Case Report

Benign papillomatosis of common bile duct in children: A rare case report

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ABSTRACT

In children benign neoplasms are rare events in the extrahepatic biliary ducts and scanty literary references on the subject are available. We are reporting a rare case of a 3-year-old female child having biliary papillomatosis in lower common bile duct.

KEY WORDS: Extrahepatic biliary papillomatosis, lower common bile duct, polyp

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INTRODUCTION

Benign neoplasms in the extrahepatic biliary ducts are rare in children. Their importance lies in their ability to mimic malignant lesions in these locations. Commonest presenting symptoms are pain, jaundice, and acute cholangitis. These cases are often diagnosed as intraoperative surprises. We are presenting our experience with one such case.

CASE REPORT

Our case was a 3-year-old female child who presented to our pediatric surgical outpatient department with complaints of pain in the upper part of the abdomen along with jaundice and fever for last 6 months. On per abdominal examination no lump or hepatosplenomegaly was found. With the clinical possibility of a choledochal cyst, ultrasonography (USG) along with liver function test and routine hematological investigations were advised. USG showed impacted worm-like appearance in the lower common bile duct (CBD) with proximal dilatation [Figure 1]. Choledochal cyst was ruled out. In view of the deepening jaundice, child was advised endoscopic retrograde cholangiopancreatography (ERCP) to clear the worm load in the lower end of CBD and to place the stent for establishing the bile flow. Thus, endoscopic sphincterotomy with placement of 10 Fr stent was done [Figure 2]. There were no worms in the CBD in this patient. The child also received anthelmintics. Following stent placement child recovered from jaundice within 2 weeks. As there was a doubt of persistent worm in the distal CBD, exploration was planned; and after obtaining informed written consent, the child was explored under general anesthesia. Right upper transverse incision was given, abdominal cavity was opened, liver mobilized, gall bladder identified, and CBD traced using stent as a guide. On opening the CBD multiple polyps were found with stent *in situ* [Figure 2] without stone or worm. In view of possible malignancy a frozen section was sent and when confirmed to be benign, polyps were removed completely along with stent and CBD was repaired over a T-tube. Postoperatively T-tube was removed after 3 weeks. The child recovered uneventfully and presently she is in close follow-up and doing well. Specimen sent for histopathological examination showed adenomatous polyps with colonic metaplasia.

DISCUSSION

Benign tumors and tumor-like lesions of the gallbladder and bile duct have a wide spectrum and despite this

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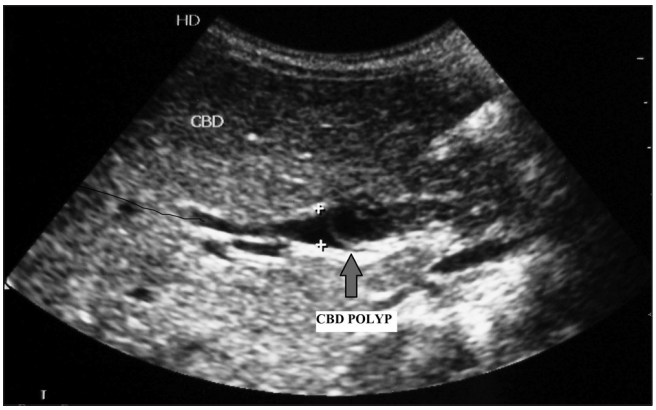

Figure 1: Ultrasound showing filling defect in the lower common bile duct (CBD) polyp

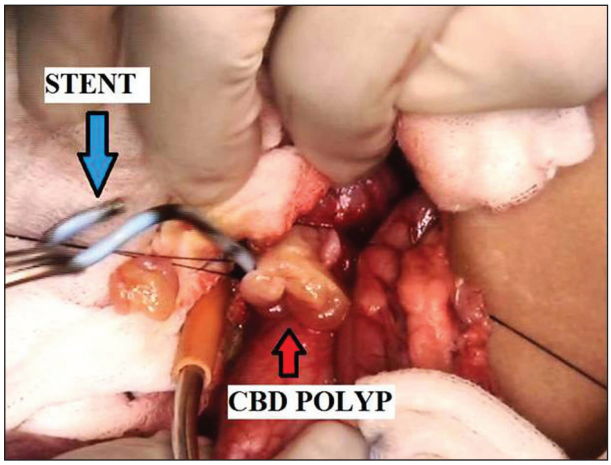

Figure 2: Intraoperative photograph showing CBD polyp and stent

diversity these lesions share common embryologic origins and histological characteristics. Although in children these lesions are relatively uncommon, their importance lies in their ability to mimic malignant lesions in these locations. Benign neoplasms are derived from the epithelial and nonepithelial structures that compose the normal gallbladder and bile ducts. The epithelium gives rise to adenomas, cystadenomas, and the unusual condition of biliary papillomatosis.[1] Granular cell tumors, neurofibromas, ganglioneuromas, paragangliomas, and leiomyomas are examples of benign tumors that may originate from nonepithelial structures.[1] Biliary papillomatosis is rare with only 140 cases reported in literature that too in adults.[1] Chappet in 1894 reported the first case of biliary papillomatosis.[2] The malignant potential of benign polypoid lesions of the extrahepatic biliary system is controversial and not fully understood. The current literature supports the notion of malignant transformation of these benign epithelial lesions.[3-5] Reported male:female ratio is 2:1 with peak incidence in the 4th decade of life.[3,4] Commonest presenting symptoms are pain, jaundice, and acute cholangitis.[3,4,6] Diagnosis of these cases predominantly is intraoperative with few cases being diagnosed through ultrasound.[7] Most of them are isolated case reports only.[8] All case reports are from adult literature. Despite extensive PubMed and Medline search we were not able to find a single case report on this topic in children.

CONCLUSION

From this case report we can conclude that although rare, biliary papillomatosis should be kept in mind when examining a patient with obstructive jaundice, acute cholangitis, and abdominal pain in children.

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