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A rare case of congenital bronchoesophageal fistula in an adult

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ABSTRACT

INTRODUCTION: When congenital bronchoesophageal fistulas exist without atresia of the esophagus, the diagnosis can be delayed, although symptoms may occur early following fistula development. Therefore, while they are usually found in infants, they can be extremely rarely found in adults. We herein report a rare case of bronchoesophageal fistula without atresia in an adult.

CASE: An 69-year-old male presented to the outpatient clinic with a decades-long history of cough with expectoration immediately after taking food, especially liquids.

Computed tomograph, esophagoscopy, and esophagography revealed the fistulous communication between the mid-esophagus and right lower lobe bronchus, with consolidation in the right lower lobe. We performed right lower lobectomy with the closure and excision of the fistula. The histopathology of the fistula revealed the mucosa to be lined by stratified squamous epithelium. There was no evidence of inflammation, granuloma, or carcinoma.

CONCLUSION: In conclusion, despite the benign nature of this malformation, if left untreated, it can cause long-term debilitating respiratory symptoms associated with the fistula. Therefore, the diagnosis should be considered in the evaluation of recurrent lung infection.

1. Introduction

Congenital bronchoesophageal fistulas have a reported incidence of 1 in 3000–4000 live births. They are usually found in infants, whereas they can be extremely rarely found in adults in cases of congenital bronchoesophageal fistulas which exist without atresia of the esophagus. We herein report a rare case of bronchoesophageal fistula without atresia in an adult, based on Surgical Case Report (SCARE) Guidelines [1].

2. Case

An 69-year-old male presented to the outpatient clinic with a decades-long history of cough with expectoration immediately after taking food, especially liquids.

Based on computed tomography (CT) findings (Fig. 1A and B), we suspected the fistulous communication between the mid-esophagus and right lower lobe bronchus and consolidation in the right lower lobe, especially S6 segment. Upper gastrointestinal endoscopy showed a fistulous opening in the mid esophagus without any evidence of malignancy, granulomatous disease or any other acquired basis for the fistula (Fig. 1C), although bronchoscopy showed edema of the right lower bronchus and failed to show the fistula. Esophagography confirmed the presence of a bronchoesophageal fistula (Fig. 1D). We planned to perform right lower lobectomy to close the fistula. Based on the intraoperative findings, although extensive adhesions were found around the right lower lobe, the adhesion was only mild around the fistulous site (Fig. 2A), which was confirmed by intraoperative endoscopy and identified above the level of the inferior pulmonary vein. The tract was closed with silk thread and excised (Fig. 2B), and the esophageal mucosa was closed by inverted sutures with 4–0 PDS (Fig. 2C). Subsequently, right lower lobectomy was performed. There were no intraoperative complications, and the patient had an uneventful recovery. The histopathology of the fistula revealed the mucosa to be lined by stratified squamous epithelium (Fig. 2D). There was no evidence of inflammation, granuloma, or carcinoma.

3. Discussion

When congenital bronchoesophageal fistulas exist without atresia of the esophagus, the diagnosis can be delayed, although symptoms may occur early following fistula development. Typical symptoms include a cough, and recurrent or chronic lung infec-

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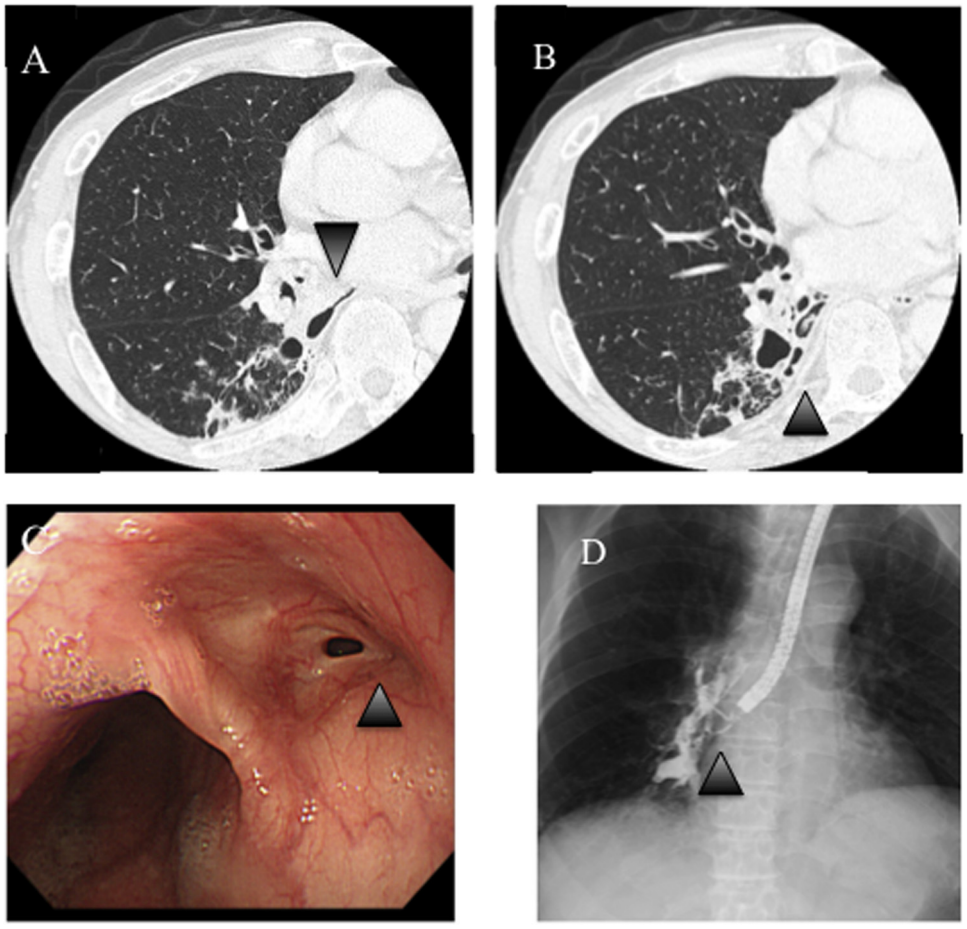

Fig. 1. (A): CT scan showing suspicion of the fistula between the right B6 peripheral bronchus and esophagus (arrow heads). (B): CT scan showing demonstrating consolidation in the right lower lobe (arrow heads). (C): Fistula in the right lateral wall of the esophagus (arrow heads). (D): Esophagography confirming the presence of the bronchoesophageal fistula (arrow heads).

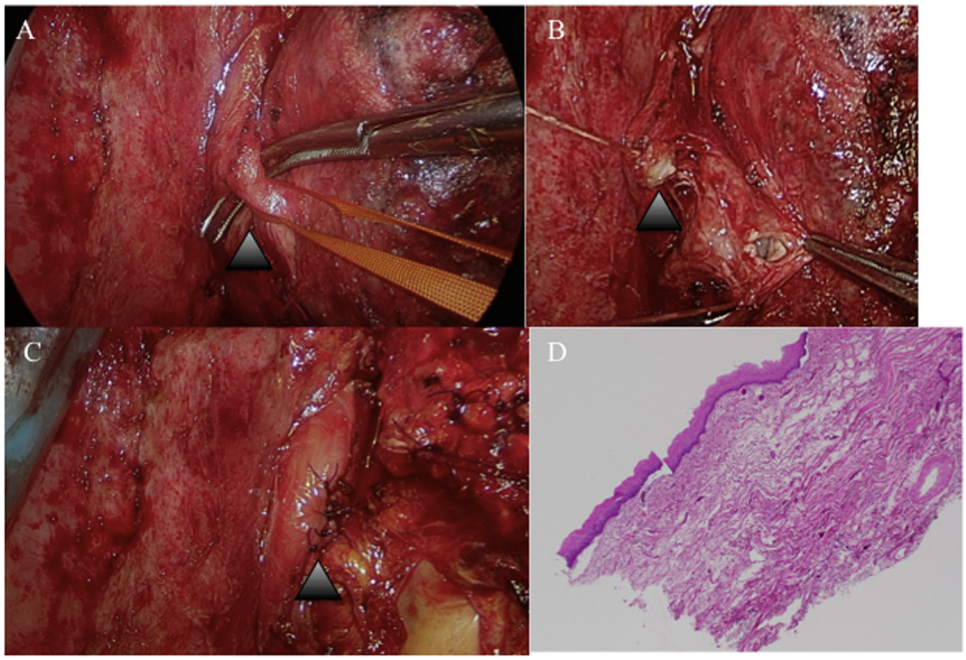

Fig. 2. (A): Intraoperative view of the tracheoesophageal fistula (arrow head). (B): Closure and division of the fistula tract (arrow head). (C): Intraoperative view at the end of the operation (arrow head). (D): Microscopic appearance of the wall of the fistula, showing that it is covered with striated squamous epithelium. There are no evidence of inflammation, granuloma, or carcinoma. (hematoxylin and eosin [HE] stain).

tions [2–6]. Chest CT may not confirm the fistula in the case of ling tissue destroyed by chronic lung infection. Therefore, examinations, such as bronchoscopy or esophagoscopy, are recommended for evaluating cases of recurrent lung infection. Esophagography, especially when performed with the patient lying prone, is essential for confirming the presence of a bronchoesophageal fistula. The differential diagnoses of an acquired fistula, which include malignancy, chronic inflammatory process, or even tuberculosis, should be excluded [7]. In our case, upper gastrointestinal endoscopy revealed a fistulous opening in the mid-esophagus and esophagography confirmed the presence of a fistula in the right bronchus B6. The most common communicated location of the fistula is the right bronchus B6, according to previous reports [8].

Surgical treatment is generally recommended for controlling symptoms and preventing pulmonary damage. With regard to the method, damaged lung resection is frequently needed in addition to ligation and division of the fistula tract. It may occasionally be difficult to locate the fistula, due to intrathoracic adhesion. In our case, intraoperative esophagoscopy helped detect the fistula. The prognosis after surgical treatment is typically excellent, according to previous reports [9,10].

In conclusion, despite the benign nature of this malformation, if left untreated, it can cause long-term debilitating respiratory symptoms associated with the fistula. Therefore, the diagnosis should be considered when evaluating cases presenting with recurrent lung infection.

Conflict of interest statement

Naohiro Taira and the other co-authors have no conflicts of interest and relevant financial interests to declare in this manuscript.

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Ethical approval

All approval has been given.

Consent

The consent has been given.

Authors contribution

Naohiro Taira: writing the paper, Hidenori Kawasaki: study concept.

Eriko Atsumi: dataanalysis, Tomonori Furugen: data collection.

Takaharu Ichi: data collection, Kazuaki Kushi: data collection, Tomofumi Yohena: study design.

Tomoo Baba: data analysis, Tsutomu Kawabata: study design.

Registration of research studies

This is not human research.

Guarantor

Naohiro Taira, Hidenori Kawasaki.

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