

Esophageal diverticulum serves as a unique cause of bronchoesophageal fistula in children

A case report

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Abstract

Rationale: Most of the esophageal diverticula are congenital traction instead of in childhood. In most conditions, esophageal diverticula are free of any symptoms. As one of the rare entity, esophageal diverticulum can also result in bronchoesophageal fistula.

Patient concerns: A 10-year-old girl was admitted due to a 2-month history of cough and choking after drinking, and fever for 3 days. No symptoms when taking solid food were found.

Diagnoses: By esophagogram, 3-dimensional computed tomography and esophagoscopy, an esophageal diverticulum was demonstrated in the middle esophagus with a bronchoesophageal fistula visualized. Then the diagnoses of esophageal diverticulum and bronchoesophageal fistula were established.

Interventions: A regular trans-antrolateral thoracotomy was carried out under general anesthesia with patient lying on the right side. The diverticulum was then removed and the fistulous tract was closed.

Outcomes: The girl discharged on the 14th postoperative day and received a regular monthly follow-up, at present, no recurrence was found.

Lessons: Bronchoesophageal fistula may be a complication of esophageal diverticula, and should be considered in cases of unexplained cough or recurrent pneumonia.

Keywords: children, esophageal diverticulum, esophageal fistula

1. Introduction

Most of the esophageal diverticula are traction instead of congenital in childhood. In most conditions, esophageal diverticula are free of any symptoms. As one of the rare entity, esophageal diverticulum can also result in bronchoesophageal fistula.^[1–4] At present, <50 cases of this nature have been reported so far, most of patients are adults instead of children, esophageal diverticulum with bronchoesophageal fistula in the

adult cases are mostly acquired.^[5–7] In this study, esophageal diverticulum with bronchoesophageal fistula in a pediatric case was reviewed and presented.

2. Ethics and consent

The use of history records and clinical data was reviewed and approved by the ethics committee of the Shengjing Hospital of China Medical University. There were no discernibly informations included in the figures and photos as a result that the written informed consent was not obtained from the patient's guardian.

3. Case presentation

A 10-year-old girl was admitted due to a 2-month history of cough and choking after drinking, and fever for 3 days. No symptoms when taking solid food were found. The physical and laboratory examination tests were both normal. The computed tomography scans of chest showed that the leaf segment bronchus opening unobstructed, with a piece of patchy shadow of high-density locating in the left inferior lung. Three-dimensional computed tomography presented a piece of air image located in the mediastinum near the left middle bronchus, which linked the left wall of the esophagus. A fistula between the esophagus and bronchus was clearly demonstrated. After taking oral contrast media of gastrografin, the bilateral principal bronchus and back segment of the left inferior lung were presented which provided a strong evidence for bronchoesophageal fistula existence (Fig. 1A–C). The esophagoscopy revealed a diverticulum 32 cm from the incisor line, 0.5 × 0.5 cm in size, in

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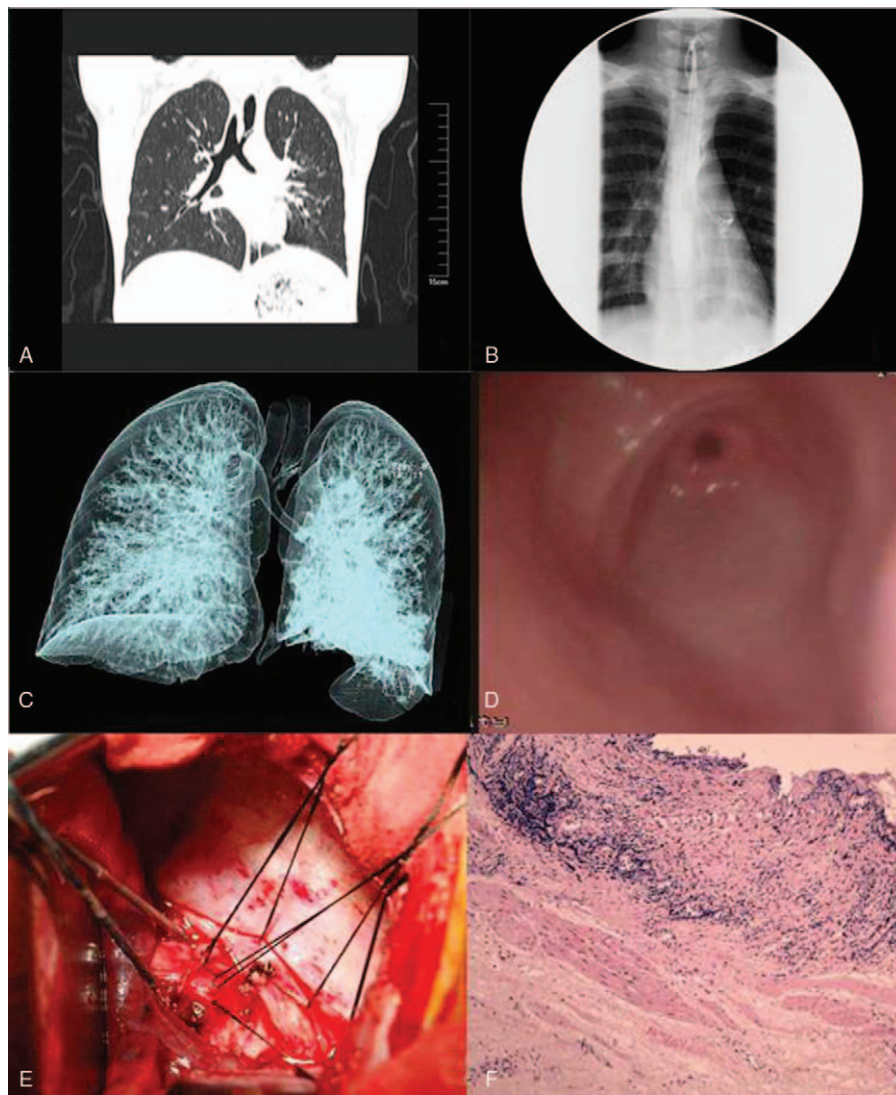


Figure 1. A–C, Esophageal radiography and 3-dimensional computed tomography showed that there was a fistula in the left antetheca of the mid-esophageal wall which communicated with the bronchus. D, Esophagoscopy revealed a diverticulum with a small orifice at the bottom of esophageal left anterior wall. E, A diverticulum with bronchoesophageal fistula was confirmed between the middle esophagus and the left bronchus. F, Histological diagnosis demonstrated the mucosa, submucosa, and muscular layer of the diverticulum.

the left lateral esophagus, and a fistula, 0.2×0.2 cm in size, with oyster white secreta nearby (Fig. 1D). A thoracotomy was finally carried out after the consent from the parents. The thoracotomy was transanterolateral under general anesthesia with patient lying on the right side. During the thoracotomy, there was a diffuse pleural adhesion in the thoracic cavity. After blunt dissection the pleura, a fistula between the esophagus and the back segment of the left inferior lung was verified (Fig. 1E). The diverticulum was then removed and the fistulous tract was closed. This case was finally diagnosed as esophageal diverticulum with bronchoesophageal fistula. The histological pathology further confirmed the diagnosis (Fig. 1F). The girl discharged on the 14th postoperative day and received a regular monthly follow-up, at present, no recurrence was found.

4. Discussion

Bronchoesophageal fistula is an abnormal communication between the esophagus and trachea. Most of bronchoesophageal

fistula in children is congenital and often associated with esophageal atresia or other malformations of esophagus.^[8–10] The overall incidence of congenital bronchoesophageal fistula ranges from 1 in every 2500 to 4000 live births.^[11] Only a few bronchoesophageal fistulas are acquired. In the present case, only 2 months have the symptoms of cough and choking been found for, and the girl was always disease free in most of time after birth indicating bronchoesophageal fistula is prone to acquired. The acquired bronchoesophageal fistula maybe caused by trauma, the inflammatory, the neoplastic, and the ingested foreign body.^[5–7] The battery or coins ingestion may cause delayed onset burns in the trachea and esophagus leading to the acquired esophagus diverticulum with bronchoesophageal fistula. Nevertheless, the risk factors of bronchoesophageal fistulas in this case were not the elements mentioned above.

As a unique cause of acquired bronchoesophageal fistula, the esophageal diverticulum has been seldom reported.^[3,5,8] By reviewing the literature, esophageal diverticulum is an evagination of the mucous projecting from the lumen of the esophagus,

with a prevalence of approximately 0.06% to 4%, and it primarily affects the elderly.^[2] Esophageal diverticulum can be characterized according to their pathogenesis as either traction or pulsion or congenital. Most of the reported diverticula are traction diverticula, it is an external inflammatory reaction in neighboring mediastinal ganglia that adhere to the esophagus and retract the wall in a way that contracts and scars the tissue. As a result of inflammatory necrosis, a middle esophageal diverticulum can cause complications when it grows into neighboring structures with the posterior appearance of fistula to the respiratory tree.^[3] The esophageal diverticulum can also be pulsion diverticulum, which occur with increased pressure against the upper and lower esophageal sphincters, respectively. In this case, the history was carefully reviewed and none of the mentioned acquired factors were obtained; therefore, we inferred that diverticulum in this child might be congenital. The bronchoesophageal fistula in this case can be secondary to esophageal diverticulum due to the inflammatory and food accumulation, which are extremely rare among children.

The anatomy of bronchoesophageal fistula can be shown through the preoperational examinations. Conventional barium esophagography is considered to be the most sensitive test for diagnosing bronchoesophageal fistula. The same happened in the present girl. As soon as the barium esophagography was carried out, the bronchoesophageal fistula was clearly demonstrated. However, barium esophagography can give little clue for the cause of bronchoesophageal fistula besides its existence. As a result, other valuable examinations such as 3-dimensional computed tomography of the mediastinum and esophagoscopy must be carried out. Three-dimensional computed tomography and esophagoscopy have recently become popular clinical examination tools with significant improvement, which was made on quality of image due to a rapid progress in computer technology.^[4,7,11] By these 2 examinations, the bronchoesophageal diverticulum with a fistula could be clearly demonstrated providing a direct evidence for the diagnosis in this case. Consequently, 3-dimensional computed tomography and esophagoscopy should be used as routine methods in the patients of bronchoesophageal fistula because of their noninvasive features. Usually, a thoracotomy with the diverticulectomy is advocated in the case of esophagus diverticulum with bronchoesophageal fistula.^[12,13] The same was followed in this case after

the consent from the parents. The result was satisfied after a regular follow-up.

Limited and tentative conclusions are now possible, based on our experience and that of others, clinicians should be aware of esophageal diverticulum as a unique entity of bronchoesophageal fistula, although encountered rarely in clinical settings, and consider it in each case of unexplained cough and recurrent pneumonia.

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