Role of mini-invasive surgery in the treatment of enteric duplications in paediatric age: a survey of 15 years.

Il ruolo della chirurgia mini-invasiva nel trattamento delle duplicazioni intestinali: l'esperienza di 15 anni

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Key words: enteric duplications, laparoscopic surgery, intestinal duplications.

Abstract

Background: Gastrointestinal tract duplications (ATD) are rare malformations that occur with an incidence of 1 in 4000-5000 live births, with heterogeneous clinical pictures in relation to the different location and size. The purpose of this study was to analyze the role of minimally invasive surgery in the management of ATDs, through a critical analysys of 15 years of experience of the Department of Paediatric Surgery, University of Bologna.

Methods: The medical records of 22 children diagnosed with ATDs were retrospectively reviewed (January 1995–August 2010). The study analyses: clinical presentation, preoperative diagnosis, site, anatomic type, treatment, and outcome.

Results: Children were 16 males and 6 females, with age ranged from 1 day to 10 years. 20 ATDs (91%) were cystic type, while 2 cases (9%) were tubular one. During the first period of our experience, 10 (45.5%) cases were approached with an open surgery. Then subsequent 8 (36.4%) cases were treated with a diagnostic laparoscopy. This approach permitted also to perform a minilaparotomy, close to the site of the malformation, with a short length of the scar. In 2 cases (9%), we realized an ileal resection with end-to-end anastomosis with a trans-umbilical video-assisted procedure.

In 2 cases (9%), we performed a complete removal of the lesions, after complete ligation of the vascular pedicle through a laparoscopic approach.

10 cases (45.5%) were located in the ileum, 6 cases (27.3%) were esophageal duplication, 3 cases (13.6%) were gastric duplications, 2 cases (9%) were located in the colon-rectum.

The postoperative course was uneventful in all the cases.

Discussions and conclusions: this study shows how, in the management of intestinal duplications, a mininvasive approach, is increasingly taking the field, along with increasing the "learning curve" with laparoscopy. In experienced hands, the laparoscopic approach allows an accurate definition of the exact site of duplication and a miniinvasive treatment with similar principles of open techniques.

Introduction

Alimentary tract duplications (ATDs) are rare anomalies. They frequently present a diagnostic challenge because they often mimic other disease entities at presentation, even after radiological investigation (1). Often they are discovered incidentally either prenatally or postnatally (2). Therapy continues to evolve thanks to minimally invasive surgical techniques (Schleef and Schalamon 2000) (1). The initial aim of laparoscopy is to locate the cyst exactly and to obtain a better idea of its nature. The procedure may then be continued as a therapeutic laparoscopic procedure in which the cyst is removed with a totally laparoscopic approach or with a video-assisted technique. We report the experience of the Department of Paediatric Surgery, University of Bologna, in the treatment of ATDs in order to study the role of mini-invasive surgery in the management of these kinds of malformations.

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Methods

The medical records of 22 children diagnosed with ATDs were retrospectively reviewed (January 1995–August 2010). Children were 16 males and 6 females, with age ranged from 1 day to 10 years. Releant data were: clinical presentation, preop diagnosis, localization, anatomic type, treatment and outcome.

Results

Table 1 shows the clinical characteristics of our patients:

- 5 cases (22.7%) were antenatally diagnosed by an ultrasound evaluation, followed by a fetal magnetic resonance imaging (f-MRI) in two cases.
- 5 cases (22.7%) presented with recurrent abdominal pain, while 2 (9%) cases presented with acute abdominal pain, a small bowel occlusion and a hemoperitoneum respectively. In 6 (27.3%) cases dysphagia in relation to the ingestion of solid food was the main complaint.

In one case we detected a colo-rectal duplications in a baby with an history of recurrent episodes of fever with recurrent abdominal pain. The other 2 (9%) cases of colo-rectal duplication presented episodes of rectal bleeding.

In all patients, an US examination of the abdomen was performed; 4 (18.2%) patients had a chest X-ray, 4 (18.2%) patients had an abdominal MRI, 12 (54.5%) patients performed an upper-GI series and 3 (13.6%) barium enemas. In 3 (13.6%) cases, a flexible esophagoscopy was performed and in 1 (4.5%) case a colonoscopy.

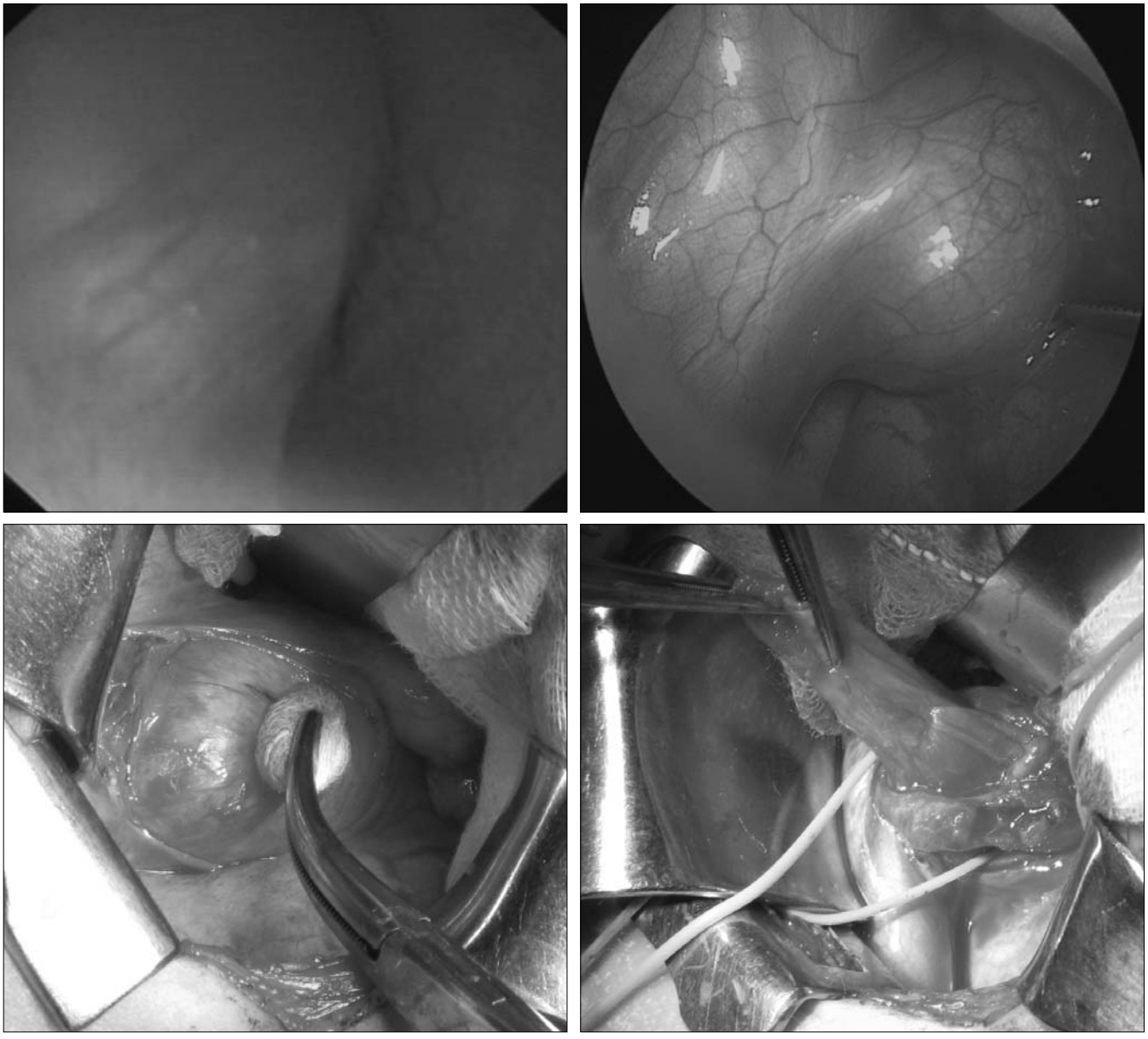

Figure 1.
Combined esofagoscopy, thoracoscopy and thoracotomy for excision of an esophageal duplication.

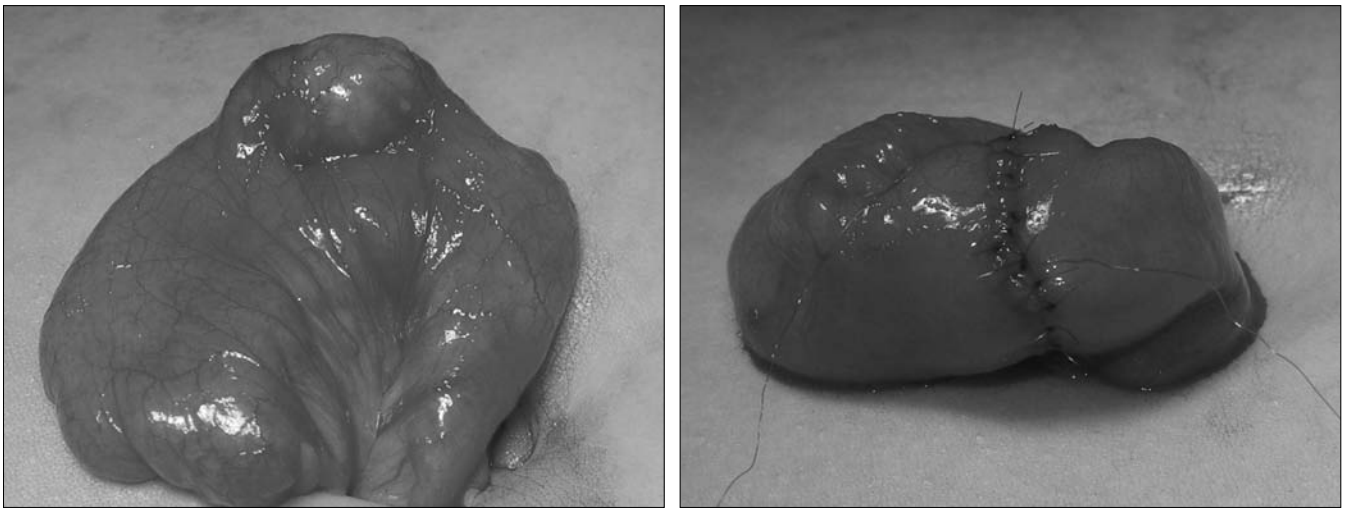

Figure 2.

Transumbilical videoassisted resection of ileal duplications with subsequent anastomosis.

20 ATDs (91%) were of the cystic type, while 2 (9%) cases were tubular.

During the first period of our experience, 10 (45.5%) cases were approached with an open surgery. Then, subsequent 8 (36.4%) cases were treated with a diagnostic laparoscopy. This approach permitted also to perform a minilaparotomy, directly close to the site of the malformation, with a shorter length of the scar (Figure 1). In 2 cases (9%), we performed an ileal resection with end-to-end anastomosis with a trans-umbilical video-assisted procedure (Figure 2). In 2 cases (9%), a complete removal of the lesions was done, after ligation of the vascular pedicle completely by a laparoscopic approach (Figure 3). As showed in the Figure 4, the evolution of the surgical treatment was characterized from the absence of only open surgery in the last five years.

Table 1

CHARACTERISTICS OF THE STUDY POPULATION.

Patient	Age at the treatment	Localization	Clinical presentation	Diagnostic exams	Treatment	Year of treatment
1	5 ys	Jejuno-ileal	Rettorragy ES + CS + Barium enema + Videocapsule	Abdo-US + Upper-GI X-Ray +	VLS + laparotomy	2001
2	4 ys	Esophageal	Dysphagia Chest X-Ray	Abdo-US + Upper-GI X-Ray +	TS + thoracotomy	2003
3	2 mm	Jejuno-ileal	Prenatal diagnosis Upper-GI X-Ray	Abdo-US + Barium enema +	Laparotomy	2004
4	10 ys	Jejuno-ileal	Recurrent abdominal pain	Abdo-US	TUARA	2006
5	1 ys	Gastric	Prenatal diagnosis	Abdo-US + Upper-GI X-Ray + ES	VLS + laparotomy	2004
6	1 ys	Esophageal	Dysphagia Chest X-Ray	Abdo-US + Upper-GI X-Ray +	Thoracotomy	1995
7	10 ys	Colo-rectal	Rettoragy	Abdo-US + Barium enema	Laparotomy	1995
8	5 ds	Colorectal	Rettoragy	Abdo-US	Laparotomy	2005
9	5 ys	Jejuno-ileal	Small bowel occlusion	Abdo-US + plain abdo- X-Ray	Laparotomy	1996
10	2 ys	Jejuno-ileal	Recurrent abdominal pain	Abdo-US	VLS + Mini-laparotomy	1998
11	1 ys	Jejuno-ileal	Recurrent abdominal pain	Abdo-US	VLS + Mini-laparotomy	1998
12	5 mm	Esophageal	Prenatal diagnosis MRI + Chest X-Ray	Abdo-US + Upper-GI X-Ray +	TS + thoracotomy	2009
13	2 ys	Colo-rectal abdominal pain	Recurrent fever and	Abdo-US + MRI	VLS	2010
14	4 ys	Gastric	Recurrent abdominal pain	Abdo-US + MRI	VLS + laparotomy	2000
15	3 ys	Jejuno-ileal	Dysphagia	Abdo-US + Upper-GI X-Ray	TUARA	2007
16	8 ys	Duodenal	Dysphagia	Abdo-US + Upper-GI X-Ray	Laparotomy	1998
17	10 ys	Duodenal	Dysphagia	Abdo-US + Upper-GI X-Ray	Laparotomy	1995
18	1 ys	Gastric	Prenatal diagnosis ES + MRI	Abdo-US + Upper-GI X-Ray +	VLS	2010
19	10 ds	Esophageal	Polidramnios Prental diagnosis ES + Chest X-Ray	Abdo-US + Upper-GI X-Ray +	TS + thoracotomy	2010
20	3 ys	Jejuno-ileal	Recurrent abdominal pain	Abdo-US + MRI	Laparotomy	1997
21	3 ys	Esophageal	Dysphagia Chest X-Ray	Abdo-US + Upper-GI X-Ray +	Thoracotomy	1998
22	6 ys	Jejuno-ileal	Hemoperitoneum	Abdo-US	Laparotomy	1995

Es: esophagoscopy, Cs: colonoscopy, Vls: videolaparoscopy, Ts: thoracoscopy, TUARA: transumbilical assisted resection and anastomosis

10 cases (45.5%) were located in the ileum, 6 cases (27.3%) were esophageal duplication, 3 cases (13.6%) were gastric duplications, 2 cases (9%) were located in the colon-rectum.

Ileal and caecal duplications were treated by resection of the involved bowel with subsequent primary anastomosis. Gastric and esophageal duplications were treated by enucleation of the cyst.

The postoperative course was uneventful in all the cases. One patient with ileal duplication was readmitted was complicated, in the postoperative time, by several episodes of small bowel bacterial overgrowth, treated by medical therapy.

Discussion

ATDs are rare anomalies.[1] The most commonly affected site is the small intestine (47% of all cases), followed, in order, by the large intestine (20%), esophagus (17%), stomach (8%), and duodenum (5%).[3]

The origin of duplications is uncertain. Ladd coined the term alimentary tract duplications in 1937, which more effectively described the clinical and pathologic aspects of these lesions.[4] In 1944, Bremer suggested that duplications were the result of errors of canalization.[5] Because the stomach lumen does not go through a period of occlusion and recanalization, he suggested the adherence of longitudinal folds. McLetchie's theory of a neurenteric band is based on the observation that during the third week of life, the primitive gut is developing and anomalies may occur in the separation of the notochord, resulting in a diverticulum of the foregut.[6]

Macroscopically, they are spherical cysts or tubular structures located in, or immediately adjacent to, part of the gastrointestinal tract. Microscopically, alimentary tract duplications contain smooth muscle in their walls and are lined with alimentary tract mucosa. The lining mucosa, however, is not necessarily that of the adjacent segment of the gastrointestinal tract. Ectopic tissue, such as gastric, squamous, transitional, and ciliated mucosa, pancreas, and ganglion cells, can be found in its wall.[7] Malignant changes are a rare complication of alimentary tract duplications in adults.[8]

They frequently present a diagnostic challenge because they often mimic other disease entities at presentation, even after radiological investigation. Often they are discovered incidentally either prenatally or postnatally.[2] Therapy continues to evolve thanks to minimally invasive surgical techniques.[1] The initial aim of laparoscopy is

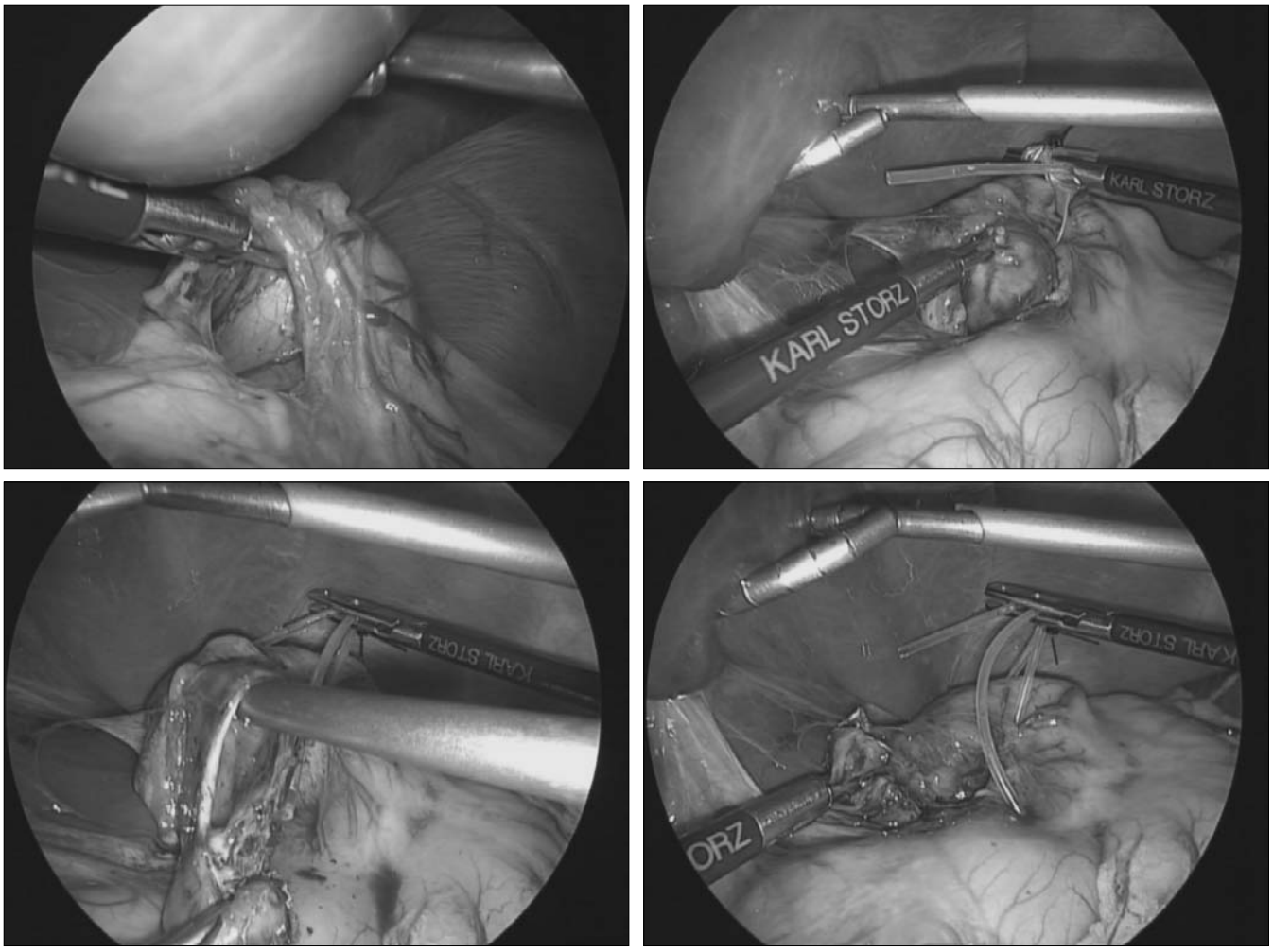

Figure 3.

Laparoscopic enucleation of a gastric duplication.

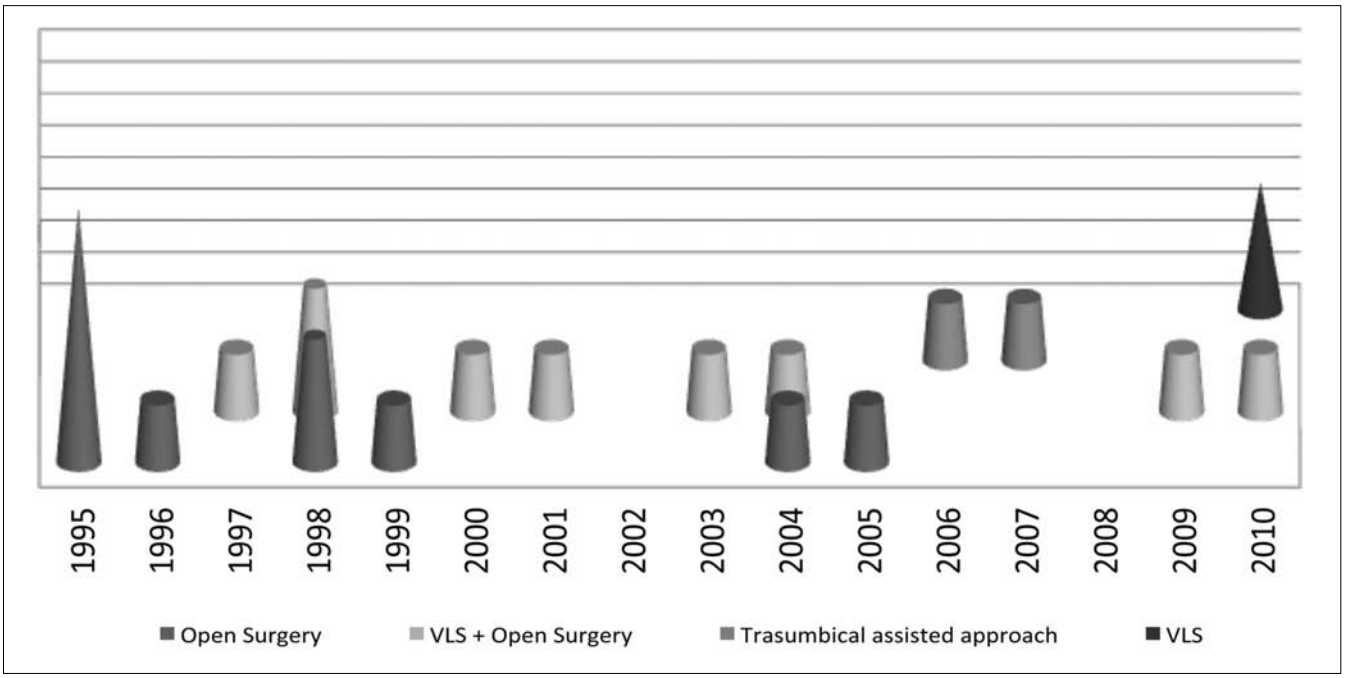

Figure 2.

Transumbilical videoassisted resection of ileal duplications with subsequent anastomosis.

to locate the cyst exactly and to obtain a better idea of its nature. The procedure may then be continued as a therapeutic laparoscopy to remove the cyst.

There appears to be a discrete increase in the incidence of this type of pathology due to an increased detection by ultrasound, and especially by prenatal ultrasound. When a prenatal diagnosis of an intra-abdominal cyst is made, the operation is best scheduled in the second half of the first year.[1] Intra-abdominal cysts should not be left in place as they can give rise to potentially lethal complications.[2]

A topographical diagnosis is important before deciding how to remove the lesion. Computed tomography (CT) or magnetic resonance imaging (MRI) are more helpful in determining the exact position of the lesion than in diagnosing the exact nature of it.[9] Laparoscopy not only confirms the diagnosis, but also identifies the exact localization.[10]

This diagnostic procedure may be extended into a therapeutic one according to the surgeon's experience.[9,10] Depending on the localization and nature of the cyst, many surgical approaches are possible. Complete removal is the approach of choice, but is not always possible without damaging adjacent organs. Enucleation of the cyst may endanger the blood supply of other organs, which is the reason why the lesion is often resected together with the adjacent bowel during open surgery.[11] Laparoscopy, in contrast, gives a much better view of the vascularization of the lesion and adjacent organ allowing for resection of the lesion only without jeopardizing the blood supply of the remaining organ. Laparoscopic enucleation is therefore almost always possible with minimal blood loss.[12]

However, when the tension in the cyst decreases the plane of separation is less easy to find.[13]

Sometimes, however, removal of enteric duplications or other abdominal cysts may require the closure of an accidentally opened bowel, or resection of the lesion together with a tract of bowel with subsequent anastomosis.[12]

In the past, for these reasons we treated these lesions directly by an open surgery. As the laparoscopic learning curve grows, we began to perform a laparoscopic approach in order to better focus the entity of the problem. Then the resection of the lesion was done outside through a minilaparotomy, first, and then laparoscopically inside the abdomen.

If a laparotomy is required, a much smaller incision will suffice when compared to a laparotomy without prior laparoscopic exploration and/or dissection.[14] Endolinear staplers may be useful but we are concerned about leaving metallic clips in a child's abdomen for a lifetime.

According to our experience it is probably better to perform a transumbilical videoassisted procedure, which many authors consider an evolution of the totally laparoscopic approach. Transumbilical approach, in fact, allows an easy access to entire small bowel, especially in the newborn, and permit to perform a safe bowel resection with a subsequent anastomosis under direct vision, outside the peritoneum. The advantages of the totally laparoscopic approach are present in the video-assisted technique, without impeding the effectiveness of the planned operation.

Anyway, we reported the evolution of the treatment in our population, showed in the Figure 4, and we noted that the distribution of the surgical treatment was characterized by the absence of only open surgery in the last five years; this fact, the increasing of the laparoscopic learning curve and the development of mini-invasive surgical approaches, as the transumbilical videoassited technique, show the real role of mini-invasive surgery in the management of ATDs.

Conclusion

In conclusion, ATDs are increasingly detected by ultrasound both postnatally as well as antenatally. The exact nature is impossible to define without pathological examination. Preoperative ultrasound is useful for the identification of a cyst, but MRI and CT scan are necessary for topographical localization. Possible complications of such cysts warrants their excision. Laparoscopy gives an exact topographical diagnosis and often allows enucleation with preservation of the remaining organ. If a laparotomy is required, a much smaller incision will suffice when compared to a laparotomy without prior laparoscopic exploration and/or dissection.

Finally, the development of the transumbilical videoassisted surgery gives the possibility to perform the resection of the lesion together with a tract of bowel and subsequent anastomosis directly through the umbilicus, with good esthetic results.

Our experience shows as, nowadays, laparoscopic approach seems to have a diagnostic and therapeutic role in the management of ATDs.

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