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Case Report

Mesenchymal Hamartoma in a 3-Year-Old Child: Case Report

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Abstract: Liver tumors account for 1 to 4% of solid tumors in children and are mainly represented by hepatoblastoma, hepatocellular carcinoma, and hemangioendothelioma. Cystic mesenchymal hamartoma is a rare benign tumor of the liver in children, accounting for 5 to 8% of primary hepatic tumors. Very few cases have been reported in the literature. We report the case of a 3-year-old girl who presented with abdominal pain. MRI suggested the diagnosis by revealing a large solid-cystic mass in the right lobe of the liver, and histological examination following a CT-guided biopsy confirmed the diagnosis of mesenchymal hamartoma of the right liver. An extended right hepatectomy including segment IVb was performed. The aim of our article is to describe the diagnostic and therapeutic approach to this very rare liver tumor.

Keywords: Hepatic mesenchymal hamartoma, cystic liver tumor, hepatic mass, benign tumor, hepatectomy in children.

INTRODUCTION

Pediatric liver tumors pose diagnostic and therapeutic problems due to their rarity: they represent 1 to 4% of solid tumors [1].

Among these tumors, six more frequent forms must be well known: hepatoblastoma, fibrolamellar carcinoma, and undifferentiated sarcoma for malignant tumors; focal nodular hyperplasia, hepatocellular adenoma and mesenchymal hamartoma for benign tumors [2].

The mesenchymal hamartoma is a benign lesion most often discovered before the age of 2 years old. Its origin and pathogenesis are poorly understood, and its management remains controversial.

We report a case of right liver hamartoma in a 3-year-old girl and discuss the clinical, radiological, anatomopathological, and therapeutic aspects of this benign hepatic tumor.

CASE

This is the patient K.N., aged 3 years, scored WHO 0, weighing 14 kg for a height of 98.5 cm [BMI: 14.6 kg/m 2]. She comes from a non-consanguineous marriage and is the fourth child of a sibling with no particular pathological history.

She consulted for right hypochondriac pain associated with vomiting, progressing for a month.

The biological assessment objectified a discreet hepatic cytolysis [TGP 51 IU/L, TGO 54 UI/L] and moderate cholestasis [GGT 99 IU/L], with a PAL level within the norms for age. Total bilirubin was 3.5 mg/L, with normal direct and unconjugated fractions. Albumin [43.6 g/L] and prothrombin [92%] were retained. The complete blood count showed a hemoglobin at 12.4 g/dL, white blood cells at 4.98 G/L and thrombocytosis at 583 000/mm 3. The CRP was slightly elevated at 11.2 mg/L.

Viral serologies [HBsAg, HIV 1/2, anti-HCV] were negative, as was the hydatic serology. Lipasemia and amylase were normal. Tumor markers [AFP, ACE, β-hCG] were negative.

The front chest X-ray revealed an ascent of the right diaphragmatic hemigpot associated with a discrete homolateral pleural effusion.

The abdominal ultrasound revealed a shocked-cystic mass of the right hepatic lobe, measuring 80 78 70 mm. The lesion was well limited, not encapsulated, with heterogeneous echostructure, comprising internal

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hypoechoic areas and presenting a low-resistance arterial and venous Doppler flow, orienting towards a mesenchymal hamartoma.

The hepatic MRI revealed a voluminous predominantly cystic formation located at segments VII and VIII, with extension to segment V. The lesion was in close contact with the right and median suprahelic veins as well as with the right portal branch, without any sign of vascular invasion. The aspect initially evoked a mucinous cystadenoma.

A scanno-guided biopsy was performed and the histological study confirmed the diagnosis of hepatic mesenchymal hamartoma.

After preoperative evaluation and hepatic volumetry, a right hepatectomy enlarged to segment IVb was performed by laparotomy through a Makuuchi incision.

Histologically, it is a benign tumor, characterized by an proliferation of mesenchymal tissue of variable maturity, tending to organize around pseudocysts or cysts lined with mesothelial or endothelial cells.

The postoperative outcomes were simple, marked by a gradual normalization of the liver function test. The patient was allowed to leave on the 8th postoperative day.

Written informed consent from parents was obtained for the publication of clinical and operative data. The patient's anonymity was strictly respected. In accordance with the principles of the Helsinki Declaration, data collection was carried out in compliance with the ethical rules in force. Local ethics committee approval was not required for this type of case report in our institution.

DISCUSSION

After its description first by Albrecht in 1903 and its recognition in 1956 by Edmondson as a clinical entity [Motiwale *et al.,* 1996], the mesenchymal hamartoma is defined as "*a tumour-like malformation in which there is an abnormal mixture of normal constituents of the organ in question, the abnormality may take the form of a change in quality, grouping or differentiation of the original elements* ".

Mesenchymal hamartoma of the liver is a rare benign tumor in early childhood that occurs mainly in children and more rarely in adults. Occasionally, it may occur in the large child [2].

It is observed in children with a median age of 16 months, some cases having been described in infants and older children [3]. In our case, the diagnosis was made at the age of 2 years and 6 months.

Based on the absence of mesenchymal mitotic activity, Stocker and Ishak [4] suggested that tumor proliferation activity occurs before or just after birth, while cyst growth is responsible for increasing tumor volume in the postnatal period.

Aberrations involving the chromosomal region 19q13.4 have also been implicated, resulting from an excess of proliferation and a lack of coordination of the primitive mesenchyme [5]. Thus, several cases with cytogenetic analyses have been described [6].

Clinically, the liver mass is often palpable [2]. In our case, it is the pain of the right hypochondrium that attracted attention and motivated the radiological examinations.

The diagnosis is essentially radiological. The lesion is well limited, presenting peripheral solid areas and central cystic areas with septa. CT scan allows to confirm the hepatic origin of the mass and to show a solid and multiloculated cystic image, in particular when ultrasound does not allow, in some cases, to define the intra- or retroperitoneal origin of the mass [7]. In our case, the diagnosis was mentioned on the ultrasound, but the MRI pointed towards a mucinous cystadenoma.

The lesion can be biopsied, with histological aspects close to those described in focal nodular hyperplasia. However, the young age of the patient and the myxoid character of the mesenchyme allow to distinguish with HNF [1]. For our patient, the biopsy and anatomopathological examination confirmed the diagnosis.

Differential diagnosis arises with fetal hepatoblastoma, especially when the serum level of AFP is increased and the hepatocytic cells contain AFP in immunohistochemistry. In our case, the AFP rate was not increased [8].

Complete excision is the treatment of choice for mesenchymal hamartoma [9]. In our case, the mass was in contact with the right branch of the portal vein and the right hepatic vein; a right hepatectomy enlarged to the IVb segment was therefore performed.

Excision can be performed by conventional liver resection or non-anatomical excision with a small healthy liver margin. Laparoscopic resection may be considered in case of pediculated lesions [10]. Other non-surgical methods have also been described in the literature.

CONCLUSION

An elevated serum AFP must be interpreted as a function of age; a significant elevation is in favor of a hepatoblastoma. A small to moderate increase may be observed in some hepatoblastomas, yolk sac tumour, hepatocellular carcinoma, as well as in some benign

tumours [mesenchymal hamartoma, focal nodule hyperplasia and infantile haemangioendothelioma].

Differential diagnosis between fetal hepatoblastoma, hepatocellular adenoma and mesenchymal hamartoma can be difficult on a biopsy. In the event of a certain diagnosis of mesenchymal hamartoma, surgery represents the treatment of choice.

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