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...
common. Laboratory results are noncontributory and nonspecific, though abdominal pain is the most neoplasm. The patient’s symptoms are typically premalignant lesion that presents as a solid/cystic is extremely rare in adults older than five years is rare (about 5% of cases) and a slight male predilection. Its occurrence in children infants and children in the first two years of life, with mesenchymal tumor affecting almost exclusively Mesenchymal hamartoma of the liver (MHL) was INTRODUCTION DOI: http://dx.doi.org/10.3748/wjg.v21.i20.6409 URL: World J Gastroent of the liver in an adult: ZY . Liver transplantation for a giant mesenchymal hamartoma Li J, Cai JZ, Guo QJ, Li JJ, Sun XY , Hu ZD, Cooper DKC, Shen 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 MHL.

Key words: Liver; Mesenchymal hamartoma; Adult; Organ donor; After cardiac death; Transplantation © The Author(s) 2015. Published by Baishideng Publishing Group Inc. All rights reserved.

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°C 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 intraoperative induction with basiliximab and high-dose methylprednisolone, and postoperative maintenance with tacrolimus, mycophenolate mofetil and prednisone. No acute rejection episode was documented. The
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
on the age of the patient. Most pediatric patients present with painless abdominal enlargement, normally appreciated by their parents. 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. In severe cases, there may be compression of the diaphragm and lungs causing respiratory difficulties. 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 nonspecificity. Liver function tests and AFP values for MHLs are usually within normal limits. 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.

MHLs have premalignant potential, particularly in adult patients. 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. Incomplete resection or marsupialization must be avoided because of the possibility of recurrence.

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).
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[(36)] | 1976 | F   | 22       | NA                     | 24 × 19 × 8 | Cystic                             | Both                 | NA                |
| 2   | Grases et al[(37)] | 1979 | F   | 19       | Abdominal pain, jaundice, hepatomegaly | 17 × 10 | Cystic                             | Right                | Hemihepatectomy |
| 3   | Li et al[(38)] | 1983 | F   | 21       | Asymptomatic           | 6 × 8      | Cystic                             | Left                 | Left hepatic lobectomy |
| 4   | Kawata et al[(39)] | 1984 | F   | 43       | NA                     | 16 × 16 × 7.7 | Cystic                             | Both                 | NA                |
| 5   | Ishizuka et al[(40)] | 1985 | M   | 59       | NA                     | 22 × 15 × 10 | Solid                             | Left                 | NA                |
| 6   | Kawakami et al[(41)] | 1986 | M   | 67       | NA                     | 30 × 28 × 12 | Cystic                             | Right                | NA                |
| 7   | Jennings et al[(42)] | 1987 | F   | 32       | Asymptomatic           | 14 × 11    | Cystic                             | Left                 | Left hepatic lobectomy |
| 8   | Kato et al[(43)] | 1988 | M   | 66       | Asymptomatic           | 18        | Both                               | Left hepatic lobectomy |
| 9   | Gutierrez et al[(44)] | 1988 | F   | 30       | NA                     | 18        | Both                               | Non-resectable       | NA                |
| 10  | Gramlich et al[(45)] | 1988 | F   | 28       | Abdominal distention, weight loss | 30 × 20 × 14 | Cystic                             | Right                | Lateral segmentectomy |
| 11  | Alcen et al[(46)] | 1989 | F   | 20       | Asymptomatic           | 6 × 8      | Cystic                             | Left                 | Left hepatic lobectomy |
| 12  | Ito et al[(47)] | 1989 | F   | 43       | Asymptomatic           | 16 × 16 × 7.7 | Cystic                             | Both                 | NA                |
| 13  | Urabe et al[(48)] | 1990 | F   | 39       | Asymptomatic           | 1.2       | Solid                             | Left                 | Left hepatic lobectomy |
| 14  | Drachen et al[(49)] | 1991 | F   | 69       | Asymptomatic           | 26 × 20 × 11.3 | Cystic                             | Left                 | NA                |
| 15  | Wada et al[(50)] | 1992 | M   | 62       | Asymptomatic           | 6 × 6 × 4.5 | Solid                             | Left                 | Hepatectomy |
| 16  | Chua et al[(51)] | 1994 | M   | 53       | Abdominal pain         | 28 × 14 × 10 | Cystic                             | Right                | NA                |
| 17  | Megremis et al[(52)] | 1994 | F   | 56       | Abdominal pain         | 7.5       | Cystic                             | Both                 | NA                |
| 18  | Yamamoto et al[(53)] | 1994 | M   | 52       | Abdominal distention, weight loss | 6 × 4 × 3.5 | Cystic                             | Left                 | Lateral segmentectomy |
| 19  | Chung et al[(54)] | 1999 | F   | 57       | Abdominal distention, weight loss | 6 × 4 × 3.5 | Solid                             | Right                | Right hepatectomy |
| 20  | Papastratis et al[(55)] | 2000 | F   | 21       | Abdominal pain, abdominal mass | 17 × 10 | Cystic                             | Right                | Right hepatectomy |
| 21  | Cook et al[(56)] | 2002 | F   | 46       | Abdominal pain         | 6 × 4 × 5  | Cystic                             | Right                | Right hepatectomy |
| 22  | Cook et al[(56)] | 2002 | F   | 46       | Abdominal pain         | 5 × 4 × 2  | Cystic                             | Right                | Right hepatectomy |
| 23  | Cook et al[(56)] | 2002 | F   | 63       | Abdominal pain         | 11 × 16 × 24 | Solid                             | Left                 | Left hepatic lobectomy |
| 24  | Mao et al[(57)] | 2002 | M   | 44       | Abdominal pain         | 2 × 2      | Solid                             | Left                 | Hepatectomy |
| 25  | Mao et al[(57)] | 2002 | M   | 76       | Abdominal pain         | 4 × 5 × 4  | Cystic                             | Right                | Right hepatectomy |
| 26  | Biric et al[(58)] | 2003 | M   | 38       | Abdominal pain         | 8 × 5      | Solid                             | Right                | Right hepatectomy |
| 27  | Kim et al[(59)] | 2003 | M   | NA       | Asymptomatic           | 5         | Both                               | Right                | NA                |
| 28  | Yesim et al[(60)] | 2005 | F   | 54       | NA                     | 2.5 × 2.5 × 1.5 | Cystic                             | Left                 | Total cystectomy |
| 29  | Yesim et al[(60)] | 2005 | F   | 51       | NA                     | 6 × 7 × 8  | Cystic                             | Right                | Unroofing procedure |
| 30  | Kim et al[(61)] | 2006 | F   | 40       | Asymptomatic           | 5 × 5      | Cystic                             | Right                | Right hepatectomy |
| 31  | Ayadi-Kaddour et al[(62)] | 2006 | F   | 21       | NA                     | 11 × 5    | Cystic                             | Left                 | NA                |
| 32  | Hernández et al[(63)] | 2006 | M   | 51       | NA                     | 19 × 13   | Solid                             | Right                | Liver transplantation (4th reported) |
| 33  | Chang et al[(64)] | 2006 | M   | 79       | Asymptomatic           | 2 × 2      | NA                                 | Right                | NA                |
| 34  | Chang et al[(64)] | 2006 | F   | 39       | Asymptomatic           | 5 × 5      | Cystic                             | NA                                 | NA                |
| 35  | Li et al[(65)] | 2007 | F   | 33       | Abdominal distention, abdominal pain | 16 | Both                               | Both                 | NA                |
| 36  | Mori et al[(65)] | 2008 | F   | 36       | Abdominal distention, abdominal pain | 20 × 15 × 10 | Cystic                             | Right                | Right hemihepatectomy |
| 37  | Gianippero et al[(66)] | 2009 | M   | 87       | Abdominal distention, abdominal pain | 20 × 20 | Cystic                             | Right                | Hemihepatectomy |
| 38  | Nakajo et al[(67)] | 2009 | M   | 38       | Asymptomatic           | 5 × 5      | Solid                             | Right                | Right hepatectomy |
| 39  | Klaassen et al[(68)] | 2010 | F   | 53       | NA                     | 9 × 9 × 7.5 | Cystic                             | Right                | Hepatectomy |
| 40  | Kulkarni et al[(69)] | 2010 | F   | 20       | Abdominal mass, abdominal pain, | 14 × 11 | Abdominal distention, abdominal pain | Right                | Mass resection |
| 41  | Tucker et al[(70)] | 2012 | W   | 74       | Abdominal distention, abdominal pain | 18 × 15 × 13 | Cystic                             | Left                 | Left hepatectomy |
| 42  | Liu et al[(71)] | 2013 | M   | 42       | Asymptomatic           | 1.5 × 1.0 × 1.0 | Solid                             | Left                 | Hepatectomy |
| 43  | Lakk et al[(72)] | 2014 | M   | 44       | Asymptomatic           | 2.9 × 3.1 × 3.5 | NA                                 | Left                 | Hepatectomy |
| 44  | Sharma et al[(73)] | 2014 | M   | 81       | Abdominal distention, abdominal pain | 21.8 × 12.3 × 18.6 | Cystic                             | Left                 | Hepatectomy |

**Notes:**
- abdominal pain includes nausea, distention, and weight loss.
- left hepatic lobectomy includes left hepatic lobectomy and left hepatic hemihepatectomy.
Laparoscopic liver resection for MHLs has been reported with successful outcomes\(^\text{[22]}\).

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

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