

An Epigastric Mass? And what if it was an Intra-Abdominal Esophageal Duplication Cyst

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Abstract

Duplication of the alimentary tract are uncommon congenital malformations that may be found anywhere from mouth to anus. Most of duplications cause symptoms in infancy or early childhood. Digestive duplication revealed at adulthood is a condition considered rare. Esophageal cystic duplication is a congenital cystic malformation of the alimentary tract consisting of a duplication of the segment of the esophagus to which it is adjacent. Here we describe a case of intra-abdominal esophageal duplication cyst in adult patient. We report the case of a 29-year-old female who had complained of intermittent epigastric pain and post prandial nausea for several weeks. Physical examination was without particularity except for the presence of abdominal epigastric mass. Transabdominal sonography combined with CT scan showed an epigastric cyst with no topographic relation to the pancreas measuring about 80 mm in diameter. Because of persistence of the epigastric pain and the nausea we decided to operate the patient. Histological exam than showed that the cystic mass was in fact an esophageal cystic duplication with no histological signs of malignancy.

Introduction

Duplication of the alimentary tract are uncommon congenital malformations that may be found anywhere from mouth to anus. By definition, they are located within or adjacent to the wall of part of the gastrointestinal tract, have smooth muscle in their walls, and are lined by alimentary tract mucosa. The most common duplication is cystic and located on the mesenteric aspect of the small or the large intestine. Most of duplications cause symptoms in infancy or early childhood. Digestive duplication revealed at adulthood is a condition considered rare. Esophageal cystic duplication is a congenital cystic malformation of the alimentary tract consisting of a

duplication of the segment of the esophagus to which it is adjacent. Here we describe a case of intra-abdominal esophageal duplication cyst in adult patient.

Case Presentation

We report the case of a 29-year-old female who had complained of intermittent epigastric pain and post prandial nausea for several weeks. Physical examination was without particularity except for the presence of abdominal epigastric mass of two centimeters. Transabdominal sonography combined with CT scan showed an epigastric cyst with no topographic relation to the pancreas measuring about 80 mm in diameter. Since Tunisia is an endemic country for echinococcus, hydatid cyst was a diagnosis that we had consider, but all blood tests performed gave normal results including a negative echinococcus serology. There was no evidence of elevated tumor markers or malignancy. We then completed by an esophageal gastric endoscopy which came without abnormalities.

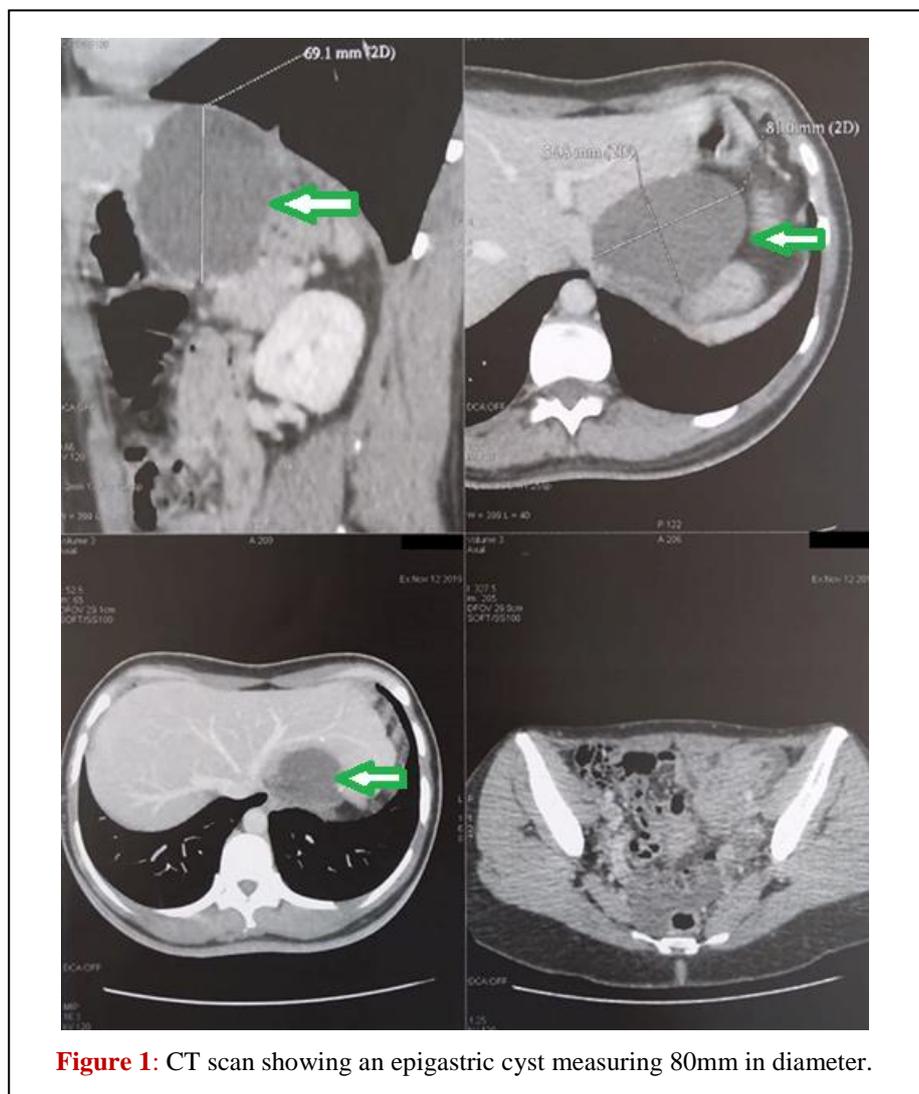


Figure 1: CT scan showing an epigastric cyst measuring 80mm in diameter.

Because of persistence of the epigastric pain and the nausea we decided to operate the patient. Surgery was performed. Subumbilical explorative laparotomy was undertaken and a large cystic mass was found located in the gastroesophageal junction, adjacent to left liver lobe, with no evidence of malignancy per operatively. The

cystic mass was freed and resected with no incident. Histological exam than showed that the cystic mass was in fact an esophageal cystic duplication with no histological signs of malignancy. The course was marked by good evolution and the patient was discharged at day five post operative.

Discussion

Alimentary tract duplication is a rare congenital malformation that develops potentially anywhere in the gastrointestinal tract, from the root of the tongue to the anus. They are usually a hollow structure that have a muscular coat, and are lined with epithelium similar to the one found in the gastrointestinal tract. These lesions are generally contiguous to some portion of the alimentary tube. The type of epithelium lining in the duplication cyst does not necessarily correspond to that in the alimentary tract to which it is adjacent. In more than 80% of cases, they are diagnosed before the age of two years old when the patient experiences acute abdominal or bowel obstruction or other associated complications. A minority of cases remain asymptomatic until adulthood. Foregut duplication cysts are uncommon and esophageal duplication cysts are even rarer especially those discovered in the adult age such as the case of our patient. Only few cases were reported in which the duplication concerns the intra-abdominal part of the esophagus. Surprisingly most cases reported in the literature are adult patients. **Table 1** shows a recap of the intra-abdominal esophageal duplications as described in the literature so far. Seventy six percent of the reported cases were adults and their ages ranged from 18 years old to 70 years old. In 24 % of the cases, they were children and their ages ranged from eight weeks to 13 years old. In 48% of the cases the cyst was discovered incidentally, in 29 % of the cases it manifested by an epigastric pain, and in the 23% of cases it had various manifestation. In 57% of the cases surgical resection of the duplicated cyst was made laparoscopically and in the other 43% it was made by open surgery. Fifty two percent of the cysts had a size less than five cm and 48% were equal five cm or more.

Table 1: Outline of intra-abdominal esophageal duplication cysts case reports as described in literature so far (December 2021).

year	Author	institution	Age at diagnosis	sex	Clinical presentation	Localisation of the cyst	size	treatment
1989	Ruffin and al [1]	Department of general surgery, Naval Hospital, Millington, Tennessee, USA	38	F	Epigastric pain and nausea	Distal esophagus	4 cm	Resection with surgical repair

1996	Harvell and al [2]	Department of Pathology, University of California, USA	57	F	Epigastric pain	Superior border of the pancreas	2.2 × 1.7 × 1.5 cm	Laparoscopic resection
1997	Karahasano glu and al [3]	Department of Surgery, Istanbul University Cerrahpasa Medical School, Turkey	51	M	Epigastric pain, dysphagia	Sub-diaphragmatic	11.0 × 9.0 × 8.0 cm	Esophagogastrectomy
1998	Janssen and al [4]	Department of Radiology, Hospital of St. Raphael, New Haven, USA.	56	F	Incidental finding at a CT scan made for rectal cancer investigation	Superior to left kidney	8.0 × 6.0 × 4.5	Open resection
2000	Rathaus and al [5]	Department of Diagnostic Imaging, Sapir Medical Center, Kfar Saba, Israel.	5	F	Epigastric pain	Distal esophagus, between liver and cardia	1 cm	Open resection
2002	Nelms and al [6]	Carolinas Medical Center, Charlotte, NC, USA.	44	M	incidental at CT	Diaphragmatic crura	7	Laparoscopic resection
2002	Vijayaraghavan and al [7]	Rajmahal Vilas Hospital, Sanjayanagar, Bangalore	70	F	Incidental at ultrasound	Midline between stomach and liver	7.5	Open resection, combined with cholecystectomy

2003	Noguchi et al [8]	Department of Surgery, Rinku General Medical Center, Izumisano Municipal Hospital, Osaka, Japan	26	F	incidental at ultrasound	Anterior wall of distal esophagus	4.0 × 3.0 × 3.0	Laparoscopic resection with esophageal repair (nissen)
2003	Kin et al [9]	Department of Surgery, Rinku General Medical Center, Izumisano Municipal Hospital, Osaka, Japan.	51	F	Incidental finding at CT scan	Distal esophagus	4.5 × 4.0 × 3.5	Laparoscopic resection
2007	Martin et al [10]	Department of Surgery, Thomas Jefferson University, Jefferson Medical College, Philadelphia, USA.	50	F	Left flank pain	Inferior to pancreatic tail	6.5 × 5.5 × 4.2	Open resection
2007	Martin et al [10]	Department of Surgery, Thomas Jefferson University, Jefferson Medical College, Philadelphia, USA.	60	M	Gastric outlet obstruction	Retro-duodenal	10.0 × 10.0	Open resection

2011	Aldrink et al [11]	Division of Pediatric Surgery, Nationwide Children's Hospital, Columbus, USA	2	M	Incidentally No symptoms. Incidental at fundoplication	Gastroesophageal junction	3	Laparoscopic resection, with fundoplication
2011	Gümüş et al [12]	Department of General Surgery, Dicle University Faculty of Medicine, Diyarbakır, Turkey.	18	F	Incidentally At CT for dyspeptic complaints	Lower end of the esophagus, adjacent to liver	4.2 × 3.6	Open resection
2013	Bhamidipati et al [13]	Department of Surgery, State University of New York Upstate Medical University, Syracuse, New York, United States	69	M	incidental at CT scan for diverticulitis	Gastroesophageal junction	4.4 × 3.7 × 3.9	Laparoscopic resection
2013	Pujar et al [14]	Jawaharlal Nehru Medical College, Belgaum, Karnataka, India	13	F	Acute pain in the epigastric region	Gastroesophageal junction, adjacent to left liver lobe	4.0 × 5.0	Laparoscopic resection
2013	Mori et al. [15]	Department of Surgery, Teikyo University Chiba Medical Center, Ichihara, Japan	9	M	Incidental	Ventral surface of the abdominal esophagus	2 cm	Laparoscopic resection

2014	Castelijns and al. [16]	Catharina Hospital, The Netherlands	20	M	Right abdominal pain	gastro-esophageal junction	2.3*3	Laparoscopic resection
2015	Watanobe and Al [17]	Juntendo University Nerima Hospital, 3-1-10 Takanodai, Nerima, Tokyo 177-8521, Japan	50	M	Epigastric Pain	Intra-abdominal esophagus extending to the distal thoracic esophagus	3.5 cm	Laparoscopic resection with intraoperative esophagoscopy
2015	Huang and al [18]	The First Affiliated Hospital of Wenzhou Medical University, Zhejiang, P. R. China	20	F	Abdominal mass discovered incidentally	near the spleen	13.8*9.5*8.5 cm	Laparoscopic resection
2018	Shinnick and al. [19]	Children's Healthcare of Atlanta, Atlanta, Georgia	8 weeks	F	Difficulties to thrive	Esogastral junction associated with gastral duplication	Not precized	Open resection
2020	Mori and Al [20]	Teikyo University Chiba Medical Center, Ichihara, Japan	64	F	Incidental	Gastro intestinal junction	7 cm	Laparoscopic restion

The indications for treating esophageal duplication cysts include symptoms such as dysphagia, abdominal pain, hemorrhage, an increase in cyst size, and suspicion of malignancy. Since the diagnosis of its condition remains doubtful until the pathology confirmation, surgical resection of the cyst remains the standard treatment. The approach to the cyst should be determined based on its size, location, and relationship with adjacent organs.

Conclusion

Esophageal duplication cysts are uncommon developmental lesions arising from the primitive foregut, when diagnosed or encountered incidentally; they should be completely excised because of the potential complications such as bleeding, infection or conversion to a malignancy.

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