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RESEARCH ARTICLE

DIAGNOSTIC ACCURACY OF DIFFERENT RADIOLOGICAL MODALITIES FOR EVALUATION OF CHOLEDOCHAL CYST- EXPERIENCE AT A TERTIARY CARE CENTER

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ABSTRACT

Background: Choledochal Cysts are congenital anomalies which present as either isolated or combined dilatations of the extra and intra-hepatic biliary tree. The condition typically presents in infancy and childhood. The different radiological investigations used to diagnose choledochal cyst have different diagnostic accuracies. **Methods:** It is a prospective study carried out in the Department of Surgical Gastroenterology, and Department of Radiodiagnosis and Imaging Sheri-Kashmir Institute of Medical Sciences, Srinagar from October 2011 to October 2014. A total of 40 patients were taken for the study. Clinical profile, different radiological imaging details and intra operative findings were collected for each patient and the radiological reports were compared with intra operative findings. **Results:** Out of 40 patients 10(25%) were children upto 18 years of age and 30(75%) were adults with a male female ratio of 1:3. 29(73%) patients had type 1, 11(27%) patients had type 4 choledochalcyst. All type 1 choledocal cysts were type 1A. Hepatobiliary calculi was the most common associated feature found in around 18(45%) patients. The diagnostic accuracy for USG, CECT,MRCP,ERCP was 87%,74%,97%,93% with a sensitivity of 88%,75%,97%,91% respectively. **Conclusion:** Of all the investigations used in the diagnosis of choledochal cyst MRCP is the diagnostic tool of choice.

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INTRODUCTION

Choledochal Cysts (CDC) are congenital anomalies which present as either isolated or combined dilatations of the extra and intra-hepatic biliary tree (Banerjee Jesudason *et al*., 2006). Estimates of actual clinical incidence range from 1 in 13,000 to 1 in 2 million patients (O'Neill *et al*., 1987; Olbourne, 1975). Biliary duct cysts account for approximately 1% of all benign biliary disease (Saxena *et al*., 1988). The condition typically presents in infancy and childhood (Nogorrey *et al*., 1984). Twenty five percent of the patients are diagnosed within the first year of life and 60% before the age of 10 years (Alonso-Lej *et al*., 1959) and the adult presentation is quite uncommon. Female to male ratio as high as 8:1 has been observed (Stain *et al*., 1995). In neonatescholedochal cyst usually presents as abdominal mass or abdominal pain (Stringer *et al*., 1995) whereas children present with the classical triad of jaundice, abdominal pain and abdominal mass, adults present with common biliary or infective complications (Omar *et al*., 2009). Biliary tract malignancy has been reported to occur in 2.5% to 28% of patients with choldochal cyst, representing a risk at least 20 times greater than that of normal population[10] necessitating proper and timely diagnosis. Choledochal cysts were first described by Vater in 1723. In 1959 Alonso-Lej *et al* described 3 types of choledochal cysts, which were later modified by (Alonso-Lej *et al*., 1959; Todani *et al*., 1977). There are many proposed theories about etiology of choledochal cyst but the most favored one is the "Long Common Channel" (LCC) theory, first described by Babitt (Babitt *et al*., 1969). Ultrasonography (USG) can diagnose choledochal cysts with a specificity of 97% in children (Chang *et al*., 2000). It is first line investigation of neonatal jaundice persisting for >2 weeks after birth (Kim *et al*., 1998) and may help to differentiate choledochal cyst (CDC) from biliary atresia.[15]However ultrasound is limited in adults in identifying the choledochoceles because of the frequency of bowel gas overlying the terminal common bile duct and the small size of these cysts. Endoscopic retrograde cholangiopancreatography (ERCP) is an excellent tool for defining biliary anatomy (Irie *et al*., 1998). It is an invasive procedure with therapeutic capability. ERCP is the gold standared for diagnosis of

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Anamalous Pancreaticobiliary Ductal Junction (APBDJ). Morbidity with ERCP ranges from 2% -8% in children and 1% -2% in adults which rises to 10% when combined with sphicterotomy and mortality estimates is estimated between 0.05-0.90%. (Lee *et al*., 1997; Hekimoglu *et al*., 2008; Prasil *et al*., 2001). Magnetic resonance cholangiopancreatography (MRCP) represents the current 'gold standard' in the imaging of CDC (Sugiyama *et al*., 1998). Lee and Lee *et al*. (1997) compared MRCP and ERCP in 46 patients with various extrahepatic biliary diseases, including choledochal cysts, found the overall diagnostic accuracy rates to be similar between the groups. Irie and associates (Irie *et al*., 1998) concluded in a study that MRCP is an important noninvasive diagnostic study for choledochal cysts but that it should not replace ERCP, especially in children. Kim *et al*. (1998) concluded that MR cholangiography is equivalent or superior to conventional cholangiography in the evaluation of choledochal cysts. The aim of our study was to evaluate the diagnostic accuracy of different radiological modalities in the diagnosis of choledochal cyst for better understanding of the disease and its treatment.

MATERIAL AND METHODS

This study was carried out in the Departments of Surgical Gastroenterology and Department of Radiodiagnosis and Imaging Sheri-kashmir Institute of Medical Sciences Soura Srinagar from October 2011 to October 2014. This was prospective study. All patients admitted as choledochal cyst and who fulfilled the inclusion criteria were included in the study. Clinical characteristics, imaging details and operative reports were collected for each patient. The radiological investigations were compared with intra operative findings.

Inclusion criteria were

- Patients with clinical diagnosis of CDC.
- Informed consent of patient.

Exclusion criteria

- Patients not giving consent to participate in the study.
- (2)Patients who were not willing for surgery.
- (3)Patients with severe comorbidities.

After a thorough history, examination, the investigations included all baseline investigations like complete hemogram, liver function test (LFT), kidney function test (KFT), in addition to the specific investigations like USG, CECT abdomen, endoscopic retrograde cholangiography (ERCP), magnetic resonance cholangiopancreatography (MRCP), for confirmation of diagnosis and comparison with operative notes.

Statistical analysis: The data was analysed using spss statistical package, version 13(Chicago, IL) by constructing Reciever operating characteristic curve, sensitivity specificity and accuracy were calculated for various tests against intraoperative findings. P<0.05 was considered as statistically significant.

RESULTS

Out of total 40 patients, children up to 18 years of age were 10 (25%) and adults 30 (75%).

Table 1. Demographic profile of studied subjects

			Number	Percent
AGE	0-18 year	Male	2	5%
		Female	8	20%
	>18 year	Male	8	20%
		Female	22	55%
GENDER		Male	10	25%
		Female	30	75%

Table 2.Types of choledocal cyst seen in our patients

	Cyst Type	Number	%age	Total
Children	Type I	8	80%	10
	Type IV	2	20%	
	Type V	0		
	Type III	0		
	Type II	0		
Adults	Type I	21	72%	29
	Type IV	8	28%	
	Type V	0		
	Type III	0		
	Type II	0		
				39

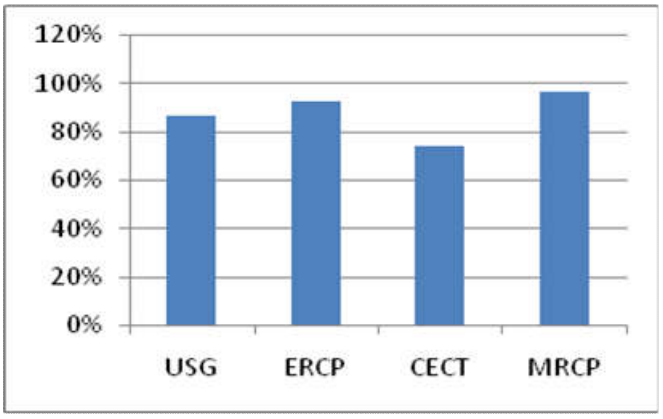

Fig 1. Showing diagnostic accuracy of various investigations

Majority of patients in our study were females (75%) with a male: female ratio of (1: 3). Among the Children, 2 were males and 8 were females and among adults, 8 were males and 22 were females [Table 1]. 29 (74%) patients had type I choledochal cyst, 10(26%) patients had type IV choledochal cyst. In one adult patient, though ultrasonography was suggestive of type I choledochal cyst however MRCP showed stricture in distal common bile duct which was later confirmed by intra operative findings. In children 80% were type I and 20% were type IV choledocalcysts, while as in adults 72% were type I, and 28% type IV. In total, 29/39 (76%) were type I and 10/39 (24%) were type IV (Table 2).

Among the type I patients, type I a were found in 22/29 (76%) and type Ic in 7/29 (24%) patients. 18/40(45%) patients with choledocal cysts had stone formation in the liver, gall bladder and cyst itself. Stricture was present in 1/40(2.5%) patients in the distal part of CBD. Aberrant Right hepatic artery was orginating from superior mesenteric artery in one patient (Table 3). Among the children stones were found in one female child and among the adults, stones were found in 17patients, of these 13 were females and 4 were males. 2(7%) adult patients had stones in the liver and 8 (27%) {6 females and 2 males} had stones in Gall bladder. Cystolithiasis was present in 7 (22%) adult patients {5 females and 2 males}. Ultrasonography was done in all patients and it diagnosed the disease in 35 patients. CECT was positive in 14 patients out of 19 patients.

Table 3. Associated features of choledocal cysts

			Number	%age	P. value
Hepatic calculi	Children	Present	0	0%	>0.05
		Absent	10		
	Adults	Present	2	7%	
		Absent	28		
Gall stones	Children	Present	1	10%	>0.05
		Absent	9		
	Adults	Present	8	27%	
		Absent	22		
Stricture	Children	Present	0	0%	>0.05
		Absent	10		
	Adults	Present	1	3.3%	
		Absent	29		
Pericholedochalinflamation	Children	Present	0	0%	>0.05
		Absent	10		
	Adults	Present	5	17%	
		Absent	25		
Abarrent Rt hepatic artery	Children	Present	0	0%	>0.05
		Absent	10		
	Adults	Present	1	2.5%	
		Absent	28		
Cystolithiasis(stone in cyst)	Children	Present	0	0%	
		Absent	10		>0.05
	Adults	Present	7	22%	
		Absent	23		

Table 4. Diagnostic accuracy of different radiological investigations with respect to the intraoperative findings

S.No.				
1	USG	Positive	35	87%
		Negative	5	13%
2	ERCP	Positive	13	93%
		Negative	1	7%
	MRCP	Positive	39	97%
3		Negative	1	3%
4	CECT	Positive	14	74%
		Negative	5	26%

ERCP was done only in 14 patients and in 13 patients it was positive. MRCP was done in all patients. I none patient ultrasound was suggestive of choledochal cyst however MRCP showed distal common bile duct stricture which was in conformity with the intraoperative findings. In this patient proximal CBD was grossly dilated. In second patient MRCP was suggestive of a small pseudocyst pancreas however intraoperatively type I choledochal cyst was found. So MRCP was accurate with respect to the intraoperative findings in 39/40 (97%) patients (Table 4). MRCP is having maximum sensitivity of 97% followed by ERCP (91%), USG (88%), CECT (75%) (Figure 2). MRCP has picked up the Choledochal cysts accurately in 38 patients out of 39 patients with a sensitivity and specificity of 100 and 67% (Figure 3).

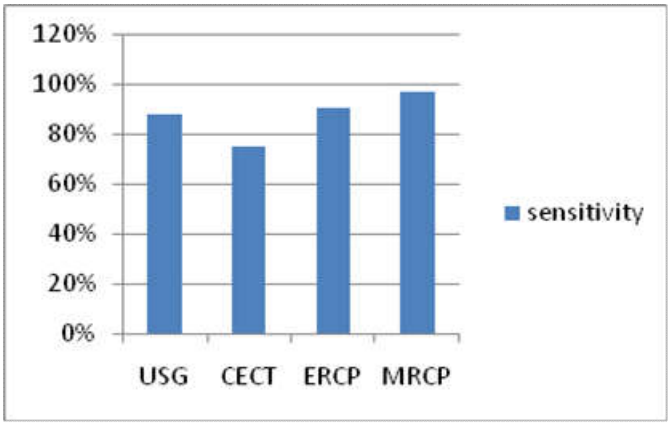

Figure 2. Showing sensitivity of different investigations

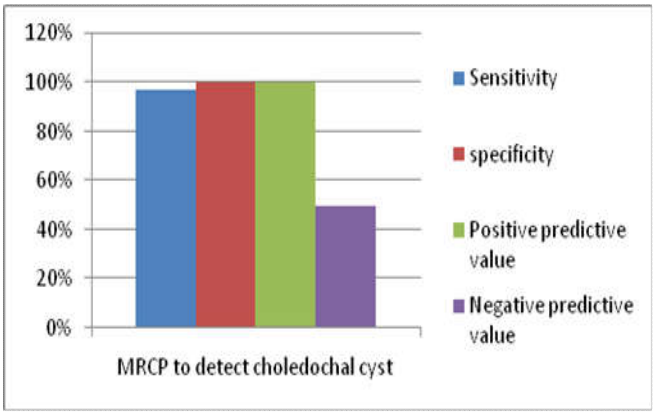

Figure 3. Diagnostic value of MRCP in detecting Choledochal cyst

DISCUSSION

Congenital choledochal cysts, described initially in 1952[21]were classified into 3 types by Alanzo-Lej *et al* in 1959. Todani[11] and colleagues modified this classification in 1977 adding types IV and V. The present study emphasis on the study of various imaging techniques for the management of choledochal cysts for better understanding and management of the disease. Although most of the patients with choledochal cysts are diagnosed during the first decade of life however with improved hepatobiliary imaging, adult patients with the disease are being increasingly seen. Adult patients are underreported as the cases present to various general hospitals whereas

pediatric patients are concentrated in pediatric surgical centers reflecting an institutional referral bias. In our series of 40 patients, 30(75%) were adults and 10 (25%) were children.30 (75%) were males and 10 (25%) were females with a male to female ratio of 1:3. Almost all other studies are showing similar picture. The presentation is often vague and non-specific resulting in delayed diagnosis. However the diagnosis is facilitated by modern imaging techniques and may be made at any time from antenatal period to adulthood (Kim *et al*., 2002; Miyazaki *et al*., 1998). The classical triad of jaundice, abdominal mass and pain, originally described as a feature in most patients, was rarely seen in adults. Shi *et al*. (2001) has also reported that adult patients tend to have non specific symptoms resulting in delayed diagnosis. The type of choledochal cysts seen in our patients were, type 1 seen in 74% patients- most common type, type IV in 26% patients. In children 80% were type I and 20% were type IV while as in adults 72% were type I and 28% were type IV. So most common choledochal cysts found in both children and adults in our study were type I. Among the type I cysts 76% were type Ia and 24% were type Ic. All the type IV cysts were IVa subtype. Type II, III,and V were not seen in any patient in our study. Similar figures have been shown in other studies as well (Mathew *et al*., 2003).

In one adult patient though ultrasonography was suggestive of type I choledochal cyst however MRCP showed stricture in distal common bile duct which was later confirmed by intraoperative findings. Cyst stones and cholethiasis are a frequent accompanying conditions occurring in over 75% of adults with bile duct cyst.[26]In our study 18/40(45%) patients with choledocal cysts had stone formation in the liver, gall bladder and cyst itself.10% of children were associated with cholithiasis whereas 57% of adults were associated with different types of stones including hepatic calculi, gall stones and cystolithiasis ($p>0.05$). Similarly, Shah *et al*[9] had reported high incidence of stone formation (45%) in their series. All these MRCP findings were in concordance with the intraoperative findings. In 1959 preoperative diagnosis of choledochal cyst was possible in only 30% of cases (Alonso-Lej *et al*., 1959). But now it is possible in almost all of cases with the help of imaging studies. Ultrasonography is a useful screening test and is considered to the first investigation for diagnosis of choledochal cyst. It is safe, cheap and noninvasive investigation with high degree of reliability. However other investigations like MRCP, CECT, ERCP are required to demonstrate anatomy and co-existant pathology such as cholelithiasis, cystolithiasis, post-op biliary stricture with greater clarity. In the presence of cystolithiasis these cysts can be mistaken for simple cholidocholithiasis. In our study Ultrasound was done in all patients and has accurately diagnosed choledochal cyst in 87%of our patients .Illiker[27]also supports use of ultrasound with 70% accuracy. Ultrasound can diagnose choledochal cysts with a specificity of 97% in children (Chang *et al*., 2000). It is an excellent first line investigation of neonatal jaundice presenting for 2 weeks after birth[14] and may help to differentiate choledochal cyst from biliary atresia (Akhan *et al*., 1994). The ultrasound features of bile duct cyst have been well defined for type I bile cysts and variant of caroli's disease (Chang *et al*., 2000). Although there are reports of computed tomography(CT) scans diagnosing choledochal cysts (Grobliy Meyer and Tschantz, 2000) but it has been found that cysts are missed on CT scan and picked up on MRCP (DeBacker *et al*., 2000). However CT scanning was shown to be superior to MRCP in locating the biliary-enteric anastomosis and defining stenosis in the postoperative period. CT combined with intravenous cholangiography is useful for demonstration of cyst communication with biliary tree (Hoglund *et al*., 1990). The diagnostic accuracy of CECT in our study was 74%.CT scan is a useful imaging test for detecting choledochal cysts but it is difficult to delineate pancreatic and bile duct union. Multidetector computed tomography (MDCT) allows very thin collimation with a high quality multiplanarreformation (MPR), which provides detailed information on the pancreatic and bile ducts. CT cholangiography (CTC) can delineate the biliary tree with a sensitivity of 93% (Sajjad *et al*., 1999) Lam *et al*. (1999). Investigated the usefulness of CTC verses MR cholangiography in the diagnosis of choledochal cysts and CT cholangiography can detect choledochal cyst with an accuracy of 91% where as MR cholangiography visualizes 100% of cyst. ERCP is an excellent tool for defining biliary anatomy (Irie *et al*., 1998). It is an invasive procedure with therapeutic capability. Adults without previous cystenterostomy probably are best evaluated by ERCP because it permits a focused view of the pancreaticobiliary ductal junction through the papilla (Komi, 1991; Savader *et al*., 1991(b)). The procedure of choice for type III cyst or choledochocele is ERCP because endoscopic papillotomy is potentially therapeutic (Venu *et al*., 1984). In our study we did ERCP in 14 patients and it was positive in 13 patients.

The diagnostic accuracy of ERCP for exactly defining the anatomy of choledochal cyst was 93% in our study. Similar results were shown by Kimura K *et al*[36] in his study. Mobidity with ERCP ranges from 2% -8% in children and 1% -2% in adults which rises to 10% when combined with sphicterotomy and mortality estimates is estimated between 0.05-0.90% (Lee *et al*., 1997; Hekimoglu *et al*., 2008; Prasil *et al*., 2001). MRCP is non-invasive diagnostic imaging and it can avoid serious complication associated with ERCP.[37]MRCP is able to diagnose choledochal cysts with an accuracy of 82% -100% (Lam *et al*., 1999; Miyazaki *et al*., 1998; Sugiyama *et al*., 1998). In our study MRCP was done in all patients and it was accurate in 38 patients, with an accuracy of 97%. In one patient ultrasound was suggestive of choledochal cyst however MRCP showed distal common bile duct stricture which was later found to be in conformity with the intraoperative findings. In this patient proximal CBD was grossly dilated. In second patient MRCP was suggestive of a small pseudocyst pancreas however intraoperatively type I choledochal cyst was found. So MRCP was accurate with respect to the intraoperative findings in 39/40 (97%) patients. The diagnostic accuracy of MRCP was 97% for choledochal cyst with a sensitivity and specificity of 97% and 67% respectively. The detection rate of MRCP in other studies in pediatric patients ranges from 40% -69.2% (Lam *et al*., 1999; Miyazaki *et al*., 1998; Irie *et al*., 1998). Whereas that in adults ranges from 82%-100% (Irie *et al*., 1998; Coackleyfv and Qayyum, 2002). MRCP is considered as the first choice modality for diagnosing choledochal cysts in pediatric patients because it is noninvasive and it does not require breath holding.

Conclusion

MRCP is gold standard for diagnosing choledochal cyst type and is helpful in diagnosing related pancreatobiliary anomalies. Given its relatively moderate risk profile and lower cost, MRCP should be the diagnostic test of choice, after an initial ultrasound, in both children and adults, when

preoperatively evaluating choledochal cysts and their associated anomalies.

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Declarations

Conflict of interest: There is no conflict of interest with any other orginisation.
Ethical approval: Ethical approval was sought from Institute Ethical community.

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