CASE REPORT

TJG
The Turkish Journal of Gastroenterology

Congenital hepatic artery aneurysm: A case report in early infancy

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Hepatic artery aneurysm in children is a very unusual pathologic entity. As most patients with this anomaly are asymptomatic, the diagnosis is usually made as an incidental finding on imaging studies performed for other reasons. Here, we report a 35-day-old infant with hepatic artery aneurysm, which was diagnosed accidentally as a liver mass during echocardiography. Considering the age of diagnosis, she most likely had a congenital type of hepatic artery aneurysm. To the best of our knowledge, this is the youngest case to be reported in the literature.

Key words: Hepatic artery aneurysm, congenital, infant, liver mass

Konjenital hepatik arter anevrizması: Erken çocukluk çağındaki bir olgunun sunumu

Çocuklarda hepatik arter anevrizması görülmesi alışılmadık bir durumdur. Hastaların büyük çoğunluğu asemptomatiktir ve bu anomalisi olan hastalar genellikle başka nedenler üzerine yapılan görüntülemeler ile tesadüfen tanı alırlar. Burada, ekokardiyografi sırasında tesadüfen tespit edilen ve karaciğer kitlesi olarak tanı konulan 35 günlük hepatik arter anevrizması hastası sunulmuştur. Tanı yaşı dikkate alındığında, hepatik arter anevrizmasının konjenital olduğu sonucuna varılabilir ve bildiğimiz kadarı ile bu hasta literatürde yayınlanmış en genç vakadır.

Anahtar kelimeler: Hepatik arter anevrizması, konjenital, enfant, karaciğer kitlesi

INTRODUCTION

Hepatic artery aneurysm (HAA) is a rare but clinically important phenomenon (1). The hepatic artery is the fourth most common site of intraabdominal aneurysm, following infrarenal aorta, iliac artery and splenic artery (2). HAA represents approximately 20% of all visceral aneurysms, of which 80% are extrahepatic and 20% are intrahepatic (1-3).

Most of the reported cases with HAA are in adults or older children, and were secondary to mycotic aneurysm, trauma or atherosclerosis (1-8). A few cases with congenital HAAs detected during infancy or in older ages have been reported (9-12).

CASE REPORT

A 35-day-old female infant was referred to our clinic due to a liver mass, which was detected by a pediatric cardiologist during echocardiography. She was the product of a full-term pregnancy born via normal vaginal delivery in good condition.

The physical examination after birth revealed a

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Manuscript received: 13.08.2010 **Accepted:** 02.03.2011

Turk J Gastroenterol 2012; 23 (1): 72-74
doi: 10.4318/tjg.2012.0318

normal acyanotic newborn with soft systolic murmur over the pericardium without any other abnormal finding. The echocardiography was normal, but a mass in the liver was detected at the time of the evaluation of the hepatic veins. The patient was referred to the pediatric hepatology clinic for further evaluation. Upon arrival in our center, she appeared normal with no organomegaly or any significant abnormal finding.

Hepatobiliary ultrasound was done, which showed a normal liver size and echo pattern, with a cystic mass of about 20 mm in the internal segment of the right lobe and mild dilatation of intrahepatic bile duct.

Color Doppler sonography revealed pulsatile flow within the mass of vascular origin. These findings were in favor of arteriovenous malformation or HAA.

Computed tomography (CT) angiography revealed severe dilatation of the celiac trunk with a huge aneurysm, 4 cm in diameter, arising from the hepatic artery and located in the hepatic artery bifurcation (Figure 1). She was referred to a pediatric surgeon. The parents refused surgical ligation, and she was discharged against medical advice. She was admitted a few weeks later in critical condition and died a few hours later.

DISCUSSION

Hepatic artery aneurysm (HAA) was first reported at autopsy in 1809 (3) and in 1959 in children by Jewett (6). HAAs are not initially diagnosed in many cases because the majority of patients are asymptomatic, and in 80% of the cases, rupture of the aneurysm is the first clinical manifestation (2). Other clinical presentations are abdominal pain in 55% and gastrointestinal hemorrhage in 46% (2). The classic triad of epigastric pain, hemobilia and obstructive jaundice are only present in one-third of the cases (2).

There are various etiologies for HAAs. Historically, mycotic aneurysms accounted for most cases, while atherosclerosis is present in as many as 30% of affected patients (2,3,5). Less common causes of HAA are: vasculitides such as polyarteritis nodosa, periarterial inflammation caused by cholecystitis or pancreatitis, fibromuscular hypoplasia or cystic medial necrosis, tuberculosis, and trauma (2,3). Congenital aneurysms have also been reported in children and adults (9-12).

Figure 1. CT angiography showed severe dilatation of the celiac trunk, and a huge aneurysm about 4 cm in diameter arising from the hepatic artery located in the hepatic artery bifurcation; superior and inferior mesenteric arteries are intact.

Hepatic artery aneurysms (HAAs) are reported among the anastomotic complications of orthotopic liver transplantation, and also occur after hepatic tumor embolization. The physical examination is usually normal, as in our case, although a large aneurysm may be associated with a pulsatile mass or an abdominal bruit (2,3).

A curvilinear calcification in the right upper quadrant on a plain radiography of the abdomen should raise the possibility of HAA. Ultrasound and CT scan provide the diagnosis in most of the cases. Color Doppler ultrasound can aid in differentiating vascular anomalies from other types of masses. Color Doppler ultrasound also can differentiate aneurysm from other vascular abnormalities such as arteriovenous fistulas or malformations (2,4). A three-dimensional spiral CT may allow a definitive diagnosis to be made prior to angiography (2,3,12,13). Appropriate management of HAA requires detailed angiography (2,14).

The rupture rate in HAAs can reach as high as 44% (15) and the mortality rate as high as 82% (16). The current trend is that all cases should be considered for treatment before rupture of the aneurysm (2,4). Common HAAs can be treated by either surgical ligation or embolization. Embolization is the treatment of choice for intrahepatic aneurysm (4). Most extrahepatic HAAs described in the literature have been treated surgically (3). Considering the age of our case, we believe she had congenital HAA, and to our knowledge, she is the youngest case to be reported to date. We conclude that HAA should be considered in the differential diagnosis of infants with hepatic mass.

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