

Case Report

Excision of Esophageal Duplication Cyst in Children

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ABSTRACT

We report a rare esophageal duplication cyst in a 9-year-old boy. The patient was referred to our department because of a mediastinal mass detected incidentally on a chest radiograph. He did not have symptoms or a history of disease. He underwent complete resection of the mass through a left posterolateral thoracotomy using thoracoscopy. Histopathological examination revealed an esophageal duplication cyst. Duplications of the alimentary tract are rare congenital malformations. About 10% to 20% of all duplications cysts are esophageal. We report the successful resection of an esophageal duplication cyst. (Jikeikai Med J 2013 ; 60 : 73-6)

Key words : esophageal duplication cyst, posterior mediastinal mass, congenital malformation

INTRODUCTION

Primary mediastinal tumors are rare in children. The mediastinum may be best divided into 3 parts : the anterior, middle, and posterior mediastinum. The posterior mediastinum includes the area from the back of the pericardium to the vertebral bodies and their costovertebral sulci¹. In children, tumors and other abnormalities that are usually noted in the posterior mediastinum are neurogenic neoplasms, lymphomas, esophageal duplication cysts, and bronchogenic cyst¹.

Duplications of the alimentary tract are rare congenital malformations. Ten percent to 20% of all duplications cysts are esophageal^{2,3}. Esophageal duplication is the second most common duplication of the alimentary tract. Most esophageal duplications are located in the lower segment of the right thoracic esophagus³. We report the successful resection of an esophageal duplication cyst in a 9-year-old boy.

CASE REPORT

A 9-year-old boy was referred to our department because of a mediastinal mass discovered incidentally on a plain chest radiograph (Fig. 1). The patient had no history of any disease and had no symptoms. Hematological and biochemical variables were within normal limits. Computed tomography showed a dense, well-defined 6-cm noncalcified mass in the posterior mediastinum (Fig. 2A, B). The mass was surrounded by the trachea, esophagus, aortic arch, superior vena cava, and pulmonary artery. The trachea and esophagus were shifted to the right because of the mass. Computed tomography also revealed a thin-walled cyst with homogenous, low-density contents. Sagittal T2-weighted magnetic resonance imaging confirmed a cystic lesion (Fig. 3). The mass was fluid-filled and had a clear border with surrounding tissue.

The differential diagnosis of the mass preoperatively included bronchogenic cyst and esophageal duplication.

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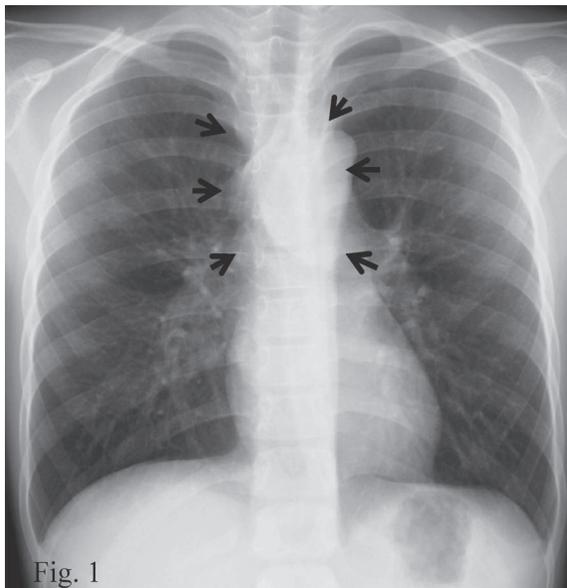


Fig. 1. Initial radiographic findings reveal a mediastinal mass (arrow) and shifted trachea.

We decided to perform surgery because if the mass were malignant it would likely enlarge and produce symptoms. Therefore, we performed thoroscopically guided left posterolateral thoracotomy, because we anticipated difficulty in separating the mass from the superior vena cava and pulmonary artery. The mass was identified with blunt dissection. Intraoperative findings revealed a well-defined, encapsulated mass with a thin wall (Fig. 4A, B). The cyst contained clear fluid that had originated from the esophagus. The mass did not communicate with the esophageal lumen. On the basis of these findings, we diagnosed an esophageal duplication cyst. We resected the cyst along with muscular tunic of the esophagus from inside the cyst. Repair of the esophagus was not necessary.

Histopathological examination showed ciliated epithelial cells with 2 layers of smooth muscle and evidence of an esophageal duplication cyst (Fig. 5). The patient was discharged after 1 week without complications. On follow-up examination 6 months later, the patient had no symptoms or recurrence.

DISCUSSION

Benign congenital cystic lesions in the mediastinum have been referred to by various names, including bronchogenic cyst, esophageal duplication cyst, enteric cyst, and

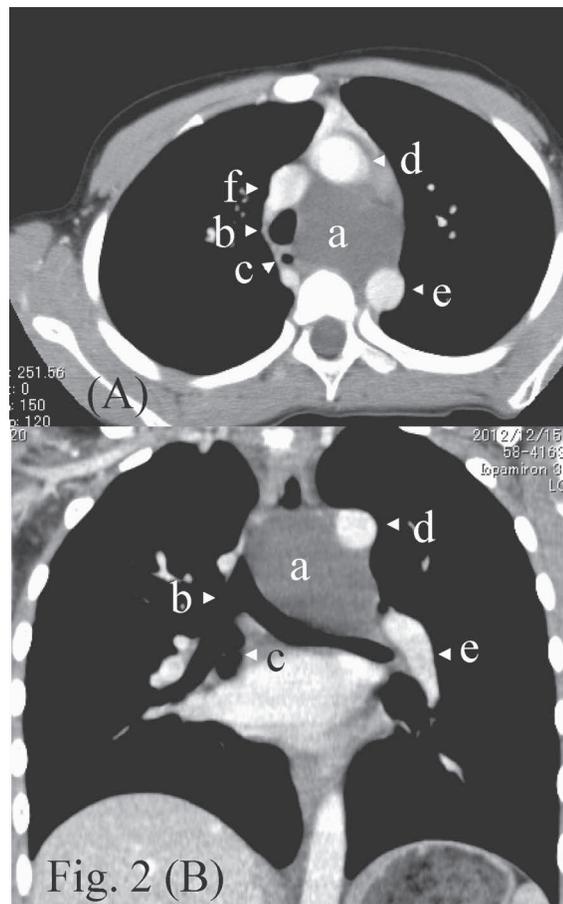


Fig. 2. A. A chest computed tomography scan demonstrates a cystic mass, measuring 6 × 6 cm. The mass is surrounded by the trachea, esophagus, aortic arch, superior vena cava, and pulmonary artery. B. A coronal image shows the trachea shifted to the right by the mass (a, mass; b, trachea; c, esophagus; d, ascending aorta; e, descending aorta; f, superior vena cava).

neuroenteric cyst. They are all believed to represent congenital abnormalities that arise during division of the primitive embryonic foregut¹. Esophageal duplication cysts result from the failure of the originally solid esophagus to become completely vacuolated to produce a hollow tube during the fifth to eighth week of embryonic life^{4,5}. Esophageal duplication is divided into 3 types: cystic, tubular, and diverticular³. Most duplications are cystic³. The overall incidence of this abnormality is estimated to be 1 per 8,200 births, with a predominance in males⁵. Approximately 80% of cysts do not communicate with the esophageal lumen⁶.

Histological criteria for diagnosis of an esophageal duplication cyst include: 1) the cyst is located within or at-

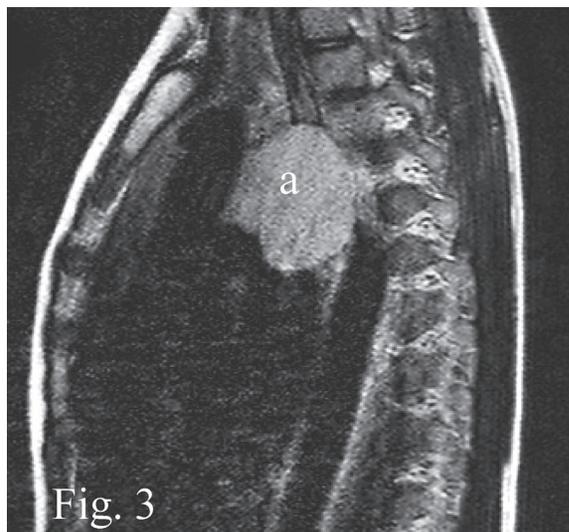


Fig. 3 Sagittal T2-weighted magnetic resonance image reveals a well-circumscribed mass in the posterior mediastinum.



Fig. 5 Histopathological examination shows ciliated epithelial cells with a smooth muscle layer and evidence of an esophageal duplication cyst (bar = 50 µm).

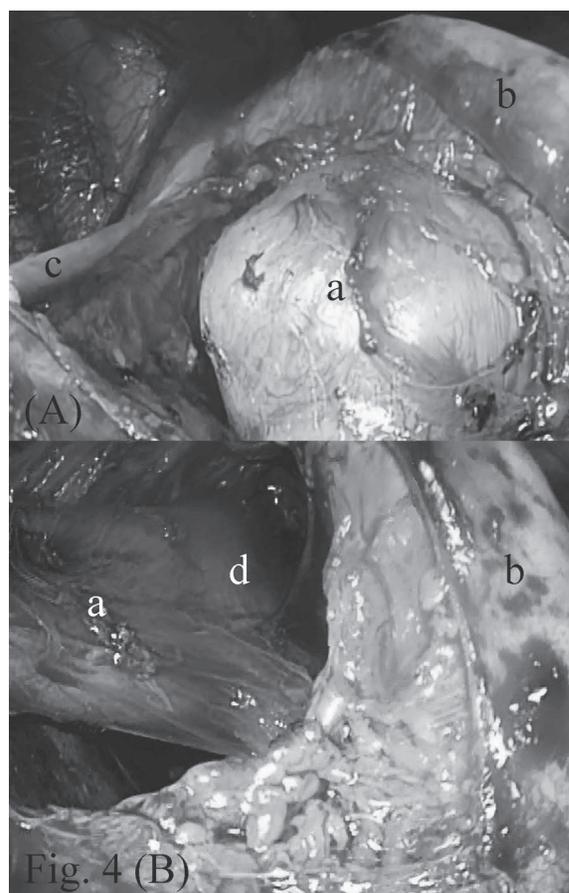


Fig. 4 Thoracoscopic view of the operative field reveals a well-defined, encapsulated mass. A. The mass is surrounded by the aortic arch. B. The mass is connected to the esophagus (a, mass ; b, aortic arch ; c, vagus nerve ; d, esophagus).

tached to the esophageal wall, 2) the cyst is covered by 2 muscular layers, and 3) the cyst contains squamous epithelium or a lining found in the embryonic esophagus^{7,8}.

Esophageal duplication can be associated with other congenital anomalies, such as small-intestinal duplication, esophageal atresia, scoliosis, hemivertebrae, and fusion⁹. In two-thirds of cases, symptoms appear with the development of complications, such as infection, mass effects, bleeding, neoplasms, and esophageal or respiratory obstruction³. Malignancy within esophageal duplication cysts is rare but has been reported^{10,11}.

Adenocarcinoma is the most common histologic type of malignant transformation^{10,11}. The preferred treatment is complete surgical resection^{12,13}. Posterior mediastinal masses are generally approached through thoracotomy or video-assisted thoracic surgery. Mediastinal tumors may involve adjacent structures, including nerves, the vena cava, or pericardium. These structures can generally be resected or reconstructed if the tumor is malignant.

Authors have no conflict of interest.

REFERENCES

- Jagers J, Balsara K. Mediastinal masses in children. *Thorac Cardiovasc Surg.* 2004 ; 16 : 201-8.
- Sethi GK, Marsden J, Johnson D. Duplication cysts of the esophagus. *South Med J.* 1974 ; 67 : 616-8.

3. Nazem M, Amouse AB, Eidy M, Khan IA, Javed HA. Duplication of cervical oesophagus : A case report and review of literatures. *Afr J Paediatr Surg.* 2010 ; 7 : 203-5.
4. Gupta B, Meher R, Raj A, Agarwal S. Duplication cyst of oesophagus : a case report. *J Pediatr Child Health.* 2010 ; 46 : 134-5.
5. Karwan WO, Walbaum PR, McCormack RJ. Cystic intrathoracic derivatives of the foregut and their complications. *Thorax.* 1973 ; 28 : 424-30.
6. Nayan S, Nguyen LHP, Nguyen VH, Daniel SJ, Emil S. Cervical esophageal duplication cyst : case report and review of the literature. *J Pediatr Surg.* 2010 ; 45 : 1-5.
7. Wootton-Gorges SL, Eckel GM, Poulos ND, Kappler S, Milstein JM. Duplication of cervical esophagus : a case report and review of the literature. *Pediatr Radiol.* 2002 ; 32 : 533-5.
8. Kolomainen D, Hurley PR, Ebbs SR. Esophageal duplication cyst : case report and review of the literature. *Dis Esophagus.* 1998 ; 11 : 62-5.
9. Nakano A, Urushihara N, Yagi T, Choda Y, Hamada M, Kataoka K, et al. Rapidly enlarging esophageal duplication cyst. *J Gastroenterol.* 1999 ; 34 : 246-9.
10. Jacob R, Hawkes ND, Dallimore N, Butchart EG, Thomas GA, Maughan TS. Case report : Squamous carcinoma in an oesophageal foregut cyst. *Br J Radiol.* 2003 ; 76 : 343-6.
11. Dai ZJ, Kang HF, Lin S, Bai MH, Ma L, Min WL, et al. Esophageal cancer with esophageal duplication cyst. *Ann Thorac Surg.* 2013 ; 96 : 15-6.
12. Hazelrigg SR, Landreneau RJ, Mack MJ, Acuff TE. Thoracoscopic resection of mediastinal cysts. *Ann Thorac Surg.* 1993 ; 56 : 659-60.
13. Martin ND, Kim JC, Verma SK, Rubin R, Mitchell DG, Bergin D, et al. Intra-abdominal esophageal duplication cysts : a review. *J Gastrointest Surg.* 2007 ; 11 : 773-7.