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Case Report

Intestinal duplication cyst in the esophagogastric junction as a cause of gastroesophageal reflux disease: a case report

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ABSTRACT

This report presents a rare case of a 42-year-old woman with refractory gastroesophageal reflux disease (GERD) attributable to a 5×4 cm enteric duplication cyst at the esophagogastric junction. Despite a year of proton pump inhibitor therapy, the patient experienced persistent symptoms, including nausea and nocturnal vomiting. Surgical intervention involved hiatal plasty, partial Dor fundoplication, and the unexpected discovery of the cystic tumor during the procedure. The cyst, firmly adhered and delimited, ruptured during dissection, leading to successful de-roofing, cauterization, and drainage. The patient's immediate postoperative course was satisfactory, demonstrating the effectiveness of this approach in managing an enteric duplication cyst. This case underscores the importance of considering congenital anomalies in the context of refractory GERD, with the enteric duplication cyst located at the esophagogastric junction representing a unique manifestation. The study contributes valuable insights into the atypical presentation, diagnostic challenges, and successful surgical management of such anomalies.

Keywords: Intestinal duplication cyst, Esophagogastric junction, Gastroesophageal reflux disease, Case report, Surgical management

INTRODUCTION

Intestinal duplication cysts constitute a rare congenital anomaly that can manifest at any level of the digestive tract. The term has been in use since 1937 with Ladd, followed by Grass in 1952, and was structurally defined in 1961 by Mellish and Koop as spherical or tubular structures containing mucosal, muscular, and serosal layers.[1]

This pathology has an incidence of 1 in 4,500 live births, representing only 0.1 to 0.3% of all congenital malformations of the digestive tract.[1] Two forms of presentation have been reported in the literature: tubular and cystic. The tubular form communicates with the intestinal lumen, while the cystic form, which is the most common presentation (80%), is separate from it. Most intestinal duplication cysts are diagnosed during the first year of life, and in around 70% of cases, they are identified before the age of 12. The most common site of presentation is the ileum (35%), while one of the less common sites is the stomach (2-9%), where the non-communicating cystic type with the gastric cavity represents up to 80% of these lesions.[2] Most gastric duplication cysts are of a singular nature and histopathologically feature a mucosal lining consisting of gastric epithelium surrounded by the proper muscular layer.[3] They are mainly located in the greater curvature of the stomach, and in most cases, they may contain heterotopic mucosa from any part of the digestive tract.[4] In the present case, a duplication cyst in the esophagogastric junction is reported, a location not described in current literature, causing symptoms of gastroesophageal reflux disease (GERD) in this patient, prompting medical attention.

The clinical presentation depends on various factors such as the portion of the digestive tract involved, the size of the cyst, associated blood vessels, compression of adjacent structures, or the cyst's growth due to the accumulation of secretions, among others. Commonly described symptoms include postprandial fullness, nausea and vomiting, abdominal pain, a palpable mass, acute intestinal obstruction, perforation or hemorrhage, and rarely jaundice, hydronephrosis, or other manifestations due to compression of nearby structures. Generally, the most common symptoms are abdominal pain and melena.[5] A presentation with GERD symptoms, as in our patient's case, was not found in the current literature.

Imaging diagnosis can be guided by a barium swallow to identify if it is communicating; however, the most effective methods are those that allow a three-dimensional view, such as computed tomography and magnetic resonance imaging.[5] The method of choice is ultrasound, which, among other findings, helps distinguish the echogenic mucosal layer and hypoechoic muscular layer (intestinal signature sign), the "Y" configuration in communicating cysts, as well as the presence of peristalsis if the static transducer is maintained long enough.[6]

The treatment of this anomaly is surgical, with two possibilities: complete cyst resection or partial resection with drainage when it is near areas with a high risk of manipulation injury, such as the bile duct. Endoscopic management has been well accepted with good results, but there is still insufficient information on it.[7] In our case, we opted for partial resection with drainage, as well as defunctionalization of the residual mucosa, adding an anti-reflux procedure.

We present the case of an adult patient who underwent laparoscopic surgery for GERD diagnosis, with a surgical plan of fundoplication and hiatal hernia repair. An intraoperative finding was the identification of an enteric duplication cyst located on the right side of the esophagogastric junction, which caused migration of the same, as well as part of the stomach, predominantly the lesser curvature. Attempted cyst resection resulted in rupture during dissection, with the release of dark brown thick material. Destechment of the cyst was performed as it was firmly adhered to the esophagus, followed by cauterization of the residual mucosa and Dor fundoplication on the affected segment.

CASE REPORT

A 42-year-old woman with a history of diabetes mellitus, open appendectomy, cholecystectomy, and hysterectomy for myomatosis, social alcohol consumption, and discontinued smoking, presented with symptoms of regurgitation and heartburn that started five years ago. She sought medical attention from a family physician, who diagnosed her with GERD and initiated treatment with a proton pump inhibitor and aluminum-magnesium combination for one year without improvement. Nausea and nocturnal vomiting developed, leading to referral to our service. Cisapride and sucralfate were added to the initial prescription, resulting in partial improvement.

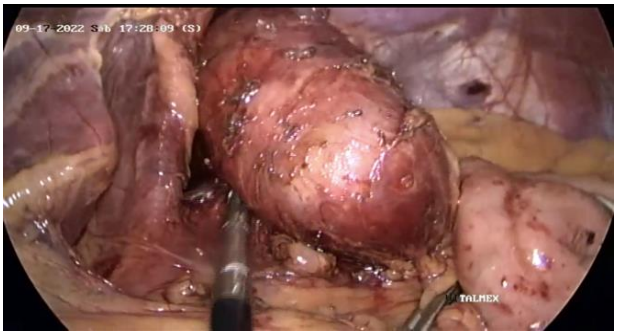

Figure 1: Intraoperative laparoscopic finding of cystic tumor at the esophagogastric junction and lesser curvature of the stomach.

Panendoscopy revealed a 5 cm type I hiatal hernia and reactive biliary gastropathy. Esophageal manometry reported a type I sliding hiatal hernia, a 3 cm difference between the esophageal sphincter and diaphragmatic crura, a short intrathoracic lower esophageal sphincter with normal intrabolus pressure, and normal esophageal peristalsis. Surgical intervention was decided for hiatal plasty and partial fundoplication.

During surgery, an extensive hiatal opening was found, along with a cystic tumor at the esophagogastric junction measuring 5×4 cm, firmly adhered and delimited. The cyst ruptured during dissection, releasing thick brown fluid. De-roofing, cauterization of residual mucosa, hiatal plasty, and partial Dor fundoplication were performed. A Penrose drain was placed with 30 cc of bleeding. The postoperative diagnosis was a cystic tumor at the esophagogastric junction and type I hiatal hernia.

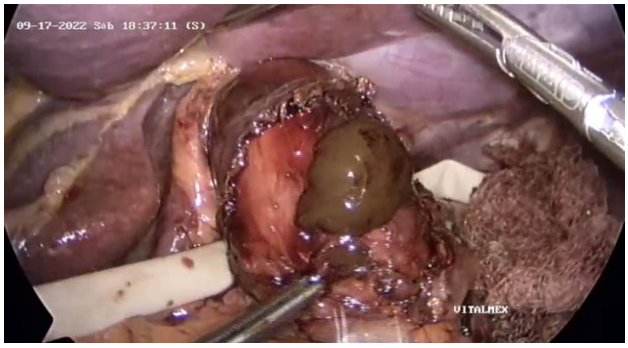

Figure 2: Rupture during cyst dissection, with release of dark brown thick material.

The patient had a satisfactory immediate postoperative course, with a contrast swallow showing no contrast medium leakage. She was discharged on the second postoperative day with outpatient follow-up. Histopathological examination revealed a wall of enteric duplication cyst with intense chronic inflammation,

evidence of old bleeding, extensively eroded epithelium, and areas lined with negative-to-malignancy ciliated columnar epithelium.

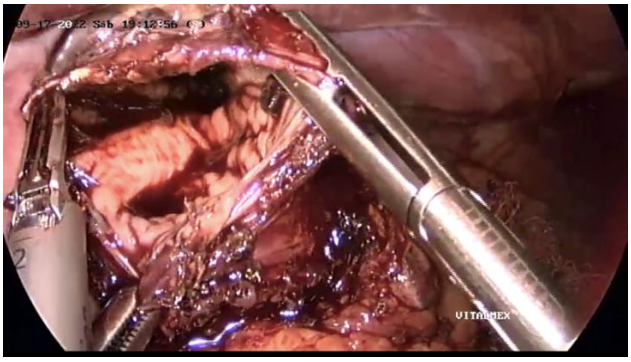

Figure 3: Laparoscopic view of de-roofing of cystic tumor at the esophagogastric junction.

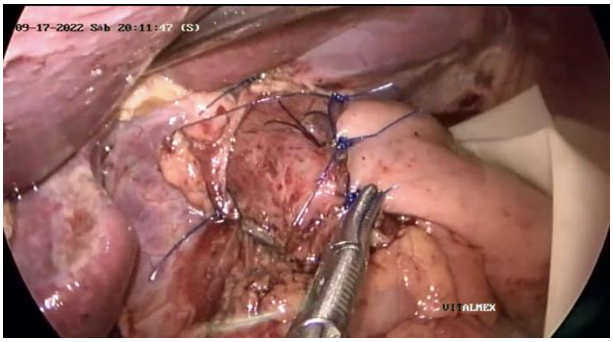

Figure 4: Dor-type partial fundoplication over de-roofed and cauterized segment.

DISCUSSION

The presented case highlights a complex medical scenario involving a 42-year-old woman with a rare intestinal duplication cyst located at the esophagogastric junction, leading to symptoms of GERD. The patient's medical history, including diabetes mellitus and prior surgeries, adds layers of complexity to the diagnostic and treatment process.

The initial medical management with a proton pump inhibitor and aluminum-magnesium combination proved insufficient, emphasizing the challenges in addressing this condition solely through pharmaceutical means.[6] The subsequent addition of cisapride and sucralfate resulted in partial improvement, underscoring the need for a multidimensional approach.

Diagnostic procedures, such as panendoscopy and esophageal manometry, played a crucial role in uncovering the extent of the pathology, including a type I hiatal hernia and a cystic tumor at the esophagogastric junction. Surgical intervention became imperative, revealing an extensive hiatal opening and a firmly adhered cystic tumor during the procedure.[2] The intraoperative findings, including the rupture of the cyst with the release of thick brown fluid, highlight the intricacies of the case.

The postoperative course was deemed satisfactory, with no contrast medium leakage observed during a contrast swallow. Histopathological examination provided valuable insights into the nature of the duplication cyst, revealing features of chronic inflammation, old bleeding, and erosion of the epithelium. These findings contribute to the understanding of the condition and inform postoperative care.

The case also sheds light on the rarity of intestinal duplication cysts, emphasizing their congenital nature and the varied presentations across the digestive tract. The unique location at the esophagogastric junction adds novelty to the literature, expanding our understanding of the potential manifestations of these anomalies.[2]

The discussion of symptoms underscores the importance of considering factors such as the portion of the digestive tract involved, size, and associated vascular structures. Moreover, the comprehensive overview of diagnostic imaging methods, including barium swallow and ultrasound, provides valuable insights for clinicians facing similar cases.[6]

The presented case advocates for surgical treatment, detailing the specific procedures employed, such as de-roofing, partial resection with drainage, and defunctionalization of residual mucosa. The incorporation of an anti-reflux procedure and hiatal plasty addresses the patient's GERD symptoms, highlighting the individualized and comprehensive approach required for optimal outcomes.

CONCLUSION

This case report not only contributes to the limited literature on esophagogastric junction duplication cysts but also serves as a valuable resource for clinicians navigating the diagnostic and therapeutic challenges associated with such rare congenital anomalies. The multidisciplinary nature of the management underscores the importance of collaboration between medical and surgical specialties to ensure comprehensive patient care.

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