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TOPIC HIGHLIGHT

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Alimentary tract duplications in newborns and children: Diagnostic aspects and the role of laparoscopic treatment

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Abstract

Alimentary tract duplications are rare congenital lesions normally diagnosed in newborns and children that can occur anywhere from the mouth to the anus and have a reported incidence of approximately 1 in 4500 life births. Symptoms and clinical presentation vary greatly. The presentation varies according to age and location. The treatment finally is surgical; total resection when possible should be the aim of the intervention. In pediatric surgery minimally invasive surgical procedures became more and more important over the last decades. In consequence the operative procedure on alimentary tract duplications changed in this manner. We review on case reports and clinical reports on minimally invasive surgery in the treatment of alimentary tract duplications, determine the importance of minimally invasive techniques in the treatment of this rare entity and rule out that further studies in the field should be performed.

Key words: Alimentary tract duplications; Gastrointestinal duplication cysts; Laparoscopic surgery; Minimally invasive surgery; Endoscopy

Core tip: Alimentary tract duplications are rare congenital lesions which treatment is surgical. In the last two decades minimally invasive surgery became very important in the field of pediatric abdominal surgery. With this review we want to give the reader an overview on recent literature reporting on minimally invasive surgery in alimentary tract duplications.

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INTRODUCTION

Alimentary tract duplications are rare congenital lesions normally diagnosed in newborns and children (although in adulthood it occurs with the same frequency). They can occur anywhere from the mouth to the anus with a reported incidence of approximately 1 in 4500 life births[1,2]. The symptoms and clinical presentation vary greatly depending on size, location, presence of gastric mucosa, and communication with the normal bowel. They may present with intestinal obstruction, abdominal pain, intestinal bleeding and intussusception. They often require surgical intervention in the neonatal, infant or older child[1,3]. About two thirds of these duplications are in the abdominal cavity, and more than half are in the jejuno-ileal segments[4]. Affected sites are the thorax or thoraco-abdomen (4%)[5], the foregut (extremely rare)[6], the gastrum (7%)[7], duodenum (5%-7%)[8,9], the pylorus (extremely rare)[10], biliary tract (extremely rare)[11],

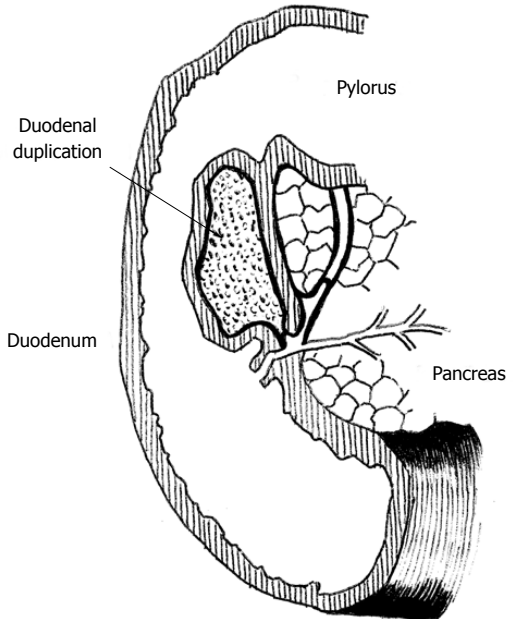

Figure 1 Duodenal duplication cyst (scheme).

the small intestine (44%)[7,12-14] the appendix (extremely rare)[15] and the colon (15%)[16]. Associated anomalies are frequently present in patients with alimentary tract duplications[12-18].

Several hypotheses about their etiology had been proposed, although no single hypothesis can explain all possible combinations of duplications, locations and associated anomalies[4]. Duplications occur before the 12th week of gestation[19]. The luminal recanalization theory explains duplications in those portions of the gastrointestinal tract that go through a "solid stage", including the esophagus, small bowel, and colon[20]. In the 5th to 6th week of life, the foregut is covered by cells similar to those in the respiratory tract. This lining grows and obliterates the lumen and later produces secretions that form vacuoles in the intercellular space. These vacuoles line up and eventually coalesce to form a new lumen. If some vacuoles failed to coalesce along this longitudinal axis, a cyst forms and becomes surrounded by the muscular layers[19]. This theory doesn't explain duplications at other levels.

The intrauterine vascular accident theory suggests that, like the small-bowel atresias, gastrointestinal duplications arise from an intrauterine vascular accident during early fetal development and may be a valid explanation for small-bowel atresias and, perhaps, their associated duplications[21].

The abortive twinning theory proposes that gastrointestinal tract duplications represent incomplete twinning[22]. This could explain the colorectal tubular duplications or duplications of the hindgut that are associated with genitourinary malformations[18].

By definition enteric duplications have an intimate attachment to some portion of the alimentary tract and they share a blood supply with the intestine[2,3] (Figure 1). The definitive diagnosis is confirmed by histological examination. They have a well-developed coat of smooth muscle and they have an epithelial lining representing some portion of the alimentary tract (usually the mucosa native to the lesion), but ectopic tissue is present in approximately 35% of specimens[12]. Gastric mucosa is the most common type, *i.e.*, 15% of duplications contain ectopic gastric mucosa followed by pancreatic tissue[3,7], which predisposes the patient to ulceration that can progress to haemorrhage and perforation[23]. They may extend into the liver or even trans-diaphragmatically[24]. These are generally diagnosed after onset of high intestinal obstruction or hemorrhage that may be accompanied by icterus or pancreatitis[13].

TYPES OF DUPLICATIONS

Three quarters of gastrointestinal duplications are cystic with no communication to the adjacent alimentary tract while the remaining are tubular, and may communicate with the intestinal lumen[25].

Thoracic and thoracoabdominal duplications

Duplications of the esophagus are the second most common duplication of the gastrointestinal tract after the ileal duplications. They account for 15%-21% of all reported duplications, whereas thoracoabdominal lesions are rare (approximately 2%). Thoracic duplications appear to be more common on the right side and have the highest incidence of associated vertebral anomalies, including anterior or posterior spina bifida, hemivertebrae, or myelomeningocele. They are twice as common in male patients, and typically present in neonates and infants[1,4,26].

The preferred treatment for both types of duplications is complete excision. These are generally straightforward for the non-communicating type, but can be technically challenging in the more complex lesions.

Thoracoabdominal lesions communicate with the intestinal tract below the diaphragm, most often through the esophageal hiatus and generally connect to the duodenum or jejunum after passing behind the stomach and pancreas. The likely presentation in an infant or neonate is respiratory distress as a result of accumulation of fluid and debris within the cyst[1,4,26].

Gastric and duodenal duplications

Duplications of the stomach are mostly non-communicating, cystic and typically located along the greater curvature[13,27] (Figure 2A). They are more common in boys than girls, and they typically present early in the first year of life with vomiting, poor feeding and weight gain, or abdominal distension. Peptic ulceration may lead to painless gastrointestinal haemorrhage and melena; it can progress to perforation. The treatment of gastric duplications is resection to prevent future gastrointestinal bleeding. Partial gastrectomy is often required due the size of the duplication[28].

Duodenal duplications are located in the medial and

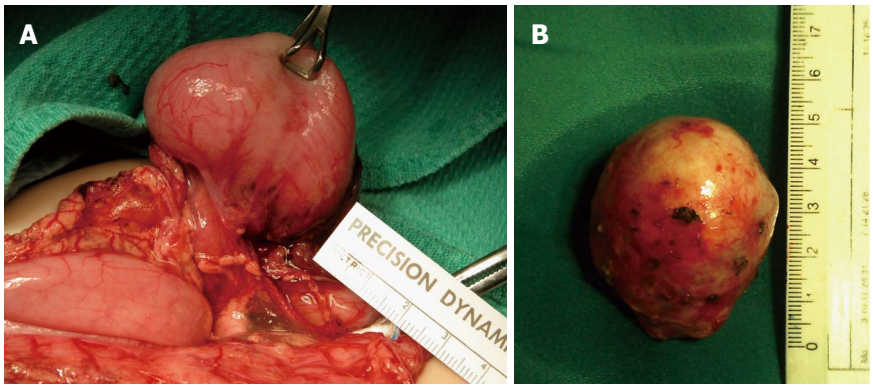

Figure 2 Gastric and duodenal duplications. A: Isolated gastric duplication cyst; B: Duodenal duplication cyst after being shelled out laparoscopically in a 6-year-old girl.

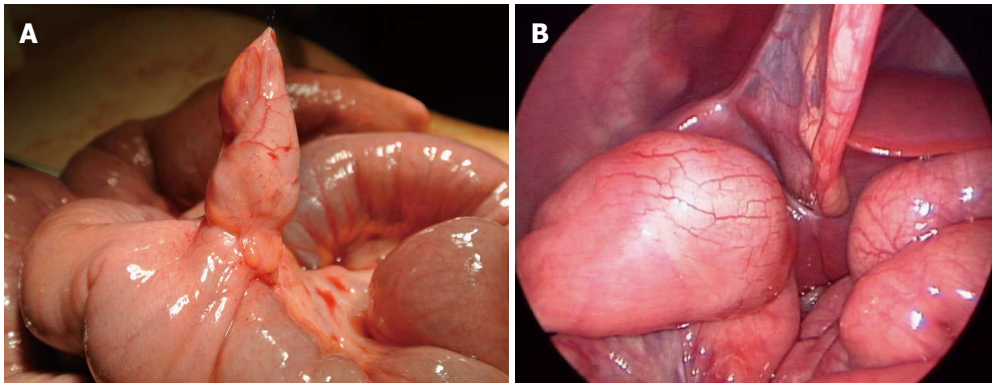

Figure 3 Small intestine duplications. A: Ileal duplication cyst (containing heterotopic mucosa causing acute intestinal bleeding in 13-year-old girl); B: Ileal duplication cyst embedded in the muscle layer wall removed laparoscopically.

posterior aspect of the second and third portions of the duodenum, normally non-communicating to the gastrointestinal tract[8,9] (Figure 2B). They may present with biliary (*e.g.*, jaundice) or pancreatic (*e.g.*, pancreatitis) symptoms. Symptoms are typically vague, consisting of upper abdominal pain, failure to thrive or early satiety. These duplications may be confused with choledochal cysts and should always be considered in the differential diagnosis. Figure 1 displays a scheme of a cystic type duodenal duplication (Figure 1).

The preferred treatment for duodenal duplications is complete excision with the attached segment of duodenum, but this is rarely possible because of proximity to the ampulla of Vater and the potential for injury to/of the biliary tree and pancreatic duct. Marsupialization or internal drainage to the duodenum or jejunum (Roux-en-Y) are adequate alternatives provided the duplication cyst does not contain gastric mucosa[9].

Gallblader duplications

Congenital duplication anomalies of the biliary tract can occur as a pure double gallbladder or biliary duplication cyst[11]. Any cystic structure lined with smooth muscle and mucosa is considered a duplication, although it may be fairly distant from the gastrointestinal tract[11]. They may contain functional heterotopic gastric mucosa lying within a biliary duplication cyst, with peptic acid secretion into the biliary system causing signs and symptoms of inflammation, ulceration, and obstruction. The patients often present pre-operatively with acute cholecystitis[29].

Small intestine duplications

These are the most common enteric duplications (50% of all reported duplications). The majority occurs in the ileum. They may be cystic or tubular; the typical ileal duplication are located on the mesenteric border[1,4] (Figure 3) sharing its muscular wall and blood supply with the adjacent intestine, and does not communicate with the associated bowel. There is an association with small-bowel atresia, and 24% of the cystic duplications contain ectopic gastric mucosa[2,4,7,30].

These cysts may cause abdominal pain from distension, leading to obstruction, volvulus, intussusception or remain asymptomatic.

The preferred treatment is resection of the duplication together with the adjacent normal intestine, followed by primary anastomosis. Long tubular duplications of the small intestine represent a surgical challenge, if the extent of the resection place the patient at risk for short-bowel syndrome, malabsorption and other nutritional deficits[7,25]. These situations can be managed with internal drainage, creating a window in the native bowel

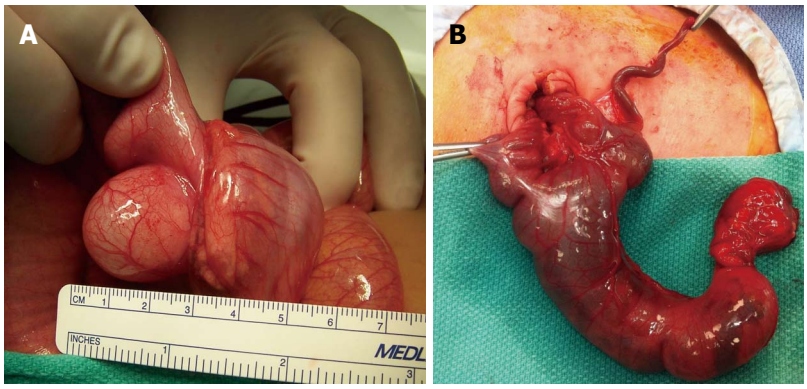

Figure 4 Colonic and rectal duplications. A: Ileoceacal duplication cyst causing obstructive symptoms; B: Cecal tubular colonic duplication.

Table 1 Clinical presentation and differential diagnosis of alimentary tract duplications

Location	Clinical presentation	Differential diagnosis
Foregut complications	Respiratory distress, obstruction	Tracheoesophageal fistula, intrathoracic mass
Gastric and duodenal duplications	Hemorrhage, obstruction (ileus)	Pyloric stenosis, gastroesophageal reflux
Gallblader duplications	Cholecystitis, peptic acid secretion into the biliary system causing signs and symptoms of inflammation, ulceration, and obstruction, Icterus	Gallblader atresia and cyst, pre-hepatic icterus
Duplications of the small intestine	Hemorrhage, obstruction (ileus), intussusception	Meckel´diverticel, intussusception
Colonic and rectal duplications	Constipation obstruction and volvulus	Constipation, Hirschsprung´s disease

lumen at the end of the duplication. This technique requires stripping of the mucosa along the common wall of the duplication to avoid complications associated with retained gastric mucosa[31,32].

Colonic and rectal duplications

Duplications of the large bowel constitute about 13% of all duplications[1,4]. They are divided into 2 subtypes: cystic duplications of the colon (Figure 4A), and colorectal tubular duplications (Figure 4B). Cystic colonic duplications present as closed spherical cysts[27]. About 40% of cystic colonic duplications involve the caecum and have imaging features similar to small-bowel duplications[4]. Ectopic gastric mucosa is occasionally present in these duplications, but associated vertebral anomalies are not a consistent finding[1,27]. Clinically, they can manifest as an abdominal mass, bowel obstruction, volvulus and constipation. Colorectal tubular duplications are double-barrelled duplications involving part or all of the large bowel[16,17]. The "twin" segment may lie on the mesenteric or antimesenteric side of the colon, usually communicating with it. Colorectal tubular duplications may be associated with rectogenital or rectourinary fistula, duplication of internal or external genitalia, or vertebral anomalies[33]. The surgical treatment of these complex hindgut duplications represents a real challenge.

Rare duplications

Since alimentary tract duplications may occur al any location from the gastrointestinal tract[1,2], there are a number of rare duplications, only reported in case reports. These include: hypopharynx[6], pylorus[10] and appendix[15].

CLINICAL PRESENTATION

Gastrointestinal duplications are generally identified during childhood (80%), although some remain silent and present later on in life. The presentation varies according to the age of the patient as well as their location. Some may be totally asymptomatic, identified on routine physical examination or during incidental investigations[2]. With the increasing use of prenatal ultrasound scan, many are now being identified in utero. Abdominal pain, vomiting and abdominal mass are the most common symptoms and signs attributable to enteric duplications located in the abdomen[14]. The mode of presentation depends on the anatomic level of the duplication, the mass effect of the lesion and whether they contain heterotopic gastric mucosa[26], with potential penetrating ulcer and severe bleeding[23]. Symptoms are often caused by obstruction caused due the raising volume of the duplication[9,14,34]. Foregut duplications may induce respiratory symptoms[28] and airway obstruction[6].

At the level of the duodenum and pancreas they can cause severe pancreatic obstructive symptoms with consecutive pancreatitis[35,36]. Dias *et al*[37] describes an intussusception arising from an ileal duplication. Colonic duplications cause obstructive symptoms or diverticulitis-like symptoms[16,17]. An infection of the cyst is also possible[38]. In adulthood, malignancies arising from alimentary tract duplications are reported[39-42]. The incidence of neoplastic change in hindgut duplications is higher than in any other location (predominantly adenocarcinoma).

Table 1 resumes the clinical presentation of alimen-

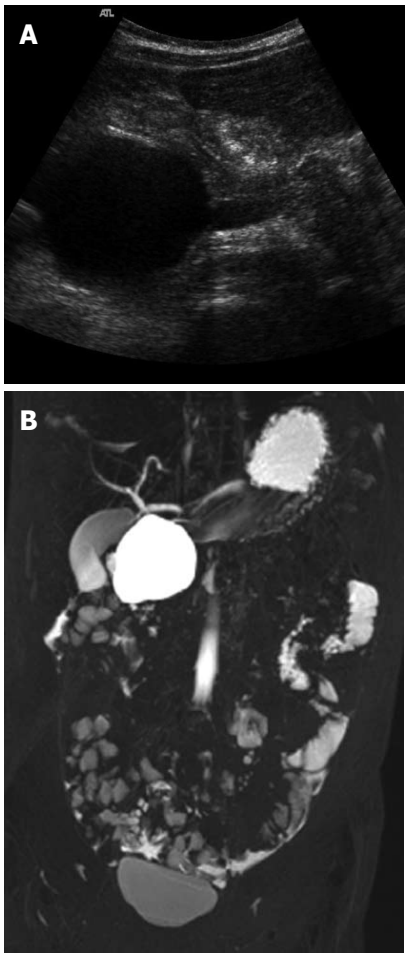

Figure 5 Diagnostic imaging. A: Ultrasound of a cystic duodenal duplication; B: Magnetic resonance scan of a cystic duodenal duplication.

tary tract duplications.

DIAGNOSTIC IMAGING

Diagnostics rely on ultrasonography, barium swallow, computed tomography (CT) or magnetic resonance (MR) imaging scans[4]. Currently GI duplications or the suspicion of a one are often seen/diagnosed in prenatal ultrasound[43]. The prenatal detection of gastrointestinal duplications has been possible since the mid 90's[44]. They rarely affect the fetal outcome or require intervention in utero[44]. Pre-operative diagnosis has changed in the last 25 years, today it lays on sonography of the abdomen normally showing a cystic structure situated next to the intestine (Figure 5A)[45]. This cystic structure differs from others because it has a "gut signature"; this term is often used by the radiologists to described an hyperechoic inner layer that is produced by the mucosa and the relatively hypoechoic outer layer produced by smooth muscle[46]. Technetium-99m pertechnetate scintigraphy can be helpful for suspected oesophageal, duodenal, and small-bowel lesions that contain ectopic gastric mucosa[30]. Regarding the age of pediatric patients, the MR scan is preferable to the CT scan[4,35], demonstrating the liquid filled cystic structure (Figure 5B)[47].

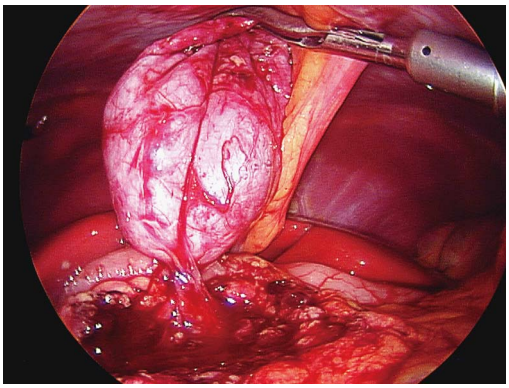

Figure 6 Duodenal duplication removed laparoscopically (stripping method).

TREATMENT

Because these lesions are rare and can present with a wide range of clinical manifestations or may even be encountered intraoperatively, the appropriate surgical management requires the surgeon to be familiar with the anatomy and clinical characteristics of these lesions. The treatment finally is surgical; total resection when possible should be the aim of the intervention, the partial excision or puncture contains high risk of recurrence[9]. Due the risk of potential complications GI duplications should be always excised, even if they are asymptomatic[1,9,14]. The operation should be performed electively as long as patient does not present with acute symptoms[14]. The resection can be indicated for relieving symptoms or preventing the malignant change, even if it is a rare complication, as the prognosis is very poor once malignancy has occurred[40,41,48].

Surgical complications are related to the size and location of the duplication, communication with the gastrointestinal tract or vertebral canal, presence of heterotopic gastric mucosa and involvement of mesenteric vessels[9,49,50]. Because of the mesenteric location of most duplications, they share a common blood supply with the normal intestine[51]. Segmental resection may be required in few cases. Otherwise, one may excise or "shell out" the cyst if an adequate plane exists[34,49]. Mucosal stripping is also possible[34,50] (Figure 6). If excision is not possible due to proximity to the biliary or pancreatic ducts, a drainage procedure may be performed[34]. However, if this is planned, one must determine whether gastric mucosa is present, and, in this case the excision of the ectopic mucosa is the surgical aim to prevent future ulceration or bleeding[51]. Mucosectomy can be a semi-conservative alternative if resection is not possible due the location or extension of the duplication[52,53].

LAPAROSCOPIC (AND THORACOSCOPIC) OPERATION ALIMENTARY TRACT DUPLICATIONS

With the rise of laparoscopy in the 1990's, one saw the first reports on laparoscopic treatment of duplication cysts. The first were on the treatment of gallbladder duplications in adults, as result of a normally planned laparoscopic gallbladder resection[29].

The first duodenal duplication cyst removal by laparoscopic technique in an adult was reported by Félix *et al*[54].

Schleef *et al*[55] published in 2000 two cases of small bowel duplications, combining diagnostic laparoscopy and definitive surgery.

Machado *et al*[56], Tayar *et al*[57] and Sasaki *et al*[58] published on successful laparoscopic resection of a gastric duplication cyst in 2003, as did Kin for a intra-abdominal esophageal duplication cyst[59]. Martínez-Ferro described a combined thoraco-laparoscopic approach for a trans-diaphragmatic thoraco-abdominal enteric duplication cyst[24].

A large retrospective multicenter study on the outcome of alimentary tract duplications operated by minimally invasive surgery (MIS) was published in 2011. 102 of 114 patients had laparoscopy (esophageal to rectal uplications) and 12 patients had thoracoscopy for esophageal duplications. The mean operative time was 90 min (range = 82-98 min). There were 32 (28%) resection anastomoses, 55 (48%) enucleations, and 27 (24%) unroofings. The conversion rate was 32%, but in a multivariate analysis, it was significantly higher, up to 41% for patients weighing < 10 kg. 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 d (range = 1-21 d) without conversion and 6 d (range = 1-27 d) with conversion (P = 0.01). The median follow-up was 3 mo (range = 1-120 mo). Eighteen of the 27 patients who under-went partial surgery had an ultrasound examination during follow-up. Five (18%) of them had macroscopic residue. In conclusion this study showed that MIS 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[47].

There are cases reported about complete endoscopic resection of duodenal duplications. Though they do not report about the further outcome on the cases[36,60-62].

Thoracic oesophageal duplication cysts are excised by thoracotomy or VATS. The intra-abdominal oesophageal duplication cysts are excised by laparotomy or laparoscopically[63,64].

Laparoscopic techniques and strategies that have to be mentioned are: (1) simple cyst excision: with closure of the muscle defect by suture or stapler[65,66]; and (2) unroofing: Partial shell-out of the cyst, optional with draining[55].

Resection with end-to-end or end to side anastomosis eventually with a trans-umbilical video-assisted procedure[49,63].

Enucleation of duplication cyst by excision of adherent mucosal layer of the cyst wall[64,67] (Figure 6).

Mesorectal excision using the prolapsing technique (for rectal duplications)[68].

Combined thoraco-laparoscopy for trans-diaphragmatic thoraco-abdominal enteric duplications[24].

Endoscopic treatment of upper duplications: An endoscopic section of the common duodenal wall, using a precut needle or sphincterotome, is used when the biliary tree is not involved. In a study by Romeo of 2011, this technique was successfully used in 4 of 6 patients, but in 2 open surgery had to be performed[62].

CONCLUSION

Alimentary tract duplications are rare congenital lesions that can occur anywhere from the mouth to the anus. Presenting with intestinal obstruction, abdominal pain, intestinal bleeding, intussusception, etc and requiring surgical intervention. Two thirds are located in the abdominal cavity. Enteric duplications have an attachment to the alimentary tract sharing a common blood supply with the intestine. The diagnosis is confirmed by histological examination. In 80% of all cases, gastrointestinal duplications are identified in childhood, the presentation depends on the age of the patient and their location. About 75% of the gastrointestinal duplications are cystic, with no communication to the adjacent alimentary tract while the remaining are tubular, sometimes communicating with the intestinal lumen.

Diagnostis relies on ultrasound, completed by barium swallow, CT or MR scans, nowadays duplication cysts are often diagnosed prenatally, in recent studies more than 50% were detected prenatally. Scintigraphy can be helpful for suspected oesophageal, duodenal, and small-bowel lesions that may contain ectopic gastric mucosa.

The treatment of alimentary tract duplications is surgical; the total resection should be the aim of the intervention, because the partial excision or puncture contains high risk of recurrence.

The operation should be performed electively, indicated for relieving symptoms or preventing further complications. Possible complications are related to the size and location of the duplication, its communication with the gastrointestinal tract or vertebral canal, presence of heterotopic gastric mucosa and the involvement of mesenteric vessels.

In review of current literature there are only few controlled studies of the outcome in laparoscopic surgery in alimentary tract duplications, as it´s a infrequent anomaly.

The largest retrospective multicenter study was published in 2011. In conclusion this study showed that minimal invasive surgery for alimentary tract duplica-

tions is feasible with a low rate of complications. These patients with a prenatal diagnosis should have prompt surgery to prevent symptoms, despite of a high rate of conversion in small infants.

In conclusion the decision if to perform a surgery in (although) suspected alimentary tract duplication relies on the experience of the surgical team. With a sufficient curriculum of the surgeon in the application of above named techniques it can be in benefit of the patient for the advantages that minimally invasive surgery (less invasive) offers to the child. Completely endoscopic techniques - until now only described in case reports - should be considered too.

Unfortunately there are no controlled studies to determine if minimally invasive surgery is beneficial for the patient in a sense of quality improvement compared to open surgery techniques, so that it has to be resumed that further studies should be performed. Regarding the frequency of alimentary tract duplications these studies should be performed as multi-center studies.

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