CASE REPORT

HEPATIC ADENOMATOSIS IN A 32-YEAR-OLD MALE WITH NO RISK FACTORS FOR HEPATIC ADENOMA

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Hepatic Adenomatosis most commonly arises in the background of hepatic adenoma in young women and less commonly in men with underlying risk factors. Rarely, it can arise in young men with none of the associated risk factors and thus can go undetected for a prolonged period resulting in delayed intervention and avoidable complications. We present a rare case of a 32-year-old asymptomatic male that presented for routine evaluation and was subsequently diagnosed with Hepatic Adenomatosis.

Keywords: Adenomas; Hepatocellular; Benign

Citation: Malik TF, Akram S. Hepatic adenomatosis in a 32-year-old male with no risk factors for hepatic adenoma. J Ayub Med Coll Abbottabad 2022;34(3 Suppl. 1):721–2.

DOI: 10.55519/JAMC-03-S1-9128

INTRODUCTION

Hepatic adenoma is an uncommon, benign, primary lesion of the liver that arises as a solitary well-circumscribed mass. The occurrence of multiple lesions (>10) throughout the normal hepatic parenchyma, results in a rare condition called Hepatic Adenomatosis (HA). The risk of development of HA in a patient with a hepatic adenoma is10–24%. This condition has a strong female predisposition and is eleven times more likely to occur in young females as compared to males.[1] The female predisposition is due to the association of the tumor with high serum estrogen states such as pregnancy and use of oral contraceptive medication and hormone replacement therapy. The tumor may also arise in males with the use of anabolic steroids[2], barbiturates, or due to underlying metabolic diseases such as glycogen storage disease type 1 and type 3[3], type 1 diabetes mellitus[4], hemochromatosis, and tyrosinemia. In rare instances, HA may arise in an otherwise healthy male with no associated underlying risk factors for hepatic adenoma. We present a case that describes this rare occurrence and highlights the importance of investigating liver lesions for the presence of hepatic adenomas regardless of age, gender, or underlying risk factors to avoid complications and offer timely intervention.

CASE REPORT

A 32-year-old male presented for routine medical evaluation. The patient denied a history of alcohol consumption, anabolic steroid use, or family history of chronic liver disease. The results from the initial blood work revealed abnormal liver function tests with elevated alkaline phosphatase = 322 (IU/L). Right upper quadrant abdominal ultrasound revealed the presence of multiple heterogeneous liver lesions. The largest lesion was present in the right hepatic lobe measuring 9.3 cm in size. Magnetic resonance imaging (MRI) with and without contrast demonstrated hepatomegaly and mild fatty infiltration of the liver along with the presence of numerous hepatic lesions. The largest lesion was present within the right hepatic lobe (Figure-1). A percutaneous liver biopsy was performed and the specimen revealed the presence of neoplastic tissue and well-differentiated hepatocytes with no cytological atypia. Immunohistochemical staining results confirmed the diagnosis of hepatocellular adenoma by demonstrating intact reticulin, low Ki-67 proliferation index, glypican-3, and arginase negativity. The specimen stained positive for C-reactive protein and serum amyloid-associated protein with retained liver-type fatty acid-binding protein expression. The specimen tested negative for beta-catenin and glutamine synthetase. A CD34 showed a strong diffuse staining pattern. These findings confirmed the presence of Inflammatory hepatocellular adenoma, the most common subtype of hepatic adenoma with the highest tendency for malignant transformation.[5]The patient was scheduled for surgery and underwent an open partial right and left lobectomy with microwave ablation. Follow-up with contrast computed tomography after 6 months revealed residual arterial enhancement in the right hepatic lobe indicating possible residual or recurrent adenomas. The patient is scheduled for a follow-up MRI.

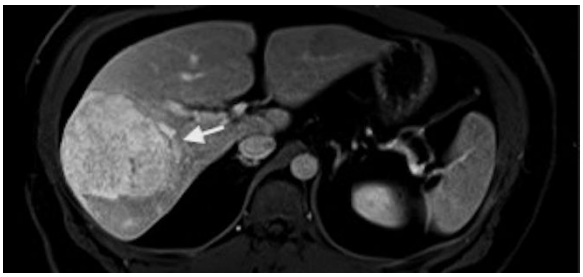

Figure-1: MRI showing a large adenomatous lesion in the right hepatic lobe

DISCUSSION

Hepatic adenomatosis was first described in 1985 by Flejou *et al* as the existence of numerous benign adenomas in an otherwise normal hepatic parenchyma.[6] Since then it has been mainly studied in the female population and less is known about the features and outcomes of this condition in men. Hepatic adenomatosis is classified into two main patterns, Massive HA is characterized by massive hepatomegaly with large adenomas and liver deformity that progresses rapidly whereas Multifocal HA is composed of small adenomas without massive hepatomegaly and liver deformity with a relatively better prognosis. Hepatic adenomas are classified into five main subtypes based on microscopy, immunohistochemical staining, and genetic analysis of the biopsy specimen.[7] The prognosis, management, and risk of complications differ for each subtype. Lesions larger than 5 cm, if left untreated can lead to spontaneous rupture and hemorrhage into the abdominal cavity. Unlike other benign tumors of the liver, there is also a mild yet significant risk of malignant transformation. Keeping in mind these complications the best management for this young, otherwise, healthy patient with large hepatic adenomatous lesions was surgical resection with regular follow-up. According to a study, tumor resection resulted in the best outcomes for young patients with hepatic adenomas regardless of tumor size as it reduces the risk of rupture and malignant transformation.[8] In the case of numerous adenomatous lesions distributed widely throughout the liver, surgical resection may not be possible and liver transplantation is recommended.

CONCLUSION

This unique case helps us understand the importance of excluding hepatic adenomatosis as a differential diagnosis in any patient presenting with liver lesions regardless of age, gender, and risk factors. Often the workup for hepatic adenomatosis is overlooked in male patients due to its widely female-predominant occurrence. Early diagnosis and intervention will likely reduce morbidity and mortality in patients as the risk of hemorrhage and malignant transformation is higher than with other benign liver tumors. Advanced studies need to be conducted to understand the natural history and clinical significance of HA in men to determine the best diagnostic and management strategies to prevent under-diagnosis and complications in the future.

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Submitted: February 6, 2021	*Revised: April 19, 2021*	*Accepted: May 2, 2021*

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