



The identification and treatment of intestinal malrotation in older children

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Abstract

Purpose Intestinal malrotation is often diagnosed in infancy. The true incidence of malrotation outside of this age is unknown. These patients can present atypically or be asymptomatic and diagnosed incidentally. We evaluate the incidence, clinical presentation, ideal imaging, and intra-operative findings of patients with malrotation over 1 year of age.

Methods Retrospective review was conducted in patients older than 1 year, treated for malrotation at a single pediatric tertiary care center from 2000 to 2015. Data analyzed included demographics, presentation, imaging, intraoperative findings, and follow-up. Patients predisposed to malrotation were excluded.

Results 246 patients were diagnosed with malrotation, of which 77 patients were older than 1 year of age. The most common presenting symptoms were vomiting (68%) and abdominal pain (57%). The most common method of diagnosis was UGI (61%). In 88%, the UGI revealed malrotation. 73 of 75 were confirmed to have malrotation at surgery. Intra-operatively, 60% were found to have a malrotated intestinal orientation and 33% with a non-rotated orientation. Obstruction was present in 22% with 12% having volvulus. Of those with follow-up, 58% reported alleviation of symptoms.

Conclusion Despite age malrotation should be on the differential given a variable clinical presentation. UGI should be conducted to allow for prompt diagnosis and surgical intervention.

Keywords Intestinal malrotation · UGI · Abdominal pain · Low lying ligament of Treitz

Introduction

Intestinal malrotation is often a diagnosis of infancy and occurs at a rate of 1 in 2500 live births and up to 1% of the general population [1]. 90% of patients with intestinal malrotation are diagnosed within the first year of life [2]. However, the true incidence outside of this age is unknown, as these patients often present atypically or are completely asymptomatic, being diagnosed intra-operatively during other procedures. Patients may have symptoms of chronic vague abdominal pain, diarrhea, early satiety, dyspepsia, obstruction, and peptic or duodenal ulcer or been diagnosed at autopsy [2–4]. We sought to evaluate the incidence, clinical presentation, ideal imaging and intra-operative findings of patients diagnosed with malrotation over 1 year of age.

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Materials and methods

Following IRB approval (#1410432), a retrospective review was conducted in patients older than 1 year of age who were treated for malrotation at a single pediatric tertiary care center between 2000 and 2015. Patient lists were obtained from our IT department utilizing International Classification of Disease Ninth Revision (ICD-9) diagnosis codes for malrotation. Patients were included if they were older than 1 year of age and treated for intestinal malrotation. Patients were excluded if they had a condition which predisposed them to malrotation such as in situs inversus, gastroschisis, omphalocele, and congenital diaphragmatic hernia. Data were collected including age at presentation, presenting symptoms, imaging performed, intraoperative findings and postoperative follow-up.

A low lying or abnormal position of the ligament of Treitz or malposition of bowel where the small bowel lies in the right abdomen and large bowel in the left abdomen was diagnostic for malrotation. Imaging results were categorized into either: diagnostic for malrotation, suspicious for malrotation, or cannot rule out malrotation. The imaging findings were then correlated to operative findings. Operative reports of those patients who underwent surgical intervention were reviewed. We evaluated for specific intestinal orientation (i.e., normal, nonrotated, malrotated, or reverse rotated), volvulus, presence of Ladd's bands, narrow mesenteric root, and obstruction.

Descriptive statistics including counts and percentages were analyzed. Statistical analysis was performed using STATA (StataCorp 2017. Stata Statistical Software: Release 15. College Station, TX, USA: StataCorp LLC) for calculations.

Results

A total of 246 patients were diagnosed with malrotation during the study period. 73 (31%) were found to be older than 1 year of age. Age distribution can be seen in Fig. 1. The median age at diagnosis was 8 years [IQR 2, 5]. Of these, 36% ($n=28$) were found incidentally during investigations for other disease processes. The most common presenting symptoms were vomiting (68%, $n=52$), abdominal pain (57%, $n=44$), feed intolerance (18%, $n=14$), and bilious vomiting (17%, $n=13$). 56% ($n=43$) of patients had recurrent symptoms and 19% ($n=15$) had chronic abdominal pain. When dividing by age group, defined according to National Institute of Child Health and Human Development pediatric terminology [6], 30% ($n=23$) were age 1–2 years, 16% ($n=12$) were age 3–5 years, 22% ($n=17$)

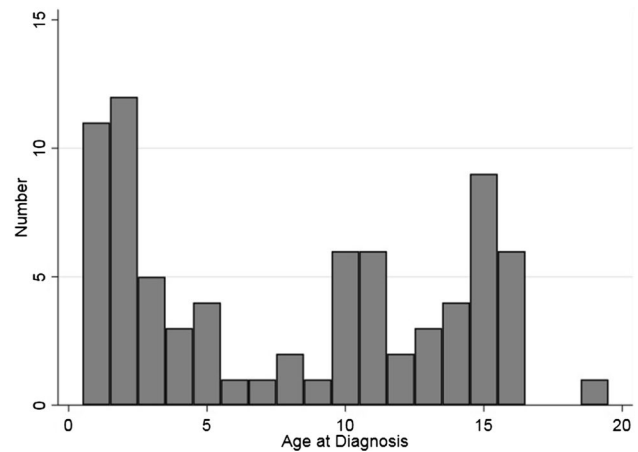


Fig. 1 Histogram displaying the age of diagnosis

were age 6–11 years, 31% ($n=24$) were age 12–18 years, and 1% ($n=1$) was age 19–21 years. The most common presenting symptom was feed intolerance in those aged 1–2 years, vomiting in age 3–5 years (although not reaching significance), and abdominal pain in those aged 12–18 years (Table 1). Abdominal pain was the only presenting symptom in the one 19-year-old patient. Recurrent episodes of presenting symptoms occurred more often in those aged 1–2 years although this was not statistically significant.

57 patients had an upper gastrointestinal series (UGI), 34 had a computed tomography (CT) scan, and nine had an ultrasound (US) during their workup. 16 had both an UGI and CT scan. Eight had both an US and an UGI and/or CT scan. The use of CT was most common in children older than 5 years (Table 2). Initial diagnosis of malrotation was reached by UGI in 61% ($n=47$), CT in 26% ($n=20$), intraoperatively in 6.5% ($n=5$), or another modality in 6.5% ($n=5$). Other modalities included barium enema, esophagram, US and CT angiogram. There was no significant age group difference in imaging modality to achieve initial diagnosis ($p=0.07$). Of the patients who were diagnosed with malrotation by UGI, 74% ($n=35$) had emesis as one of their presenting symptoms. Of those diagnosed by CT, 90% ($n=18$) had abdominal pain as the most common presenting symptom. Only one patient was diagnosed by US and the presenting symptoms were abdominal pain and emesis. Of the eight who had US plus another modality, UGI was diagnostic for malrotation in six, whereas CT was diagnostic in one, and one was diagnosed intra-operatively. Of those that had both an UGI and CT scan, the UGI led to initial diagnosis of malrotation in 63% ($n=10$) while CT led to initial diagnosis in 25% ($n=4$) and the remaining two were diagnosed by CT angiogram and barium enema. The four patients diagnosed by CT did had a follow-up

Table 1 Most common presenting symptoms by age

	1–2 years (n=23) (%)	3–5 years (n=12) (%)	6–11 years (n=17) (%)	12–18 years (n=24) (%)	19–21 years (n=1) (%)	P value
Reflux	48	0	12	4	0	<0.001*
Abdominal pain	17	58	59	92	1	<0.001*
Emesis	70	83	59	67	0	0.41
Bilious emesis	17	33	6	17	0	0.42
Chronic abdominal pain	0	17	24	38	0	0.02*
Previous episodes	70	58	41	54	0	0.35

*Statistically significant

Table 2 Imaging modalities by age

	1–2 years (n=23) (%)	3–5 years (n=12) (%)	6–11 years (n=17) (%)	12–18 years (n=24) (%)	19–21 years (n=1) (%)	P value
UGI	91	75	65	63	100	0.18
CT	4	33	71	67	100	<0.001*
BE	17	17	0	4	0	0.27
US	9	33	12	4	0	0.13

*Statistically significant

UGI study where three were confirmed to have malrotation and one was diagnostic.

Of those patients diagnosed with intestinal malrotation, 75 had surgical intervention. Two patients did not have surgery because their symptoms resolved. Surgical approach was laparoscopic in 53% (n=40), open in 36% (n=27), and laparoscopic converted to open in 11% (n=8). Of the 75, 95% (n=71) were confirmed to have malrotation. 57 of the 75 patients who had an operation had an UGI during their workup, of which 95% were confirmed to have malrotation at surgery. 88% of UGIs (n=50) had reported malrotation, while 8% (n=5) were suspicious for malrotation and the remainder could not rule out malrotation 4% (n=2). 53 of the 57 UGIs commented on an abnormal position of the proximal small bowel, of which 96% (n=51) were confirmed to have malrotation at surgery. Only 27 of the UGIs commented on the position of the ligament of Treitz and from these, 11 were reported to be low-lying. Out of these 11 patients, 10 were confirmed to have malrotation. The eleventh patient did not have surgical exploration. Of the 34 patients who underwent CT during their workup, 97% were confirmed to have malrotation at surgery. Intra-operatively, 60% had a typical malrotated intestinal orientation where the duodenojejunal segment lies within the right hemi-abdomen and the cecocolic limb has partial rotation ending up in the mid-upper abdomen. 33% had a nonrotated orientation where the duodenojejunal segment lies within the right hemi-abdomen and the large bowel lies within the left hemi-abdomen and 1% had a reverse rotation orientation where the duodenum rests anterior to the superior mesenteric artery or the transverse colon is posterior to the superior mesenteric

Table 3 Intraoperative findings

	Frequency (%)
Normal orientation	6
Nonrotated	33
Typical malrotation	60
Reverse rotation	1
Volvulus	8
Ladd's bands	93
Narrow mesenteric root	68
Obstruction	22

artery. We found no age-related differences in the type of intestinal orientation found at the time orientation. Of 73 patients who went to the operating room for a Ladd's procedure, 93% (n=68) of patients were found to have Ladd's bands at the time of surgery. Intraoperative findings for these patients are shown in Table 3. 60% (n=41) of patients with typical malrotation, 34% (n=23) with nonrotated orientation, 2% (n=1) with reverse orientation, and 4% (n=3) with normal orientation had Ladd's bands present (p=0.26). Of 65 patients where description of the mesenteric root was documented, 68% (n=44) were noted to have a narrow mesenteric root. 59% (n=26) of patients with typical malrotation had a narrow mesenteric root, as did 36% (n=16) of nonrotated patients, 2% (n=1) of reverse oriented, and 2% (n=1) of normal oriented patients (p=0.26). 22% (n=16) of patients were reportedly obstructed with no significant association with a particular orientation (p=0.53), but 38% (n=6) of those had volvulus. Volvulus was also not found to

be associated with a particular orientation ($p=0.78$). None required bowel resection at the time of operation. There were no intra-operative complications. Post-operatively, one had recurrent volvulus resulting in re-operation, one had a duodenal hematoma resulting in re-operation, one had a paraduodenal hematoma managed conservatively, two had superficial wound infections, one had prolonged ileus, and one underwent exploratory laparotomy 3 months post-operatively for adhesive small bowel obstruction.

64 patients (85%) had a median postoperative follow-up of 1 month [IQR 0.75, 13.5]. Complete alleviation of symptoms was reported in 58% ($n=37$) of patients, while 25% ($n=16$) had persistence of preoperative symptoms and 17% ($n=11$) remained asymptomatic. There were no differences in post-operative symptom resolution based on age group ($p=0.86$), imaging used to reach diagnosis ($p=0.09$), anatomical orientation found at surgery ($p=0.71$), or surgical approach ($p=0.98$). Post-operative symptom resolution was greater in patients who presented with bilious emesis ($p=0.02$).

Discussion

Malrotation can affect pediatric patients despite patient age. In our retrospective study, one-third of patients diagnosed with malrotation were older than 1 year of age. A KID database study examined 219 patients between 1 and 18 years of age, of which the majority presented in their first or second year of life [7]. The median age of our cohort was older at 8 years. Current literature suggests that presentation may be affected by patient age with several studies revealing that patients with malrotation greater than 1 year of age present with abdominal pain [2, 8, 9]. One retrospective study of 38 patients reported that even within 2 years of birth, symptoms can be consistent with infantile malrotation but that after 2 years atypical symptoms such as chronic abdominal pain and non-bilious emesis develop [3]. This is consistent with our study where greater than half of our patients presented with abdominal pain and/or vomiting. This suggests that beyond infancy, patients with persistent abdominal pain and emesis should be considered for a diagnosis of malrotation.

In many studies, UGI is considered the gold standard in diagnosing malrotation even in children greater than 1 year old [3, 10]. This is also true at our institution where 74% of the study population underwent UGI. However, 12% ($n=7$) of the UGIs did have questionable malrotation. Five patients had UGIs that were suspicious for malrotation of which, four were confirmed to have malrotation at surgery and one was found to have cecal volvulus. Four of these five had follow-up and reported symptom resolution. Two patients had an UGI that could not rule out malrotation. Both were confirmed to have malrotation at surgery

and both reported symptom resolution at follow-up. Thus, of the seven patients with questionable imaging, six had post-operative follow-up and reported symptom resolution. This suggests that even in the face of questionable UGI imaging, these patients benefit from surgical intervention.

We found that 34 patients had a CT during their workup and CT use increased with increasing age. Of the 20 patients diagnosed by CT, four underwent UGI where three confirmed malrotation and one was non-diagnostic. The patient with non-diagnostic UGI had undergone previous fundoplication and gastrostomy tube placement and literature suggests that previous abdominal surgeries or indwelling tubes can distort relevant anatomy so it can be difficult to assess for an anomaly of rotation in these cases [10]. In the context of diagnosing malrotation, much of the literature discusses the relationship of the mesenteric vessels and the “mesenteric swirl” sign that can be indicative of volvulus and a surrogate for malrotation [11–13]. However, in several studies including ours, not all malrotated patients present with volvulus. Furthermore, in a study by Sodhi et al. [12], counterclockwise rotation of the superior mesenteric vein on the superior mesenteric artery gives an appearance of a mesenteric swirl in otherwise normal mesenteric vessels due to variation in the branching pattern of mesenteric vessels and can be misinterpreted as midgut volvulus resulting in advertent surgical intervention. More recent studies are exploring an abnormally positioned duodenum as a more reliable imaging finding compared to an abnormality in the positioning of the mesenteric vessels in order to identify malrotation [12, 14]. The position of the duodenum and proximal small bowel can be readily seen with an UGI and with less reliance on the positioning of mesenteric vessels for identification of malrotation and the increased radiation delivered with CT, UGI should be the first imaging modality considered for diagnosing malrotation.

There has been some suggestion that US could be utilized to diagnose malrotation with observation of a retromesenteric 3rd portion of the duodenum to exclude malrotation [11]. In our study, of the nine patients who underwent US, eight had additional imaging to confirm malrotation, the majority as UGI. Only one patient was diagnosed with malrotation by US. The US described a malpositioned appendix winding around a vessel at the level of the umbilicus rather than the classic US findings of malrotation such as the inversion of the superior mesenteric vessels or whirlpool sign. Consistent with an American Pediatric Surgical Association systematic review by Graziano et al. [15], we continue to use an UGI as the primary method of malrotation diagnosis. As we continue to try to reduce radiation exposure in children, there may be a role in detailed ultrasonography for diagnosing malrotation in the future, but further larger studies should be undertaken to confirm this.

Of those patients radiologically diagnosed with intestinal malrotation, four patients were found to not have malrotation at the time of their operation. Interestingly, all four of these patients had imaging from another facility and three of these had no repeat imaging done at our institution. However, all imaging was reviewed by our radiologists. Surgical procedures performed in these patients include duodenocolic dissociation with cecopexy, repair of cecal volvulus with cecopexy, lysis of adhesive duodenal bands and appendectomies. If a patient is already going to the operating room due to an acute abdomen or volvulus and repeat imaging would not change that plan, one could argue there is no need for repeat imaging. However, in a non-acute setting diagnosis should be confirmed. Thus, imaging should be repeated or confirmed by radiology when accepting patients from non-pediatric hospitals, hospitals without adequate resources or low volume centers where malrotation is not encountered frequently.

At our institution, we offer surgery for malrotation patients who are symptomatic. There are a number of patients who are found to have malrotation incidentally and depending on the comorbidities of the patient, the severity of symptoms and whether those symptoms could be attributed to the malrotation determines the need for urgent surgical intervention versus elective surgical intervention to reduce risk of volvulus. We have on rare occasion chosen observation for incidental malrotation found on imaging. In this study, two patients were observed, both were poor surgical candidates and both had eventual resolution of their symptoms with treatment of another medical problem. In these now asymptomatic patients, it was felt that the risk of an operation outweighed the benefit.

Over the course of this study, our surgical approach is primarily laparoscopy, particularly if there is a question of diagnosis as we saw that 6.5% of patients in our study were diagnosed intra-operatively. The systematic review by Graziano et al. [15] reviewed evidence on open versus laparoscopic approach that indicates the laparoscopic approach is safe for treating and diagnosing malrotation, particularly when distinguishing between typical malrotation with a narrow mesenteric base and nonrotation or atypical malrotation. We report greater than half of surgically treated patients undergoing a laparoscopic procedure during which any volvulus found was detorsed, the bands were taken down and an appendectomy was performed. The root of the mesentery was widened in most cases and in those where it was not, the operating surgeon noted in the operative reported that the base was already very broad and did not necessitate widening. We did find an 11% conversion to open rate due to inability to define anatomy, inadequate visualization, or difficult dissection. Therefore, unless there is a contraindication, our surgical approach is traditionally laparoscopic.

After correction of malrotation long-term complications can occur. One review of 196 cases examining the incidence of adhesive bowel obstruction following surgery for malrotation suggested that up to 14.2% of patients may develop adhesive small bowel obstruction [16]. Another retrospective study comparing 131 children, with 27 older than 12 months, found that older children had a higher emergency reoperation rate after a Ladd's procedure [17]. In our study, four patients with persistent symptoms did return to the operating room. The first had persistent bilious emesis and was found to have narrowing at D2 for which he underwent duodenostomy. The second was taken back for obstructive symptoms and was found to have a duodenal hematoma and had a temporary jejunostomy tube placed which was eventually removed 3 months later. The third had bilious emesis 3 months after Ladd's procedure and was found to have recurrent volvulus. She was de-torsed, the base of the mesentery was widened and the duodenum was pexied to the right renal fossa. The fourth went back for bilious gastrostomy tube output and abdominal pain and was found to have adhesive obstruction for which he underwent adhesiolysis and additional widening of the mesentery. Despite these rare complications, correction of malrotation improves symptoms in greater than 50% of patients [3]. Thus, while there are a variety of reasons that patients may have chronic abdominal pain, it appears that with a diagnosis of malrotation a Ladd's procedure provides relief in around half of patients.

Similarly, we saw symptom resolution in 58% of patients who had post-operative follow-up ranging from 3 weeks to 13.5 months. Symptom improvement following surgery was significantly greater in children presenting with bilious emesis. However, 30% were still symptomatic after surgery. This has been seen in other studies, with up to 40% of patients > 1 year having persistent symptoms post-operatively [3]. Many of these patients underwent additional work up of persistent symptoms and were given another diagnosis such as esophagitis, duodenal eosinophilia, dysmotility, gastroenteritis, constipation, cyclical vomiting, or inborn metabolism error. Our data suggest that the children who will benefit most from surgical intervention are those presenting with bilious emesis even though this is least common presenting symptom in older children. The most common presenting symptom identified in this study was vomiting not reported as bilious followed by abdominal pain, which led to further work-up and identification of malrotation. Based on the fact that all the patients in our study had some types of presenting symptom, even if atypical, we agree with others that patients who are symptomatic should undergo surgery due to the risk of a catastrophic event such as midgut volvulus, ischemic bowel, possible short bowel syndrome or even death [18]. There is still debate on operative intervention in asymptomatic patients as we do not know who is at the

highest risk of volvulus in the absence of symptoms. However, the systematic review by Graziano et al. [15] gives a grade C recommendation based on level 3–4 evidence to consider operative intervention in younger asymptomatic patients but that observation may be appropriate in older asymptomatic patients, and an asymptomatic patient reliably diagnosed with atypical malrotation can be observed. Based on this and our study, we advocate for surgical correction of malrotation unless the risk of surgery outweighs the benefit, such as in an asymptomatic patient with major medical comorbidities.

There are several limitations to our study, mainly the small patient population, retrospective nature, and short follow-up. Despite the small patient population, this is the largest single institution study to date of patients with malrotation over the age of 1 year. To improve the conclusions that can be drawn, a larger multicenter study with longer follow-up should be performed. However, our results are similar to previous studies on this topic and add to the body of evidence supporting the consideration of malrotation in older children with atypical symptoms, the use of UGI for diagnosis, and pursuance of surgical correction to alleviate these symptoms and avoid future intra-abdominal catastrophe.

Conclusion

Malrotation should be on the differential despite age of child given the variable clinical presentation. An UGI should be conducted first for a prompt diagnosis and surgical correction should be undertaken despite atypical presenting symptoms.

Author contributions CD: review of original data, statistical analysis and interpretation, re-drafting of article, approval of article. JS: drafting article, critical revision of article, approval of article. KW: concept and design, data collection, data analysis and interpretation, critical revision of article, approval of article. SS: data collection, data analysis. KG: data collection, data analysis and interpretation, approval of article. SDP: concept and design, data analysis and interpretation, critical revision of article, 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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