominal cavity, with other abdominal organs being compressed and displaced.

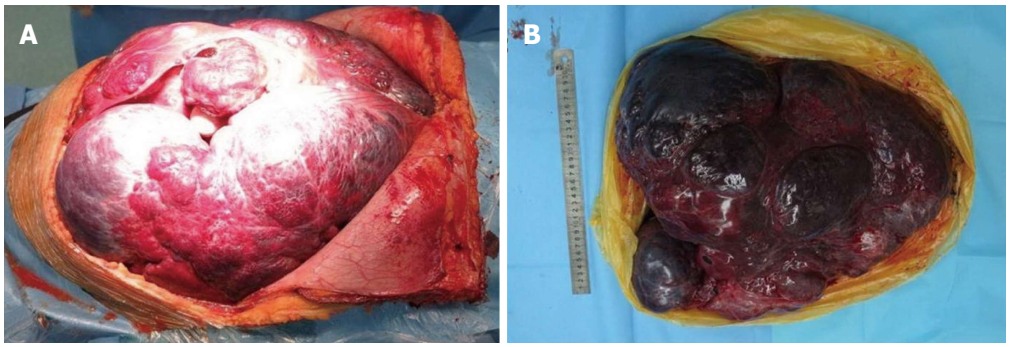

Figure 2 Intraoperative view of the tumor mass (A) and the excised liver (B).

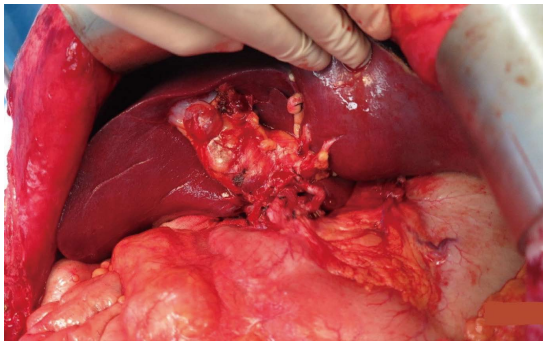

Figure 3 View of the operative field after liver transplantation, demonstrating the well-perfused liver graft.

patient was discharged home on postoperative day 20, at which time all laboratory tests were within normal limits. Three months after the operation, the immunosuppressive regimen was reduced to tacrolimus monotherapy, and the T-tube was removed after cholangiography showed no abnormalities. After 12 mo, the patient remains well and is carrying out all normal activities.

Pathologic examination of the excised diseased native liver was carried out. It contained multiple well-circumscribed masses, ranging in diameter from 2-16 cm. All masses were cystic in the central portion and contained 20-50 mL of muddy yellowish or bloody fluid. The liver mass contained dilated bile ducts with connective tissue forming multiple cysts. Histologically, corresponding to the cystic areas noted grossly, myxoid stroma and spindle cells showed smooth muscle differentiation, confirmed by positive staining for vimentin and smooth muscle actin. Benign dilated bile ducts were confirmed by positive staining for cytokeratin 7. In peripheral areas, only small amounts of liver tissue remained, with a lack of lobular architecture. There was a clear boundary between the liver parenchyma and proliferating connective tissue (Figure 4). The diagnosis of MHL was based on the typical morphological appearance, as described above.

DISCUSSION

MHL was first reported by Maresch in 1903[9]. Until relatively recently, this disease was known by different names, such as cavernous lymph adenomatoid tumor, bile cell fibroadenoma and benign mesenchymoma. The first definitive description of MHL was provided by Edmondson[1]. While the precise pathogenesis of MHL is uncertain, the most common theory relates to aberrant mesenchymal development in the portal tract, likely related to the bile ducts[10,11].

The clinical presentation of MHL appears to depend

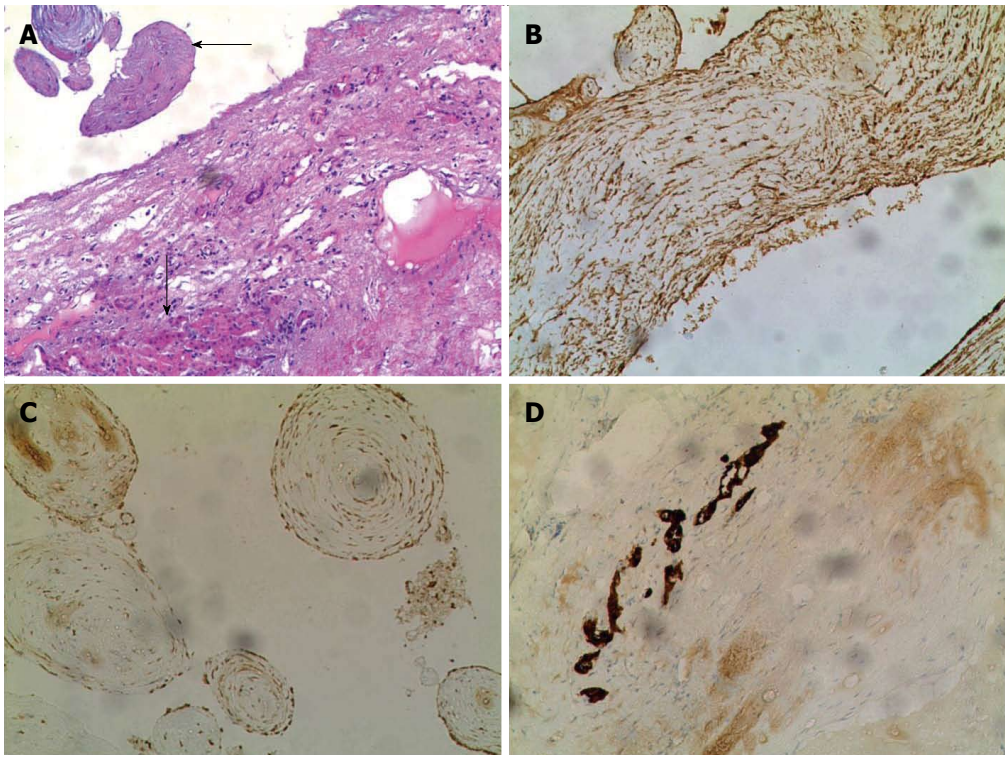

Figure 4 Clear boundary between liver parenchyma and proliferating connective tissue. A: The mass consisted of loose connective tissue full of myxoid matrix forming visible cysts (upper arrow). Small amounts of remaining liver tissue, with a lack of lobular architecture, were located in peripheral areas (lower arrow) (HE, original magnification × 100); Myxoid stroma with spindle cells showing smooth muscle differentiation were confirmed by positive staining for vimentin (B) and smooth muscle actin (C) (original magnification × 100); Benign dilated bile ducts were confirmed by positive staining for cytokeratin 7 (D) (original magnification × 100).

on the age of the patient. Most pediatric patients present with painless abdominal enlargement, normally appreciated by their parents[5]. However, in adult patients (age range: 19-87 years; females 62%, mean age 39 years; males 40%, mean age 60 years (Table 1), clinical features included hepatomegaly, and diffuse abdominal pain or pain in the right hypochondrium or left upper quadrant[12-14]. In severe cases, there may be compression of the diaphragm and lungs causing respiratory difficulties[4]. In the present case, the patient suffered from progressive abdominal distention and respiratory distress caused by the expanding multiple cystic masses distributed throughout the liver.

Concerning the localization and structure of the tumor, pediatric and adult populations have different characteristics. MHLs are more common in the left liver lobe in children. In adults, 17 cases (38%) were localized to the left lobe, 22 (49%) to the right lobe, and in six (13%) extended into both lobes (Table 1). All six cases of MHLs involving both lobes occurred in females. Among 45 cases of MHLs, 30 (67%) presented with cystic lesion, 12 (26%) with solid lesions, and three (7%) with both types. Of 30 cases of cystic MHLs, 21 (70%) were reported in females and only nine (30%) in males (Table 1).

MHLs are difficult to diagnose by laboratory tests or other investigations because of its non-specificity. Liver function tests and AFP values for MHLs are usually within normal limits[15]. Additionally, all imaging methods, including ultrasonography, CT and magnetic resonance imaging (MRI), provide nonspecific findings. The differential diagnosis of a cystic MHL includes simple liver cysts, hydatid cysts, biliary cystadenocarcinoma, and cystic metastases. If a lesion consists of a solid mass, the differential diagnosis includes focal nodular hyperplasia, hepatic adenoma, cavernous hemangioma, angiomyolipoma and hepatocellular carcinoma. In the present case, the initial abdominal enhanced CT scan revealed multiple liver cysts, which could easily have been misdiagnosed as a polycystic liver.

The diagnosis of MHL often relies on histological examination of tissue obtained by biopsy or by tumor resection; however, the histological appearance of the stromal component of an MHL can be variable. Hematoxylin and eosin (HE) staining, as well as immunohistochemical studies, have indicated MHLs as having spindle cells positive for vimentin and smooth muscle actin and negative for CD31, CD34 and S100 proteins, while the ducts stain positive for cytokeratin 7 and negative for cytokeratin 20[13,16].

MHLs have premalignant potential, particularly in adult patients[17]. The potential malignant evolution of a subset of MHLs into embryonal sarcoma or angiosarcoma supports the necessity for complete surgical excision both in children and adults[4,18]. Incomplete resection or marsupialization must be avoided because of the possibility of recurrence[19-21].

Table 1 Cases of adult mesenchymal hamartomas of the liver reported in the literature

No.	Ref.	Year	Sex	Age (yr)	Clinical manifestation	Size (cm)	Gross appearance (cystic or solid)	Liver lobe(s) affected	Surgical treatment
1	Yamamura *et al*[26]	1976	F	22	NA	NA	Cystic	Both	NA
2	Grases *et al*[27]	1979	F	19	Abdominal pain, jaundice, hepatomegaly	24 × 19 × 8	Cystic	Left	Left hepatic lobectomy
3	Li *et al*[28]	1983	F	21	Asymptomatic	17 × 10	Cystic	Right	Hemihepatectomy
4	Kawata *et al*[29]	1984	F	43	NA	22 × 15 × 10	Solid	Left	NA
5	Ishizuka *et al*[30]	1985	M	59	NA	30 × 28 × 12	Cystic	Right	NA
6	Kawakami *et al*[31]	1986	M	67	NA	NA	Cystic	Right	NA
7	Jennings *et al*[32]	1987	F	32	Asymptomatic	14 × 11	Cystic	Left	Left hepatic lobectomy
8	Kato *et al*[33]	1988	M	66	Asymptomatic	NA	Solid	Left	Left hepatic lobectomy
9	Gutierrez *et al*[34]	1988	F	30	NA	18	Both	Both	Non-resectable
10	Gramlich *et al*[35]	1988	F	28	Abdominal distention, hepatomegaly	30 × 20 × 14	Solid	Right	Right hepatic trisegmentectomy
11	Alanen *et al*[36]	1989	F	20	Asymptomatic	6 × 8	Cystic	Left	Left hepatic lobectomy
12	Ito *et al*[37]	1989	F	43	NA	16 × 16 × 7.7	Cystic	Both	NA
13	Urabe *et al*[38]	1990	F	39	Asymptomatic	1.2	Solid	Left	Left hepatic lobectomy
14	Drachenb *et al*[39]	1991	F	69	Asymptomatic	26 × 20 × 11.5	Cystic	Left	NA
15	Wada *et al*[40]	1992	M	62	Asymptomatic	6 × 6 × 4.5	Solid	Left	Hepatectomy
16	Chau *et al*[41]	1994	M	53	Abdominal pain	20 × 14 × 10	Cystic	Right	NA
17	Megremis *et al*[42]	1994	F	56	Abdominal pain	7.5	Cystic	Both	NA
18	Yamamoto *et al*[43]	1994	M	52	Abdominal discomfort, weight loss	6 × 4 × 3.5	Cystic	Left	Lateral segmentectomy
19	Chung *et al*[44]	1999	F	57	Abdominal discomfort, weight loss	6 × 4 × 3.5	Solid	Right	Right hepatectomy
20	Papastratis *et al*[45]	2000	F	21	Abdominal pain, abdominal mass	17 × 10	Cystic	Right	Right hepatectomy
21	Cook *et al*[13]	2002	F	46	Abdominal pain	6 × 4 × 5	Cystic	Right	Right hepatectomy
22	Cook *et al*[13]	2002	F	66	Cough and shortness of breath	5 × 4 × 2	Cystic	Right	Right hepatectomy
23	Cook *et al*[13]	2002	F	63	Abdominal pain	11 × 16 × 24	Solid	Left	Left hepatic lobectomy
24	Mao *et al*[46]	2002	M	44	Abdominal discomfort	2 × 2	Solid	Left	Hepatectomy
25	Mao *et al*[46]	2002	F	43	Asymptomatic	3 × 4 × 4	Cystic	Right	Right hepatectomy
26	Mao *et al*[46]	2002	M	76	Abdominal pain	4 × 5 × 4	Cystic	Right	Right hepatectomy
27	Brkic *et al*[47]	2003	M	38	Abdominal pain	8 × 5	Solid	Right	Right hepatectomy
28	Kim *et al*[48]	2003	M	NA	Asymptomatic	5	Both	Right	NA
29	Yesim *et al*[12]	2005	F	54	NA	2.5 × 2.5 × 1.5	Cystic	Left	Total cystectomy
30	Yesim *et al*[12]	2005	F	51	NA	6 × 7 × 8	Cystic	Right	Unroofing procedure
31	Kim *et al*[49]	2006	F	40	Asymptomatic	5 × 5	Cystic	Right	Right hepatectomy
32	Ayadi-Kaddour *et al*[50]	2006	F	21	NA	11 × 5	Cystic	Left	NA
33	Hernández *et al*[25]	2006	M	51	NA	19 × 13	Solid	Right	Liver transplantation (4th reported[1])
34	Chang *et al*[51]	2006	M	79	Asymptomatic	2 × 2	NA	Right	NA
35	Chang *et al*[51]	2006	F	39	Asymptomatic	5 × 5	Cystic	NA	NA
36	Li *et al*[17]	2007	F	33	Abdominal distention	16	Both	Both	NA
37	Mori *et al*[52]	2008	F	36	Abdominal distention	20 × 15 × 10	Cystic	Right	Right hemihepatectomy
38	Giunippero *et al*[53]	2009	M	87	Abdominal distention	20 × 20	Cystic	Right	Hemihepatectomy
39	Nakajo *et al*[54]	2009	M	38	Asymptomatic	5 × 5	Solid	Right	Right hepatectomy
40	Klaassen *et al*[5]	2010	F	53	NA	9 × 9 × 7.5	Cystic	Right	Hepatectomy
41	Kulkarni *et al*[55]	2010	F	20	Abdominal mass, abdominal pain	14 × 11	cystic	Right	Mass resection
42	Tucker *et al*[56]	2012	W	74	Abdominal distention, abdominal pain	18 × 15 × 13	cystic	Left	Left hepatectomy
43	Liu *et al*[57]	2013	M	42	Asymptomatic	1.5 × 1.0 × 1.0	solid	Left	Hepatectomy
44	Lakić *et al*[58]	2014	M	44	Asymptomatic	2.9 × 3.1 × 3.5	NA	Left	Hepatectomy
45	Sharma *et al*[59]	2014	M	81	Abdominal distention	21.8 × 12.3 × 18.6	cystic	Left	Hepatectomy

46	Current case	2014	F	34	abdominal discomfort, dyspnea	41 × 32 × 31	cystic	Both	Liver transplantation (5th reported[1])

[1]The first three cases of liver transplantation for mesenchymal hamartomas of the liver were reported in pediatric patients. NA: Not available.

Laparoscopic liver resection for MHLs has been reported with successful outcomes[22].

Very rarely an MHL is non-resectable, even in an experienced center, and liver transplantation may have to be considered. Tepetes *et al*[23] reported two children who underwent liver transplantation following partial resections for MHLs. One died from intraoperative bleeding and the other survived. Bejarano *et al*[24] described a neonate with a recurrent MHL (after resection) who underwent successful liver transplantation. Hernández *et al*[25] reported the first case of an MHL in an adult that was treated by liver transplantation.

In conclusion, giant MHLs in adults are extremely rare. Clinical features, laboratory results and radiographic imaging are often nonspecific and inconclusive. Surgical excision of the whole lesion is imperative for both definitive diagnosis and cure. Liver transplantation should be considered as an option in the treatment of non-resectable MHLs.

COMMENTS

Case characteristics

A 34-year-old female with a history of progressive abdominal distention and respiratory distress.

Clinical diagnosis

Physical examination revealed a grossly distended abdomen without evidence of ascites, a firm and massively enlarged liver extending below the umbilicus, and tenderness in the upper quadrant.

Differential diagnosis

Polycystic liver, hydatid cyst, biliary cystadenocarcinoma and cystic metastases.

Laboratory diagnosis

Laboratory test results were unremarkable and non-diagnostic.

Imaging diagnosis

Abdominal computed tomography scan showed multiple liver cysts, with the diameter of the largest cyst being 16 cm × 14 cm. The liver hilar structures were not clearly displayed. The adjacent organs were compressed and displaced.

Pathological diagnosis

Histological examination showed dilated bile ducts and extensive connective tissue in the liver mass, while immunohistochemical staining showed positivity for vimentin, smooth muscle actin and cytokeratin 7.

Treatment

The patient underwent orthotopic liver transplantation.

Related reports

Mesenchymal hamartoma of the liver is a rare disease in adults and only 45 patients with this condition have been reported; the references are cited.

Term explanation

Mesenchymal hamartoma of the liver is a rare and potentially premalignant lesion that presents as a solid/cystic neoplasm. The pathogenesis remains incompletely understood; however, these lesions have generally been considered to represent a developmental abnormality in bile duct plate formation.

Experiences and lessons

This case report represents a successful application of liver transplantation for adult giant mesenchymal hamartomas of the liver, which could not be treated by conventional partial hepatectomy. We recommend that liver transplantation should be considered as an option in the treatment of non-resectable benign hepatic tumors.

Peer-review

This paper is a case report of a 34-year-old woman with liver transplantation for a giant mesenchymal hamartoma of the liver. Mesenchymal hamartoma of liver is a rare disease in adults and only 31 patients have been reported to date worldwide.

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