Gallbladder Polyp Complicated by Hemobilia in a Child: A Case Report

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Abstract

Gallbladder polyps are the result of mucosal proliferations. Presence of gallbladder polyps may be the first clue to diagnose Metachromatic Leukodystrophy (MLD) in children. Serious complications of this rare condition is their malignant potential and haemobilia. We describe a 6 years old boy with MLD who presented with upper gastrointestinal bleeding and was eventually found to have haemobilia secondary to the presence of a gallbladder polyp. Our case has highlighted the importance of systematic and timely approach to such patients. A review of literature showed no current available guidelines adressing such conditions in children. In this report we propose a suggested approach to the management of gallbladder polyps and haemobilia in children.

Keywords: Leukodystrophy, Metachromatic, Gallbladder, Polyps, Hemobilia

Introduction

Gallbladder polyps form when its mucosa proliferates.[1,2] It occurs in almost 4.5% of the adults,[3,4] while it is rarely reported in children.[3] They can present at different age groups in children, including infancy.[1] In adults, features such as growth rate, irregularity, or association with a gallstone may signify malignant transformation.[1] No similar criteria is specified in children.[1] In fact, gallbladder polyps in children show a variable course, such as increasing or decreasing in size.[1] They may also bleed and cause hemobilia and its consequences.[2]

We present a rare case of haemobilia and gallbladder polyp in a child with Metachromatic Leukodystrophy (MLD).[2,5]

Case Report

A 6 years old boy with MLD, gastroesophageal reflux disease and gastrostomy status (GT), presented with 2 days history of large-volume coffee-ground drainage from his GT. He was tachycardiac, tachypneic and had a soft abdomen. He had no melena or hematemesis. His Haemoglobin dropped from 140 gm/L to 110 g/L (over 4 hours). Urgent esophagogastroduodenoscopy (EGD) was performed. Fresh Blood oozing from the ampulla of Vater was seen. (Figure 1)

Subsequent Computed Tomography (CT) scan with angiography showed a distended gallbladder with an internal hematoma and a dilated inflamed cystic duct wall. His condition improved, initially, with conservative management. However, he continued to drop his Haemoglobin level.

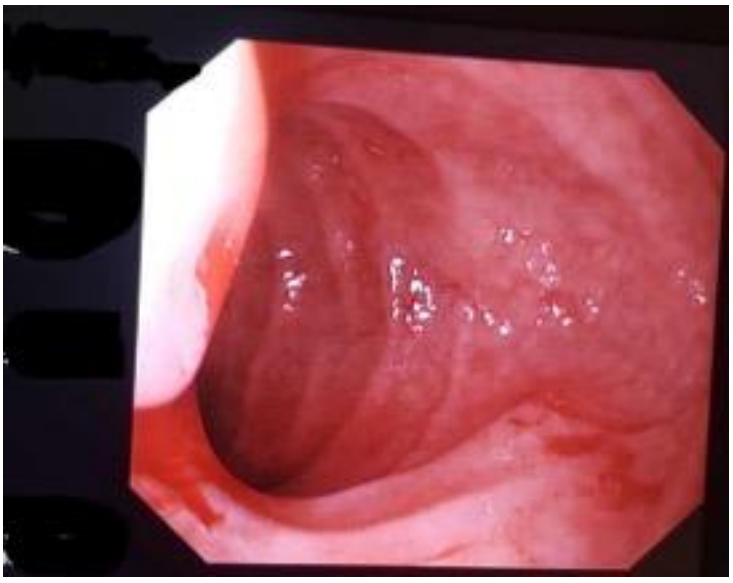

Figure 1: Endoscopic view showing blood oozing from the ampulla of Vater

Concomitantly, he developed features of cholangitis (ALT 304 (5.08 microkat/L), AST 191(3.19 microkat/L), GGT 675 (11.27 microkat/L), total bilirubin 79 micromol/L, direct bilirubin 76 micromol/L and INR of 1.13 which worsened later to 1.46). Magnetic Resonance Imaging (MRI) showed a dilated gallbladder with a polypoid structure within (measuring up to 20 x 40 mm), fed by cystic artery. The common bile duct (CBD) was 7 mm. (See Figure 2) Despite antibiotic coverage and gradual normalization of his liver function tests, his fever and abdominal pain persisted. Urgent laparotomy and cholecystectomy were then performed. It showed signs of intraabdominal and gallbladder inflammation along with dilated CBD.

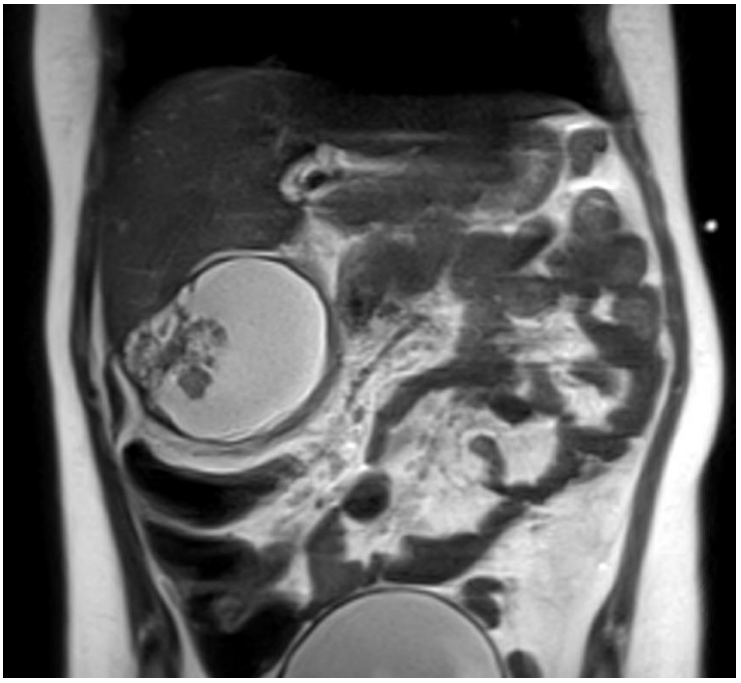

Figure 2: Abdominal MRI images of the child. It shows a mass (polyp) inside the distended gallbladder.

Gallbladder histopathology showed features of chronic cholecystitis. The polyp was typical for benign papillary hyperplasia of the gallbladder with no high-grade dysplasia features (See Figure 3).

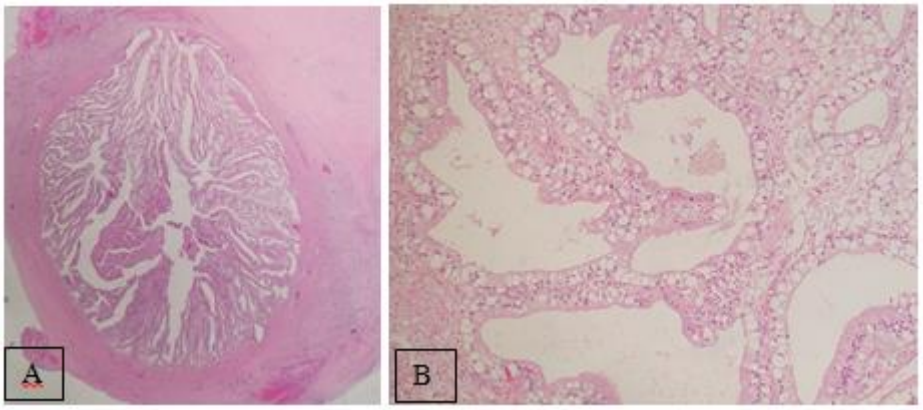

Figure 3: A: Gallbladder papillary hyperplasia - (1. x 4 Magnification): Histology shows a tubulopapillary growth comprising multiple papillae with fibro vascular cores. **B**: The lining epithelium of papillary lesion comprises eosinophilic to clear cytoplasm with bland monomorphic nuclei and inconspicuous nucleoli.

Post operatively, patient's fever and abdominal pain subsided and he was discharged home 5 days later. The patient was seen in the clinic after discharge and was doing well.

Discussion

Gallbladder pathologies in children may result in chronic abdominal pain.[6] The wide use of ultrasonography (US) lead to an increasing detection rate reaching up to 79% in different series.[1-3] Ultrasound can detect gallbladder polyps larger than 10 mm with a sensitivity of 80% and a specificity of 99.3%.[1]

Polypoid lesions in the gallbladder (PLGs) of adults may be of different nature including cholesterol polyps that has no malignant potential (60% of adults' PLGs).[3,4] In Children, PLGs are either primary or secondary.[1,3,4] Primary PLGs in children may be a cholesterol polyp, as well.[4] Similarly, our patient's polyp was of papillary hyperplasia and had a core of cholesterolosis. Despite the above, literature reported an increased malignant transformation risk in cases with underlying MLD.[1,5]

Another determinant of malignant transformation, in adults, is size.[1,4] Thus, cholecystectomy is advised for adults having PLGs larger than 10 mm.[1,4,7] Our patient's polyp size was exactly 10 mm in size. (See figure 3). Presence of concomitant gallstone, a sessile lesion, irregular and thickened adjacent gallbladder wall, and a rapid increase in size are other predictors of malignant nature.[1,4,7,8] Literature review showed no such criteria is set for children so far.[1] In fact, PLGs in children show different courses, as some may disappear on follow up, making the prediction of malignant transformation a challenging task in children.[1]

Nonetheless, among conditions associated with secondary PLGs in children is MLD,[3,4] similar to our patient. Seventy six percent of patients with MLD have gallbladder abnormalities (1/4 have polyps).[5] MLD, a lysosomal storage disease, results in diffuse metachromatic sulfatides (a glycolipid) accumulation in various tissues including the gallbladder.[2,4,5,9-11] It results in subsequent wall thickening and polypoid mass formation. It usually consists of papillary hyperplasia and macrophages containing metachromatic material, typical to our patient.[4,5,12] In fact, gallbladder polyps may be the first clue to the diagnosis of MLD, preceding neurological symptoms.[9] Despite the rarity of PLGs and non-traumatic haemobilia in children, MLD is considered the main cause for PLGs in children.[2,6]

Haemobilia results from a communication between the hepatic vascular system and the biliary system (25% mortality rate).[2,13,14] In our patient a feeding vessel to the polyp was identified. The rare non traumatic haemobilia may present with classical biliary colic, obstructive jaundice and gastrointestinal bleeding,[6,13,15] which is not seen in our patient. In the literature, a proposed diagnostic approach is to first exclude infections and perform an US before proceeding with endoscopy.[13,15] However, due to the rapidity of symptoms, rarity of haemobilia in children and the rarity of parasitic causes in our region, an EGD was attempted first to exclude more common etiologies. Presence of blood oozing from the ampulla of Vater during EGD confirmed haemobilia.[6,13,15]

Clots, secondary to haemobilia, may persist in the biliary tree, resulting in biliary obstruction and cholangitis (similar to our patient).[13,14] Clots can mimic a mass, stone or sludge.[1,6,13] They may appear as mobile or hypoechoic masses as well as non-shadowing filling defects on US.[1,6] A biliary CT with contrast and CT with angiography, are useful tools to demonstrate haemobilia, and to plan management.[6,13] However, MRI and MRCP are best imaging modalities for haemobilia in children, as less radiation is involved and bleeding can be demonstrated as mixed signal intensity.[6,13]

So far, treatment plans for PLGs in children is extrapolated from adult studies.[1] Literature has suggested different approaches to managing haemobilia in children [Figure 4].[4,6,8]

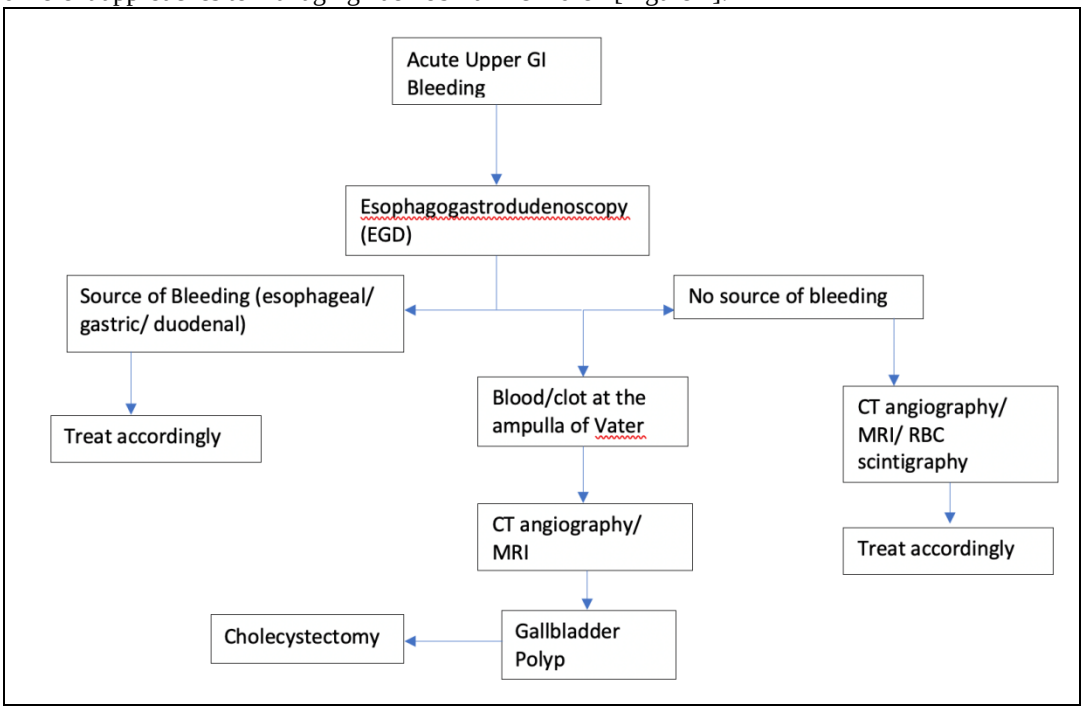

Figure 4: Suggested diagnostic approach for a child presenting acutely with upper gastrointestinal bleeding secondary to a gallbladder polyp and haemobilia. GI: gastrointestinal, CT: Computed Tomography, MRI: Magnetic resonance imaging, RBC: Red blood Cell.[1,4,7]

in 2013, the American college of radiology recommended a yearly follow up of polyps sized 7-9 mm in adults. A surgical consultation is needed if they continued to increase in size.[4] Furthermore, other studies recommended cholecystectomy for all polyps greater than 10 mm, those larger than 5 mm but with risk factors and for those growing 2 mm or more in a year.[1,5] Otherwise serial US is recommended to avoid unnecessary interventions in children, especially that many may disappear over time.[1,5]

Conclusion

Although the primary concern about PLGs may be their malignant potential, they may, also, result in the rare serious haemobilia and hemodynamic instability. Thus, early diagnosis is of paramount importance. Adequate follow up and timely management is crucial. Cholecystectomy should be attempted if patient shows signs of malignant transformation or uncontrolled bleeding. This report raises awareness and proposes a needed approach to these patients.

Acknowledgment

All authors contributed to the data collection, management, data analysis and preparation and review of the manuscript.

Disclosure

The authors declared no conflicts of interest, such as financial or any other relationship/affiliations with the subject. Written consent was obtained from the patient/kin of the patient.

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