ORIGINAL RESEARCH

Surgical management of enteric duplication cysts in children

¹Dr. Ratnakar, ²Dr. Abhijit Indrasingh Morwal, ³Dr. Pritam Paul, ⁴Dr. Jaydeep Bariya

¹Assistant Professor, Department of Surgery, Lord Buddha Koshi Medical College and Hospital, Saharsa, Bihar, India

²Assistant Professor, Department of Paediatrics, KM Medical College and Hospital, Mathura, UP, India ³Assistant Professor, Department of Surgery, KM Medical College and Hospital, Mathura, UP, India ⁴Assistant Professor, Department of Pathology, KM Medical College and Hospital, Mathura, UP, India

Corresponding Author

Dr. Jaydeep Bariya

Assistant Professor, Department of Pathology, KM Medical College and Hospital, Mathura, UP, India

Received: 11 June, 2022

Accepted: 14 July, 2022

ABSTRACT

Background: Enteric duplication cysts (EDCs) are rare congenital anomalies that are typically found along the gastrointestinal tract, most commonly in the small intestine. The present study was conducted to assess cases of enteric duplication cysts in children. **Materials &Methods:** 24 patients who were clinically diagnosed and proved on histopathological examination to be of enteric duplication cysts were enrolled. Clinical presentation, radiological features, operative findings and histopathology report were recorded. All cases were managed surgically. **Results:** Out of 24 patients, 16 were males and 8 were females. Out of 24 cases, age group 2-4 years had 1, 4-7 years had 10 and 7-10 years had 13 cases. The difference was significant (P< 0.05). Clinical presentation was abdominal pain in 20, bilious vomiting in 15, jaundice in 7, abdominal lump in 13. Surgical procedure performed was cyst excision with end to end anastomosis in 5, excision of the cyst along with excision of the segment of sigmoid colon in 7, excision of the cystic mass in 6 and excision of the cyst along with excision of the ileal segment in 6 cases. Microscopic examination showed cyst wall lined by gastric mucosa in 5, colonic duplication cyst in 5 cases. The difference was non- significant (P> 0.05). **Conclusion:** Since this rare congenital defect can manifest in a variety of clinical manifestations and can result in serious morbidity and even death if treatment is not received, it is crucial to recognize it and make a conclusive diagnosis.

Keywords: Abdominal lump, Children, Enteric duplication cysts

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-Non Commercial-Share Alike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

INTRODUCTION

Enteric duplication cysts (EDCs) are rare congenital anomalies that are typically found along the gastrointestinal tract, most commonly in the small intestine.¹ These cysts are usually lined by gastrointestinal epithelium, such as mucosa resembling the stomach, small intestine. or colon.EDCs can occur anywhere along the gastrointestinal tract, but they are most often located in the ileum, jejunum, or stomach.²These cysts are often spherical or cylindrical in shape, and they may contain fluid, mucus, or occasionally food material.Many cases are asymptomatic, especially if the cyst is small. However, large or complicated cysts can lead to symptoms such as abdominal pain, nausea, vomiting, and distention.³ In some cases, these cysts may cause intussusception, bowel obstruction, or perforation.Diagnosis is typically made through imaging techniques, such as ultrasound, CT scan, or

MRI, which can reveal a cystic mass with characteristic features. Endoscopy and sometimes histopathological examination after surgical removal are used to confirm the diagnosis.⁴

Enteric duplication cysts may be an incidental finding intraoperatively but in the majority of cases preoperative radiological diagnosis is possible. Appropriate surgical management is required, for this the attending surgeon should be familiar with the pathology and clinical characteristics of these rare cysts.⁵ Due to the rarity of this condition the vast majority of literature on enteric duplication cysts is in the form of case reports.⁶The present study was conducted to assess cases of enteric duplication cysts in children.

MATERIALS & METHODS

The study was carried outon 24patients who were clinically diagnosed and proved on histopathological

examination to be of enteric duplication cysts.All gave their written consent to participate in the study. Data such as name, age, gender etc. was recorded. clinical presentation, radiological features, operative findings and histopathology report were recorded. All cases were managed surgically. Results thus obtained were subjected to statistical analysis. P value < 0.05 was considered significant.

RESULTS

Table I Distribution of patients

Total- 24				
Gender	Male	Female		
Number	16	8		
1	1.0	C 1		

Table I shows that out of 24 patients, 16 were males and 8 were females.

Table II Age wise distribution

Age group (years)	Number	P value
2-4	1	0.05
4-7	10	
7-10	13	

Table II shows that out of 24 cases, age group 2-4 years had 1, 4-7 years had 10 and 7-10 years had 13 cases. The difference was significant (P < 0.05).

Table III Assessment of parameters

Parameters	Variables	Number	P value	
Clinical	Abdominal pain	20	0.84	
presentation	Bilious vomiting	15		
	Jaundice	7		
	Abdominal lump	13		
Surgical	Cyst excision with end to end anastomosis	5	5 0.61	
procedure	Excision of the cyst along with excision of the segment of	7		
	sigmoid colon			
	Excision of the cystic mass	6		
	Excision of the cyst along with excision of the ileal segment	6		
Microscopic	Cyst wall lined by gastric mucosa	5	0.57	
examination	Colonic duplication cyst containing large intestinal mucosa	7		
	Cyst wall lined by rectal mucosa	4		
	Cyst lined by ileal mucosa	8		
Final	Pyloroduodenal duplication cyst	4	0.82	
diagnosis	Colonic duplication cyst	7]	
	Ileal duplication cyst	8]	
	Rectal duplication cyst	5		

Table III shows that clinical presentation was abdominal pain in 20, bilious vomiting in 15, jaundice in 7, abdominal lump in 13. Surgical procedure performed was cyst excision with end to end anastomosis in 5, excision of the cyst along with excision of the segment of sigmoid colon in 7, excision of the cystic mass in 6 and excision of the cyst along with excision of the ileal segment in 6 cases. Microscopic examination showed cyst wall lined by gastric mucosa in 5, colonic duplication cyst containing large intestinal mucosa in 7, cyst wall lined by rectal mucosa in 4, and cyst lined by ileal mucosa in 8 cases. Final diagnosis was pyloroduodenal duplication cyst in 4, colonic duplication cyst in 7, ileal duplication cyst in 8 and rectal duplication cyst in 5 cases. The difference was non- significant (P > 0.05).

DISCUSSION

Duplication of gastrointestinal tract is a rare congenital anomaly found in about 0.2% of all children. Although enteric duplication cysts can present at any age the vast majority of patients present during infancy.^{7,8}Duodenal duplications can be cystic or tubular, communicating or non-communicating, but the most common type is cystic and non-communicating.⁹ These are generally located at the medial border of the first and second parts of the duodenum and extend to the anterior or posterior

side.¹⁰ Duodenal duplication observed in our case was cystic and located in the first and second parts of the duodenum, but it was of the communicating type and located on the antimesenteric side.¹¹A variety of clinical manifestations have been reported that are determined by the type, site and size of the duplication. Generally, patients present with a palpable mass in the abdomen, signs of intestinal obstruction, or abdominal pain. Bleeding or perforation caused by peptic ulcer and jaundice, and pancreatitis caused by biliary obstruction may also be

the manifestations.12The present study was conducted to assess cases of enteric duplication cysts in children. We found that out of 24 patients, 16 were males and 8 were females.Sharma et al13presented six cases of enteric duplication cysts with diverse clinicopathological features. Six children between age range of 3 days to 10 years had enteric duplication cysts. Two had ileal and one each were of pyloroduodenal, colonic and rectal duplication cyst. In one patient a presumptive diagnosis of enteric duplication cyst was made. Radiology played an important contributory role in diagnosis of these cysts in all the patients but histopathology proved to be gold standard for its confirmation. All these patients were managed by surgical excision. The postoperative and follow up period in all the cases was uneventful.

We found that out of 24 cases, age group 2-4 years had 1, 4-7 years had 10 and 7-10 years had 13 cases. Uzun MA et al¹⁴reported a cases in a 38-year-old woman who had occasional abdominal pain with a clinical diagnosis of gastric outlet obstruction. The epigastrium was mildly sensitive on physical examination. Laboratory findings were normal but abdominal ultrasonography (US) showed gastric distension. Upon abdominal computerized tomography (CT), a cystic lesion of 5 cm \times 8 cm \times 9 cm in diameter was observed, which extended along the lateral wall of the first and second parts of the duodenum. Remnants of food and orally taken contrast media were found within the lesion, and we observed the nasogastric tube entering the lesion through a defect between the duodenum and the cyst. They operated on the patient and found a cystic dilatation, $10 \text{ cm} \times 12 \text{ cm}$ in diameter, anterolateral to the first and second parts of the duodenum. They performed cystotomy and observed it make contact with the normally located duodenum at the posteromedial side of the cyst, through a defect of 2 $cm \times 2$ cm in diameter. The diagnosis was confirmed histopathologically by identifying a separate mucosa with its own muscularis mucosa on both sides of the wall between the cyst and duodenum and intervening connective tissue fibers. The patient was without complaint after 9 months follow-up.

We found that clinical presentation was abdominal pain in 20, bilious vomiting in 15, jaundice in 7, abdominal lump in 13. Surgical procedure performed was cyst excision with end to end anastomosis in 5, excision of the cyst along with excision of the segment of sigmoid colon in 7, excision of the cystic mass in 6 and excision of the cyst along with excision of the ileal segment in 6 cases. Microscopic examination showed cyst wall lined by gastric mucosa in 5, colonic duplication cyst containing large intestinal mucosa in 7, cyst wall lined by rectal mucosa in 4, and cyst lined by ileal mucosa in 8 cases. Final diagnosis was pyloroduodenal duplication cyst in 4, colonic duplication cyst in 7, ileal duplication cyst in 8 and rectal duplication cyst in 5 cases. Schalamon J et al¹⁵ studied paediatric patients with

intestinal duplications in order to analyse the influence of prenatal sonography and laparoscopy on the clinical course. Thirteen duplications of the alimentary tract in 12 patients were treated over a 10year period from 1989 to 1999. Six patients were diagnosed prenatally by ultrasound and were free of symptoms until surgery, except for one patient who had meconium-ileus owing to cystic fibrosis. In another five patients, the diagnosis was made on the basis of symptoms with signs of obstruction. In one child, the duplication was found incidentally during an operation for an anorectal malformation. The location of the 13 duplications was the stomach in three cases, the duodenum in one case, the jejunum in two cases, the ileum in six cases and the rectum in one case. Laparotomy was performed in ten patients. Two cases were treated by laparoscopic-assisted resection.Early diagnosis and treatment of uncomplicated intestinal duplications by means of prenatal sonographic screening and laparoscopicassisted resection, respectively, are desirable in this congenital malformation. Resection of the duplication with or without minimal resection of the adjacent normal intestine should be mandatory.

The shortcoming of the study is small sample size.

CONCLUSION

Authors found that since this rare congenital defect can manifest in a variety of clinical manifestations and can result in serious morbidity and even death if treatment is not received, it is crucial to recognize it and make a conclusive diagnosis.

REFERENCES

- 1. Macpherson RI. Gastrointestinal tract duplications: clinical, pathologic, aetiologic, and considerations. Radiographics. 1993;13:1063–80.
- Cavar S, Bogovic M, Leutic T, Antabak A, Batinica S. Intestinal duplications – experience in 6 cases. EurSurg Res. 2006;38:329-32.
- Puligandla PS, Nguyen LT, St-Vil D, Flageole H, Bensoussan AL, Nguyen VH, et al. Gastrointestinal duplications. J Pediatr Surg. 2003;38(5):740-44.
- Abdur-Rahman L, Abdulkadir A, Nasir A, Ibrahim O, Adeniran J, Adesiyun O. Gastrointestinal Duplications: Experience in Seven Children and a Review of the Literature. Saudi J Gastroenterol. 2010;16(2):105-09.
- 5. Murty TV, Bhargava RK, Rakas FS. Gastroduodenal duplications. J Pediatr Surg. 1992;4:515-17.
- Uzun MA, Koksal N, Kayahan M, Celik A, Kilicoglu G, Ozkara S. A rare case of duodenal duplication treated surgically. World J Gastroenterol. 2009;2:882-84.
- Fitz RH. Persistent omphalomesenteric remains: their importance in the causation of intestinal duplication, cyst formation and obstruction. Am J Med Sci.1884;88:30-57.
- Ladd WE. Duplications of the alimentary tract. South Med J.1937;30:363. [10] Gross RE. Duplications of the alimentary tract. In: The Surgery of Infancy and Childhood. 1953:221-45.
- 9. Ladd WE, Gross RE. Surgical treatment of Duplication of the Alimentary Tract: Enterogenous Cysts, Enteric

Cysts, or Ileum Duplex. Surgery, Gynaecology &Obstretrics .1940;70:295-307.

- 10. Parker BC, Guthrie J, France NE, Atwell JD. Gastric duplications in infancy. J Pediatr Surg. 1972;7:294-98.
- 11. Rai BK, Zaman S, Mirza B, Hanif G, Sheikh A. Duodenal duplication cyst having ectopic gastric and pancreatic tissues. APSP J Case Rep. 2012;3:15.
- Patel MP, Meisheri IV, Waingankar VS, Ramesh S, Naregal AM, Muthaal PB. Duplication cyst of the pylorus - A rare cause of gastric outlet obstruction in the newborn. J Postgrad Med. 1997;43:43-45
- Sharma S, Yadav AK, Mandal AK, Zaheer S, Yadav DK, Samie A. Enteric duplication cysts in children: A clinicopathological dilemma. Journal of Clinical and Diagnostic Research: JCDR. 2015 Aug;9(8):EC08.
- Uzun MA, Koksal N, Kayahan M, Celik A, Kilicoglu G, Ozkara S. A rare case of duodenal duplication treated surgically. World J Gastroenterol. 2009;2:882-84.
- 15. Schalamon J, Schleef J, Hollwarth ME. Experience with gastro-intestinal duplications in childhood. Langenbecks Arch Surg. 2000;385(6):402-05.