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Case Report

Pediatric Hepatic Hemangiomas: A report of 5 Cases.

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ABSTRACT:
Infantile hepatic hemangiomas are the most common benign liver tumors in children. They can be solitary in most cases or multiple. We report the cases of 5 patients presenting with infantile hepatic hemangiomas. The mean age at diagnosis was 4 months. Clinical presentation was marked by abdominal distension in 3 cases, while two patients were asymptomatic. the diagnosis was determined through the findings of hepatic Doppler ultrasound, which revealed a large, heterogeneous, multinodular hepatomegaly occupying the entire liver parenchyma in 4 cases. Asymptomatic hypothyroidism was found in two patients. All 5 patients were treated with Propranolol, resulting in disappearance of the hemangiomas in two cases and stability of the lesions in two cases.

Keywords: infantile hepatic hemangiomas, hepatic Doppler ultrasound, hypothyroidism, Propranolol

INTRODUCTION:

Infantile hepatic hemangiomas are the most common benign liver tumors in children. They arise from mesenchymal tissue, consisting of masses of blood vessels with atypical or irregular arrangement and size. The etiology of these lesions remains unknown. They can occur as solitary or multiple hemangiomas. Typically, they are asymptomatic; however, in some cases, they can lead to complications of varying degrees of severity.

Patients and Methods:

We conducted a retrospective study on medical records of patients under the age of 5 years with hepatic hemangiomas who were followed up in the hepatology outpatient clinic of the Department of pediatrics at Mustapha Bacha university hospital-from January 2017 to January 2023. We analyzed epidemiological, clinical, therapeutic, and outcome data of the patients.

RESULTS:

We included 5 children; 1 girl and 4 boys. The average age at diagnosis was 4 months [range: 3-5 months]. Clinical presentation was characterized by abdominal distension in 3 cases, while two cases were asymptomatic. In one patient, the diagnosis was made by abdominal ultrasound to look for another location of a cutaneous hemangioma on the left arm, and in another patient, the diagnosis was made incidentally during radiological exploration for infantile colic. There were no growth delays, digestive disorders, or signs of liver involvement.

Abdominal ultrasound was performed in all patients, and it was sufficient for a positive diagnosis in 3 patients. It was complemented by abdominal CT scan in one case and abdominal MRI in another patient to rule out other differential diagnoses. In 4 cases, hepatic Doppler ultrasound revealed a large, heterogeneous, multinodular hepatomegaly occupying the entire liver parenchyma, while in one case, hepatic Doppler ultrasound showed a liver of normal size with 4 hyperechoic nodules distributed across different segments. One patient with a cardiac murmur underwent echocardiography, which revealed an arterioportal fistula. Biologically, no disturbances in liver function tests were noted. Systematic thyroid assessments in all patients revealed asymptomatic hypothyroidism in 2 cases. Biological assessment of renal and pancreatic function, and also hematological parameters were normal.

Four patients were treated with Propranolol at 3 mg/kg in two divided doses as first-line therapy, which acts not as a beta-blocker but as an anti-angiogenic agent. One patient initially received corticosteroid therapy with prednisolone at 2 mg/kg with gradual tapering over 3 months, and then was switched to Propranolol after the failure of corticosteroid therapy. After an average treatment duration of 14 months [range: 4-28 months], complete disappearance of the lesions was observed in two patients, stability in number and size of the lesions in two patients, while one patient exhibited the appearance of new lesions. Regarding hypothyroidism, both patients received hormonal replacement therapy with Levothyroxine.

DISCUSSION:

Various terminologies have been used in the literature to describe these hepatic vascular lesions, including infantile hepatic hemangioendothelioma, hepatic angioma, and others. However, the most commonly used term currently is infantile hepatic hemangioma (IHH).

Infantile hepatic hemangiomas (IHH) are the most common benign tumors in infants, affecting approximately 5% of children under one year of age [1]. They typically appear after birth and increase in size during the first weeks of life. Following a variable growth phase, which can range from 3 to 12 months, spontaneous regression is the natural course [2]. The liver is the second most common location for infantile hemangiomas after the skin. There are solitary and multinodular forms [3,4]. It is believed that hemangiomas result from an anomaly in angiogenesis regulation, with vascular endothelial growth factors potentially playing a role in tumor development [1]. They can be focal, multifocal (involving 4-20 lesions), or diffuse when the number of lesions exceeds 20 [5]. Focal forms are generally asymptomatic, while most symptomatic hemangiomas are multifocal or diffuse [6]. Screening abdominal ultrasound is recommended for infants under 6 months of age who present with more than 5 cutaneous hemangiomas [5].

Solitary forms may appear on ultrasound as rounded lesions primarily composed of clearly perfused and tortuous cavities, while multifocal lesions present as solid tumors [6], hence the need for better assessment with CT or MRI, as was the case with 2 patients in our series, to rule out other differential diagnoses.

First-line treatment for cutaneous infantile hemangiomas and solid organ hemangiomas traditionally relied on corticosteroid therapy, followed by interferon and vincristine for resistant lesions [5]. However, severe side effects have been reported in patients treated long-term [5]. The effectiveness and good tolerability of Propranolol have been documented [4,5], leading to its suggestion as a first-line treatment for infantile hemangiomas [5]. A series of 8 patients, including 4 with multifocal and 4 with diffuse forms, was reported by Mazereeuw-Hautier et al. [7]. Among them, 3 patients initially received corticosteroid therapy combined with vincristine, followed by Propranolol, while the other 5 received Propranolol as initial treatment. Evaluation revealed complete disappearance of lesions in 3 cases and a reduction in tumor size by more than 50% in 5 patients. In our patient series, the failure of corticosteroid therapy as initial treatment in one case led us to use Propranolol. The other 4 patients received Propranolol as first-line treatment, with a favorable outcome in 80% of cases.

In this study, hypothyroidism was found in two patients. Indeed, infantile hepatic hemangiomas can be associated with a type of hypothyroidism known as consumptive hypothyroidism. This complication occurs almost exclusively in diffuse forms or when hemangiomas are large. It is due to increased catabolism of thyroxine by the tumor. Specifically, there is described destruction of T3 by secretion of deiodinase by the hemangioma. This phenomenon was first described by Huang et al. [6,8].

Multifocal forms can also be complicated by secondary congestive heart failure due to arteriovenous shunting, which could be the initial ~~revealing~~ sign of the clinical picture, suggesting a congenital heart defect [6]. We noted the presence of an arterioportal fistula in only one of our cases, detected by a cardiac murmur and confirmed by echocardiography, without heart failure. In case of heart failure, an mTOR inhibitor with anti-angiogenic effects such as Sirolimus or Everolimus is recommended. In extreme cases of uncontrolled major life-threatening failure, radiological embolization may be attempted to reduce flow, or if feasible, liver transplantation may be considered.

CONCLUSION:

Infantile hepatic hemangioma is a common vascular tumor characterized by its perinatal presentation, rapid growth during the first year of life, and subsequent involution. Diagnosis is typically straightforward and relies on a combination of clinical symptoms and radiographic findings. Despite the benign nature of IHH, some of these lesions may require medical intervention, especially for multiple and diffuse forms. Propranolol remains the first-line treatment. The prognosis is usually favorable except for multifocal forms, which can be complicated by high-output heart failure; therefore, close monitoring is necessary until complete lesion involution.

Conflict of Interest: All authors declare no conflict of interest.

REFERENCES:

[1] Imteyaz, Hejab; Karnsakul, Wikrom; Levine, Michael A; Burrows, Patricia E; Benson, Jane; Hsu, Stephanie; Schwarz, Kathleen B. Unusual Case of Hypothyroidism in an Infant with Hepatic Hemangioma. Journal of Pediatric Gastroenterology and Nutrition 54(5): p 692-695, May 2012.

[2] Mathivon F, Enjolras O, Escande JP. Hemangiomatose neonatale miliaire multiple cutanee et hepatique. Quatre observations d'evolution favorable. Ann Pediatr 1994 ;41 :337-45

[3] Gandolfi L, Leo P, Solmi L, Vitelli E, Verros G,Colecchia A. Natural history of hepatic

haemangiomas: clinical and ultra-sound study. Gut 1991; 32:677-80
[4] Reading NG, Forbes A, Nunnerley HB, Williams R.Hepatic hemangioma: a critical review of diagnosis and management. Q J Med 1988; 253:431-45
[5] Avagyan S, Klein M, Kerkar N, Demattia A, Blei F, Lee S, Rosenberg HK, Arnon R. Propranolol as a first-line treatment for diffuse infantile hepatic hemangioendothelioma JPGN Volume 56, Number 3, March 2013.
[6] Zimmermann A, Tumors and Tumor-Like Lesions of the Hepatobiliary Tract. Springer International Publishing Switzerland 201. DOI 10.1007/978-3-319-26956-6_52.
[7] Mazereeuw-Hautier J, Hoeger PH, Benlahrech S, et al. Efficacy of propranolol in hepatic infantile hemangiomas with diffuse neonatal hemangiomatosis. J Pediatr 2010; 157:340–2
[8] C. Eschard, Hémangiomes infantiles : quand explorer et actualités thérapeutiques. Annales de dermatologie et de vénéréologie (2015) 142, 476—482.