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Management of Fetal Mesenchymal Hamartoma of the Liver: A Case Report

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Abstract

Introduction
Hepatic mesenchymal hamartomas are rare benign tumours of the liver. Although most cases are discovered after birth, a few antenatal diagnoses have been reported in the literature.

Case Presentation
This is an 8-month-old girl with a prenatal diagnosis of a non-vascular anechogenic intra-abdominal mass, 30 mm in size, without septum, discovered during the third-trimester ultrasound. She was born at term by caesarean section due to macrosomia and gestational diabetes.

No Palpable Mass on Clinical Examination was Found
Abdominal ultrasound revealed the presence of a cystic mass in anterior of the left hepatic lobe with transsonic content, without clean wall or tissue component, with cloisons, some of which are thickened, measuring 79*40*70mm. A CT scan showed an exophytic cystic mass of the left liver in segments 2, 3 and 4. It measured 83*37*84 mm. It was next to the liver's left portal branch and away from the hepatic veins. On a clinical X-ray, the left liver's mesenchymatous hamartoma appears. Tumour markers were negative.

The patient underwent surgery at 8-month-old via a right subcutaneous incision and was found to have a thin-walled cystic mass. This mass was exophytic in relation with segments II and III, measuring 8cm with a cleavage plane, and the entire mass was resected.

Conclusion
Mesenchymal hamartoma is a rare benign pathology that can be diagnosed antenatally.

Complete resection is the recommended therapeutic approach, with a favorable outcome.

Conclusions
This study showed that HDL-C, lipid metabolites, including hexadecanedioate, tetradecanedioate, and 1-arachidonoylglycerophosphoinositol, had causal effect with PBC risk. Targeting ABCG5/ABCG8 may reduce PBC risk.

Keywords: Mesenchymal Hamartoma, Liver, Fetus, Case Report

Introduction

Hepatic mesenchymal hamartomas are rare benign tumors of the liver. They are defined as a focal proliferation of mature normal cells and native stroma that are epithelium-free, well circumscribed, and remote from the bile ducts. They typically manifest in early childhood, with patients aged between 1 month and 5 years [1].

Only a handful of cases have been identified prenatally, with ultrasound findings exhibiting considerable variability [2].

Complete resection is the recommended therapeutic approach, with a favorable outcome [2].

We report here a case of prenatally diagnosed hepatic mesenchymal hamartoma that was successfully treated.

Aim

A thorough understanding of the natural history of these tumors and skillful surgical treatment are indispensable elements of care.

Case Report

This is an 8-month-old girl with a prenatal diagnosis of a non-vascular anechogenic intra-abdominal mass, 30 mm in size, without septum, discovered during the third-trimester ultrasound. She was born at term by caesarean section due to macrosomia and gestational diabetes. The infant demonstrated effective adaptation to life outside.

No Palpable Mass on Clinical Examination was Found.

Abdominal ultrasound revealed the presence of a cystic mass in anterior of the left hepatic lobe with transsonic content, without clean wall or tissue component, with cloisons, some of which are thickened, measuring 79*40*70mm (figure1).

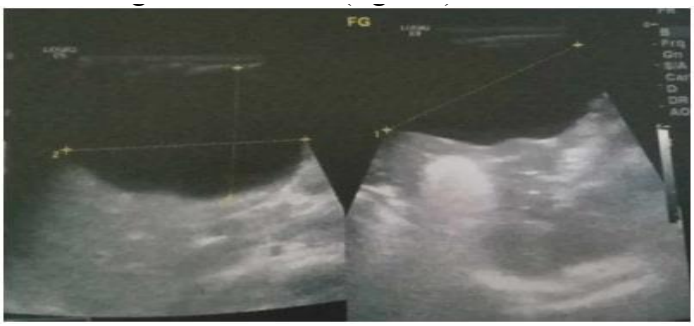

Figure 1: An Abdominal Ultrasound Showed Cystic Mass

in front of the left hepatic lobe with transsonic content

A Computed tomography (CT) scan showed an exophytic cystic mass of the left liver in segments 2, 3 and 4. It measured 83*37*84 mm (figure2). It was next to the liver's left portal branch and away from the hepatic veins. On a clinical X-ray, the left liver's mesenchymatous hamartoma appears.

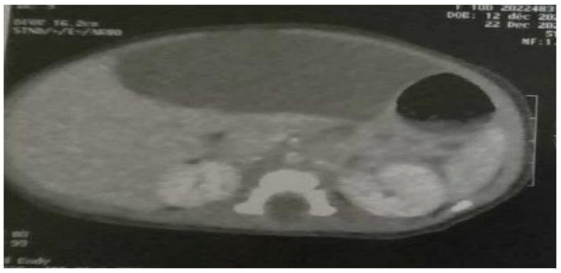

Figure 2: CT scan of the abdomen showed

an exophytic cystic mass of the left liver.

Tumour markers were negative.

The patient underwent surgery at 8-month-old via a right subcutaneous incision and was found to have a thin-walled cystic mass with translucent fluid content that was not infected on aspiration. This mass was exophytic in relation with segments II and III, measuring 8cm with a cleavage plane, and the entire mass was resected (figure3: A andB).

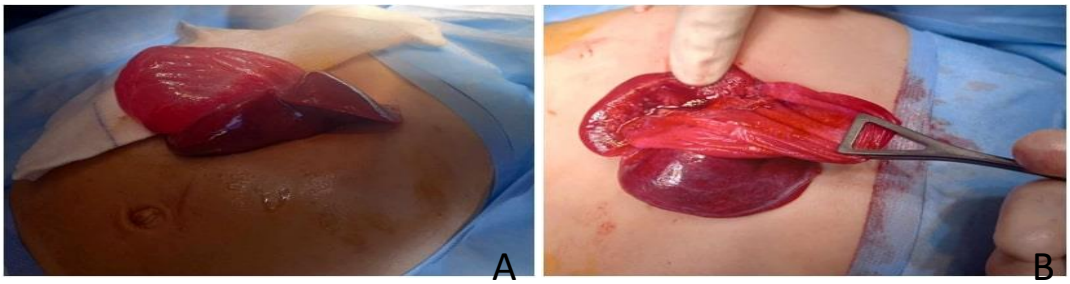

Figure 3: A) B) Intraoperative Photography: Hepatic Mesenchymal Hamartomas.

Pathological examination confirmed the diagnosis of mesenchymal hamartoma.
After one year on follow-up, ultrasound revealed a normal liver with no residual or recurrent tumour.

Discussion

Mesenchymal hamartomas were first described in the literature in 1903 by Mares [2]. Initially, these lesions were variously referred to as hepatic lymphangiomas and cavernous lymphangiomatoid tumors. They were subsequently grouped together under the term "mesenchymal hamartoma." These rare benign hepatic tumors that represent 5-8% of primary hepatic tumors [3].

They are the most frequent hepatic tumors in children after hemangiomas [4].

Some author have suggested that it is an anomaly of connective tissue development during fetal life, rather than a true neoplasm [5,6].

They also showed that this pathology could be the consequence of biliary obstruction, ischemia or disordered hyperplasia.

Kapur identified genetic alterations in mesenchymal hamartomas, including androgenetic-biparental mosaicism (ABM) and chromosomal rearrangements, which activate the chromosome 19q microRNA cluster (C19MC) [6].

A few cases of antenatal diagnosis have been described in the literature (approximately 25 cases) [4].

It may manifest as a cyst or solid mass at antenatal ultrasound. In general, fetal liver hamartomas are mostly avascular lesions on Doppler [1]. While histologically benign, it can cause heart failure due to arteriovenous shunting, as well as compression of the inferior vena cava and umbilical vein [1].

It is important to be careful when doing ultrasounds during pregnancy, especially if the mother's alpha-fetoprotein levels are high as not to break up.

Some factors were associated with a poor outcome: early onset, tumor size, rapid growth rate, the presence of hydrops, and compression of arteries by hydramnios [1,5,7].

Liver hamartomas are usually not associated with any abnormality. However, they can be linked to other conditions, including heart disease, intestinal problems, and kidney disease [8,10].

In regard to the mode of delivery, a cesarean delivery is not recommended unless there is a large tumor or fetal distress [1]. After birth, the smallest hamartomas are usually without symptoms. For larger tumors, the most common signs are a swelling in the abdomen and a noticeable mass.
The tumor can make the aFP levels in the blood higher, but they are usually normal. Sometimes the Gamma-glutamyl Transferase (GGT) levels are higher [11].

Given that ultrasound is the preferred initial assessment modality for children, findings can range from small cyst to large septate cyst, with varying degrees of associated echogenic solid tissue. On color Doppler, hepatic mesenchymal hamartomas are typically avascular or have low blood flow.

Abdominal computed tomography (CT) scan showed a multiloculated cystic tumor with a variable amount of solid tissue. Cysts are frequently septated. This multi-cystic configuration is commonly referred to as the "Swiss cheese appearance".

CT scan is an effective method for investigating the extent of the lesion and its relationship to other organs.

MRI can show the different parts of MHL and how they fit into the tissue around them. The MRI appearance of MHL depends on the protein content of the fluid and the presence of stromal elements [12].

Although some authors have proposed fetal aspiration [13,14]. The results have been unsatisfactory. Thus, prenatal

treatment is only a temporary decompression.

After birth, Non-operative management has been suggested in literature given that spontaneous regression of these lesions has been documented [8].

However, complete surgical excision is the only curative option to avoid the risk of recurrence and malignant transformation [2,8].

At section, there are multiple cysts containing a gelatinous serous fluid, but never bile. The composition of this fluid is similar to that of plasma, apart from a lower concentration of protein, cholesterol and glucose [15].

Although the prognosis after resection is excellent, clinical and radiological follow-up is recommended for at least 5 years to avoid long-term complications [8,10,16].

Conclusion

Mesenchymal hamartoma (MHL) is a rare, benign condition that could be better managed if diagnosed antenatally. Postnatally, radiological investigations (ultrasound and abdominal CT scans) are indicative of the diagnosis, with characteristic MHL findings including solid, cystic, non-vascularised or poorly vascularised liver masses. Complete resection of the tumour post-natally is an effective treatment with an excellent long-term prognosis.

Limits

It's a case report so we can't draw any relevant conclusions also, lacks precise data on the evolution of the mass during the 8 months prior to surgery.

• **Parental Consent for Minors:** Written informed consent was obtained from the patient's parents/ legal guardian for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.
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• **Corresponding Author:** jarray Leila : jarray.leila0@gmail.com
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