



Enteric duplication in children

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Abstract

Introduction Enteric duplication is a congenital anomaly with varied clinical presentation that requires surgical resection for definitive treatment. This had been approached with laparotomy for resection, but has changed with minimally invasive technique. The purpose of our study was to determine the demographics, natural history, operative interventions, and outcomes of pediatric enteric duplication cysts in a contemporary cohort.

Methods With IRB approval, we performed a retrospective chart review of all patients less than 18 years old treated for enteric duplication between January 2006 and August 2016. Demographics, patient presentation, operative technique, intra-operative findings, hospital course, and follow-up were evaluated. Descriptive statistical analysis was performed; all medians were reported with interquartile range (IQR).

Results Thirty-five patients underwent surgery for enteric duplication, with a median age at surgery of 7 months (2.5–54). Median weight was 7.2 kg (6–20). Most common patient presentations included prenatal diagnosis 37% ($n = 13$). Thirty-four patients (97%) had their cyst approached via minimally invasive technique (thoracoscopy or laparoscopy) with only three (8%) requiring conversion to an open operation. Median operative time was 85 min (54–133) with 27 (77%) patients requiring bowel resection. Median length of bowel resected was 4.5 cm (3–7). Most common site of duplication was ileocecal ($n = 15$, 42%). Postoperative median hospital length of stay was 3 days (2–5) and median number of days to regular diet was 3 (1–4). No patients required re-operation during their hospital stay. Median follow-up was 25 days (20–38).

Conclusion In our series, most enteric duplication cysts were diagnosed prenatally. These can be managed via minimally invasive technique with minimal short-term complications, even in neonates and infants.

Keywords Enteric duplication · Laparotomy · Bowel resection

Introduction

Enteric duplication is a rare congenital anomaly with varied clinical presentations that requires surgical resection for definitive treatment [1]. Historically, this has been approached with laparotomy for resection, but, with the advent of minimally invasive techniques (thoracoscopy or laparoscopy), this is changing [2–4]. Case series in the literature has a limited patient population and predominately focuses on open resections [5, 6]. The purpose of our study was to determine the patient demographics, natural history, operative interventions, and outcomes of pediatric enteric duplication cysts in a contemporary cohort. We hypothesized that these patients can be successfully managed with minimally invasive interventions.

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Materials and methods

Following IRB approval (#17010016), a retrospective chart review of all patients less than 18 years old treated for mediastinal or abdominal enteric duplication based on postoperative diagnosis between January 2006 and August 2016 was performed. Patients with preoperative diagnosis of enteric duplication without this finding intraoperatively were excluded. Patient lists were obtained from our IT department utilizing International Classification of Disease Ninth Revision (ICD-9) diagnosis codes. Demographics, patient presentation, diagnostic testing, operative technique, intraoperative findings, hospital care, and follow-up were reviewed.

Data were collected including presenting symptoms, timing of diagnosis, diagnostic imaging modality, operative details including use of thoracoscopy/laparoscopy with or without conversion, and need for bowel resection. Postoperative course was also reviewed including complications, time to regular diet, duration of hospital stay, and length of follow-up. Resections were characterized as esophagus, gastric, duodenum, jejunum, terminal ileum, ileum, and cecum, or colon. The characteristics of the duplication included the shape of the duplication, tubular or cystic, and whether it had a common wall with the bowel or had a luminal connection with the bowel.

Descriptive statistics including counts and percentages were analyzed. Statistical analysis was performed using STATA (StataCorp 2017. Stata Statistical Software: Release 15. College, Station, TX: StataCorp LLC) for calculations, all medians are reported with interquartile range (IQR).

Results

Thirty-five patients underwent surgery for enteric duplication during the study period. Thirteen (37%) patients were male and twenty two (62%) were female. Median age at time of surgery was 7 months (2.5–54); 62% ($n=22$) were less than 1 year old. Median weight was 7.2 kg (6–20). The most common patient presentation was prenatal diagnosis in 37% ($n=13$). Other presentations included abdominal pain 25% ($n=9$), bilious emesis 14% ($n=5$), intraoperative finding 5% ($n=2$), respiratory symptoms 5% ($n=2$), abdominal mass 2% ($n=1$), and constipation 2% ($n=1$) (Table 1). The diagnostic imaging modality used most commonly was ultrasound 51% ($n=18$). Other common diagnostic tools included computed tomography (CT) 25% ($n=9$), non-barium contrast enema or contrast upper gastrointestinal series 5% ($n=2$), diagnostic laparoscopy 5% ($n=2$), CXR 5% ($n=2$), and open operative exploration 3% ($n=1$) (Table 2).

Table 1 Presenting symptoms

	<i>n</i> (%)
Prenatal diagnosis	13 (37%)
Abdominal pain	9 (25%)
Bilious emesis	5 (14%)
Intraoperative finding	2 (5%)
Respiratory symptoms	2 (5%)
Abdominal mass	1 (2%)
Constipation	1 (2%)
Nonbilious emesis	1 (2%)
Incidental finding	1 (2%)

Table 2 Primary diagnostic tests

Primary diagnostic test	<i>n</i> (%)
Ultrasound (US)	18 (51%)
Computed tomogram (CT)	9 (25%)
Barium enema/upper GI	2 (5%)
Diagnostic laparoscopy	2 (5%)
Chest X-ray (CXR)	2 (5%)
Abdominal X-ray (KUB)	1 (2%)
Open operation	1 (2%)

In the two patients who underwent diagnostic laparoscopy, one of these patients was discovered to have a duplication cyst as part of a laparoscopic appendectomy. The second was a prenatally diagnosed cyst that was unable to be confirmed on repeat ultrasound. As a result, the patient underwent laparoscopy to evaluate, and subsequently resect this mass when it was discovered. Patients who were diagnosed prenatally had their duplication resected at a median of 4.3 months (1.5, 7). Those who were diagnosed postnatally had their duplication resected at a median of 23 months (3, 149). (Table 3).

Thirty-four patients had their cyst resected via minimally invasive technique with only three requiring conversion to an open operation. One patient was approached open. Median operative time was 85 min (54–133) with 27 of 35 patients requiring bowel resection. Median length of bowel resected was 4.5 cm (3–7). The most common site of duplication was ileocecal ($n=15$, 42%), followed by jejunum ($n=6$, 17%). Other sites of resection included esophagus ($n=5$, 14%), gastric ($n=3$, 8%), duodenum ($n=2$, 5%), ascending colon ($n=1$, 2%), and descending colon ($n=1$, 2%) (Table 4). Two patients had cysts located within the small intestine, although the exact location could not be determined.

Thirty-one (88%) of the duplications were cystic in shape with the remaining 11% ($n=4$) being tubular. Twenty-five (71%) of the duplications had a common wall with the GI tract, whereas only 13 (37%) had a luminal connection with

Table 3 Prenatal vs postnatal diagnosis characteristics

	Prenatal (n = 12)	Postnatal (n = 23)
Time to resection, months ^a	4.3 (1.5, 7)	23 (3, 149)
Laparoscopic resection	12 (100%)	22 (95%)
Location of duplication		
Not specified	1 (8%)	1 (4%)
Esophagus	2 (16%)	3 (13%)
Stomach	1 (8%)	2 (8%)
Duodenum	1 (8%)	1 (4%)
Jejunum	0 (0%)	6 (26%)
Ileum	3 (25%)	5 (21%)
Ileocecal	4 (33%)	3 (13%)
Right colon	0 (0%)	1 (4%)
Left colon	0 (0%)	1 (4%)
Cyst characteristics		
Tubular	2 (16%)	2 (8%)
Cystic	10 (83%)	21 (91%)
Bowel common wall	9 (75%)	16 (69%)
Connected bowel lumen	4 (33%)	9 (39%)

^aMedian (IQR)

Table 4 Duplication location

Duplication location	n (%)
Esophagus	5 (14%)
Gastric	3 (8%)
Duodenum	2 (5%)
Jejunum	6 (17%)
Ileum	8 (22%)
Ileocecal	7 (20%)
Ascending colon	1 (2%)
Descending colon	1 (2%)
Not specified	2 (5%)

Table 5 Duplication characteristics

Duplication characteristics	n (%)
Common wall	25 (71%)
Lumen connected	13 (37%)
Cystic	31 (88%)
Tubular	4 (11%)

the bowel (Table 5). Five (14%) of the patients had the evidence of obstruction intraoperatively and only one (2%) showed the signs of infection.

Postoperative median hospital length of stay was 3 days (2–5) and median number of days to regular diet was 3 (1–4). The only reported complication was a single surgical site infection. The median length of follow-up was 25 days (20–38).

Discussion

Our study demonstrated that minimally invasive techniques are a viable means to treat enteric duplications with 91% (n = 32) not requiring conversion to an open procedure. All enteric duplications approached open or requiring conversion to an open operation were located in the abdomen. Our patient population was similar to past populations with some interesting differences. Whereas, in many studies, there was a male predominance [2–7], in our study, females predominated at 62%. This could indicate that, with greater study, the actual rate of duplication is closer to 1:1 between sexes or that this difference is due to random population variance. The demographics of our study is consistent with the other previous studies in that patients presented primarily at less than a year of age [2–7].

Duplication cysts have a variable morphology and location throughout the GI tract. Our study is consistent with the previous studies in that the majority of our morphology was cystic, shared a common wall with the bowel, and were predominately ileocecal [2–7]. The most common method of operative intervention in our study was bowel resection. The length of resection was small at only 4.5 cm and none of our patients had complications from resection. Rate of resection and anastomosis ranges from 9 to 75% in other studies [2–7]. Some studies have indicated that enucleation or other options exist for the removal of the duplication, but this was most common in the esophagus and stomach [2].

The largest study about laparoscopic management of enteric duplication was a multicenter retrospective study that examined 114 patients undergoing minimally invasive surgery for treatment of enteric duplication cyst [2]. This study examined patients between the years of 1994–2009. In this study, there was a 32% rate of conversion to open surgery, most commonly secondary to the inability to separate the duplication cyst from the digestive tract, difficulty with visualization, or need for bowel resection. In addition, they reported a 7% rate of complications in their cohort. In comparison, while our group is smaller, it represents a more contemporary cohort. Eight percent of cases (n = 3) were laparoscopic assisted, in that when the cyst was discovered and the involved intestine mobilized, the umbilical incision was extended a few centimeters. The cyst and involved intestine were easily eviscerated through the small umbilical incision, and the remainder of the case was completed open. This conversion rate was much less than the previous study in spite of our population having a median weight less than 10 kg, which was shown in multivariate analysis to be a predictor of conversion. These findings continue to indicate that minimally invasive techniques represent a viable strategy when managing enteric duplication.

With improvement in perinatal monitoring in the past decade, it is no surprise that a large part of our sample was diagnosed prenatally with ultrasound [8, 9]. Other common symptoms included abdominal pain and bilious emesis which is consistent with the past studies, as well. In the previous studies of laparoscopic removal of enteric duplication cysts, 22% [3] and 33% [4] were diagnosed prenatally. Our prenatal diagnosis rate of 37% continues to show an upward trend in the ability to diagnosis enteric duplication cysts prenatally.

Limitations to our study include its retrospective nature and our small patient population. Though our population was larger in comparison to past studies, it is still a small cohort with a significant possibility of bias in our results. Enteric duplication does not also have a specific diagnostic code, so it is possible that these patients were not collected as a part of our protocol. To mitigate this, when collecting data, we confirmed all codes with our hospital's coding department to limit missing patients.

Conclusion

In our series, most enteric duplication cysts are diagnosed prenatally. These can be managed via minimally invasive techniques with minimal short-term complications, even in neonates and infants.

Author contribution Joseph Sujka: concept and design, data collection, data analysis and interpretation, drafting article, critical revision of article, approval of article, and statistics. Justin Sobrino: data collection, data analysis and interpretation, and drafting article. Leo A. Benedict: drafting article, critical revision of article, and statistics. Hanna Ale-mayehu: concept and design, data collection, data analysis and interpretation, drafting article, critical revision of article, approval of article, and statistics. Shawn D. St. Peter: concept and design, data collection, data analysis and interpretation, drafting article, critical revision of article, approval of article, and statistics. Richard J. Hendrickson: concept and design, data collection, data analysis and interpretation, drafting article, critical revision of article, and approval of article.

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Compliance with ethical standards

Conflict of interest The authors have no conflict of interest to disclose.

Ethical approval This article does not contain any studies with human participants or animals performed by any of the authors.

Informed consent Informed consent was waived by our IRB due to the fact that the data collected for this study were retrospective and de-identified.

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