RESEARCH ARTICLE

THORACOSCOPIC EXCISION OF SYMPTOMATIC ESOPHAGEAL DUPLICATION CYST IN ADULT POPULATION-EXPERIENCE FROM A TERTIARY CARE CENTER

Sankar Subramanian, Niket Shah, Neelendra Yesaswy and Suresh Kumar P.

Abstract

Gastrointestinal duplications are rare congenital anomalies in children. The most common site being jejunum and ileum, followed by mediastinum, colon, stomach, duodenum and rectum. Esophageal duplication cysts accounts for twenty percent. Majority of them are detected in childhood and treated. A very small proportion can present in adult population posing diagnostic challenges due to the clinical presentations. Minimal access surgery has obviated the need for thoracotomy. Herein we share our experience of treating four symptomatic esophageal duplication cysts in adults, by minimally invasive approach.

Introduction:

Gastrointestinal duplications are rare congenital anomalies and most of them manifest with symptoms in childhood. Small intestine is the most common site for the duplications, which is followed by esophagus, colon, stomach, duodenum and rectum(1). Nearly one fifth of the duplication cyst involve the esophagus. Majority of the esophageal duplications are detected in childhood and treated. A very small proportion can present in adult population which can pose diagnostic challenges, considering the myriad of mediastinal pathologies. Most of the literature on thoracoscopic management of esophageal duplication cysts are no more than case reports. Herein we share our experience of treating four symptomatic esophageal duplication cysts in adults, by minimally invasive approach.

Materials and Methods:

This was an observational study of retrospective analysis of the prospectively maintained database of all the patients who were diagnosed with esophageal duplication cyst in the department of surgical gastroenterology of Sri Ramachandra medical center, a university hospital and a tertiary referral center located in south India. The period of study was 10 years (September 2010 to August 2020)

Results:

There were totally four patients diagnosed in this period. All the four patients were men in the third and fourth decade of life. The most common clinical presentation was mild to moderate dysphagia which warranted evaluation. The other associated symptoms include, regurgitation, chest discomfort, chronic dry cough. All the four patients had the duplication cyst located in the thoracic esophagus, two involving the infra-carinal portion and two involving the retro cardiac segment of esophagus. The average size of the cyst was 5 centimeters. The diagnosis was achieved in all the four cases with CT scan and flexible esophagoscopy (picture 1,2,3). None of the patient underwent EUS as; imaging findings on CT was unambiguous. All the four patients underwent thoracoscopic excision using the standard technique of right thoracic approach in prone position (picture 4). Our policy is to resort to double lung

Corresponding Author: Sankar Subramanian
ventilation, and carbondioxide insufflation to a pressure of 10 mm of mercury, achieved the lung collapse needed for the working surgical space. Standard three ports (5th, 7th and 9th intercostal space, in the mid-clavicular line, posterior axillary line and scapular line) were used to achieve triangulation. All the patients underwent complete excision of the cyst. As these cysts are intra mural in location and forms the integral part of the muscular layer of the esophagus, intra operatively they were decompressed before dissection (picture 5). This technical step facilitated complete excision of the cyst without opening the esophageal mucosa. However, in all patients, an intra operative leak test was performed post excision. In two patients where the cyst was projecting into the right hemithorax (picture 6), post-excision myotomy was sutured, whereas in the remaining two patients the cyst was eccentrically projecting into the left hemi thorax (picture 7), the myotomy was left unsutured. Postoperative period was uneventful. ICD tube was removed on the first post-operative day after performing a gastrograffin study demonstrating integrity of the mucosa. Oral liquids started on the first day and gradually advanced to solid diet. All the patients stayed in the hospital for three days. Histological confirmation was achieved in all four patients by the characteristic double layer of muscle, lining epithelium(squamous or pseudostratified columnar) and the absence of cartilaginous elements(picture 8)

**Discussion:**
Gastrointestinal duplications are rare congenital anomalies in children. The most common site being jejunum and ileum, followed by mediastinum, colon, stomach, duodenum and rectum (1).

Esophageal duplication cysts accounts for twenty percent. Majority of them are detected in childhood and treated. A very small percentage can present in adult population posing diagnostic challenges due to the clinical presentations.

Clinical presentation includes dysphagia, chest pain and regurgitation. Some cases can be picked up as incidental findings. Esophageal duplication cyst may be cystic or tubular. It can also be intramural and extramural. The most common location of these esophageal duplication cysts is the lower thoracic region.

Rarely it may involve the cervical and abdominal esophagus. The embryogenesis is the faulty development of the posterior division of the foregut. The important differential diagnosis includes bronchogenic cyst, leiomyoma, cystic degeneration of tumors and hydatid cyst. CT findings are very characteristic which include a well-defined lesion with homogenous density (2).

However, with the advent of endoscopic ultrasound, the diagnostic accuracy has increased tremendously, making it a very important tool in the imaging.

The main usefulness of EUS is it helps to differentiate duplication cyst from bronchogenic cyst, by the close proximity to esophagus, double layer of muscle forming the cyst wall and absence of cartilaginous elements (3). The duplication cyst may appear anechoic, hypoechoic or mixed echoic depending on the content like, thick fluid, blood or pus.

EUS guided FNAC is generally avoided for fear of introducing infection and resorted only when there is a diagnostic dilemma. Esophageal duplication cysts warrant surgery even in asymptomatic patient as it is more prone for complications like infection, hemorrhage, perforation.

Esophageal duplication cyst can also masquerade as tumors (4). There are reports of malignant transformation occurring in duplication cyst of the esophagus (5). With the advent of minimally invasive approach, surgical excision of esophageal duplication cyst has become less morbid procedure (6) and most of the large series of thoracoscopic excision of esophageal duplication cysts are reported in children.

When it comes to adult population there are only case reports regarding thoracoscopic management. Herein we are reporting our experience of managing four cases of esophageal duplication cysts managed thoracoscopically.

**Conclusion:**
Esophageal duplication cyst is a very rare congenital anomaly and presentation in adult population is even more rare. Thoracoscopic excision is the standard of care with minimal morbidity.
Picture 1: CT axial section.

Picture 2: CT sagittal section.

Picture 3: CT images.

Picture 4: Port Positions.
Picture 5:- Intra-Operative Decompression.

Picture 6:- Thoracoscopic Dissection.

Picture 7:- Myotomy.

Picture 8:- Two Layers Of Muscles.
References:
1. Karnak I, Ocal T, Senocak ME, et al. Alimentary tract duplications in children: Report of 26 years’ experience. Turk J Pediatr 2000; 42: 118-25.
2. Jang KM, Lee KS, Lee SJ, et al. The spectrum of benign esophageal lesions: Imaging findings. Korean J Radiol 2002; 3: 199-210.
3. Wiechowska-Kozłowska A, Wunsch E, Majewski M, et al. Esophageal duplication cysts: Endosonographic findings in asymptomatic patients. World J Gastroenterol 2012; 18: 1270-2.
4. Chaudhary V, Rana SS, Sharma V, Sharma AR, Nada R, Gupta R, Dutta U, Singh K, Bhasin DK. Esophageal Duplication Cyst in an Adult Masquerading as Submucosal Tumor. Endosc Ultrasound 2013; 2(3): 165-167.
5. Dai ZJ, Kang HF, Lin S, Bai MH, Ma L, Min WL, Lu WF, Wang XJ. Esophageal cancer with esophageal duplication cyst. Ann Thorac Surg. 2013 Jul;96(1):e15-6. doi: 10.1016/j.athoracsur.2013.01.019. PMID: 23816110.
6. Michel JL, Revillon Y, Montupert P, Sauvat F, Sarnacki S, Sayegh N, et al. Thoracoscopic treatment of mediastinal cysts in children. J Pediatr Surg. 1998;33:1745–8.