

Regression of Large Infantile Multifocal Hepatic Hemangioma after Propranolol

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Abstract

Recent literature describes several cases of successful response of infantile liver hemangioma to propranolol treatment. Some multifocal hepatic hemangioma can involve a massive area of the liver in children and carry a high risk of development of cardiovascular compromise, requiring a fast approach and monitoring.

This case report describes a 3-months infant with a very large multifocal hepatic hemangioma for whom was proposed to start propranolol. Treatment was completed in a 20 months period with no report to side effects and a surprisingly fast response.

Keywords: Propranolol; Infantile; Hemangioma; Liver

Introduction

The authors present the following case to highlight the efficacy and safety of blocker- β propranolol treatment in young children even for very large liver hemangiomas.

Case Presentation

A healthy 3-months girl was brought for treatment with irritability and underwent an abdominal ultrasonography (US) for exclusion of intussusception diagnosis. The exam showed hepatomegaly and a solid lesion in the liver measuring 66 × 38 mm. The infant was born at 38 weeks of gestation, neither polyhydramnios nor abdominal mass were identified on prenatal ultrasonography and examination at birth was normal (Figure 1).

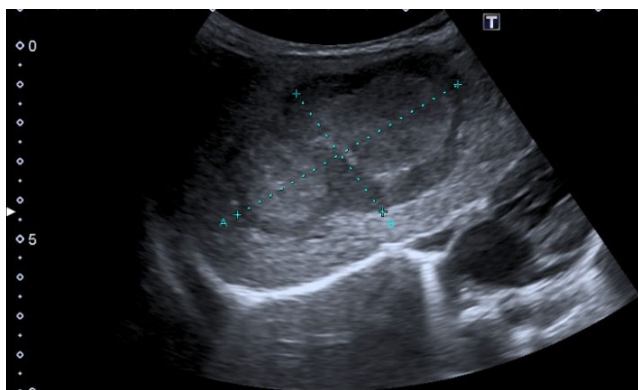


Figure 1: Abdominal US in 3-months child showing a solid lesion of liver.

She was referred to consultation and laboratory evaluation was performed at that time, including liver enzymes, hemoglobin and thyroid hormones that were normal. The serum alpha-fetoprotein (AFP) level was elevated (516 ng/ml). A MRI examination was performed at 3 months of age and multiple T1-hypointense and T2-hyperintense focal lesions of varying sizes were seen in the liver, mostly in segment VII (the largest two with 47 × 39 × 34 and 29 × 29 × 27 mm, which together measured 66 × 36 × 40 mm) – (Figures 2A and 2B). These findings suggested a multifocal hepatic hemangioma (MHH) diagnosis. Cardiac echogram mentioned no high-output congestive heart failure.

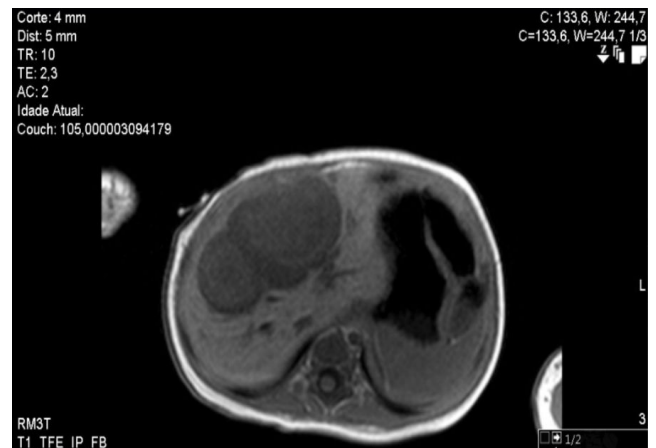


Figure 2A: MRI of the liver before initiate propranolol treatment - axial T1 image of the liver shows hypointense lesion (3 months of age).

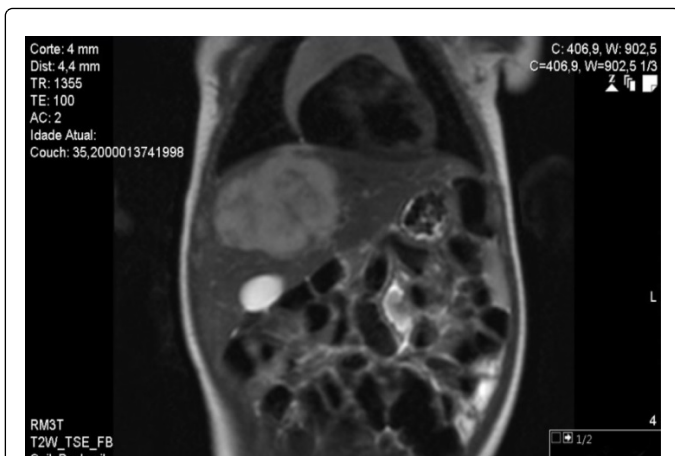


Figure 2B: MRI of the liver before initiate propranolol treatment - Coronal T2 image of the liver shows hyperintense lesion measuring about 66 × 36 × 40 mm (3 months of age).

The hypothesis of hepatoblastoma seemed to be remote but it could not be ruled out safely at beginning because of the high AFP level. Its levels significantly decreased and became normal during the following months (<1.3 ng/ml) and meanwhile the patient was proposed for treatment with propranolol. The parents did not accept the treatment immediately scared about eventual side effects, so that its start was postponed.

Propranolol was introduced at 7 months-age and a dose of 3 mg/kg per day was started. Before starting treatment, an US was repeated and found a similar size lesion compared to the previous study. The patient was hospitalized and monitored in the first 24 hours of treatment to evaluate adverse effects. During that period blood pressure, heart rate and glycemia were normal.

Sonographic follow-up examinations showed a gradual and fast decrease of tumor size, getting half of dimension at the end of the first week of treatment. The other lesions were no longer visible in this ultrasonography. After 7 months of treatment the lesion measured 17 mm as shown in Figure 3 and after 20 months only areas of heterogeneity, poorly defined, were visible, corresponding to a successful and almost complete regression as shown in Figure 4.

Discussion

The child was clinically asymptomatic during follow-up. The dose was gradually increased according to the weight. Some episodes of bronchiolitis occurred and resolved without discontinuation of treatment. The authors propose a follow-up consultation every 3 months performing abdominal ultrasound and eventually a second MRI.

Infantile hepatic hemangioma is the most common benign liver tumor during infancy. It is classified in focal, multifocal or diffuse [1,2]. Mortality can reach 18% of cases and is dependent of the presence of heart failure due to high volume of arteriovenous shunting [3]. Fortunately, often these lesions are asymptomatic. MRI plays an important role in the correct diagnosis [2,3]. In the last years the use of propranolol has scattered and actually many practitioners have adopted it as first line of therapy [3-5].

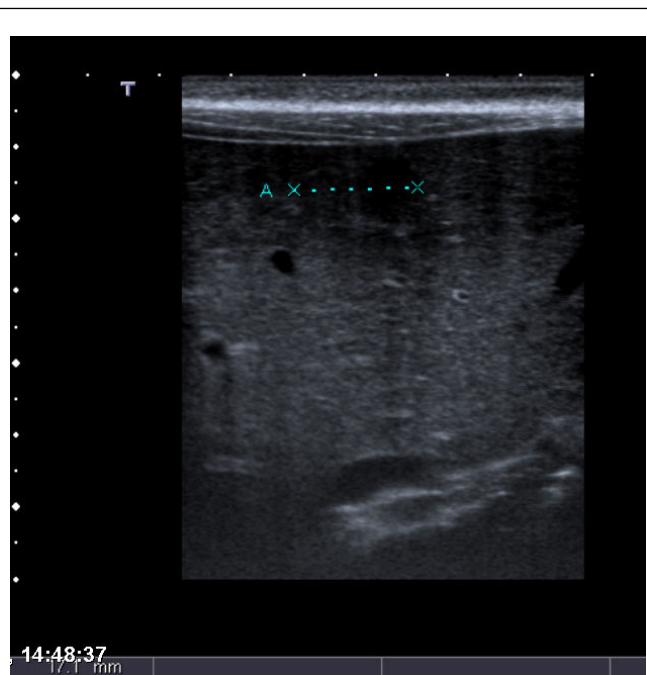


Figure 3: Abdominal US after 7 months of treatment (17 mm of lesion).



Figure 4: Abdominal US after 20 months of treatment (areas of heterogeneity, poorly defined and very small size).

Conclusion

The authors described a case of an infant with a multifocal hepatic hemangioma, with massive involvement of the liver carrying high risk of development of cardiovascular compromise. It was successfully treated with propranolol during 20 months with no report to side

effects and a surprisingly fast response. The efficacy of treatment is partially attributed to the size of hemangioma and few cases in liver are described having these initial dimensions and responding so well to blocker- β treatment.

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