PST | PEDIATRIC SURGERY IN TROPICS

PAINLESS OBSTRUCTIVE JAUNDICE IN A CHILD: A RARE CHASE OF HEPATIC ARTERY ANEURYSM

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Keywords	**Abstract**
Painless obstructive jaundice Hepatic artery aneurysm Sarcoidosis Biliary stricture	**Introduction:** Hepatic artery aneurysms are rare visceral aneurysms, predominantly reported in adults, with limited literature in children. Although rare, this condition poses a considerable diagnostic and therapeutic challenge and is associated with substantial morbidity and mortality. **Case Presentation:** We report on a case of an eleven-year-old boy presenting with painless obstructive jaundice and anemia due to a large hepatic artery aneurysm (HAA). The patient was also found to have sub-mitral and left common carotid artery aneurysms, with an underlying diagnosis of sarcoidosis. The HAA was managed by surgical ligation. Postoperatively, the patient developed an extrahepatic bile duct stricture that was managed through external and internal drainage by means of percutaneous transhepatic biliary drainage (PTBD) and endoscopic stenting. Despite these complications, he has shown steady recovery and is

	currently receiving medical treatment for sarcoidosis. **Conclusion:** Hepatic artery aneurysm (HAA), though rare, should be considered a potential cause of painless obstructive jaundice in pediatric patients, particularly those with underlying risk factors. Prompt investigation and a multidisciplinary approach are essential to guide individualized treatment, given the risk of rupture and the need to balance immediate and long-term outcomes.
Abbreviations	HAA: Hepatic Artery Aneurysm CT: computed tomography CHA: Common Hepatic Artery MR: Magnetic Resonance PTBD: percutaneous transhepatic biliary drainage

INTRODUCTION

Hepatic artery aneurysm (HAA) is a rare condition accounting for 14-20% of all visceral artery aneurysms and predominantly affects adults. [(1)] HAA may be congenital or acquired and is frequently associated with underlying conditions such as hypertension, malignancy, trauma, vascular disease, or connective tissue disorders. Advancement in cross-sectional imaging has resulted in earlier detection of incidentally identified HAA in asymptomatic patients. Among symptomatic cases, 60–80% present with the most severe complication—aneurysmal rupture—characterized by the classical triad of pain, jaundice, and anemia. [(2)]

In the pediatric population, HAAs are exceptionally rare, with few documented cases and even fewer presenting without the hallmark symptoms of rupture. We present a rare case of an 11-year-old boy with painless obstructive jaundice due to a HAA. This case adds to the limited literature by describing a unique presentation in a child with systemic sarcoidosis and outlines the complex diagnostic and interventional course.

CASE REPORT

An 11-year-old boy presented with a four-week history of progressive jaundice, abdominal distension, dark urine, pale stools and anorexia. Previously, he had been well with no significant medical or surgical history. Physical examination revealed tense ascites, hepatomegaly and marked jaundice with signs of systemic wasting, bilateral basal crepitations and a grade II pansystolic murmur. No discrete abdominal mass was palpable.

Initial biochemistry showed significant hepatic dysfunction and normocytic anemia. Cross-sectional imaging with contrast-enhanced computer tomography (CT) revealed a large aneurysm of the common hepatic artery measuring 4.7cm x 6.4cm x 6.9cm containing a mural thrombus. Significant biliary dilation was also noted due to the compressive effect of the aneurysm. Angiography revealed a common hepatic aneurysm that was not amenable to endovascular intervention due to the short neck of the affected vessel and inability to cannulate the affected vessel beyond the aneurysm. (Fig 1) Initial biliary decompression was achieved through percutaneous transhepatic biliary drainage.

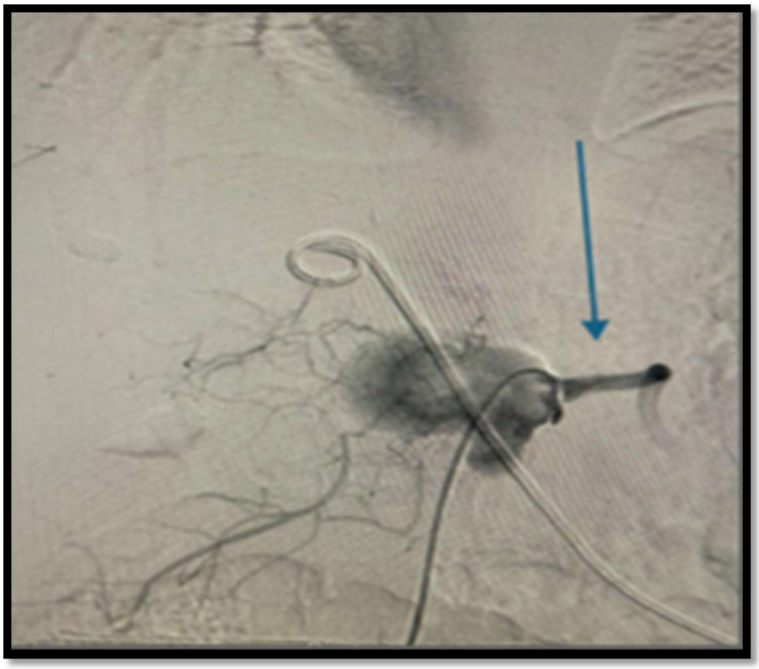

Fig 1. Angiogram showing common hepatic artery aneurysm and left gastric artery (indicated with an arrow)

Following multidisciplinary discussion, the patient underwent exploratory laparotomy. Intraoperatively the aneurysm was successfully ligated distal to the origin of the gastroduodenal artery. Intraoperative doppler ultrasound confirmed cessation of flow. Follow up imaging performed 1-month after surgery demonstrated a small residual mass (1.7cm x 2.1

cm x 2.7cm) with no intraluminal flow, regional calcification, and significant left gastric and peribiliary collateralization.

Despite initial recovery, the patient developed a long biliary stricture in the months following surgery. The stricture is seen in Figure 2 and was likely due to ischemic injury from hepatic artery ligation and postoperative fibrosis. (Fig 2) The stricture is being managed by means of a self-expanding metal stent inserted through a Rendezvous procedure. (Fig 3)

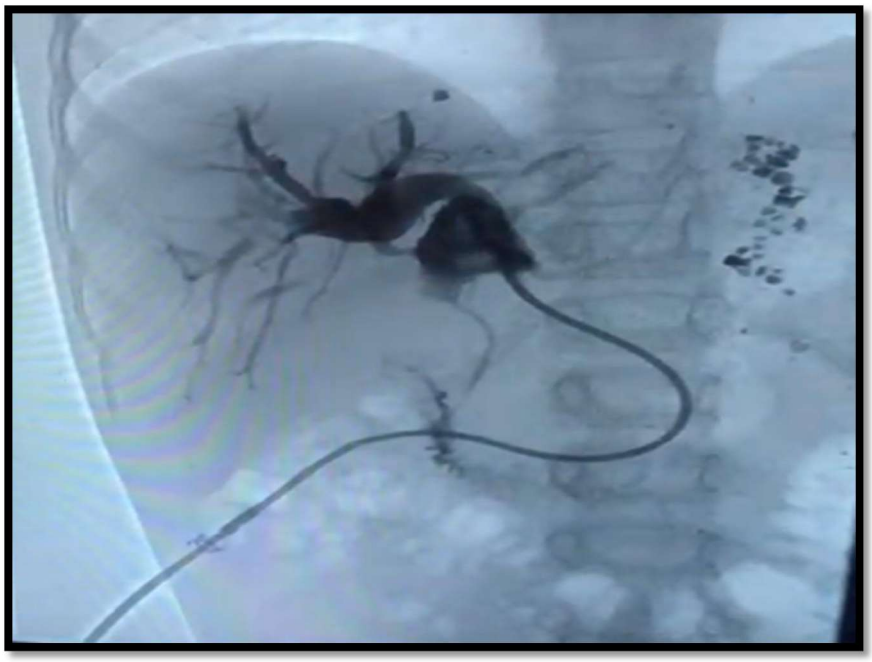

Fig 2. Postoperative tomogram demonstrating a long segment stricture of the extrahepatic bile duct (indicated with an arrow)

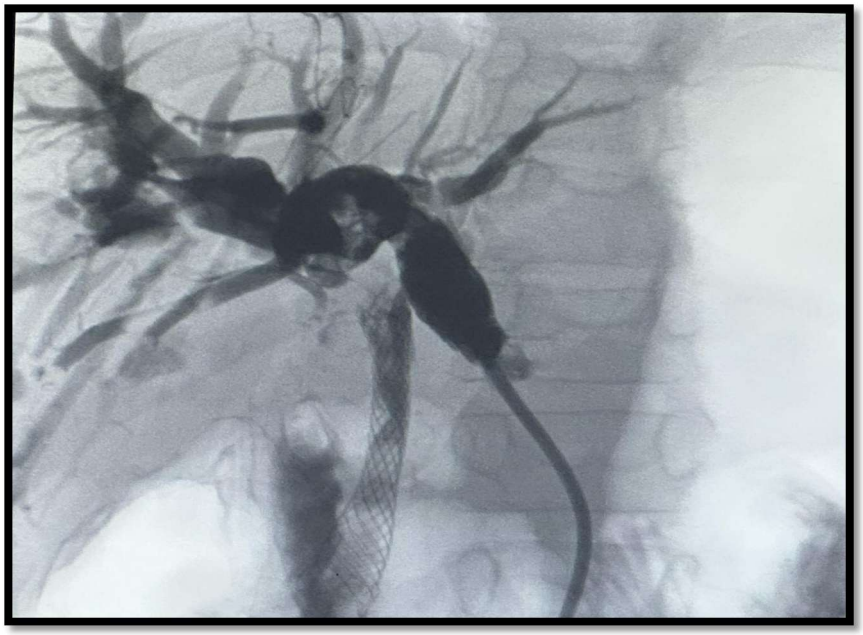

Fig 3. Self-expanding metal stent positioned through a Rendezvous procedure

Histological analysis of excised lymph nodes revealed a non-caseating granulomatous lymphadenitis while microbiological studies were negative for acid-fast bacilli and fungi. Given these results, and based on the patient's entire clinical picture, a diagnosis of sarcoidosis was made and therapy initiated by the pediatric rheumatological team.

DISCUSSION

Hepatic artery aneurysms (HAA) are rare visceral aneurysms primarily documented through case reports. [1] In children, literature on HAA is extremely limited. HAA exhibit a male predominance with a 2:1 ratio, and most commonly present in the sixth decade of life. [3] Extrahepatic aneurysms are more frequently observed than intrahepatic lesions, with two-thirds of extrahepatic cases originating from the common hepatic artery (CHA). [4] The underlying etiopathogenic mechanisms are not fully understood. However, proposed contributing factors in acquired HAA include arteriosclerosis, medial degeneration disease, vasculitis, fibromuscular dysplasia, secondary periarterial inflammation and trauma. [5,6] Congenital HAA has been associated with genetic and connective tissue disorders including Ehlers-Danlos syndrome, Marfan syndrome, Osler-Weber-Rendu syndrome and polycystic liver disease. [2]

Clinically, HAA are often asymptomatic until rupture with most cases detected incidentally through imaging. However, among symptomatic patients, 14-80% present acutely with aneurysmal rupture, commonly exhibiting the classical Quincke's triad of pain, jaundice and

anemia. [2] Although rare, obstructive jaundice can occur because of extrinsic compression of the biliary tree by the aneurysm. In our case the patient presented with painless obstructive jaundice and anemia without evidence of aneurysmal rupture.

Reported cases of HAAs have increased, likely due to the widespread use of cross-sectional imaging modalities. Computed tomography (CT) angiography, magnetic resonance (MR) angiography, and formal aortography with celiac artery angiography have all been documented as effective diagnostic tools. Angiogram remains the gold standard for diagnosis of HAA, offering the additional advantage of guiding decision on therapeutic embolization and facilitating precise surgical planning in appropriate cases.

Unlike aortic aneurysms, the correlation between size of HAA and risk of aneurysmal rupture remains debatable. While intervention is universally recommended for ruptured aneurysms the management of asymptomatic cases remains controversial. Abba et al. recommend intervention in any patient with non-atherosclerotic aneurysms, multiple HAAs, and aneurysm exceeding 5cm in diameter due to the increased risk of rupture. [1] Management strategies for HAA range from conservative observation to endovascular or surgical resection. The selection of an appropriate intervention should consider factors such as aneurysm location, size, surrounding anatomical structures and the feasibility of radiological intervention techniques. Endovascular approaches include aneurysm embolization, stenting across the parent vessel and embolization of the common hepatic artery, with embolization being the preferred treatment for intrahepatic aneurysms. [7] Surgical techniques include hepatic resection for a large intrahepatic aneurysm, excision with arterial or venous jump graft placement, bypass using a jump graft and surgical ligation provided that sufficient collateral circulation is maintained. [9] However, in instances where collateral circulation is inadequate arterial reconstruction may be necessary. [8]

CONCLUSION

Although rare, HAA should be recognized as a potential but unusual cause of painless obstructive jaundice in pediatric patients, more especially, in those with underlying risk factors. Prompt and thorough investigation is essential to guide appropriate treatment modality given the significant risk of aneurysmal rupture, the most severe and life-threatening complication. Treatment should be tailored to the patient, and available resources and surgical expertise. A multidisciplinary approach is crucial in determining the optimal management strategy and carefully weighing both immediate and long-term complications to ensure the best possible outcome for the patient.

For pediatric surgeons, the key lesson is vigilance: even rare vascular lesions such as HAA can masquerade as biliary obstruction, and early diagnosis is essential to prevent life-threatening rupture.

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