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WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

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Retrospective Study

Surgical perspectives of symptomatic omphalomesenteric duct remnants: Differences between infancy and beyond

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Conflict-of-interest statement: The

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Abstract

BACKGROUND

The clinical manifestations of omphalomesenteric duct remnant (OMDR) can vary with the age at diagnosis, from asymptomatic incidental findings to symptoms related to gastrointestinal complications. The lifelong complication rates are reported as 4%-34%, and complications are more common in patients younger than 2 years of age. The authors attempted to identify different clinical features and management for the various pediatric age groups.

AIM

To find surgical perspectives for the pediatric age-related variants of OMDR and make recommendations for optimal management.

METHODS

The medical records of pediatric patients diagnosed with OMDR were reviewed retrospectively. Fifteen patients diagnosed based on incidental findings during other surgeries were excluded. The patients were divided into two groups based on age: < 12 mo (infants) and > 12 mo (beyond infancy). We analyzed the demographic characteristics, clinical manifestations, diagnostic tools, surgical procedures, and clinical outcomes of the patients and compared them for the age groups. Chi-squared and Fisher's exact tests were used for nominal scales and a Mann-Whitney test was used for ratio scales.

RESULTS

A total of 35 patients (7 infants, 28 children beyond infancy) were finally included.

authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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In both groups, Meckel's diverticulum (MD) was the most common type of OMDR, while umbilical lesions were more common in the infant group ($P = 0.006$). Hematochezia and abdominal pain were common in the beyond infancy group, while umbilical lesions were the most frequent symptoms in the infant group. Several diagnostic tools were used, but Meckel's scan was most useful in diagnosing OMDR in patients with painless rectal bleeding. Minimally invasive surgery was more commonly performed for children than for infants ($P = 0.016$). Single-incision laparoscopic surgery (SILS) was performed for fifteen patients who underwent laparoscopic surgery. There were only three cases of postoperative complications, and all patients survived in good condition.

CONCLUSION

The clinical type of OMDR varies with age, umbilical lesions in infants, and MD beyond infancy. SILS is effective for managing children with MD regardless of age.

Key Words: Omphalomesenteric duct remnant; Age; Surgical; Meckel's diverticulum; Single-incision laparoscopic surgery; Children

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Core Tip: This is a retrospective study aimed at identifying surgical perspectives for variants of the omphalomesenteric duct remnant (OMDR) manifesting in different age groups. Meckel's diverticulum was the most common type of OMDR in all the subjects, while umbilical lesions were more common in the infant group. For management, minimally invasive surgery was more common beyond infancy, and single-incision laparoscopic surgery may be considered the preferred surgical procedure.

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INTRODUCTION

The omphalomesenteric duct is an embryologic connection between the yolk sac and primitive midgut. It normally disappears between the fifth and seventh weeks of fetal life[1,2], failure of this process results in various anomalies collectively called the omphalomesenteric duct remnant (OMDR), including Meckel's diverticulum (MD), umbilical fistula, sinus, polyp, and omphalomesenteric cysts[1-3]. The most common type of OMDR is MD (67%), which is also one of the most common congenital anomalies of the digestive tract in children (2%-4%)[1-4].

The clinical manifestations of OMDR can vary with age at diagnosis, from asymptomatic incidental findings to symptoms related to gastrointestinal complications[1-3]. Lifelong complication rates are reported as 4%-34%, and complications are more common in patients younger than 2 years of age[5,6].

This study aimed to identify surgical perspectives of OMDR by assessing its variants in different age groups (infants and children beyond infancy). The findings of this study may also provide useful information for the optimal management.

MATERIALS AND METHODS

We retrospectively reviewed the medical records of pediatric patients aged < 18 years old with a histopathological diagnosis of OMDR after surgery who were treated in Pusan National University Children's Hospital between November 2008 and



December 2019. Fifty patients were histologically diagnosed with OMDR. We excluded 15 patients diagnosed with OMDR incidentally during other surgeries.

The patients were divided into two groups based on the age at surgery: Infants (< 12 month old) and children beyond infancy (> 12 month old). A total of 35 patients (7 infants and 28 children beyond infancy) were finally included.

We analyzed the data of the following variables: Sex, type of delivery, weight at birth and surgery, gestational age, and age at symptom onset, diagnosis, and surgical procedure. In addition, we investigated the symptom durations, associated anomalies, type of OMDR (MD, patent duct, umbilical polyp, duct cyst, and fibrous cord), clinical manifestations, diagnostic tools and sensitivity, surgical procedures performed, complications, and the final outcomes. We used medians with standard deviations (SDs) for representative values for ratio scales. Pain was measured using the Face-Legs-Activity-Cry-Consolability scale for the patients aged < 3 years old and the Wong-Baker Faces Pain scales for the older children; based on the instruction for each scale, scores of ≥ 1 were interpreted as abdominal pain[7,8].

All analyses were completed using IBM SPSS Statistics version 25 (SPSS Inc, Chicago, IL, United States). Chi-squared and Fisher's exact tests were used for nominal scales and a Mann-Whitney test was used for ratio scales. A $P < 0.05$ was considered statistically significant.

This study was approved by the Pusan National University Yangsan Hospital Institutional Review Board (IRB No. 05-2020-111) and carried out according to the recommendations of the IRB committee.

RESULTS

The subjects comprised 27 males and 8 females (male:female ratio = 3.4:1). There were no significant differences in delivery, birth weight, and symptomatic duration among the age groups (Table 1).

In both groups, MD was the most frequent type of OMDR. While the infant group had other types of OMDR, most cases in the beyond infancy group were MD ($P = 0.006$). Hematochezia and abdominal pain were common in the beyond infancy group, while umbilical lesions were the most frequent in the infant group. Gastric-type ectopic mucosa was most common in the pathologic reports, but the presence of ectopic mucosa was not reported for several cases (Table 2).

The diagnoses were based on the chief complaint (CCs) of the patients. The CCs of our patients were classified into four features: Painless rectal bleeding, abdominal pain, vomiting, and umbilical lesions. Patients with rectal bleeding underwent various diagnostic modalities, including abdominal ultrasonography (US), computed tomography (CT), endoscopy, or colonoscopy; most of these failed to show the bleeding focus. Technetium-99m pertechnetate scintigraphy, known as Meckel's scan (MS), helped diagnose these patients with OMDR. For abdominal pain, US and CT were performed first. A MS was performed in patients with rectal bleeding-related conditions, such as anemia or melena. US was first performed for obstruction symptoms such as vomiting, but it could not be used to diagnose OMDR. Two of 3 patients with vomiting had CT findings of MD. The other patient underwent emergency operation for reduction-failed intussusception, and MD was found as the leading point of intussusception. Umbilical lesions can be diagnosed as OMDR based on US only. One child with umbilical discharge underwent only physical examinations before surgery. Figure 1 shows a flowchart for the diagnostic process of OMDR according to the CCs of patients.

The median operation time was not significantly different between age groups. The most common surgical procedure was segmental resection of the small bowel (24 patients, 68.6%). There were no significant differences between the surgical procedure for the groups. Minimally invasive surgery (MIS) was more commonly performed for children than for infants ($P = 0.016$). Fifteen patients who underwent laparoscopic surgery underwent a single-incision laparoscopic surgery (SILS) (Table 3).

The durations of hospitalization were similar for both groups. Complications were rare in both groups. One patient in the infant group, who underwent open segmental resection of the small bowel for an omphalomesenteric fistula, had a complication of superficial wound infection on the umbilicus. Two children in the beyond infancy group had a small bowel obstruction and pseudomembranous colitis, respectively. All patients survived and were discharged in good condition after surgery.

Table 1 Patient characteristics

	Total (n = 35) ¹	Infants (n = 7) ¹	Children beyond infancy (n = 28) ¹	P value
Gender				0.180
Male	27 (77.1)	4 (57.1)	23 (82.1)	
Female	8 (22.9)	3 (42.9)	5 (17.9)	
Male:female ratio	3.4:1	1.3:1	4.6:1	
Delivery				0.170
NSVD	22 (62.9)	6 (85.7)	16 (57.1)	
Caesarean section	13 (37.1)	1 (14.3)	12 (42.9)	
Preterm				0.171
Preterm	4 (11.4)	2 (28.6)	2 (7.1)	
Full term	31 (88.6)	5 (71.4)	26 (92.9)	
Weight (kg)				
Birth	3.40 ± 0.51 (2.23–4.30)	3.18 ± 0.34 (2.90–3.73)	3.41 ± 0.55 (2.23–4.30)	0.825
Operation	18.40 ± 21.53 (2.95–77.45)	6.64 ± 2.17 (2.95–8.85)	22.55 ± 21.39 (7.25–77.45)	N/A
Age (mo)				N/A
Symptom onset	53.0 ± 67.3 (0–201)	2.3 ± 4.2 (0–11)	85.4 ± 65.1 (0–201)	
Diagnosis	53.2 ± 65.9 (0–202)	2.4 ± 4.2 (0–11)	85.6 ± 62.5 (12–202)	
Operation	53.3 ± 65.9 (0–202)	2.6 ± 4.2 (0–11)	85.8 ± 62.5 (12–202)	
Symptomatic duration (d)	3.0 ± 292.0 (1–1540)	7.0 ± 3.9 (2–12)	3.0 ± 325.1 (1–1540)	0.246

¹Data is presented as the median ± SD or n (%). NSVD: Normal spontaneous vaginal delivery.

DISCUSSION

Fifteen cases were excluded due to incidental findings during other surgeries because their clinical course may have been affected by the underlying disease and not OMDR. The underlying problems of these patients included gastrointestinal (jejunal atresia, anorectal malformation, and Hirschsprung's disease) and cardiovascular (coarctation of aorta with ventricular septum defect and total anomalous pulmonary venous connection) anomalies and other gastrointestinal diseases (acute appendicitis, small bowel obstruction, gastric polyp, hiatal hernia, and intussusception).

Previous investigators found that more than 50%-60% of the patients with symptoms were younger than 2 years of age[5,6,9]. Based on the previous studies, we categorized our patients by age into the infancy and beyond infancy groups. There were various types of OMDR in the infants, while MD was the most common clinical type in the children beyond infancy. Some previous studies reported that umbilical abnormalities associated with OMDR are usually present in infants[3,10-12]. In this study, three children in the beyond infancy group with umbilical lesions had symptoms onset during their infancy. Umbilical lesions developing after one year of age may have other diagnoses rather than OMDR. It is necessary to consider the age of diagnosis as well as the age of symptom onset for children with umbilical lesions. US also may be helpful for OMDR with umbilical lesions for the differential diagnosis of other umbilical lesions (umbilical granuloma, polyp, or urachal remnant), evaluation of connection with the bowel, and planning for surgical excision[1].

Table 2 Clinical types and features of omphalomesenteric duct remnants

	Total (n = 35) ¹	Infant (n = 7) ¹	Children beyond infancy (n = 28) ¹	P value
Clinical type				0.006
MD	31 (88.6)	4 (57.1)	27 (96.4)	
Patent duct	2 (5.7)	2 (28.6)	0	
Duct cyst	2 (5.7)	1 (14.3)	1 (3.6)	
Umbilical polyp	0	0	0	
Fibrous cord	0	0	0	
Clinical features ²				
Hematochezia	20 (57.1)	2 (28.6)	18 (64.3)	0.101
Abdominal pain	12 (34.3)	0	12 (42.9)	0.036
Fever	11 (31.4)	2 (28.6)	9 (32.1)	0.619
Bilous vomiting	8 (22.9)	1 (14.3)	7 (25.0)	0.484
Umbilical lesion	7 (20.0)	4 (57.1)	3 (10.7)	0.018
Abdominal distension	4 (11.4)	1 (14.3)	3 (10.7)	0.609
Diarrhea	3 (8.6)	1 (14.3)	2 (7.1)	0.499
Ectopic mucosa (in MD)				0.283
Gastric	13 (41.9)	0	13 (48.1)	
Pancreatic	3 (9.7)	1 (25.0)	2 (7.4)	
Coexistent	2 (6.5)	0	2 (7.4)	
Absent	1 (3.2)	0	1 (3.7)	
Not mentioned	12 (38.7)	3 (75.0)	9 (33.3)	

¹Data is presented as the median ± SD or n (%).²Multiple symptoms were presented in some patients.

MD: Meckel's diverticulum.

Besides umbilical lesions, the other main symptoms of OMDR included painless rectal bleeding, abdominal pain, and vomiting. Diagnostic tools were selected case by case according to the CCs. On evaluating the diagnosis process for patients in our study, MS was identified as the most useful modality for the diagnosis of OMDR. CT, endoscopy, and colonoscopy failed to detect OMDR. US may be helpful in infants, but it did not yield satisfactory results in this study, except for umbilical lesions. As described in previous studies, imaging studies such as CT or US, may help detect associated features such as bowel obstruction or perforation, but can seldom be used to locate the OMDR lesion itself[1-3,11]. The common causes of painless rectal bleeding in children aged > 12 month old include anal fissures, juvenile polyps, and MD. It is highly recommended that MD should be considered as the cause of pediatric lower gastrointestinal bleeding[13,14]. MS is a noninvasive diagnostic tool with high sensitivity for MD[1-4,11,15]. Its sensitivity in detecting MD is reported to be between 50% and 100% with an increased rate if bleeding is a presenting symptom[3]. In this study, MS had a high positive predictive value for the diagnosis of MD. Therefore, we recommend MS as the first diagnostic tool for prompt management of rectal bleeding in children. Symptoms such as abdominal pain or vomiting are nonspecific, as they can be associated with various diseases as gastroenteritis, intussusception, and other surgical abdomen. Several patients with CCs of abdominal pain and associated features of anemia or melena were finally diagnosed as OMDR using MS. Therefore, MS should be performed before other modalities such as colonoscopy or CT if the history and clinical features are suspicious of OMDR. Figure 2 shows an example of the evaluation process for pediatric lower gastrointestinal bleeding.

There was only one patient with negative findings on MS who was finally diagnosed with MD by capsule endoscopy. MS may have been negative if there was no ectopic mucosa in the lesion, but the gastric glandular epithelium was highlighted

Table 3 Characteristics of surgical procedure

	Total (n = 35) ¹	Infant (n = 7) ¹	Children beyond infancy (n = 28) ¹	P value
Time (min)	80.0 ± 34.1 (15-205)	78.0 ± 25.7 (45-120)	80.0 ± 35.3 (15-205)	0.171
Operative method				0.278
Segmental resection of small bowel	24 (68.6)	4 (57.1)	20 (71.4)	
Diverticulectomy	7 (20.0)	1 (14.3)	6 (21.4)	
OMDR resection only	4 (11.4)	2 (28.6)	2 (7.1)	
Minimal invasive surgery				0.016
Open	15 (42.9)	6 (85.7)	9 (32.1)	
Laparoscopic	20 (57.1)	1 (14.3)	19 (67.9)	
Single port (SILS)	15 (42.9)	1 (14.3)	14 (50.0)	
Multiport	5 (14.3)	0	5 (17.9)	

¹Data is presented as the median ± SD or n (%). OMDR: Omphalomesenteric duct remnants; SILS: Single-incision laparoscopic surgery.

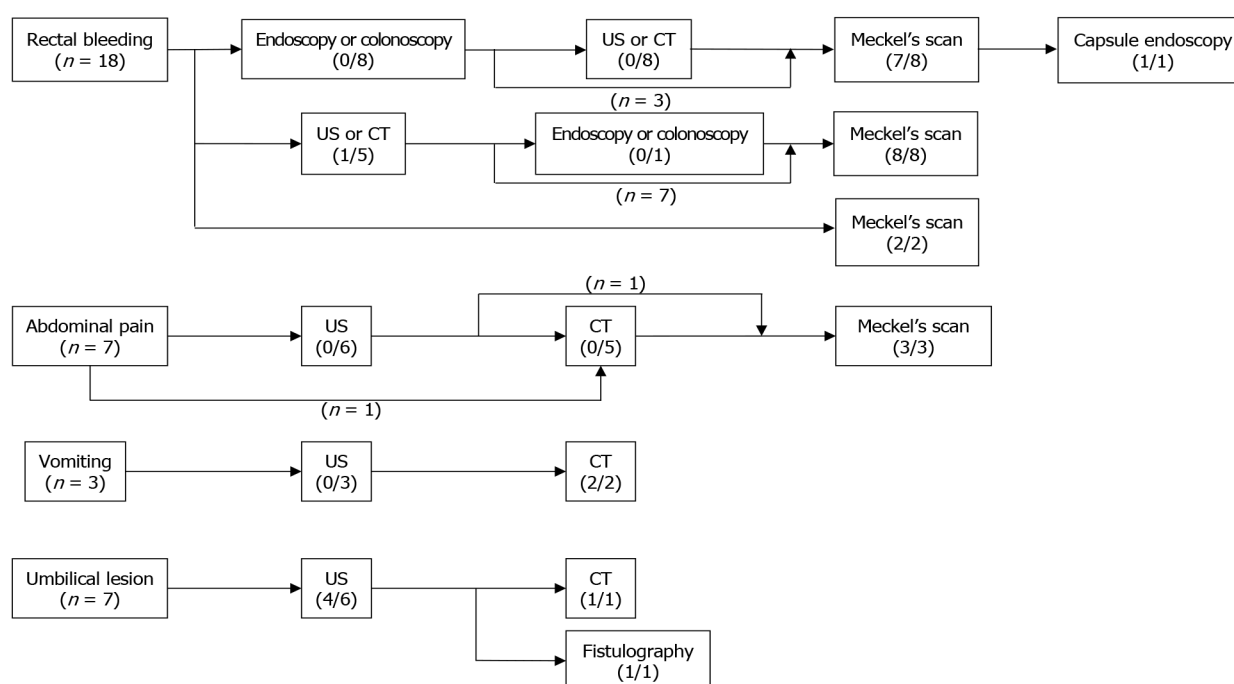


Figure 1 Flowchart of the diagnostic process for omphalomesenteric duct remnants based on chief complaints. (1) Eighteen patients had painless rectal bleeding. Eight patients underwent endoscopy or colonoscopy initially, and all of them had negative findings; five of them underwent ultrasonography (US) or computed tomography (CT), and the remaining three patients underwent a Meckel's scan (MS). One patient had negative findings on MS and proceeded to capsule endoscopy. Eight other patients had US or CT as the initial imaging study. Only two patients underwent a MS initially; (2) Seven patients presented with abdominal pain; of them, six patients initially underwent US and one underwent CT. All the patients had negative findings for omphalomesenteric duct remnant. Three patients underwent a MS, while four other patients underwent surgery; (3) Three patients presented with vomiting and underwent US initially, while only two patients underwent CT afterward. The patient who did not undergo CT received air reduction for intussusception followed by surgery for failed reduction; and (4) Seven patients presented with umbilical lesions. Six patients initially underwent US, while the other patient did not undergo additional imaging studies. Two patients who underwent secondary studies after US; one underwent CT and the other underwent fistulography. Figures under the modality show the number of diagnosed cases/number of performed cases. CT: Computed tomography; US: Ultrasonography.

in the histopathologic report of this patient. We could not determine why MS revealed negative findings here.

Recently, MIS has become popular for the surgical treatment of OMDR. Several previous studies had demonstrated the advantages of MIS: Diagnosis without delay, fast recovery, and fewer complications[2,16,17]. In pediatric patients, especially small infants, there are restrictions for manipulating laparoscopic instruments due to the limited working space. Therefore, we prefer SILS with extracorporeal bowel resection

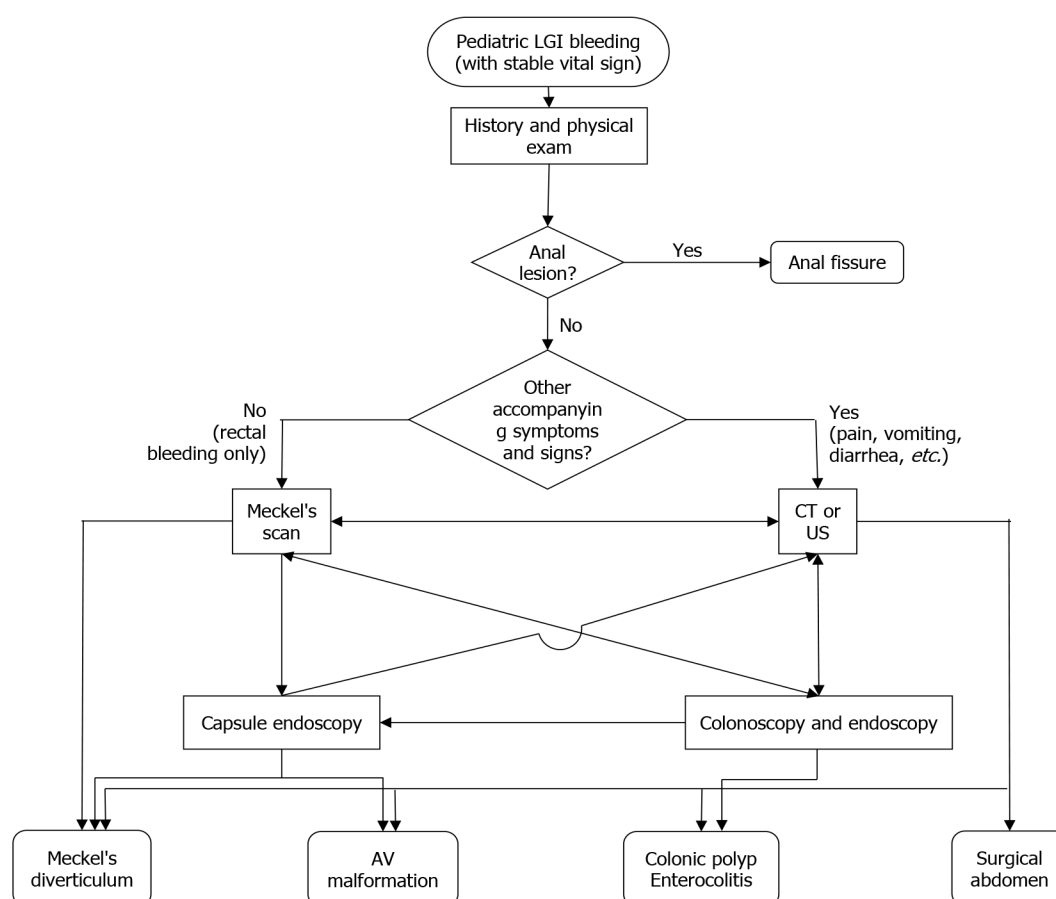


Figure 2 Flowchart for the initial evaluation of pediatric lower gastrointestinal bleeding. LGI: Lower gastrointestinal; CT: Computed tomography; US: Ultrasonography; AV: Arteriovenous.

to totally laparoscopic surgery using staplers. SILS also has the advantage of being able to directly modulate the lesion and achieve complete resection[17]. Our previous study showed that most lesions of the small bowel in pediatric patients can be managed with SILS, especially for MD and small bowel masses causing intussusception[18]. Patients in this study, especially those in the beyond infancy group, also had good outcomes of MIS. We, therefore, suggest MIS, especially SILS, as the first choice for the surgical treatment of OMDR.

This study has some limitations. It was a single-center retrospective observational study, and the sample of subjects was small. Further prospective multicenter studies with a large sample of patients will be needed.

We propose a work up protocol for the early diagnosis of OMDR and ruling out of other causes. Some highlights of the protocol are as follows. For pediatric massive rectal bleeding, MS should be the first diagnostic modality rather than US, CT, or endoscopy. For umbilical lesions in infants, US should be performed. This will be helpful for the rapid diagnosis and management of pediatric OMDR.

CONCLUSION

The most common type of OMDR differs among age groups; umbilical lesions are common in infancy, while MD is common in children with symptoms beyond infancy. US will be helpful for the differential diagnosis OMDR of with umbilical lesions. MS, a less invasive and accurate modality for OMDR, is recommended for patients with painless rectal bleeding for diagnosing MD. MIS, especially a SILS, is effective for managing children with MD regardless of age.

ARTICLE HIGHLIGHTS

Research background

The omphalomesenteric duct remnant (OMDR) shows variable clinical manifestations according to the age at diagnosis with a lifelong complication rates of 4-34 which more common in patients younger than 2 years of age.

Research motivation

Identify the distinct clinical features and its management according to different age groups.

Research objectives

Variant clinical types of OMDR according to pediatric age were assessed and its results were analyzed, then tried to suggest a useful information for proper management by figuring out surgical perspectives.

Research methods

A total of 35 patients (7 infants, 28 children beyond infancy) were reviewed. The patients were divided into two groups, infant and beyond infancy. The patients' demographic characteristics, clinical manifestations, diagnostic tools, surgical procedures, and clinical outcomes were compared between two groups.

Research results

There were two different clinical patterns, Meckel's diverticulum (MD) was the most common clinical type and umbilical lesions were significantly common in the infant group ($P = 0.006$). Umbilical lesions were the most commonly presented symptom in infants, whereas hematochezia and abdominal pain were beyond infancy group. Meckel's scan was most useful in diagnosing OMDR in patients with painless rectal bleeding. Minimally invasive surgery (MIS), especially a single-incision laparoscopic surgery (SILS), was performed more frequently in children than infants ($P = 0.016$).

Research conclusions

Considering the different clinical types, umbilical lesions in infants and MD beyond infancy, a MIS is effective for managing MD regardless of age.

Research perspectives

SILS could be considered as a preferred method for managing OMDR regardless of age.

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