

Haemobilia: secondary to micro aneurysms of hepatic artery

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Abstract

Hepatic artery is the fourth most common site of the intra-abdominal aneurysm, after infra renal aorta, iliac artery and splenic artery aneurysms. Rupture of the aneurysm may lead to the upper gastrointestinal haemorrhage. Here we report a 5 years old boy, who presented with fever, abdominal distension and unexplained upper GI bleed. Upper GI endoscopy revealed a normal esophagus and stomach with clear evidence of haemobilia with blood oozing from the ampulla. Fluoro- guided angiography followed by embolization of hepatic artery branches with 5 metallic coils was performed in this case by an interventional radiologist.

Keywords: Microaneurysms of Hepatic artery, Haemobilia, Fluoro guided angiography, Coil embolization.

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Introduction

An Aneurysm is a ballooning greater than 50% of the vessel's normal diameter or a bulging, weakened area in the wall of a blood vessel. Abnormal communication between hepatic arterial and biliary systems leads to Haemobilia. In 1959 Hepatic artery aneurysm (HAA) was first reported in children by Jewett.¹ As the majority of patients are asymptomatic hence were not initially diagnosed in many cases. Rupture of the aneurysm is the first clinical manifestation in 80% of the cases.² Other clinical presentations include abdominal pain in 55% and gastrointestinal haemorrhage in 46% of patients.² Epigastric pain, Haemobilia and obstructive jaundice is a classic triad present in one third of the cases.² Here we describe a case of Microaneurysms of Hepatic Artery in a 5 years old boy, who presented with fever, abdominal distension and unexplained upper GI bleed.

Case Summary

We are reporting a 5 year old boy resident of Waziristan,

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who presented at Gastroenterology & Hepatology Department of The Children's hospital & The Institute of Child Health, Lahore in September 2015. Consent to enroll him in this report was given by his parents. He was admitted in hospital with history of fever and abdominal distention for 5 months, along with history of copious amount of haematemesis for one month, followed by bleeding per rectum 2 months later requiring frequent blood transfusions. Neither was there a previous history of trauma, nor was there any history of joint, skin, or systemic involvement. At the time of presentation in his native tertiary care city hospital, he was sick, febrile with profuse bleeding per rectum where he was extensively worked up. On general physical examination his height was 92cm and weight was 13 kg, both were at 25th centiles. He was pale with no jaundice or peripheral stigmata of chronic liver disease. Systemic examination revealed distended, tender abdomen with laparotomy scar mark, liver total span was 12 cm, firm in consistency, smooth surface regular borders, with no ascites. Rest of the systemic examination was unremarkable. He was stabilized by giving IV fluids, packed cells and antibiotics.

Initial lab data of our index case with the haemobilia is

Table: Laboratory data of our index case with Hemobilia.

Investigations	Laboratory findings	Normal Range
Haemoglobin	9.38g/dl	11.5-13.5g/dl
TLC*	15.7x 10 ⁹ /l	5.0-17.0x 10 ⁹ /l
DLC(Neutrophils)**	70%	54-62%
ESR***	68mm/hr	3 - 13 mm/hr
ALT (SGPT)****	76 U/dl	5-20 U/dl
Serum Amylase	23U/L	23 - 85 U/L
LDH *****	226U/L	60 - 170 U/L
Ascitic fluid Analysis		
TLC*	730/μL (Polys: 39%, Lymphocytes: 61%,)	<250/ μL
RBC*****	3250/μL	>100/μL (Malignancy, TB), >100,000/μL (Intra- Abdominal trauma, None=Normal
Total Protein	4.9gm/dl	0.3-4gm/dl
SAAG*****	0.62g/dl	>1.1g/dl=Transudate, <1.1g/dl=Exudate

*Total Leucocyte Count, **Differential Leucocyte Count, ***Erythrocyte Sedimentation Rate, ****Alanine Transaminase, *****Lactate dehydrogenase, *****Red Blood Cell Count, *****Serum-Ascites Albumin Gradient.

mentioned in Table. His viral screening, autoimmune panel was negative. Radioisotope scan done was found to be normal as well. His initial abdominal ultrasound and CT scan showed hepatosplenomegaly with moderate ascites and bilateral effusion. He was discharged on first line ATT with no improvement in symptoms. He developed frequent episodes of haematemesis and bleeding per rectum. His upper and lower GI endoscopies were inconclusive. By now (over a period of 5 months) he had received 28 whole blood and 13 fresh frozen plasma transfusions. He underwent Exploratory Laparotomy to find the source of bleeding, which showed two bluish spots over the anti-mesenteric border of jejunum one foot from DJ junction, No haemangioma was found. About 20 cm jejunum was resected and end to end anastomosis was done. Resected jejunum showed no evidence of dysplasia or malignancy. However, his symptoms persisted and the patient was referred to our unit. We reviewed clinical details and labs and re-sent his vascular parameters which showed Hb: 6.8 gm/dl (11.5-13.5gm/dl), TLC=25.5x10⁹/l (5.0-17.0x10⁹/l) with 65% Neutrophils (Normal range: 54-62%), Plt=584x10⁹/l (150-400 x10⁹/l), with normal liver, renal and coagulation profile. Urine and blood culture were sterile, abdominal USG revealed mildly enlarged liver with altered echo texture with periportal fibrosis, the periphery of the liver showed few hypo echoic areas, the impression was an inflammatory or fibrotic disease with no definite biliary dilatation seen. Upper GI endoscopy was performed again, which revealed a normal oesophagus and stomach with clear evidence of Haemobilia with spurts of blood from ampulla. With these endoscopy findings diagnosis of hepatic artery aneurysm was suspected. Magnetic resonance cholangiopancreatography (MRCP) was done. Anti-phospholipid antibodies & c ANCA were sent which were negative. Case was discussed with the interventional radiologist. In order to confirm we performed fluoro-guided angiography followed by embolization of hepatic artery branches with 5 metallic coils. Post embolization abdominal ultrasound showed no sonographic evidence of any collection in liver and gall bladder. Magnetic Resonance Angiography (MRA) of brain was also normal. The patient came for follow up once and is now doing well on follow-up of referring paediatrician.

Discussion

Hepatic artery aneurysm (HAA) is clinically important and

uncommon.³ Following infrarenal aorta, iliac artery and splenic artery aneurysms, hepatic artery constitutes the fourth most common site of the intra-abdominal aneurysm. It represents approximately 20% of all visceral aneurysms, of which 80% are extra hepatic and 20% are intrahepatic.³ The majority of the cases reported with HAA are in adults or older children and with most common etiologies being a mycotic aneurysm, trauma or atherosclerosis, vasculitis such as polyarteritis nodosa, periarterial inflammation caused by cholecystitis or pancreatitis, fibro muscular hypoplasia or cystic medial necrosis.⁴ A couple of instances with congenital HAAs detected during infancy or in older ages have been reported.⁵

Our patient presented more with fever, abdominal distension with an evidence of fluid in the peritoneal cavity and pleural cavity initially, followed by haematemesis and melena requiring multiple blood transfusions. We did not find any definite reason for these findings except systemic infection, which was evident in the form of rising ESR, as a part of his early work up done at some different hospital. Anti-phospholipid antibodies & c ANCA was later also found to be negative, excluding any vasculitis.⁶ Although these initial clinical findings do not fit into the classic triad of haemobilia, obstructive jaundice and epigastric pain, which can only be seen in one third of such patients, there is also no literature report supporting these initial clinical features.

Mortality and rupture rate of hepatic aneurysms are high 76% & 25- 40% simultaneously in HAA.⁷

Diagnostic procedures incorporate upper GI endoscopy, which could unmask rare diagnosis like haemobilia. Dilatation of the bile ducts with blood within the ducts and the gall bladder may be evident on abdominal US & CT scan. CT scan findings are of mixed or uniform high-attenuation blood within the lumen of the gallbladder in patients of Haemobilia although, it is not diagnostic as this may be seen with gallstones, vicarious excretion of intravenous contrast, biliary sludge, and milk of calcium bile.⁸

Our patient presented with fever, abdominal distension followed by haematemesis, upper GI endoscopy revealed haemobilia. MRCP gave a positive pointer towards diagnosis of multiple micro aneurysms in branches of the hepatic artery. It was confirmed and dealt by fluoro-guided angiography. To the best of our knowledge, our

case is the first of its kind in children and has not been reported before. Unlike our case, there is one case of congenital hepatic artery aneurysm reported by Mahmood Haghghat⁹ & colleagues, where a normal acyanotic neonate was found to have cystic mass of about 20 mm in the internal segment of the right lobe and mild dilatation of intrahepatic bile duct. Colour Doppler ultrasonography revealed pulsatile flow within the mass of vascular origin. There was a severe dilatation of the Coeliac trunk with a huge aneurysm, 4 cm in diameter, arising from the hepatic artery and located in the hepatic artery bifurcation, evident on CT angiography.

CT angiography can help to guide a treatment by determining the size, morphology, and location of the aneurysm. For example, intrahepatic aneurysms can be embolized with coils, microspheres, or glue. In 1977, Cho et al.¹⁰ described experimental hepatic artery embolization in four cases with favourable outcome as well. As in our case fluoro-guided angioembolization of hepatic artery branches with 5 metallic coils was performed. Embolization or surgical ligation can be performed for the aneurysms at the level of the common hepatic artery. For aneurysm involving the origin of the gastroduodenal artery, surgery may be easier than a percutaneous approach. Saccular aneurysms with a good neck can be treated with embolization. Saccular or fusiform aneurysms without a good neck require surgical intervention.¹¹

In summary, this case study highlights the importance of considering rare disorders in children who present with common signs and symptoms like upper GI bleed requiring multiple transfusions where basic work-up has failed. We should consider rare entities like Haemobilia and prompt targeted work up should be done to minimize morbidity and mortality. This case would help fellow peers

to consider rare cause of upper GI bleed.

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