

Outcome of alimentary tract duplications operated on by minimally invasive surgery: a retrospective multicenter study by the GEICI (Groupe d'Etude en Coeliochirurgie Infantile)

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Abstract

Background Alimentary tract duplications (ATD) are a rare cause of intestinal obstruction in childhood. There are many case reports but few series about laparoscopy or thoracoscopy for ATD. The aim of our study was to report the outcome of minimally invasive surgery (MIS) for ATD.

This study was conducted on behalf of the GEICI (Groupe d'Etude en Coeliochirurgie Infantile). The members of this study group are listed in the appendix.

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Methods This was a retrospective multicenter study from the GEICI (Groupe d'Etude en Coeliochirurgie Infantile). We reviewed the charts of 114 patients operated on by MIS for ATD from 1994 to 2009.

Results Sixty-two patients (54 %) had a prenatal diagnosis. Forty-nine patients (43 %) were symptomatic before surgery: 33 of those patients (63 %) with postnatal diagnosis compared to 16 (25 %) with prenatal diagnosis ($P < 0.01$). In this last group, the median age at onset of symptoms was 16 days (range = 0–972). One hundred and two patients had laparoscopy (esophageal to rectal

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duplications) and 12 patients had thoracoscopy for esophageal duplications. The mean operative time was 90 min (range = 82–98). There were 32 (28 %) resection anastomoses, 55 (48 %) enucleations, and 27 (24 %) unroofings. The conversion rate was 32 %, and in a multivariate analysis, it was significantly higher, up to 41 % for patients weighing <10 kg ($P < 0.01$). Ten patients (8 %) had unintentional perioperative opening of the digestive tract during the dissection. Eight patients had nine postoperative complications, including six small bowel obstructions. The median length of hospital stay was 4 days (range = 1–21) without conversion and 6 days (range = 1–27) with conversion ($P = 0.01$). The median follow-up was 3 months (range = 1–120). Eighteen of the 27 patients who underwent partial surgery had an ultrasound examination during follow-up. Five (18 %) of them had macroscopic residue. **Conclusion** This study showed that MIS for ATD is feasible with a low rate of complications. Patients with prenatal diagnosis should have prompt surgery to prevent symptoms, despite a high rate of conversion in small infants.

Keywords Gastrointestinal tract · Duplication · Laparoscopy · Thoracoscopy · Prenatal diagnosis

Alimentary tract duplications (ATD) are uncommon congenital lesions that can occur anywhere from the mouth to

the anus and have a reported incidence of 1 in 4,500 [1]. Some ATD may be totally asymptomatic and discovered only incidentally. However, more than three quarters of ATD give rise to symptoms in the first 2 years of life, presenting as an abdominal mass, symptoms of intestinal obstruction, intussusceptions, or gastrointestinal bleeding [2]. Various malignant transformations in the mucosa of the duplication have been reported in adults [3, 4]. Therefore, complete surgical removal of the duplication is advised, but removal cannot always be radical as in large thoracoabdominal or long tubular duplications.

Prenatal ultrasonography (US) can lead to diagnosis of ATD in asymptomatic neonates. Early surgical correction in the neonatal period is advocated to avoid potential morbidity and mortality but it could increase surgically related morbidity in asymptomatic patients [5].

Advances in laparoscopy and thoracoscopy in small infants and neonates had led to the publication of short case series or reports of ATD treated successfully by minimally invasive surgery (MIS) [6–9]. However, large series assessing conversion rates and outcomes of MIS for ATD are lacking, especially in neonates [5, 7].

Our hypothesis is that prenatal diagnosis can influence the outcome of the patients operated on by MIS for ATD. The aim of this study was to assess the morbidity rate of patients with ATD operated on by thoracoscopy or laparoscopy and the factors that induce conversion or complications, and to compare the outcome of prenatal and postnatal diagnoses of ATD.

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Materials and methods

A retrospective multicenter study was conducted by the surgeons of the GECEI (Groupe d'Etude en Coelochirurgie Infantile). We reviewed the operative protocols, pathology reports, and hospitalization charts of children under 18 years of age who were operated on by thoracoscopy or laparoscopy for ATD from January 1994 to December 2009.

Definitions

Alimentary tract duplications were defined in pathology reports as a well-developed coat of smooth muscle, lined with alimentary tract epithelium, and attached to some part of the alimentary tract [10]. We excluded from the study bronchogenic cysts which are lined by respiratory epithelium and cartilage in the cyst wall. The prenatal diagnosis group comprised patients with a cystic intra-abdominal or intrathoracic formation discovered during prenatal US and for which postnatal diagnosis was an ATD. Symptomatic patients were defined by the appearance of symptoms

before surgery. Emergency surgery was defined by the need to perform surgery in symptomatic patients at admission or the day after a unplanned admission.

We defined three groups according to the procedures:

- *Full MIS group* patients with intracorporeal dissection and resection of the ATD, with extraction through a trocar site after the dissection had been completed.
- *Assisted MIS group* patients in which one trocar site had to be enlarged to extract the ATD in order to complete the dissection or resection extracorporeally, without an additional laparotomy.
- *Conversion group* patients for whom there was a need to perform an additional laparotomy or thoracotomy to complete the procedure.

The resection was complete if the duplication was removed either by resection anastomosis or total mucosal enucleation. The resection was incomplete if the duplication was unroofed, with or without mucosal coagulation.

For statistical comparisons of ATD location, we made two groups: small bowel (jejunum, ileum, ileocecal valve) versus other locations (esophagus, stomach, duodenum, colon, and rectum).

Variables of interest

We assessed demographic data, conversion rate, perioperative complications, type of resection (complete versus incomplete), peri- and postoperative complications, and residues at last follow-up.

Statistics

Qualitative data were reported as numbers (and percentages). Quantitative data with normal distribution were reported as means with confidence intervals and comparisons were performed with parametric tests (ANOVA, χ^2). Quantitative data without a normal distribution were reported as medians with extremes and comparisons were made with the Mann–Whitney test. For multivariate analyses we used the logistic regression test. Results with $P < 0.05$ were considered significant.

Results

Demographic data

Demographic data are summarized in Table 1 and the numbers of patients treated per center are displayed in Fig. 1. In seventy patients (61 %) the ATD was localized in the small bowel (Table 2). Twenty patients (17.5 %) had comorbidities, and among them 7 (6 %) had urologic anomalies and 2 (1.7 %) had vertebral anomalies.

Table 1 Demographic data

No. patients	114
No. centers	18
Median No. patients/center	5 (1–23)
No. operating surgeons	34
Sex ratio (M/F)	51/63
Median term at birth (weeks)	39 (30–42)
Median weight at birth (g)	3,200 (1,065–4,500)

Ranges are in parentheses

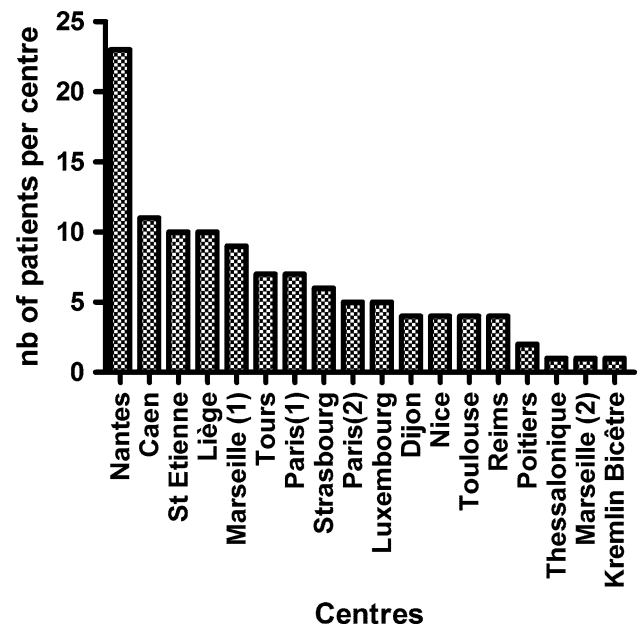


Fig. 1 Number of patients with alimentary tract duplication operated on by minimally invasive surgery per center from 1994 to 2009. Paris (1): Robert Debré Hospital, Paris; Paris (2): Trousseau Hospital, Paris; Marseille (1): La Timone Hospital, Marseille; Marseille (2): St. Joseph Hospital, Marseille

Prenatal diagnosis

Sixty-two patients (54 %) had a cystic image detected prenatally by ultrasound (US), including 40 in the second trimester of pregnancy. Duplication was mentioned as a diagnosis in 40 cases. The mean diameter of the duplication increased, not significantly, from 16 mm (range = 13–19) in the second trimester to 19 mm (range = 15–23) in the third trimester of pregnancy ($P = 0.3$).

Postnatal events

The main symptoms of ATD consisted of emesis/occlusion for 16 patients (34 %) and pain for 16 patients (34 %) (Table 2). Among patients with emesis/occlusion, one had intussusception and another had a volvulus (both for small

Table 2 Number of alimentary tract duplications, proportion of symptomatic patients, and emergency surgeries according to location

	Total	Type of symptoms (No. patients)	Emergency surgery (No. patients)
Esophagus	16	Dyspnea/cough (4) GERD/dysphagia (2) Fever, pain (1) Pain (1)	Respiratory distress (1)
Stomach	14	Pain (1)	Pain (1)
Duodenum	7	Occlusion/emesis (3)	Occlusion (3)
Jejunum-Ileum	47	Occlusion/emesis (6) Pain (10) Abdominal mass (5)	Occlusion (6) Pain (4) Increase in size (4) Abdominal trauma (1)
Ileocecal	23	Occlusion/emesis (6) Pain (3) Abdominal mass (1)	Occlusion (5)
Appendix	2	Fever, pain (1) Abdominal mass (1)	Suspicion of appendicitis (2)
Colon	4	Pain (2) Occlusion (1)	Pain (1) Occlusion (1)
Rectum	1	Constipation (1)	
Total	114	49	29

Table 3 Differences between pre- and postnatal diagnosis groups

	Prenatal (<i>n</i> = 62)	Postnatal (<i>n</i> = 52)	<i>P</i>
Symptoms	16	33	<0.01
Median age at symptoms (days)	16 (0–972)	1268 (0–3,202)	<0.01
Median age at diagnosis	NR	29 (0–157)	
Mean diameter of the duplications at diagnosis (mm)	30 (24–36)	35 (28–42)	0.3
Small bowel duplication	27	20	0.7
Median age at surgery (days)	102 (1–1,356)	1041 (6–4,872)	<0.01
Mean weight at surgery (kg)	6.4 (6–6.8)	16.8 (13.5–20.1)	<0.01
Mean diameter of the duplication at surgery (mm)	34 (26–42)	38 (31–45)	0.5
Emergency surgery (No)	12	17	0.1

NR not relevant

Ranges are in parentheses

bowel duplication). For the others, the cause was presumably lumen obstruction by the duplication. Differences between prenatal and postnatal groups are summarized in Table 3.

For patients with occlusion, the diameter of the duplication was the same as for patients without occlusion ($P = 0.8$). The preoperative imaging workup was made by US in 74 cases, CT scan in 25 cases, contrast studies in 9 cases, and MRI in 7 cases. ATD was mentioned as a preoperative diagnosis in 101 cases (88.5 %). The mean diameter of the duplication increased significantly from 32 mm at diagnosis to 36 mm (range = 30–42) before surgery ($P = 0.01$). Twenty-nine patients (25 %) underwent emergency surgery (Table 2). The mean diameter of the duplication was 46 mm (range = 36–56) in the emergency surgery cases versus 33 mm (range = 25–39) in

nonemergency surgery cases ($P = 0.04$). In a multivariate analysis comprising post/prenatal group, age at surgery (<1 year versus others), duplication size, and location (small bowel vs. others), only the postnatal diagnosis group was significantly associated with the risk of being symptomatic before surgery ($P < 0.01$).

Surgery

There were 102 laparoscopies and 12 thorascopies (7 right, 5 left). All the duplications, except one, were cystic. The two patients with multiple duplications were counted once because only one of the surgeries was performed by MIS. There were two thoracoabdominal duplications: one operated on by thoracoscopy for the thoracic component (the patient had had an earlier laparotomy for the abdominal

Table 4 Number and type of full, assisted, or converted MIS according to the location of the duplication

	Full MIS	Assisted MIS	Conversion
Esophagus	13	0	3
Stomach	12	1	1
Duodenum	4	0	3
Jejunum/ileum	7	23	17
Ileocecal	7	5	11
Appendix	2	0	0
Colon	3	0	1
Rectum	1	0	0
Total	49 (43 %)	29 (25 %)	36 (32 %)

Table 5 Conversion rates according to the presence of prenatal diagnosis, age, weight, emergency surgery, and location

	Conversion rates		<i>P</i>
	Yes (%)	No (%)	
Prenatal diagnosis	42	20	<0.01
Age < 1 year	40	19	<0.01
Weight < 10 kg	41	8	<0.01 multivariate
Emergency surgery	48	26	0.04
Small bowel/ileocecal	40	18	0.02

Only the weight was considered significant in multivariate analysis

component) and the other operated on entirely by laparoscopy. Two patients underwent perioperative endoscopy to facilitate the dissection of their duplication (colon and esophagus). The mean operative time was 90 min (range = 82–98) and did not differ statistically between thoracoscopy and laparoscopy ($P = 0.8$). Thirty-six patients (32 %) had conversion to an open procedure. The rates of conversion according to the location of the duplication are listed in Table 4. The factors affecting the conversion rate are listed in Table 5. In a multivariate analysis, only weight less than 10 kg was considered significant for a higher risk of conversion ($P < 0.01$). The reasons for conversion found in the operative charts are listed in Table 6. Among the 12 patients in whom dissection from the alimentary tract was considered difficult, 5 achieved successful enucleation rather

Table 6 Number and reasons for conversion found in operative charts

Difficulty separating the duplication from the digestive tract	12
Duplication hardly visible by MIS	8
Need for intestinal resection	7
Easier by an inguinal or Pfannenstiel incision	3
Dilation of the small bowel	2
Duplication size	2
Need for adhesiolysis	1
Esophageal perforation (before the procedure)	1

Table 7 Rate of radical surgery according to the location of the duplication

	Resection anastomosis	Enucleation	Unroofing
Esophagus	0	13	3
Stomach	0	9	5
Duodenum	0	2	5
Jejunum/ileum	26	19	2
Ileocecal	3	10	10
Appendix	2	0	0
Colon	1	1	2
Rectum	0	1	0
Total	32 (28 %)	55 (48 %)	27 (24 %)

than resection anastomosis or unroofing after conversion. The size of the duplication did not statistically affect the rate of conversion ($P = 0.1$). Twenty-nine patients (25 %) had an assisted MIS. All but one (stomach) were for small bowel duplications. Among them, four patients had a laparoscopic emptying of the duplication to facilitate the extraction through the umbilical trocar site. Eighty-seven patients (76 %) underwent radical surgery: 32 resection anastomoses and 55 enucleations. Among the 27 patients (23 %) with partial surgery, 18 had coagulation of the remaining mucosa, and for the other 9 patients the mucosa was left in place.

The rate of radical surgery was 83 % if the duplication was in the small bowel compared to 66 % for other locations ($P = 0.04$) (Table 7). Radical surgery of the duplication was achieved in 83 % of the 36 patients with a conversion and in 73 % of the 78 patients without conversion ($P = 0.1$).

Outcome

The peri- and postoperative complications are displayed in Table 8. Eight patients (7 %) had nine postoperative

Table 8 Outcome, type, and number of patients with peri- and postoperative complications

Perioperative	
Unintentional opening of digestive tract	10
Before conversion	5
After conversion	5
Postoperative < 30 days	
Occlusion	5 ^a
Trocar cellulitis	1 (colonic)
Chylothorax	1 (thoracoscopy)
Gastroparesis	1
Late complications: 2 months (30 days–4 years)	
Small bowel obstruction managed conservatively	4

^a One patient had a surgical band adhesiolysis

Table 9 Patients with small bowel adhesions, according to the type of surgery, type of resection, and the perioperative events

Location	Opening digestive tract	Type of MIS	Type of resection	Need for adhesiolysis
Esophagus	No	Full Previous laparotomy	Enucleation	No
Duodenum	Yes	Full	Unroofing	No
Ileum	No	Assisted	Enucleation	Yes
Ileum	No	Conversion	Resection/anastomosis	No
Ileum	No	Conversion	Enucleation	No

Table 10 Presence of ectopic mucosa according to the location of the duplication

Location	Ectopic mucosa	No ectopic mucosa
Esophagus	3 ^a	13
Stomach	0	14
Duodenum	3	4
Small bowel	26 ^{a,b,c}	21
Ileocecal	15	8
Appendix	0	2
Colon	0	4
Rectum	0	1

^a One small bowel and one esophageal duplication had respiratory ectopic mucosa

^b One small bowel ATD had pancreatic ectopic mucosa

^c One small bowel ATD had colonic ectopic mucosa

complications, which are listed in Tables 8 and 9. The rate of complications did not statistically differ according to the presence of a conversion, complete resection of the duplication, age at surgery, and emergency ($P > 0.1$). Only one small bowel obstruction occurred after a full MIS. The rates of preoperative and postoperative adverse events were not different between thoracoscopic procedures (25 %) and laparoscopic procedures (14 %) ($P = 0.4$). The median length of hospital stay was 3 days (range = 1–21) for those without conversion compared to 6 days (range = 1–27) for those who had conversion ($P < 0.01$) and was not different between thoracoscopy and laparoscopy patients ($P = 0.4$).

Pathology

An ectopic mucosa was found in 47 cases (41 %) of pathologic resection specimens. The rate of ectopic mucosa according to the location of the duplication is listed in Table 10. Ectopic mucosa was found in 9 of the 27 patients (33 %) with partial resection and in 38 of the 87 patients (46 %) with complete resection ($P = 0.5$).

Follow-up

Fifteen patients (13 %) were lost to follow-up. The median follow-up was 3 months (range = 1–120). Thirty-seven

patients (32 %) had an US during follow-up. Eighteen of the 27 patients (66 %) who underwent partial surgery had an US during follow-up. Five of them (18 %) had a residue of 15 mm (range = 10–25) and two had ectopic mucosa.

Discussion

This study showed that MIS for ATD is feasible and safe with a low rate of adverse events. This is, to date, the largest published study about MIS for ATD. In our study, only one of our centers had more than 12 patients in 15 years (Fig. 1), which explains why reports on large series of MIS for ATD is scarce [5, 7–9, 11]. The latest two-center large study of 73 patients with ATD was published in 2003, but none of the patients underwent a minimally invasive procedure [11]. In our study, the distribution of the ATDs along the digestive tract is comparable to already published series of ATD [2, 11], showing that the location of the ATD did not influence the choice of MIS over open surgery. Although thoracoscopy obviously differs from laparoscopy, with specific adverse events (e.g., chylothorax), we wanted to include the outcome of thoracoscopy for thoracic esophageal duplications as part of MIS. Moreover, mean operative time, length of hospital stay, and the number of adverse events in the cases of thoracoscopy were not different from those of laparoscopy, and thoracoscopic resection results in a shorter hospital stay than thoracotomy as previously found by other studies [8, 12]. In our study, the rate of prenatally detected cases was higher than that in published series, and our study is the first that tries to address both the symptoms and the outcome in this group of patients [2, 7, 11–12]. Prenatal ultrasonographic features of ATD have been identified to improve diagnostic accuracy. These include intra-abdominal or intrathoracic cystic mass detected in the second trimester of gestation (unlike ovarian cysts, which appear at the third trimester), with a hyperechoic mucosa surrounded by hypoechoic muscular layers, and the presence of debris within the lesion [7] (Fig. 2).

In our study, the prenatal diagnosis group had significantly fewer symptoms than the postnatal group. This



Fig. 2 Gastric duplication in a fetus, at 26 weeks of gestational age, presenting as a cystic mass adjacent to the stomach (S), with hyperechoic mucosa surrounded by hypoechoic muscular layer (*white arrow*)

shows that the ability to accurately diagnose ATD prenatally has provided an opportunity to treat these lesions before they produce symptoms or complications. On the other hand, one quarter of prenatally detected patients had symptoms before surgery, which occurred from birth to almost 3 years after prenatal detection. Moreover, half of symptomatic patients in the prenatal group had symptoms before 1 month of age. In our series, the median age at surgery for the prenatal group was almost 3 months. In a recent series published by Laje et al. [7], although the rate of symptomatic patients was the same as ours, the age at surgery was 1 month. Moreover, the number of emergency surgeries was the same in the prenatal group as in the postnatal group, which shows that the benefit of prenatal diagnosis can be lost by waiting too long before operating on asymptomatic patients. An interesting finding not found in other series with prenatal diagnosis [5, 7, 12] is that the duplication sizes in prenatal and postnatal groups were the same. This may be attributed to a faster growth pattern in the prenatal group, which is why, despite generalized and systematic prenatal ultrasound in France and Western Europe, some duplications are detected prenatally and others later in infancy. The fast growth in the prenatal group could explain why the emergency rate is the same in the prenatal and postnatal groups in a multivariate analysis. Moreover, a 3-cm-diameter duplication may lead to occlusions more often in neonates than in older children. All these results and hypotheses indicate that there was no benefit to waiting longer than 1 month before surgery for prenatally detected patients.

This study is the first one to report and analyze the rate of conversion in MIS for ATD. The rate of conversion was significantly higher among patients who weighed less than

10 kg and was not reported in other studies about ATD [7, 8, 11]. This is in contrast with other publications that show that complex endoscopic procedures in small neonates are safe and feasible [6, 13]. The high rate of conversion in small infants could reflect the lack of working space to clearly identify or dissect the duplication from the normal alimentary tract. Moreover, in small infants or neonates, there is the opportunity to perform a small and more cosmetic incision by laparotomy. Other factors of conversion included emergency surgery and location of the ATD in the small bowel but they were not significant in multivariate analysis (Table 5). Hence, 42 % of patients with prenatal diagnosis underwent conversion to an open procedure, although they had an earlier diagnosis and they were less symptomatic than postnatal diagnosis patients. However, we still believe that MIS should be attempted first in this subgroup, because almost two thirds of these patients could benefit from the advantages of MIS (less postoperative bowel obstruction, pain, and scarring) over open procedures. These statements should be confirmed by prospective studies comparing MIS with open surgery for ATD. Moreover, in our series, conversion did not provide a higher rate of complete resection and the length of hospital stay was longer than in assisted or full MIS patients.

Although this study showed some variation in surgical technique used to treat ATD (use of endoscopy to dissect the duplication, or emptying the duplication first to facilitate its extraction), almost all centers used assisted MIS for small bowel duplication (Table 4). In this location, assisted MIS was preferred because small bowel can be easily extracted through an umbilical or a trocar incision, whereas it is not possible when the ATD is located in the nonmobile alimentary tract (esophageal or duodenal duplications). Thus, assisted MIS for small bowel duplication could be a good alternative if complete resection is not possible by full MIS (Fig. 3).

Partial resections can be justified for duodenal or esophageal ATD in order to decrease the morbidity of a complete resection (Table 7). Moreover, ectopic mucosa is rarely found in these locations (Table 10). Conversely, given the high rate of ectopic mucosa in small bowel duplications, partial resection in these locations is not recommended. A good alternative could be a resection anastomosis or total enucleation through assisted MIS. Complete resection rates were not different between conversions and full or assisted MIS. This result tends to show that the choice of MIS over an open procedure might not affect the rate of complete resection. Perioperative complications were minor: only one opening of the digestive tract could have led to a further small bowel adhesion. For the other five patients with postoperative small bowel adhesions, we could implicate either assisted MIS or conversion (Table 9) and most of the patients were treated without additional surgery.

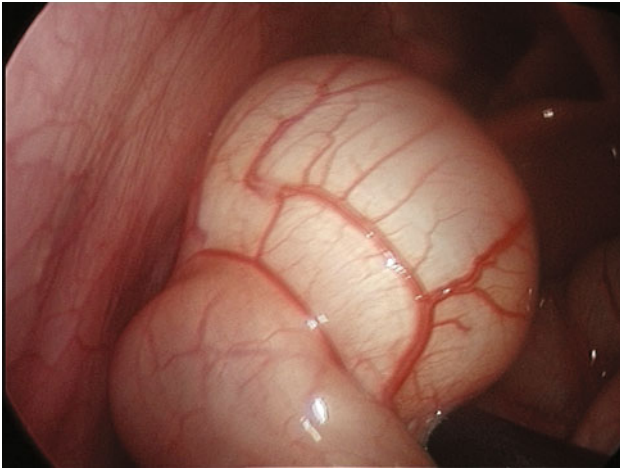


Fig. 3 Laparoscopic view of asymptomatic and uncomplicated ileal duplication in an 11-month-old boy, who had to be treated by an assisted laparoscopy to complete enucleation

The rate and distribution of ectopic mucosa are consistent with those of other series of ATD [2, 5]. The high rate of ectopic mucosa in small bowel duplications reinforces the need to resect completely the mucosa in these locations. If total resection is not possible without morbidity (long tubular duplications, thoracoabdominal, esophageal, or duodenal ATD), sequential US follow-up of incomplete resection is mandatory because of the risk of malignancy in adulthood. In our study, 23 % of our patients had incomplete resection, and among them one third had ectopic mucosa. Only two thirds of patients with incomplete resection had imaging during their follow-up, which was inadequate. For 18 % of patients who had an US evaluation, macroscopic residue was present and required a close follow-up given the risk of malignancy in adulthood.

This study is the largest attempt to focus on MIS for ATD, and it provides reliable conversion rates and addresses the outcome of prenatally diagnosed patients with ATD. Although this study is retrospective, it shows that MIS can be the preferred approach for ATD because of low morbidity and a short length of hospital stay. Surgeons and parents should be aware that the risk of conversion is higher in small infants or neonates. Whenever it is possible, and especially for small bowel duplication, resection should be complete; therefore, assisted MIS offers a good alternative in this location.

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Philippe Montupet, Martine Demarche for the GECI (Groupe d'Etude en Coeliochirurgie Infantile) have no conflicts of interest or financial ties to disclose.

Appendix: GECI (Groupe d'Etude en Coeliochirurgie Infantile)

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