Editorial

Esophageal duplication cyst

Chikara Kunisaki

Department of Surgery, Gastroenterological Center, Yokohama City University, Yokohama, Japan

Correspondence to: Chikara Kunisaki, MD, PhD. Professor, Department of Surgery, Gastroenterological Center, Yokohama City University, 4-57 Urafune-cho, Minami-ku, Yokohama 232-0024, Japan. Email: s0714@med.yokohama-cu.ac.jp.

Comment on: Nguyen ND, Pham DH, Fingerhut A. Video-assisted thoracoscopy or laparoscopy for excision of esophageal duplication cyst: case report. Ann Laparosc Endosc Surg 2022;7:39.

Received: 01 August 2022; Accepted: 21 September 2022; Published: 30 October 2022.

doi: 10.21037/ales-22-43

View this article at: https://dx.doi.org/10.21037/ales-22-43

Nguyen *et al.* reported a rare congenital case of esophageal duplication cyst excised using video-assisted thoracoscopy and laparoscopy in *ALES* (1). Therefore, I write an editorial for this fine case report.

Esophageal duplication is a rare congenital malformation originating from the embryonic foregut (bronchopulmonary and alimentary tracts) (2). Of alimentary tract duplications, esophageal duplication forms 10% to 20% (3). In embryologically, the bronchogenic cyst, which is the most common foregut cysts, generates from abnormal budding of vertebral primitive foregut whereas the esophageal duplication represents abnormal budding of the dorsal primitive foregut (4). In 1944, Bremer firstly reported enteral duplication cyst (5). In 1885, Roth focused on these cysts by dividing into two categories: simple epithelial-lined cysts and foregut cysts (6). In 1960, Bentley and Smith advocated a most acceptable theory for enteric cyst "split notochord syndrome" related with persistence of a neuroenteric canal (7). According to this theory, the notochord fuses with the embryonic endoderm and at the next stage, the endoderm separates from the notochord during the third to fourth week of gestation. If adhesions or a neuroenteric band persists during this separation time, traction diverticula and duplication cysts develop.

Although esophageal duplication cyst is a rare congenital malformation, some cases were diagnosed prenatally by ultrasonography (8). Moreover, about 80% of duplication cyst are diagnosed in childhood (9). The remaining is diagnosed in adult as shown in this case. The incidence of esophageal duplication cyst is reported to be one in 8,200 live new born with a 1:1 sex equal ~2:1 male predominance (10-12).

Although esophageal cyst embryologically occurs anywhere along the esophagus, about 60–80% of the cyst originates in the lower third of the esophagus (12). Generally, 90% of esophageal duplication cyst does not communicate with the esophageal lumen and the remaining cysts communicate with the esophageal lumen accompanied by the normal esophagus (13). Approximately 60% of the cysts originate from the lower third of the esophagus and this type of the cysts induces dysphagia and arrhythmias. By contrast, the remaining cysts occur in the upper/middle third of the esophagus frequently produces chest pain, and respiratory symptoms (cough, stridor and wheezing) (14,15). In adult cases, dysphagia was most frequently observed in >50% cases followed by chest pain (3,11). In the current report, both two cases showed dysphagia in spite of good general condition. These cases showed typical symptoms of esophageal duplication cyst among adult cases.

Of esophageal duplications, the cystic type is the most common type followed by the tubular type. Histologically, the lining of the cysts varies and encompasses stratified squamous, simple columnar, cuboidal, pseudostratified, and ciliated. Moreover, a double layer of surrounding smooth muscle lined by enteric epithelium (16). According to Palmer's pathologic criteria (17), this type of cyst is classified as follows: (I) attachment to the esophageal wall; (II) presence of gastrointestinal tract epithelium; non-keratinizing squamous or ciliated columnar epithelium; and (III) existence of two layers of smooth muscle. However, we have to pay attention to non-applicability of this criterion for some intra-abdominal esophageal duplication cysts. In another study, heterotopic tissue including gastric and pancreatic mucosae along with mucosae resembling Peyer's patches were reported (18). The cases in this report

are accurately diagnosed as esophageal duplication cyst according to pathologic criteria.

As previously mentioned, most cases were diagnosed in early childhood (9). Of these, some cases were reported to accompany with the congenital abnormalities related with the vertebrae. In a subset of population with the duplication cyst in the lower third of the esophagus, dysphagia, vomiting, arrhythmia or heart failure were commonly observed, and chest pain, cough, dyspnea were frequently observed in patients with duplication cyst in the middle/upper third of the esophagus due to the extrinsic compression or displacement by the larger cyst (11,19). Albeit rare cases, hematemesis in the cases with peptic ulcer from ectopic gastric mucosa, neurological deficiency in the cases with nerve root compression, failure to thrive, and occurrence of malignancy were observed (20-22). Other than those symptoms mentioned above, heartburn, reflux esophagitis, rupture, obstruction, and infection are also observed (11).

At least, chest radiography, upper gastrointestinal series with barium swallow, and computed tomography (CT) are basically mandatory to diagnose esophageal duplication cyst. Chest radiology and barium swallow studies reveal a mediastinal mass compressing the esophagus accompanied with the displacement or narrowing of the trachea (23,24). CT also shows the cystic mass communicating with and without the esophageal lumen (25). CT enables us to distinguish esophageal duplication cyst from bronchial and vertebral anomalies. Upper gastroenterological endoscopic examination rules out the esophageal intraluminal mass or extrinsic compression of the esophageal lumen by the duplication mass (2).

Abdominal ultrasound examination for an expectant mother identifies the cystic mass in the embryo during pregnancy (8). Prenatal diagnosis of this rare entity may provide us the useful strategy for a newborn. Endoscopic ultrasound (EUS) is a distinguished imaging modality to diagnose the esophageal duplication cyst. We can realize the origin of the mass, anatomical orientation with the surrounding organs and property of the mass (9,19). In addition, it is possible to perform fine needle aspiration biopsy (FNAB) using EUS and subsequently, we can obtain useful information to diagnose the esophageal duplication mass (26).

It is necessary to differentially diagnose esophageal duplication cyst from bronchogenic cyst cervical duplication cyst, pericardial cyst, anterior meningocele, neuroenteric cyst, thyroglossal duct cyst, lipoma, hemangioma, hydatid cyst, and Mullerian cysts using the useful imaging modalities as mentioned above. In a report of adult series, CT was most frequently employed followed by upper gastrointestinal (GI) series, endoscopy, EUS (11). Magnetic resonance imaging was just used in 16%. In the current case report, CT, endoscopy and EUS were appropriately performed and these imaging modalities led correct preoperative diagnosis. Performance of preoperative diagnostic imaging supposing a rare disease based on the abundant knowledge induces a correct diagnosis. This case report is a good role model.

Surgical resection stands on the main stay for this disease regardless of symptoms. A long time ago, a posterolateral thoracotomy was the first choice for the resection of the duplication cyst (11,27). According to the development of the optical technology, video-assisted thoracoscopic surgery (VATS) (28,29) and more recently, robotic-assisted thoracoscopic surgery (RATS) have been employed for the treatment of this entity (30). Meticulous manipulation by these techniques provides safety and steady resection of the cyst and additionally, cosmetic satisfaction. We can easily suppose that a combination of thoracoscopic and laparoscopic technique make us to resect huge cystic mass in the lower thoracic to abdominal esophagus successfully. Moreover, endoscopic submucosal tunnel dissection (ESTD) is a newest and challengeable treatment option (31). We can anticipate the short- and long-term favorable outcomes under the sufficient number of endoscopic treatments for this disease. Long-term outcomes are generally favorable after complete resection of the duplication cyst. Therefore, it is better to make effort to prevent postoperative complications by the minimally invasive surgery.

We have to gain the knowledge even for a rare case and to master the surgical skill of the newly developed method for this purpose.

In the current cases, authors successfully performed thoracoscopic and laparoscopic surgery for this entity based on their distinguished experiences. As authors concluded, their surgical therapeutic strategy is optimal according to their many experiences and resulted in favorable outcomes even for this rare entity.

Acknowledgments

Funding: None.

Footnote

Provenance and Peer Review: This article was commissioned

by the editorial office, *Annals of Laparoscopic and Endoscopic Surgery*. The article did not undergo external peer review.

Conflicts of Interest: The author has completed the ICMJE uniform disclosure form (available at https://ales.amegroups.com/article/view/10.21037/ales-22-43/coif). The author has no conflicts of interest to declare.

Ethical Statement: The author is accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

References

1. Nguyen ND, Pham DH, Fingerhut A. Video-assisted thoracoscopy or laparoscopy for excision of esophageal duplication cyst: case report. Ann Laparosc Endosc Surg 2022;7:39.
2. Chan M, Zavala SR. Esophageal cyst. StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing, 2022.
3. Bagheri R, Asnaashari AM, Afghani R. Esophageal duplication cyst. Asian Cardiovasc Thorac Ann 2015;23:332-4.
4. Berrocal T, Torres I, Gutiérrez J, et al. Congenital anomalies of the upper gastrointestinal tract. Radiographics 1999;19:855-72.
5. Bremer LJ. Diverticula and duplications of the intestinal tract. Arch Pathol 1944;38:132-40.
6. Daum R, Roth H. Cystic duplications of the cervical esophagus. Langenbecks Arch Chir 1985;366:175-8.
7. Bentley JF, Smith JR. Developmental posterior enteric remnants and spinal malformations: the split notochord syndrome. Arch Dis Child 1960;35:76-86.
8. Khatib N, Beloosesky R, Blumenfeld Z, et al. Early prenatal diagnosis of intraabdominal esophageal duplication cyst. Ultrasound Obstet Gynecol 2016;48:796-7.
9. Liu R, Adler DG. Duplication cysts: Diagnosis, management, and the role of endoscopic ultrasound. Endosc Ultrasound 2014;3:152-60.
10. Cuch B, Nachulewicz P, Wieczorek AP, et al. Esophageal Duplication Cyst Treated Thoracoscopically During the Neonatal Period: Clinical Case Report. Medicine (Baltimore) 2015;94:e2270.
11. Gonzalez-Urquijo M, Hinojosa-Gonzalez DE, Padilla-Armendariz DP, et al. Esophageal Duplication Cysts in 97 Adult Patients: A Systematic Review. World J Surg 2022;46:154-62.
12. Castelijns PS, Woensdregt K, Hoevenaars B, et al. Intra-abdominal esophageal duplication cyst: A case report and review of the literature. World J Gastrointest Surg 2014;6:112-6.
13. Wootton-Gorges SL, Eckel GM, Poulos ND, et al. Duplication of the cervical esophagus: a case report and review of the literature. Pediatr Radiol 2002;32:533-5.
14. Birmole BJ, Kulkarni BK, Vaidya AS, et al. Intrathoracic enteric foregut duplication cyst. J Postgrad Med 1994;40:228-30.
15. Bhatia V, Tajika M, Rastogi A. Upper gastrointestinal submucosal lesions--clinical and endosonographic evaluation and management. Trop Gastroenterol 2010;31:5-29.
16. Sonthalia N, Jain SS, Surude RG, et al. Congenital Esophageal Duplication Cyst: A Rare Cause of Dysphagia in an Adult. Gastroenterology Res 2016;9:79-82.
17. Martin ND, Kim JC, Verma SK, et al. Intra-abdominal esophageal duplication cysts: a review. J Gastrointest Surg 2007;11:773-7.
18. Ildstad ST, Tollerud DJ, Weiss RG, et al. Duplications of the alimentary tract. Clinical characteristics, preferred treatment, and associated malformations. Ann Surg 1988;208:184-9.
19. Wiechowska-Kozłowska A, Wunsch E, Majewski M, et al. Esophageal duplication cysts: endosonographic findings in asymptomatic patients. World J Gastroenterol 2012;18:1270-2.
20. Sun CF, Chen CH, Ke PZ, et al. Esophageal duplication cyst presenting with stridor in a child with congenital pulmonary airway malformation: A case report and literature review. Medicine (Baltimore) 2019;98:e16364.
21. Sethi GK, Marsden J, Johnson D. Duplication cysts of the esophagus. South Med J 1974;67:616-8.
22. Singh S, Lal P, Sikora SS, et al. Squamous cell carcinoma arising from a congenital duplication cyst of the esophagus in a young adult. Dis Esophagus 2001;14:258-61.

23. Stringel G, Mercer S, Briggs V. Esophageal duplication cyst containing a foreign body. Can Med Assoc J 1985;132:529-31.
24. Kim YW, Sohn TI, Shim HS, et al. Intra-abdominal esophageal duplication cyst in an adult. Yonsei Med J 2005;46:859-61.
25. Weiss LM, Fagelman D, Warhit JM. CT demonstration of an esophageal duplication cyst. J Comput Assist Tomogr 1983;7:716-8.
26. Diehl DL, Cheruvattath R, Facktor MA, et al. Infection after endoscopic ultrasound-guided aspiration of mediastinal cysts. Interact Cardiovasc Thorac Surg 2010;10:338-40.
27. Santamaria JI, Nunez R, Sanjuan S, et al. Esophageal atresia associated with cystic duplication of the esophagus. Chir Pediatr 1988;29:293-4.
28. Herbella FAM, Tedesco P, Muthusamy R, et al. Patti1Thoracoscopic resection of esophageal duplication cyst. Diseases of the Esophagus 2006;19:132-4.
29. Da Col M, Regamey N, Szavay PO. Thoracoscopic Resection of a Foregut Duplication Cyst with the Use of a 5-mm Stapling Device in an Infant-A Case Report. European J Pediatr Surg Rep 2022;10:e30-2.
30. Obasi PC, Hebra A, Varela JC. Excision of esophageal duplication cysts with robotic-assisted thoracoscopic surgery. JSLS 2011;15:244-7.
31. Yang X, Zong Y, Zhao HY, et al. Complete excision of esophageal bronchogenic cyst by endoscopic submucosal tunnel dissection: a case presentation. BMC Gastroenterol 2019;19:155.

doi: 10.21037/ales-22-43
Cite this article as: Kunisaki C. Esophageal duplication cyst. Ann Laparosc Endosc Surg 2022;7:31.

