JGH Open Open Access An open access journal of gastroenterology and hepatology JGHF

doi:10.1002/jgh3.12119

ORIGINAL ARTICLE

Outcome of radiological interventions in pediatric gastrointestinal diseases: A large tertiary center experience

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Key words
child, gastrointestinal, intervention, radiology.

Accepted for publication 3 November 2018.

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Declaration of conflict of interest: None.

Abstract

Background: Interventional radiology (IR) is an indispensable component of multidisciplinary care in various gastrointestinal (GI) diseases. The literature on safety and utility of IR is limited in children.
Objectives: In this study, we aim to analyze the outcomes of IR in various pediatric GI diseases.
Methods: The data of children (≤18 years) who underwent radiological interventions for GI disorders (2009–2017) were analyzed, retrospectively. The indications for interventions included vascular (Budd Chiari syndrome [BCS], pseudoaneurysm) and nonvascular (pancreatic fluid collections [PFCs], cholangitis and anastomotic biliary strictures). The outcomes of radiological interventions, including success and adverse events, were assessed.
Results: A total of 93 children (mean age 13.45 ± 4.09 years) underwent radiological interventions for vascular (chronic BCS = 14, pseudoaneurysm = 28) or nonvascular (PFCs = 33, hepaticojejunostomy strictures or leaks = 12, cholangitis = 6) indications. Of 33 children who underwent drainage of PFCs, clinical success was noticed in 32 children during a mean follow-up of 32.4 ± 21.66 months; 11 children with persistent external pancreatic fistula were managed with endoscopic pancreatic ductal stenting (8 children) and internalization of transgastric drain (3 children). In children who underwent hepatic vein or inferior vena cava stenting for BCS, mean stent patency was 78.57% during a follow-up of 24.1 ± 13.78 months. In children with pseudoaneurysms, angioembolization was successfully performed in 92.8% patients. Re-bleeding was noticed in two children, one of whom required reintervention. In children who underwent percutaneous transhepatic biliary drainage, resolution of anastomotic strictures was noticed in all during a follow-up of 36.1 ± 13.73 months.
Conclusion: Interventional radiology is safe and effective in the management of various pediatric GI diseases.

Introduction

Interventional radiology (IR) plays an indispensable role in the management of various gastrointestinal (GI) diseases. IR is used either as a primary treatment modality or as an adjunct to therapeutic endoscopic interventions. Recent advancements in therapeutic GI endoscopy have minimized the morbidities associated with surgery for many of the GI diseases, like pancreatic fluid collections (PFCs), large polyps, GI bleeding, etc. On the other hand, many of the advanced therapeutic interventions may be associated with adverse events like bleeding and perforation. In these cases, IR plays a diagnostic as well as therapeutic role. The major indications for IR in gastroenterology include GI bleeding not amenable to endotherapy, PFCs, biliary obstruction, hepatic or intra-abdominal abscesses, and angiotherapy in vascular stenosis. The spectrum of GI disorders requiring IR differs in children compared to adults. Therefore, the excellent results of IR in adults may not be applicable to pediatric population. Moreover, the indications for IR in adult patients outnumber those in pediatric patients. Therefore, the published literature predominantly describes the outcomes of IR in adult patients. The data in children are limited to small studies, case series, and case reports.

In this study, we aimed to analyze the safety and efficacy of IR in a large cohort of children with different GI disorders.

Materials and methods

The data of all children (≤18) who underwent an IR procedure related to GI diseases was analyzed, retrospectively. The indications of IR procedure, technical and clinical success, adverse events were recorded. All procedures were performed by trained interventional radiologists. Majority of the procedures were conducted using local anesthesia or light sedation supervised by a pediatric anesthesiologist.

All the IR procedures were performed after obtaining detailed informed consent from the parents.

The detailed description has been provided in the supplementary data. In brief, the technique of various radiological interventions is as follows.

Nonvascular interventions

Pancreatic fluid collections. The indications for percutaneous drainage of PFCs included symptoms like pain, fever, vomiting, intolerance to oral diet, nonresponse to conservative management (antibiotics and bowel rest), and inability to perform endoscopic drainage. Percutaneous drainage of PFCs was performed under ultrasonography (USG) guidance using the Seldinger technique. The tract was sequentially dilated over a 0.035-in. J-shaped stiff hydrophilic guidewire to facilitate the placement of a catheter in the pancreatic collections. The route of drainage, that is, transgastric, transretroperitoneal, or transperitoneal, was chosen depending on the site of collection. All the children were reassessed clinically and with imaging after 48–72 h of placing the percutaneous drain to assess the response. USG abdomen was utilized in most of the cases, and noncontrast-enhanced computed tomography was used only in cases of deep-seated collections. Subsequent follow-up was performed at 2-week intervals until removal of percutaneous drainage catheter. Catheters were upsized in cases with persistence of clinical symptoms and collection on imaging. The decision to remove the catheters and stop the drainage was based on the drain output (<10 mL/day in 48 h), resolution of symptoms, and cyst cavity on USG. In cases with increased drain output with high drain fluid amylase, magnetic resonance pancreaticography (MRP) and or endoscopic retrograde pancreaticography (ERP) was performed to localize the site of pancreatic ductal leak. In children with confirmation of pancreatic ductal leak, either ERP with pancreatic ductal stenting was performed, or transgastric catheter was internalized.

Percutaneous transhepatic biliary drainage. Prophylactic antibiotics were administered 1 h before the procedure. Under USG guidance, the preferred bile duct was punctured. The level of stricture was noted, which was negotiated with a catheter guidewire combination. In case of hepaticojejunostomy (HJ) stricture, sequential balloon dilatation was performed. Finally, a silastic transanastomotic stent was placed across the HJ stricture. In patients with intraductal calculi, balloon sweeping of calculi was performed in addition to the above steps. The children were followed up every 3 months, and sequential upsizing was performed for 1 year in an outpatient setting. At the end of 1 year, a cholangiogram was conducted to ensure adequate resolution of HJ stricture (Fig. 1).

Vascular interventions

Angioembolization for pseudoaneurysms. Angiography and endovascular interventions were performed using the right transfemoral approach. After securing a 6F vascular sheath, 5F angiography catheter and 0.035-in. hydrophilic guidewire were used to cannulate celiac, superior mesenteric artery, and inferior mesenteric artery. Angiography was performed to localize any pseudoaneurysm, contrast extravasation, and vessel wall irregularity. Super selective catheterization of the involved vessel (common hepatic artery, gastroduodenal artery, left gastric artery, and splenic artery) was performed using a microcatheter. Vascular abnormalities were treated either by embolization with glue, coils, or both and stent graft placement. Check angiography was performed at the end of procedure. Successful embolization was defined as cessation of blood flow to the target vascular bed. Clinical success was defined as absence of rebleeding within 4 weeks of embolization (Fig. 2).

Budd Chiari syndrome. The diagnosis of Budd Chiari syndrome (BCS) was established using Doppler ultrasound, computed tomography, or conventional venography in all patients. The level of obstruction was classified as: isolated inferior vena cava (IVC) block, isolated hepatic vein (HV) block, and combined HV and IVC block.

In cases with IVC or HV block, balloon angioplasty followed by self-expandable metallic stent (SEMS) placement was performed. In cases with long-segment HV block (>3 cm) involving all the three or two major HV, direct intrahepatic portosystemic shunt (DIPS) was performed.

Technique of hepatic vein and inferior vena cava angiotherapy. HV angioplasty was performed by either transjugular route, percutaneous transhepatic route, or a combination of both approaches. Angiotherapy of IVC was performed by the transfemoral approach in the majority of cases. In cases with failure to negotiate the stenotic segment, a transjugular approach was also used.

After cannulating through the transjugular route (for HV angiotherapy) or transfemoral route (for IVC angiotherapy), a venogram was performed, and the site of occlusion was identified and negotiated using a guidewire–catheter combination. If there was difficulty in negotiating the site of occlusion through the transjugular route, percutaneous transhepatic route was used. The stenotic segment was dilated with a balloon. At this stage, bolus of heparin (50 IU per Kg patient's body weight) was given. If there was no significant gradient (<10 mmHg) across the stenotic segment, the procedure was considered complete. However, if there was residual stenosis or gradient >10 mmHg, stenting was performed (Fig. 3).

Technique of DIPS. After gaining access to the internal jugular vein, pressure measurement of the right atrium was performed to rule out the presence of right heart failure. We used the gun sight approach to perform DIPS. A percutaneous transhepatic puncture was made using an 18-gauge (Chiba) needle under USG guidance toward the anterior branch of the right portal vein, after which entry into the portal vein was confirmed by injection of contrast material. The needle was introduced further towards the intrahepatic IVC under the guidance of USG, and entry into the IVC was confirmed by contrast injection. A guidewire was placed through the needle into the right atrium, which was pulled out using a snare through the internal jugular vein catheter. This was followed by dilatation of the tract, hepatic, and portal entries using balloon. At this stage, a bolus of heparin (50 IU per Kg patient's body weight) was given. Polytetrafluoroethylene (PTFE)-covered stents were used in all the cases. Poststent deployment, the stent was dilated with a 10 mm balloon. The pressure gradients were again recorded with an intent to keep the

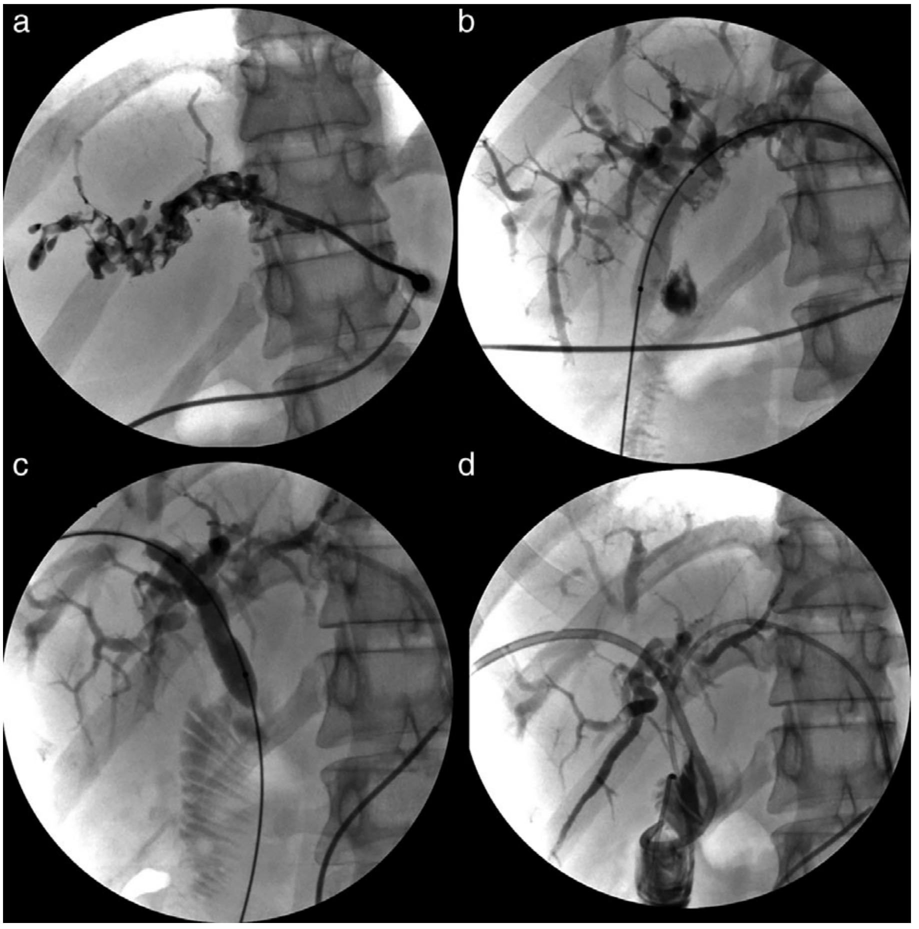

Figure 1 Hepaticojejunostomy stricture with calculi in a 9-year-old child. (a) Cholangiogram in patient with HJ anastomotic stricture shows multiple calculi in both hepatic ducts, (b) balloon dilatation of the H-J anastomosis, (c) balloon assisted sweeping of calculi across the stricture through both left and right ducts, (d) postballoon sweeping, two silastic transanastomotic stents placed across the stricture through right and left ducts with absence of calculi.

Figure 2 Angioembolization in a 17-year-old female with postcholecystectomy bleed. (a) Common hepatic artery angiography demonstrates active contrast extravasation from the gastroduodenal artery, (b) coil embolization of the gastroduodenal artery distal to the site of active extravasation by super-selective catheterization using a microcatheter, (c) check angiography shows complete occlusion of the gastroduodenal artery with absence of extravasation.

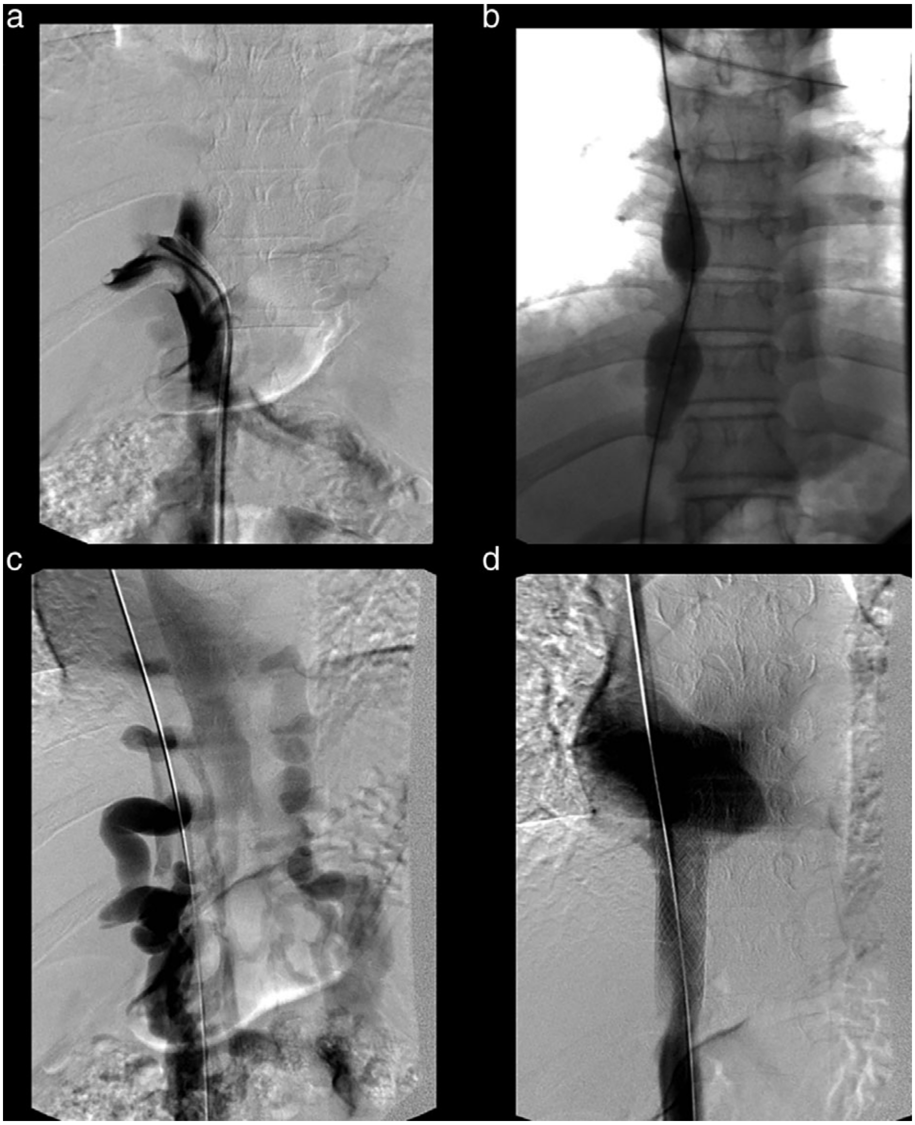

Figure 3 Inferior vena cava stenting in a 15-year-old child with Budd Chiari syndrome. (a) Inferior vena cava venogram showing complete occlusion of the inferior vena cava at the cavo-atrial junction with multiple collaterals, (b) inferior vena cava venoplasty with balloon following negotiation of the web (short segment), (c) postvenoplasty venography showing persistence of venous collaterals, (d) venography following stent deployment shows free flow of contrast across the stent with absence of collaterals.

pressure gradient between 5 and 10 mmHg. To decrease the risk of bleeding, the percutaneous tract in the abdominal wall was embolized (Fig. 4).

Postprocedure monitoring. All the children were observed in an intensive care unit for 24–48 h and started on weight-based heparin infusion every 6 h followed by oral anticoagulation, with an intent to maintain international normalized ratio between 2 and 3. Doppler USG was performed on day 3, day 7, 1 month, 3 months, 6 months, 1 year, and yearly thereafter. The clinical improvement in children with ascites was assessed, and diuretics were withdrawn in whom ascites disappeared.

Statistics. The data are presented as median (range) or mean (SD). The data were analyzed using MedCalc for Windows, version 12.2.1.0 (MedCalc Software, Ostend, Belgium).

Results

A total of 93 children (mean age, 13.45 ± 4.09 years) underwent radiological interventions for various GI disorders during the study period (2009–2017).

The radiological interventions included percutaneous catheter drainage of PFCs in 33 (35.48%), percutaneous transhepatic biliary drainage (PTBD) in 18 (19.35%), angioembolization of aneurysm/pseudoaneurysm in 28 (30.11%), and stenting of HV or IVC and DIPS in 14 (15.05%) children (Table 1).

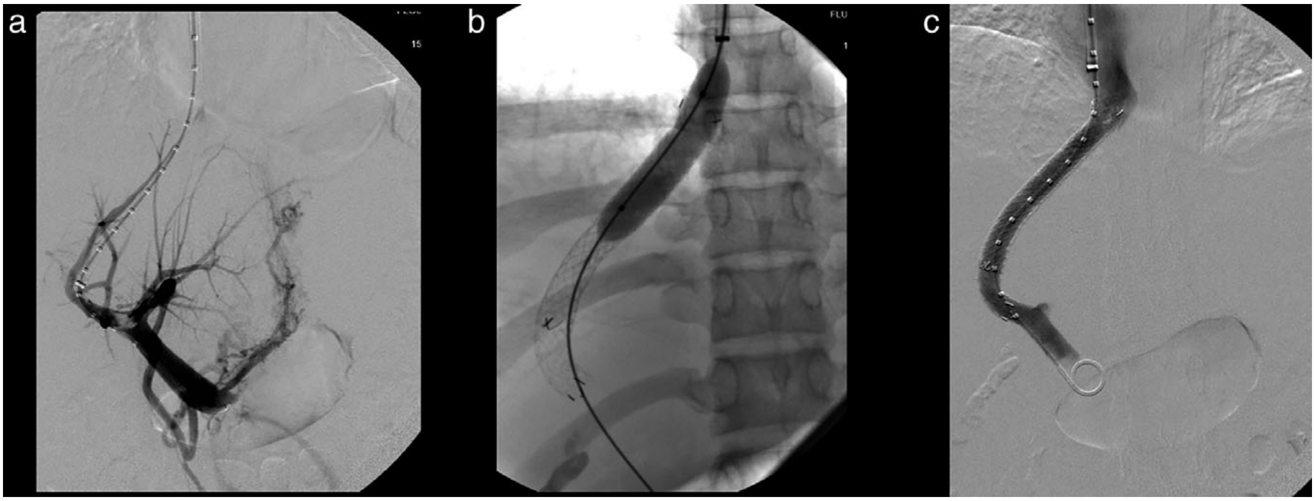

Figure 4 A case of a 16-year-old female with chronic Budd Chiari syndrome who underwent direct intrahepatic portosystemic shunt (DIPS) for refractory ascites. (a) Portal venography following access gained through the intrahepatic inferior vena cava into the intrahepatic portal vein demonstrates multiple collaterals arising from the portal vein suggesting portal hypertension, (b) covered metal stent deployed into the intrahepatic portal vein from the intrahepatic inferior vena cava followed by balloon dilatation, (c) portal venography following stent deployment shows free flow of contrast into the inferior vena cava across the stent with no collaterals.

Nonvascular radiological interventions

Pancreatic fluid collections. Percutaneous drainage of PFCs was performed in 33 children. The median size of PFCs was 8 (5–18) cm. The approach to fluid collections was transgastric in 11 children and direct in collections at other locations. The radiological resolution of PFCs was noticed in all the children. At a mean follow-up of 32.4 ± 21.66 months, one child developed symptomatic recurrence of PFC, for which single-time aspiration was performed under USG guidance. Of 33 children, 11 (33.3%) developed external pancreatic fistula. Of these, ERP with pancreatic duct stent was placed in eight children, and percutaneous transgastric drain was internalized in three children (Table 2).

Percutaneous transhepatic biliary drainage. PTBD was performed in 18 children. The indications for PTBD included benign HJ stricture in nine, HJ site leak in three, and cholangitis due to various other reasons in six children. The duration of catheter placement was 6 and 12 months in children with HJ leak and HJ strictures, respectively. Minor adverse events were noticed in three children, pericatheter leak in two, and local

Table 1 Characteristics of patients who underwent radiological intervention

Total number of patients	93
Male:female	58:35
Age in years, mean ± SD	13.45 ± 4.09
Procedure done	
PCD for pancreatic fluid collection	33
Percutaneous transhepatic biliary drainage (PTBD)	18
Vascular intervention	
Angioembolization of aneurysm/pseudoaneurysm	28
HV/IVC/DIPS for BCS	14

BCS, Budd Chiari syndrome; DIPS, direct intrahepatic portosystemic shunt; HV, hepatic vein; IVC, inferior vena cava; PCD, percutaneous drainage.

Table 2 Characteristics of patients who underwent percutaneous drainage for pancreatic fluid collections

Total patients	33
Male:female	23:10
Age in years, mean ± SD	13.24 ± 4.42
Types of pancreatitis	
Acute pancreatitis	24
Recurrent acute pancreatitis	1
Chronic pancreatitis	8
Types of collections	
Pseudocyst	24
Walled-off necrosis	9
Site of collection	
Lesser sac	11
Multiple sites	7
Pararenal and paracolic	5
Perihepatic	4
Intrapancreatic	3
Subphrenic	3
Perisplenic	1
PCD approach	
Transgastric	11
Direct	22
Median size of collection in cm (range)	8 (5–18)
No of catheters, median (range)	1 (1–4)
Follow-up in months (mean ± SD)	32.4 ± 21.66
Duration of catheter in situ	21.16 ± 14.83
Complication	
Pancreatic fistula	11
*R*eintervention	1

PCD, percutaneous drain.

Table 3 Characteristics of patients who underwent percutaneous transhepatic biliary drainage

Total number of patients	18
Male:female	10:8
Age in years, mean ± SD	12.77 ± 5.12
Indication	
HJ anastomosis leak	3
HJ anastomotic stricture without calculi	4
HJ stricture with calculi	5
Cholangitis with failed ERCP	6
Choledochal cyst	1
Lymphoma	2
Posthepatectomy LHD stricture	1
Post-whipples surgery (SPEN)	1
Intrahepatic calculi (Sickle cell anemia)	1
Duration of catheter in situ (months)	
HJ stricture	12
HJ leak	6
Lymphoma	6
Posthepatectomy LHD stricture	12
Post-whipples surgery (SPEN)	12
Intrahepatic calculi (Sickle cell anemia)	1
Complications	
Pain	10
Pericatheter leak	2
Infection at PTBD site	1
Technical success	100%
Follow-up in months (mean ± SD)	36.1 ± 13.73
Recurrence in HJ strictures	Nil
Clinical success (resolution of cholangitis/leak)	100%

HJ, hepaticojejunostomy; LHD, left hepatic duct; PTBD, percutaneous transhepatic biliary drainage; SPEN, solid pseudopapillary epithelial neoplasm.

infection in one child. Technical and clinical success was noted in all the children (Table 3).

Vascular radiological interventions. Angiographic interventions were performed in 42 children, including conventional angiography in 28 and hepatic vein/inferior vena cava stenting in 14 children.

Angioembolization for pseudoaneurysms. Of 28 children who underwent convention angiography, pseudoaneurysms were detected in 25 children. In one child, angioembolization was performed following massive GI bleed after endosonography-guided drainage of pancreatic fluid collection. One child had true aneurysm of superior mesenteric artery, and in one child, no abnormality was detected. The sites of pseudoaneurysms were splenic artery in eight, gastroduodenal artery in seven, hepatic artery in five, superior mesenteric artery in three, renal artery in one, and right gastroepiploic artery in one child. The complications of angioembolization included pain at puncture site in six, splenic infarct in two, and rebleeding in two children. Of the two children with rebleeding, one child improved with conservative management, while reintervention in the form of glue embolization was required in one child. (Table 4).

Table 4 Characteristics of children who underwent intervention (angioembolization and stent graft) for aneurysm/pseudoaneurysm

Total no. of patients	28
Male:female	17:11
Age in years, mean ± SD	13.71 ± 3.31
Indication	
Aneurysm	1
Pseudoaneurysm	27
Etiology	
Pancreatitis	19
Trauma	5
Post-EUS-guided drainage	1
Postsurgical—cholecystectomy	1
Choledochal cyst excision	1
Ehler Danlos syndrome (true aneurysm)	1 (failed to cannulate)
Site of pseudoaneurysm	
Splenic artery	8
Gastroduodenal artery	7
Hepatic artery	5
Superior mesenteric artery	3
Renal artery	1
Right Gastroepiploic artery	1
Arterioportal fistula	1
Site of true aneurysm	Superior mesenteric artery
Normal angiography	1
Size of pseudoaneurysm (average),mm	9.73 ± 3.04
Technique of angioembolization	
Conventional	28
Agents used for angioembolization	
Coil	14
Coil + glue	5
Glue + lipoidal	5
Graft	2
Complications	
Splenic infarct	2 (7.14%)
*R*ebleeding (gastrointestinal/intracystic)	2 (7.14%) 1—Self-limiting 1—Repeat direct percutaneous glue
Reintervention	1
Technical success	26/27 (failure to cannulate in 1)
Clinical success	24/27 (rebleed—2, failure to cannulate—1)

Angiotherapy for BCS. Angioplasty was performed in 14 children with chronic BCS. In majority of children, angioplasty was followed by stent placement in HV/IVC. In one child, IVC angioplasty alone was performed. IVC and HV stenting were performed in five and four children, respectively. IVC with common iliac stenting, IVC with HV stenting, IVC with DIPS, and DIPS alone were performed in one child each. During a follow-up duration of 24.1 ± 13.78 months, reocclusion was noticed in two children (14.28%). One of these children underwent successful repeat intervention, while the other child succumbed to the disease. Overall, adverse clinical outcomes were noticed in four children (28.57%), including persistence of ascites (one child),

Table 5 Characteristics of patients who underwent hepatic vein/IVC stenting

Total no. of patients	14
Male:female	7:7
Age, mean ± SD	13.61 ± 4.25
Indication	
Refractory ascites	7
GI bleeding	1
Pedal edema	6
Procedure	
IVC stenting	5
HV stenting	4
DIPS	1
IVC + Common Iliac vein stenting	1
IVC stenting with DIPS	1
IVC + HV stenting	1
IVC angioplasty	1
Size of stent (mm)	
IVC stent	18 × 40, 20 × 40, 24 × 50
HV stent	12 × 40
DIPS (PTFE)	10 × 100
IVC and DIPS (PTFE)	20 × 40, 10 × 100
Iliac stent	10 × 80
Complications	
No response	1 (DIPS graft patent at 3 months, no resolution of ascites-? Decompensation)
Reocclusion	2 (1—successfully reintervened)
Follow-up in months	24.1 ± 13.78
Death	2 1—Reocclusion (lost follow-up, died at 45 days) 1—Due to intracranial hemorrhage anticoagulant toxicity (after 3 years of intervention
Reintervention	1

DIPS, direct intrahepatic portosystemic shunt; HV, hepatic vein; IVC, inferior vena cava; PTFE, polytetrafuloroethylene.

requirement of reintervention (one child), and death (two children: reocclusion and intracranial bleeding) (Table 5).

Discussion

In this study, we comprehensively analyzed the outcomes of IR in a large cohort of children with GI diseases. The interventions were performed in children with a variety of indications for IR procedure, including nonvascular (pancreatic fluid collections, biliary obstructions) and vascular (angioembolization for pseudoaneurysms and stenting for BCS) indications.

IR was used either as a primary therapeutic modality or as a secondary intervention following failure or complications with other modalities (endoscopy/surgery).

The literature is sparse in children mainly due to the uncommon nature of these diseases in the pediatric population and lack of expertise in pediatric IR.

Percutaneous drainage of PFCs constituted the largest group of our study population (n = 33). In our study, the PFCs not amenable to endoscopic drainage were managed with percutaneous drainage. Although endoscopic drainage of PFCs is gaining momentum, it may not be feasible in all the cases.[1–4] The size of endoscopic ultrasound scope may be disproportionate, with a risk of airway compression in small children.[5] In addition, some fluid collections are distantly placed from the gastric wall rendering them unsuitable for endoscopic drainage.[6] Percutaneous drainage has equal efficacy to endoscopic drainage for pancreatic pseudocysts. Therefore, either of the modalities can be safely used for the management of symptomatic PFCs in children.[7] On the other hand, surgery is associated with higher morbidity and mortality than noninvasive interventions like endoscopic or percutaneous drainage of PFCs.[8,9] Therefore, nonoperative management of pancreatic pseudocysts is preferred in children.[10]

One potential concern with percutaneous drainage is the high occurrence of external pancreatic fistula.[11,12] In our study, nearly one-third children developed pancreatic fistula. In the majority of these cases (n = 8), we could successfully perform endoscopic transpapillary drainage. In the remaining children, we internalized the trans-gastric percutaneous drain. Percutaneous transgastric cystogastrostomy has been described in adult patients with successful outcomes.[13,14] To our knowledge, this technique of internalization of percutaneous drain catheter has not been described previously in children.

The second group of children who underwent percutaneous nonvascular radiological intervention included those with HJ stricture/leak and cholangitis for which endoscopic retrograde cholangiopancreatography (ERCP) was unsuccessful. The options in children with HJ stricture/leak include ERCP or redo surgery. However, these cases are often not amenable to ERCP due to altered anatomy. Moreover, revision of HJ is invasive and usually not preferred by patients.[15] In our study, most of the cases were not amenable to ERCP due to altered anatomy after surgery. In some cases, ERCP was attempted, but biliary cannulation was unsuccessful. Technical and clinical success was achieved in all, and none of the children required reintervention. In our opinion, sequential upsizing and placement of PTBD catheters to achieve an anastomotic diameter > 30 Fr improves outcomes in patients with HJ strictures. This is analogous to endotherapy for benign biliary strictures where sequential placement of increasing number of plastic stents has been shown to improve outcomes.[16] Therefore, percutaneous biliary drainage appears to be a valid and minimally invasive alternative to surgery in such cases.

In the present series, 14 children successfully underwent HV/IVC stenting for chronic BCS. The role of IR in pediatric BCS is not well defined, and long-term outcomes are not known. Limited data suggest that IR is safe and useful in children with BCS.[17,18] In one study, 25 children underwent therapeutic radiological intervention for chronic BCS. Similar to our study, majority of the children (82.6%) had ascites. The stent patency was 75% at a median follow-up of 6.5 months.[18] In another study, the outcomes of HV stenting and TIPSS were better than angioplasty in children with BCS.[17] For the same reason, we performed stenting in a vast majority of our patients. Compared to previous studies, the follow-up duration was longer in our study. The role of IR and impact of angiotherapy on clinical outcomes in children with BCS need to be evaluated in large comparative multicenter trials.

Angioembolization and stent graft was successfully performed in 26 and 2 children with pseudoaneurysms, respectively. The exact prevalence of pseudoaneurysm in patients with pancreatitis is not well known. However, they are associated with substantial risk of rupture and fatal hemorrhages.[19,20] Angiographic intervention is the preferred mode of treatment for visceral artery pseudoaneurysms.[21,22] To our knowledge, the present study is one of the largest describing the utility of IR in pediatric patients with pseudoaneurysms. The most common indication for angiographic embolization in our study was pancreatitis followed by blunt trauma abdomen.

The strengths of our study are manifold. This is the largest single-center series with comprehensive analysis of safety and efficacy of therapeutic radiological interventions in pediatric population. Unlike previous studies which focused on isolated GI disorders, our study population comprised a wide spectrum of GI disorders. However, certain drawbacks are noteworthy. The study was retrospective in nature with inherent flaws. The majority of study population was beyond the infancy age group, and therefore, the results may not be applicable to the same. Finally, all the cases were performed at a tertiary care center by expert radiologists with expertise in pediatric interventional radiology.

In conclusion, therapeutic radiological interventions are safe and valuable for various GI diseases in children. The role and long-term outcomes of certain interventions like stenting in BCS need to be determined further in prospective, randomized studies.

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Supporting information

Additional supporting information may be found in the online version of this article at the publisher's website:

Appendix S1. Supplementary Material