



3D LAPAROSCOPY IN IDIOPATHIC INFLAMMATORY BILIARY STRICTURE IN CHILDREN: A CASE REPORT

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ABSTRACT **Background:** Common surgical causes of obstructive jaundice in children are biliary atresia and choledochal cyst. Extrahepatic biliary obstruction as in adults is not very common in paediatric age group. Jaundice secondary to idiopathic stricture of the hepatic duct is still rarer. We report two such children with isolated idiopathic hepatic duct strictures managed successfully with 3-D laparoscopy. **Case presentation:** One girl aged 20 months and a boy 6 years, both belonging to Asian ethnicity, presented with jaundice. Investigations revealed obstructive jaundice with suspected stricture of extrahepatic biliary ducts. Both were treated by percutaneous biliary drainage as initial management to reduce the jaundice followed by 3-dimensional laparoscopic management. The girl had stricture at the confluence of hepatic duct and was managed with resection of stricture along with CBD excision and cholecystectomy with hepatico-duodenostomy. The boy had stricture in the distal common hepatic duct and was managed with excision of stricture followed by hepatico-choledochal anastomosis. **Conclusion:** Traditionally, management of these strictures is by open surgery. Recently, endoscopic management has been reported; however, laparoscopic management in children has never been reported in English literature, thus this report is the first using 3D laparoscopy and the girl who was 20 months of age is the youngest child treated with 3-D laparoscopy.

KEYWORDS : Obstructive Jaundice, Biliary Stricture, 3d-laparoscopy, Youngest Child

INTRODUCTION:

Common surgical causes of obstructive jaundice in children are biliary atresia and choledochal cyst. Extrahepatic biliary obstruction as in adults is not very common in paediatric age group. Jaundice secondary to idiopathic stricture of the hepatic duct is still rarer. We report two such children of isolated idiopathic hepatic duct strictures managed successfully with 3-D laparoscopy.

CASE PRESENTATION:

Case 1: A 20-month-girl of Asian ethnicity was admitted with history of fever, jaundice, acholic stools and pruritus for 20 days. Examination was essentially normal except icterus. Serum bilirubin levels were - total/direct: 13/ 8 mg/dL respectively. Multiple ultrasonography examinations showed bi-lobar dilation of intra-hepatic biliary ducts suggestive of Caroli's disease and one of the ultrasounds in addition even reported normal gall bladder and common bile duct (CBD). Contrast enhanced computed tomography (CECT) abdomen and magnetic resonance cholangio-pancreatography (MRCP) showed moderate bi-lobar intrahepatic biliary radical (IHBR) dilation with abrupt cut-off at the confluence; common hepatic duct (CHD), the CBD and gall bladder were not seen, suggesting possibility of proximal CHD stricture (Figure 1). HIDA scan showed no activity in the GI tract even on 24 hours follow up scan. In view of hyperbilirubinemia, percutaneous transhepatic biliary drainage (PTBD) was done to reduce jaundice followed by definitive treatment after 9 days. Percutaneous transhepatic cholangiogram (PTC Gram) showed dilated right and left hepatic ducts with abrupt cut-off at the confluence (Figure 2). In view of uncommon and uncertain diagnosis, a diagnostic 3D laparoscopy with intraoperative cholangiogram (IOC) followed by definitive, drainage procedure was planned. Meticulous, careful dissection wherein 3D laparoscopy helped tremendously which showed hugely dilated left and right hepatic ducts at porta. On further dissection, between the anterior and posterior branches of right hepatic artery, there was a narrow segment of CHD suggestive of stricture (Figure 3). The duct was traced to CBD distally. The two dilated hepatic ducts at porta were opened by hook cautery at the most

prominent part. Intra-operative contrast study through this opening showed a collapsed right ductal system and dilated left ductal system possibly because of PTBD draining the right system preferably (Figure 4). Intra-operative ultrasound was also done to confirm this anatomy. Proximal end of CHD stricture was identified just distal to the confluence, and was tied with polyglactin 5-0 and cut. The lower end of CBD was ligated and cut. The CHD stricture was excised along with cholecystectomy followed by a hepatico-duodenostomy done with polyglactin 5-0 in a single layer, with all knots kept outside. Postoperatively, bilirubin values gradually came down and on 9 months follow up, the child is anicteric and the bilirubin levels are 0.92/0.16. HIDA scan on 12 months follow up showed normal biliary to enteric drainage.

Case 2: A 6-year-old boy of Asian ethnicity presented with jaundice of 3 months duration. On examination, he was icteric without hepatosplenomegaly. Bilirubin levels were total/direct of 15.5/9mg/dL respectively. Ultrasound examination showed altered liver echotexture with IHBR dilation but gall bladder, CBD were not dilated. CECT abdomen and MRCP showed bilateral IHBR dilatation with abrupt cut-off at the confluence suggestive of CHD stricture and a 14x16mm peri-portal lymph node (Figure 5). A working diagnosis of obstructive jaundice due to CHD compression by periportal lymph node was considered. Endoscopic ultrasound guided fine needle aspiration cytology of the periportal node showed only reactive changes. Hence, PTBD followed by balloon dilatation and stenting of the CHD stricture was done. The bilirubin levels reduced to 5.4/ 3.1 mg/dL. The PTBD accidentally came out. The patient came after 3 months with palpable hepatomegaly and bilirubin levels of 3.1/1.9 mg/dL. Repeat MRCP showed narrowing of the CHD just proximal to the insertion of cystic duct with dilated CHD till 2cm distal to the confluence (Figure 6). Child was posted for diagnostic 3D laparoscopy with excision of stricture and end to end anastomosis. It showed liver with early cirrhotic changes, a distended gall bladder with dense adhesions to colon and duodenum. Extensive adhesiolysis was done to delineate the complex biliary anatomy. Aspiration of gall bladder had

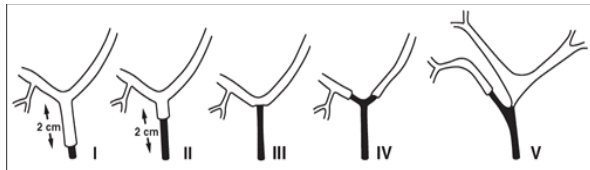
white bile, & IOC showed distal flow of contrast into duodenum with no flow into the proximal biliary tree. Cystic duct was identified with great difficulty and was traced towards CBD. There were lots of dense adhesions in the region. Careful dissection using 3D system revealed the strictured segment of distal CHD just proximal to the insertion of cystic duct, which was traced proximally to the dilated CHD. As CBD distal to stricture and the proximal CHD were of same calibre, the strictured segment was excised along with gall bladder, peri-portal lymph node and a common hepatico-common bile duct anastomosis was done using polyglactin 5-0 in single layer (Figure 7 and 8). Histopathological reports were suggestive of non-specific chronic inflammation in strictured segment and reactive lymph node. The child has remained anicteric and symptom free on 6 months of follow up. HIDA scan at 6 months follow up showed normal liver function and patent biliary-enteric pathway.

DISCUSSION:

Obstructive jaundice in children is most commonly caused by biliary atresia and choledochal cyst. Idiopathic benign non-traumatic inflammatory strictures can also cause extrahepatic biliary obstruction with a reported incidence of 2.2% as reported by Krishna et al [1] of all the causes. Other rarer causes include choledocholithiasis, post-acute pancreatitis pseudo-cyst of pancreas, idiopathic fibrosing chronic pancreatitis, post-cholecystectomy benign biliary stricture, non-Hodgkin's lymphoma (NHL) with extramural common bile duct compression and Langerhans cell histiocytosis.

Biliary strictures are classified as per Bismuth classification – Type 1 is a low CHD stricture > 2 cm distal to confluence, Type 2 is Proximal CHD stricture < 2 cm of confluence, Type 3 includes confluence involvement, Type 4 involves confluence with no communication between left and right ducts, Type 5 includes stricture of an aberrant right sectoral duct. In our 1st case, the stricture can be classified as type 3 and in our 2nd case as type 1.

Bismuth class	Location
I	>2 cm distal to hepatic confluence
II	<2 cm distal to hepatic confluence
III	At the level of the hepatic confluence
IV	Involves the right or left hepatic duct
V	Extends into the left or right hepatic branch ducts



Diagnosis of biliary stricture is based on ultrasound, MRCP and percutaneous or intra-operative cholangiography. Reported management is either endoscopic or surgical treatment.

Traditionally, management of benign biliary strictures in children was open surgery. M J Bowles et al [2] have published their experience of 7 children with benign biliary strictures who had presented with obstructive jaundice and were treated with excision of stricture in 5 patients and stent placement in 1 patient. All 7 patients had undergone open hepatico-jejunostomy as the final procedure with successful outcomes. D'Cunha et al [3] have reported a 1-year-old boy with obstructive jaundice due to extrinsic compression of CHD by anteriorly crossing right hepatic artery causing complete obstruction. Cholangiogram showed distal run-off of contrast with non-visualization of biliary tree proximal to cystic duct and CBD junction. The entire stricture along with CBD was excised and Roux-en-Y hepaticojejunostomy was done anterior to the right hepatic artery. Similarly, S. Watanabe et al [4] have reported a 5 years old girl with biliary obstruction due to extrinsic compression by Burkitt's lymphoma and was treated initially with biliary stenting and chemotherapy. After completion of chemotherapy, Roux-en-Y reconstruction was done in view of persistent stent dependent hepatic duct stricture. The resected bile duct wall showed fibrosis, thickening, and prolapse of the mucosa and muscularis propria on histology.

As per personal communication with Dr. Shah et al from B.Y.L. Nair Ch. Hospital, Mumbai, two similar cases of biliary stricture were operated by open surgery. A 12 years old boy with obstructive jaundice for 8 months underwent exploratory laparotomy. Intra-operatively,

there was stricture involving the cystic duct, common hepatic duct and common bile duct with fibrosis at porta hepatis and cirrhotic liver changes. Excision of stricture with Roux en Y hepaticojejunostomy was done. Histopathology showed ulcerative cholecystitis, non-specific inflammation in stricture and biliary cirrhosis. Another boy aged 5 years presented with obstructive jaundice for last 2 years underwent exploratory laparotomy. Intra-operatively, CHD stricture was identified just at the confluence of right and left hepatic duct (Bismuth type 3). The stricture was excised and Roux en Y hepaticojejunostomy was done. Histopathology identified non-specific inflammation in the stricture. Both patients are asymptomatic on 6 months of follow up.

Recently, there is an evolving use of endoscopic techniques for management of biliary stricture. Pooja Reddy et al [5] have reported benign biliary strictures in 2 infants who were managed with endoscopic techniques. In their first case, a 3 months infant with long segment CHD stricture was managed with serial endoscopic dilation. 2nd case was a 5 months infant with CHD stricture managed with rendezvous technique with combined use of ERCP with PTC to place a stent across stricture segment. Biopsy of the surrounding mass revealed fibrosis and collagen deposition with inflammatory cells. Both patients showed complete resolution of intrahepatic biliary dilation. J. Ferreira et al [6] have successfully treated a 26 months old male child with obstructive jaundice secondary to idiopathic isolated benign non-traumatic biliary stricture at CHD bifurcation with periportal lymph nodal mass. The child was initially treated with surgical removal of lymph node causing CHD compression followed by endoscopic balloon dilation of the narrowed CHD portion. On 11 years of follow up, the child has remained asymptomatic without any recurrent disease. Histological evaluation of the lymph nodes showed reactive hyperplasia and sinus histiocytosis.

Use of minimal access surgery i.e. laparoscopy in successful management of biliary strictures has been reported recently, only in adults. Manash Ranjan Sahoo et al [7] have published their experience of laparoscopic management of biliary strictures in adult population. However, no literature is available for laparoscopic management of biliary stricture in children. Also, there is no report available in English literature of use of 3-D laparoscopy for treatment of benign biliary stricture in children. Both our cases highlight usefulness of 3-D vision in completing the management in children. 3D laparoscopy uses two cameras to reproduce the human binocular vision by producing two differently polarised images of the same object, that are co-displayed on the screen. These can be viewed using eyeglasses containing oppositely arranged polarizing filters on each side thereby permitting depth perception by stereopsis. We were helped by our experience which was highlighted in our publication on 3D laparoscopic management of choledochal cyst in children, offering the advantages of depth perception aiding intracorporeal suturing, using small-sized instruments and thus 'bridging the gap' between conventional laparoscopy and robotic surgery [8]. 3D Laparoscopic biliary reconstruction can be safely done with improved precision, meticulous intra-operative dissection and faster intracorporeal suturing in children.

In a literature review between 2D and 3D laparoscopy, Carmen et al have concluded faster operating time and low error rates with use of 3D laparoscopy owing to better depth perception and spatial orientation which 2D system lacks [9]. In a comparative study of 2D vs 3D laparoscopic assisted anorectal pull through (LAARP) for high anorectal malformation in children, Agrawal et al have concluded that 3D laparoscopy was associated with shorter operating times, better visual parameters in terms of image quality, depth perception and precision allowing a better spatial orientation, reducing manipulation and trauma to tissues [10].

In the 2nd case, initially we tried endoscopic serial dilation of the strictured segment followed by internalization of the PTBD stent. Unfortunately, due to technical reasons internalization of stent could not be done. The PTBD stent had also slipped on follow up and so, patient had to finally undergo laparoscopic hepatico-choledochal anastomosis with excision of the strictured segment using 3D system. Our 1st case is the youngest child treated with 3D-laparoscopy for benign biliary stricture.

CONCLUSION:

Minimal Access Surgery especially with 3D system plays a vital role in diagnosis and management of biliary strictures in pediatric age group.

3D vision provides accurate delineation of the pathologic anatomy and is also best suited for intracorporeal suturing. Excision of the biliary stricture with drainage is the treatment of choice for biliary strictures in children.

DECLARATIONS:

ETHICAL APPROVAL AND CONSENT TO PARTICIPATE:

- Proper ethical approval has been taken from the Institutional ethical committee (IEC-II), Seth GS Medical College and KEM Hospital, Mumbai; Reference number: IEC(II)/OUT/574/2025.
- Consent for publication: Written informed consent was obtained from the patients' legal guardians for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

AVAILABILITY OF DATA AND MATERIALS: Not applicable.

COMPETING INTERESTS: The authors declare that they have no competing interests.

FUNDING: Not applicable.

CASE 1 - IMAGES:



Fig.1 – MRCP showing dilated right and left main hepatic ducts and intra-hepatic biliary radicals with abrupt cut-off at the confluence (arrow).



Fig.2 – PTC Gram showing dilated right and left main hepatic ducts and intra-hepatic biliary radicals with abrupt cut-off at the confluence (arrow).

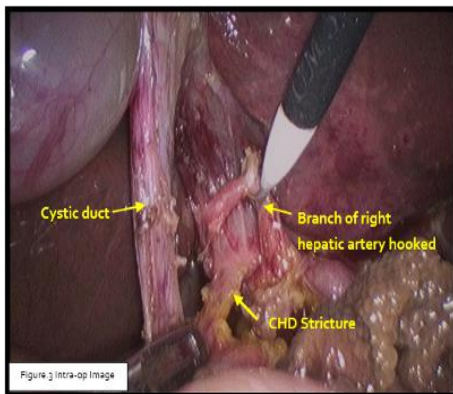


Fig.3 – Intra-operative image showing CHD stricture between anterior and posterior branches of right hepatic artery.

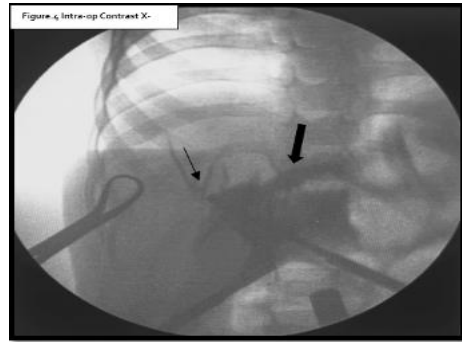


Fig.4 - Intra-op Contrast Xray showing collapsed right hepatic ductal system(arrow) and dilated left hepatic ductal system (bold arrow).

CASE 2 – IMAGES:

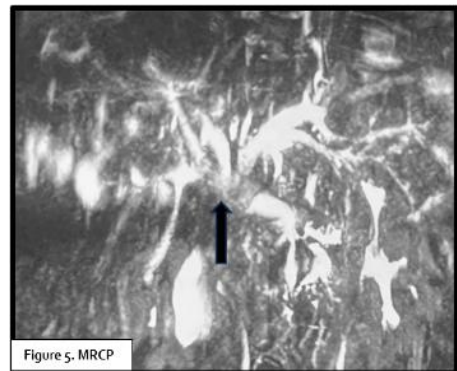


Fig. 5 - MRCP showing dilated right and left hepatic ducts with abrupt cut off at CHD confluence (Arrow).



Fig. 6 - Repeat MRCP showing CHD stricture just proximal to cystic duct insertion (Arrow).

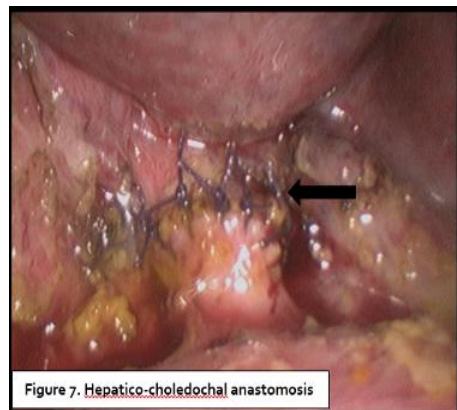
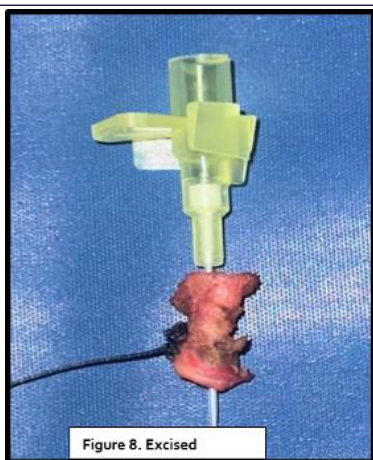


Fig. 7 – End to end hepatico-choledochal anastomosis done.

Fig.8 - Excised specimen showing passage of 24 G cannula across.



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