Laparoscopic Ladd's procedure for malrotation in infants and children is still a controversial approach.

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Abstract

Background: Open Ladd's procedure is the gold standard for the correction of intestinal malrotation and laparoscopic approach remains controversial. This study aimed to evaluate our experience in laparoscopic management of malrotation.

Methods: Single centre retrospective study including patients who underwent a laparoscopic assessment of intestinal malrotation with correction if appropriate between 2007 and 2017. *Results:* Sixty-five patients (median age 7 months) had a laparoscopic assessment with and without correction of malrotation. Forty-five (69%) were symptomatic, including 16 (25%) with a midgut volvulus. The procedure was completed laparoscopically in 55 (86%) patients in 110 minutes (30-190). Conversions happened more frequently at the beginning of the experience. With a follow-up of 12.5 months (8 days-5.3years), morbidity rate was 15% and 4 (6%) patients underwent a redo surgery, all in the first 5 months after surgery, compared with 3/53 (6%) in a contemporaneous group undergoing open Ladd's.

Conclusion: This is the largest series reported so far of the laparoscopic management of malrotation. Laparoscopic Ladd's procedure is reliable but still exposes to open conversion which may be in part due to a learning curve. A low conversion threshold is important in cases with volvulus. The redo rate is similar to that of the open procedure.

Key words: intestinal malrotation, laparoscopic Ladd's procedure, midgut volvulus

Level of evidence: Level III retrospective comparative treatment study

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Intestinal malrotation is defined by an abnormal disposition of the gut within the abdominal cavity resulting in an increased risk of midgut volvulus. One of the main factors associated with volvulus is the width of the root of the mesentery, which can only be assessed surgically. This condition most commonly becomes symptomatic during infancy but atypical symptoms may result in a delayed diagnosis until later childhood.

The surgical approach to malrotation was initially described by Ladd in the 1930s (1), and consists in detorsion of the volvulus, where present, followed by the division of abnormal peritoneal bands attached to the caecum (Ladd's bands), widening of the mesenteric root and positioning of the bowel in a stable position within the abdominal cavity, with the large bowel on the left and small bowel on the right. In addition to that, some surgeons proceed to a prophylactic appendectomy and/or a caecopexy, which was not present in the original description. These manoeuvres have been abandoned by several centres in an effort to avoid associated complications (2).

Some clinical conditions are likely to be associated with an incomplete rotation (e.g. congenital diaphragmatic hernia, exomphalos, isomerism) which may require treatment when symptomatic (intestinal obstruction secondary to volvulus, Ladd's band or kinking duodenum), thus reducing the risk of midgut volvulus and avoiding its consequences. The routine use of a prophylactic Ladd's procedure remains a matter of debate in asymptomatic patients with intestinal rotational abnormalities associated with these conditions (3,4). Two decades ago, progress in minimally invasive techniques and surgical expertise led to the development of laparoscopic Ladd's procedure, particularly in an effort to avoid the complications associated with open surgery in these patients (5). The effectiveness and the safety of this approach remains a matter of concern in particular when the patient presents with a volvulus, although the laparoscopic approach has the potential to improve long term outcomes by decreasing the risk of adhesional obstruction and improve cosmesis (6–8).

The aim of our study was to evaluate the results of the laparoscopic Ladd's procedure over a 10-year period in patients with a malrotation diagnosed on the upper gastrointestinal contrast study; either presenting with an acute volvulus or other gastrointestinal symptoms or asymptomatic with a clinical condition at risk of a rotational anomaly.

1. Patients and methods

After institutional audit approval, a retrospective single centre review was performed on patients who underwent a laparoscopic Ladd's procedure between April 2007 and August 2017. Indications to proceed were based on a clinical presentation suggesting a midgut volvulus and/or suspicion of malrotation on the upper gastrointestinal contrast study. The surgery was performed as an emergency in case of volvulus, or as elective in case of incidental finding of malrotation during a general assessment or on a contrast study performed in preparation of other surgical procedures (i.e. laparoscopic fundoplication or laparoscopic assisted gastrostomy).

Patient details, clinical presentation, operative notes, hospitalisation events, complications and outcomes were collected. The Clavien-Dindo classification was used to analyse the severity of complications (9). The operative technique used in our centre followed the Ladd's procedure previously described with its adaptation for laparoscopy reported by Van der Zee et al (5,10). Briefly, an umbilical port was inserted (open technique) and 2 to 3 ports (5 or 3mm) placed, including a liver retractor if necessary. The surgeon assessed the vitality of the gut, the rotational status of the bowel and performed the section of the bands followed by the widening of the root of the mesentery. Cecopexy was performed according to surgeon preference, and involved fixation of the caecum to the lateral abdominal wall in the left upper quadrant using non-absorbable sutures.

Results are presented as median (range) or N (%) as appropriate. Continuous data were compared using Kruskall-Wallis test with Dunn's multiple comparison test. Categorical variables were compared using Fisher's exact test, using a Bonferonni-adjusted cut-off of 0.017 for significance. A p value lesser than 0.05 was considered statistically significant (two-tailed).

2. <u>Results</u>

2.1. Patients 'characteristics (Tables 1 and 2)

Sixty-five patients had a laparoscopic exploration of an intestinal malrotation and 64 underwent a laparoscopic Ladd's procedure during the study period. Thirty-nine patients were (60%) male and 26 (40%) female. Overall, 50 (77%) presented with concomitant abnormalities.

2.2. Surgical data (Table 2)

The surgery was performed at median age of 218 days (range 0 day-15.8 years) at a median weight of 6.3 kg (2.0 -100.8).

Indications for surgery were: suspicion of volvulus (n=14), symptomatic malrotation without suspicion of volvulus (31) and prophylactic (20). Predominant presenting symptoms (Figure 1) were bilious vomiting (19), non-bilious vomiting (8), gastro-oesophageal reflux (9), abdominal pain (7) and failure to thrive (2). Four patients presenting with symptoms had a previously diagnosed situs inversus. Of the 20 (31%) patients who were asymptomatic, the indications to investigate the mesenteric rotation with an upper gastrointestinal contrast study were: exomphalos (7), suspected situs inversus or heterotaxia (6), tracheo-oesophageal fistula repair (1) and other congenital syndrome or concomitant abnormalities (6).

Operating time was 110 minutes (30-190) overall, 120 (60-160) minutes if a volvulus was present, 90 (45-130) minutes for a prophylactic procedure, and 125 (40-180) minutes when combined with another procedure.

A midgut volvulus was found in 16 cases. The Ladd's procedure was associated with a caecopexy in 3 (5%) cases and appendectomy in 7 (11%) cases (4 laparoscopic and 3 laparoscopic converted to open procedures). Additional procedures were performed during the same surgery in 17 cases: 6 gastrostomy formations, 5 fundoplications with gastrostomy, 1 fundoplication, 1 redo fundoplication, 1 excision of jejunal duplication, 1 Meckel diverticulectomy, and 2 duodeno-duodenostomies for duodenal atresia. Three of these procedures were performed in patients with volvulus: 1 gastrostomy, 1 Meckel diverticulum resection and 1 resection of jejunal duplication. No patients required a bowel resection for ischaemic bowel. In one patient, the malrotation was deemed not to require Ladd's procedure (mobile caecum but wide root of the mesentery). The Ladd's procedure was completed laparoscopically in 54 (84%) cases. Conversion to open surgery was necessary in 6 (38%) patients in the volvulus group, 2 (7%) in the symptomatic malrotation group without volvulus and 2 (10%) in the asymptomatic group.

The conversion rate was not significantly different (p=0.5) before the age of 6 months (19% versus 12%). Six (60%) of the conversions happened in the 4 first years of our experience (2007-2011).

Minor perioperative complications (serosal tear) occurred in two patients during the laparoscopic procedure (3%), one in the prophylactic group and one in a patient who underwent additional duodeno-duodenostomy.

2.3. Postoperative outcomes

The hospital stay was 6 (1-426) days overall, with a significantly shorter stay for prophylactic procedures (Table 2). Morphine was used during 1 (0-4) day. The time from surgery to full enteral feeding was 4 (1-27) days with a significantly longer time for the volvulus group (Table 2). 6 patients were lost to formal follow-up but were presumed not to require further surgery.

The 30-day morbidity rate was 12% (14% of those with follow-up), with 5 patients presenting with a Clavien-Dindo grade III complication, 2 patients a grade II complication and 1 patient a grade I complication. Two patients required redo surgery within 30 days postoperatively; one for recurrent bilious vomiting (patient 1, Table 3) and one for midgut volvulus (patient 3, Table 3). One patient presenting with bilious vomiting and severe abdominal pain 3 weeks after discharge presented with an internal hernia through the mesentery and required surgery which was initiated laparoscopically but converted to open surgery. Another patient required an abdominal drainage for a chylous collection on the 6th postoperative day. A further patient presented with a missed congenital duodenal stenosis and required laparoscopic duodeno-duodenostomy six days after the Ladd's procedure. Two patients developed pneumonia and one developed a wound infection.

The median follow-up time was 12.5 months (8 days-5.3 years). The overall morbidity rate was 15% (7 Clavien Dindo III, 2 Clavien Dindo II and 1 Clavien Dindo I) (17% of those with follow-up). The overall redo/second surgery rate was 9% (10% of those with follow-up). Second surgery was required in 2 cases for an internal hernia and for the missed congenital duodenal stenosis (see above). Redo surgery was required in 4 (6%) cases (Table 3): 3 (4.5%) patients for a midgut volvulus at day 19 (patient 3), 108 (patient 2) and 150 (patient 4) after surgery respectively and 1 (1.5%) for incomplete procedure 12 days after initial surgery (patient 1). Thus, two patients had their redo within 30 days while the other two redo occurred later. All these 4 (3 symptomatic, 1 asymptomatic) patients had an intestinal malrotation without volvulus at the time of the first procedure (laparoscopy). Redo surgeries were attempted by laparoscopy in 3 patients: 1 was achieved successfully and 2 were converted to open.

One patient had a laparotomy at a first instance. This latter patient with post Ladd's volvulus required bowel resection 2 months later for small bowel stenosis and was on parenteral nutrition at the last follow-up. All 4 patients were younger than 2 months at the first surgery and were operated on during the first six years of experience. No death or adhesive bowel obstruction occurred during the period of follow -up. While this study was not designed as a comparative between open and laparoscopic repair of malrotation, during 2006-2012, 53 patients had an open Ladd's procedure at our Institution, with 3 patients requiring redo; a redo rate of 6%.

3. Discussion

This study reports 64 cases of laparoscopic Ladd's procedures in children, of which one quarter were performed in the presence of a midgut volvulus. To the best of our knowledge, this is the largest series ever presented and, while showing the benefit of the procedure, it also demonstrates its limitations. On the positive side, the correction of malrotation even in case of volvulus is feasible and the operative time was in keeping to what has been reported by the literature, as were the perioperative complications. In the volvulus group, the percentage of conversion was higher than in the other groups but surgery was definitive in all cases. While the learning curve may explain the higher conversion rate in our early experience, we have no evidence to suggest that the learning curve could play a role on the recurrence rate as laparoscopic Ladd's procedure exposed to the same redo surgery rate than the open procedure. Laparoscopic Ladd's procedure has been adopted by a limited number of centres and it can be challenging when performed in small infants (7).

Our series showed that 60% of conversions occurred during the first part of the experience. Similarly, Ooms et al in 2015 (11) reported that 73% of the conversions happened in the first 3 years of their experience. In the literature, articles reporting large series (more than 30 patients) described a conversion rate between 8% and 45% (mean=23%) (7,11–14). Our overall conversion rate was 16%, but increased to 19% when considering patients below 6 months of life and reached 37% in patients with a midgut volvulus. This tendency for increased conversion rates in smaller children is mirrored in the literature. Hsiao et al (13) reported 50% conversion rate in neonates, but only 18% in older patients, whilst in their meta-analysis, Catania et al described a conversion rate up to 25.3% (8). Similarly, recurrence happened more frequently in infants younger than 6 months (p<0.05) in whom laparoscopy is more demanding and in whom accurate dissection of the mesentery can be difficult to achieve. In a recent series comparing 9 laparoscopic (3 neonates vs 6 older) and 17 open Ladd's procedures (11 neonates vs 6 older), Miyano et al (15) concluded that the laparoscopic approach cannot be recommended in neonates. While our study was not designed as a comparative between open and laparoscopic repair of malrotation, the redo rate after an open Ladd's procedure at our Institution was 6%. This is comparable to the incidence observed by us after laparoscopic repair and it is in keeping

with a reported incidence between 2 and 7 % of recurrence rate after open Ladd's procedure (16,17); a technique which has shown in same cases to have a recurrence rate as high as 19%(18).

All patients were operated on following the modified procedure which was initially reported by Van der Zee et al (5,10). This series emphasized some points to facilitate the progression of this demanding surgery. First, patients should be head up to ease the exposition of the gut. A Nathanson retractor is used to mobilize the liver and facilitate the visualization of the duodenum and upper abdomen. In case of ischemic and/or dilated bowel loops, surgeons should carefully mobilized the small bowel to avoid serosal tears or bowel perforation. The lateral adherence of the colon as described by Ladd should be carefully separated to mobilize completely the colon. Lateral dissection of the duodenum should be made to mobilize the duodenojejunum avoiding at all cost damage to the common bile duct or to the pancreas. At the end of the surgical dissection, surgeon has to be able to easily follow the straightened duodenum and the all small bowel with a large widened mesentery base. This major point may decrease the risk of redo for insufficient procedure. In case of opening of the mesentery, suture repair can prevents future internal hernias.

We have shown here that redo-surgery was limited to the first half of our experience, and we believe that the laparoscopic approach for the correction of an intestinal malrotation can be safely performed in infants keeping in mind that a careful widening of the root of the mesentery should be adopted. In case of difficult mobilization, there is also the possibility of fixating the caecum using non-absorbable sutures in the left upper quadrant, however this technique was only adopted for 3 patients in our series, so we are unable to draw any definitive conclusions on the safety and efficacy on this point of technique. We agree with Kalfa et al , who suggested that in neonates with suspected volvulus, this procedure should be performed only on selected cases, haemodynamically stable, with an early diagnosis after presentation and the parents should be aware of the high conversion rate. Nevertheless the majority of our patients with midgut volvulus had a laparoscopic Ladd's completed and none of them presented with a recurrence. This is different to what has been previously reported, and it reassures us that there is a potential benefit of minimally invasive surgery in case of volvulus. Indeed, in their series of 20 patients, Ferrero et al (19) showed a 30% recurrence rate in their laparoscopically treated volvulus, and Stanfill et al (12) described a 5.6% post laparoscopic Ladd's volvulus rate (n=36). The most recent meta-analysis showed a 3.5% post laparoscopic Ladd's volvulus rate (8). Similarly, we

have observed 3 (4.5%) patients operated on for malrotation without volvulus in our early experience presenting with a volvulus during the early follow-up period. This is particularly relevant in infants and we agree with what was stated by Ooms et al(11), that neonates below 6 months might represent the highest surgical risk group for redo surgery. None of the patients that had a conversion from laparoscopic to open Ladd's procedure required redo surgery, although the number of patients (0/10 converted vs. 4/54 laparoscopically completed) is too small to meaningfully statistically analyse. Although the follow-up time is limited and the risk of volvulus is lifelong, all the recurrences in our series occurred within the first 5 months after surgery. Nevertheless, extended follow up is necessary to determine the long term efficacy of the laparoscopic procedure.

The benefits of minimally invasive surgery include lower postoperative pain, decreased length of stay, better cosmesis and decreased rate of adhesive bowel obstruction. The median hospital length of stay was short in our series (6 days), which is in keeping with a prompt recovery after a laparoscopic Ladd's procedure (11,12,14,20). In their series comparing 53 laparoscopic cases (mean age of 4.4years) versus 86 open cases (mean age of 0.3 years), Huntington et al showed that the laparoscopic Ladd's approach led to a significant shorter length of stay and a tendency to less postoperative complications in the initial 30 days postoperative period (20). In our series, no patient presented with bowel obstruction during the follow-up period and this should be taken into account when counselling parents since the occurrence of post-operative adhesional obstruction after open Ladd's procedure has been reported to be up to 13% (12).

In this series, 19 (29%) asymptomatic patients with a radiological diagnosis of malrotation underwent laparoscopic Ladd's procedure. The treatment of asymptomatic malrotation remains controversial. Graziano et al (21) in their recent review concluded that it is possible that a procedure may be necessary only in asymptomatic patients younger in age, while observation may be appropriate in the older asymptomatic patient. If reliably diagnosed, atypical malrotation (ligament of Treitz at or to the left of midline but below the level of pylorus), can be observed, especially when there is associated congenital heart disease (level 3-4 evidence, grade C recommendation). Recent studies on heterotaxia and isomerism associated with intestinal rotational abnormalities suggest close observation as an alternative to prophylactic Ladd's procedure in asymptomatic patients (3,4). Considering the low complication rate in this group (5%), the significant shorter time to full enteral feeding and shorter length of stay, our team still proposes laparoscopic exploration in such cases.

The literature also supports the view that where radiological findings are equivocal, laparoscopy allows a safe and easy assessment of the width of the mesentery (22–24). Diagnosis or confirmation of the malrotation is performed by the assessment of caecal fixation, the width of the root of the mesentery and the presence of Ladd's bands crossing the duodenum. Although a multicenter randomised controlled trial might be advocated to compare outcome of laparoscopic versus open Ladd's, a large number of patients would likely be required. For example, a trial based on incidence of adhesional bowel obstruction, based on 0% (laparoscopic) vs. 13% (open) would require 106 patients (80% power, α =0.05).

4. Conclusion

The laparoscopic Ladd's procedure is feasible and safe in infants and children presenting with malrotation suspected on upper gastrointestinal contrast studies. It may also reduce the risk of adhesive obstruction, which is commonly described after the open procedure. A low threshold for conversion is important when the mesentery cannot be adequately assessed or in the presence of a volvulus. Moreover, only experienced laparoscopic surgeons should perform this procedure and families should be aware of the initial institutional learning curve which may be partially responsible to an increased incidence of conversions.

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Legends for illustration

Table 1: Patients' concomitant abnormalities.

Table 2: Patient characteristics

Continuous data were compared using Kruskall-Wallis test (p-value) with Dunn's multiple comparison test. Superscripts indicate significant differences between the indicated pair of groups: ^a p<0.05, ^b p<0.001, ^c p<0.01.

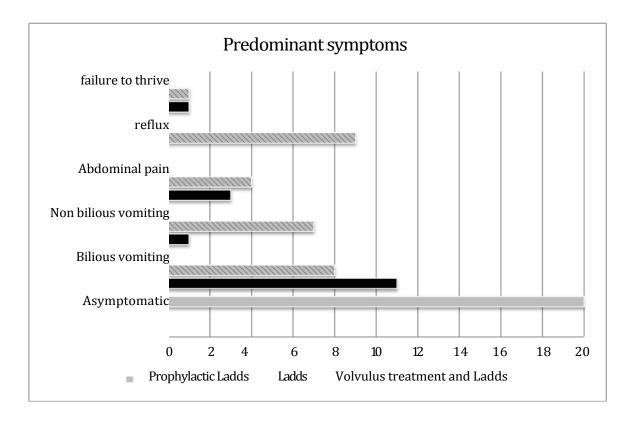
Categorical variables were compared using Fisher's exact test, using a Bonferonni-adjusted cut-off of 0.017 for significance. Significant differences between pairs of groups are indicated with d.

Table 3: Characteristic of patients requiring a redo procedure

Four patients underwent a redo procedure. All of them needed the redo surgery within the first 7 months of follow-up. Among them, 3 presented with a mid-gut volvulus and only 1 had a redo surgery completed laparoscopically.

Figure 1: Symptoms at presentation.

The surgery was performed in patients presenting with failure to thrive (2), gastrooesophageal reflux (9), abdominal pain (7), non-bilious vomiting (8), bilious vomiting (19) and 20 patients were asymptomatic. A volvulus was found in 16 patients.



Concomitant abnormalities	Ν
Syndromic isomerism	10
Exomphalos	7
Down syndrome	4
Upper urinary tract malformation or kidney disease	3
Œsophageal atresia with trachea-œsophageal fistula	2
Beta-thalassemia	2
Isolated cardiac malformation	2
Pfeiffer syndrome	2
Batten's disease	1
Goldenhar syndrome	1
Congenital CMV infection	1
Apert syndrome	1
Eosinophilic gastro-intestinal disease	1
Mitochondrial disorder	1
Noonan syndrom	1
Cerebral palsy	1
Ehler Danlos syndrome	1
Cow milk allergy	1
Hydrops fetalis	1
Unnamed syndrome with associated malformations	7

	Volvulus (16)	Symptomatic malrotation without volvulus (29)	Asymptomatic malrotation (20)	р
GA (weeks)	40 (25-41)	39 (28-41)	38 (28-40)	0.103
BW (Kgs)	3.3 (0.7-3.8)	3.1 (1.52-4.06)	3.0 (1.0-3.6)	0.118
Associated anomalies	8 (50%) ^d	22 (76%)	20 (100%) ^d	
Age at surgery (days)	43 (1-2373)	232 (3-5565)	228 (1-5780)	0.190
Weight at surgery (Kgs)	3.67 (2-20.3)	8.28 (2.55-10.8)	5.59 (2-29.2)	0.08
Conversion to open	6 (38%) d	2 (7%) d	2 (10%)	
Appendectomy	3 (19%)	2 (7%)	2 (10%)	
Operative time	120 (60-160) ^a	100 (30-190)	90 (45-130) ^a	0.04
Peroperative complications	0	1 (3%)	1 (5%)	
Time full feed (days)	7 (3-27) ^{a,b}	4 (1-23) ^a	2.5 (1-8) ^b	0.0002
Length of admission (days)	9.5 (3-90) ^c	7 (1-426) ^a	3 (1-11) ^{a,c}	0.001
Redo surgery	0 (0)	3 (10%)	1 (5%)	

	Patient 1	Patient 2	Patient 3	Patient 4
Age at first surgery (days)	1	11	36	39
Presenting symptoms	Asymptomatic but heterotaxia	Bilious vomiting	Bilious vomiting	Reflux
Findings	Malrotation	Malrotation	Malrotation	Malrotation
Conversion	No	No	No	No
Age at redo (days)	13	119	55	189
Time surgery redo (days)	12	108	19	150
Time discharge redo (days)	4	100	N/A	142
Reason for redo	Bilious vomiting after discharge	Volvulus	Volvulus	volvulus
Surgical approach	Laparoscopic Ladd converted	Laparoscopic Ladd	Open Ladd	Laparoscopic Ladd converted