CASE REPORT

Congenital tracheoesophageal fistula successfully diagnosed by CT esophagography

Koichi Nagata, Yoshito Kamio, Tamaki Ichikawa, Mitsutaka Kadokura, Akihiko Kitami, Shungo Endo, Haruhiro Inoue, Shin-Ei Kudo

Abstract

Tracheoesophageal fistula (TEF) or bronchoesophageal fistula may be congenital, inflammatory, neoplastic, or secondary to trauma. Congenital TEF or bronchoesophageal fistula is usually associated with esophageal atresia and is readily diagnosed in infancy. But if it is not associated with esophageal atresia, it may persist until adulthood. Some theories have been proposed to explain this delay in diagnosis. The long, silent interval until adulthood and the irregular character of the signs have been attributed to the presence of a membrane that subsequently ruptures, to the proximal fold of esophageal mucosa initially overlapping the orifice but subsequently becoming less mobile, and to the fact that the fistulous tract runs upward and may close during swallowing.

We present a case of a 70-year-old man with congenital bronchoesophageal fistula.

CASE REPORT

A 70-year-old man was admitted to our hospital with the complaint of severe cough and choking after swallowing liquid. The patient had done well until 7 mo prior to the admission. Then increased symptoms of cough and dysphagia were developed. Esophagoscopy showed a depressed lesion that looked as if it was punched without inflammation at a distance of 23 cm from the incisors in the middle intrathoracic esophagus, but the fistula opening was invisible (Figure 1). Histological examination of the esophageal biopsy specimens had only normal findings. A multidetector-row CT scan using Aquilion M8 (Toshiba, Tokyo, Japan) was performed with 3-dimensional reconstruction. A nasogastric tube was inserted into the esophagus during the examination, and the esophagus was inflated with oxygen flow (1L/min). The CT scanning parameters were 120 kVp, 200 mA, 8 × 0.5 mm collimation, a pitch of 10.0, and a 0.3-mm reconstruction interval. CT esophagography revealed the presence of a TEF (Figure 2). Conventional barium esophagography was not performed for the patient.

The patient underwent a right posterior lateral thoracostomy. The fistula was identified between the middle intrathoracic esophagus and the distal trachea. There was no evidence of inflammation or adherent lymph nodes around the fistula. The fistula was divided and closed using endostapler (EndoGIA Universal stapler®; US Surgical Corporation, Norwalk, Conn.), and covered with an intercostal muscle flap to avoid the recurrence of fistula.
operative esophagoscopy was found to be useful for the definitive localization and complete excision of the fistula and the avoidance of stenosis of the esophagus. Pathological examination of the resected specimen revealed that the fistula was lined by benign squamous epithelium with the muscularis mucosa, and there was no evidence of malignancy, infection, or chronic inflammation (Figure 3). The postoperative course was uneventful. At 18 mo follow-up, there was no clinical or radiographic evidence of recurrence of the fistula.

DISCUSSION

Congenital TEF or bronchoesophageal fistulas were first reported by Negus in 1929 and Gibson in 1696[8]. These congenital fistulas are still controversial and rare clinical problem in adults. Generally, the criteria for the diagnosis of a congenital fistula in adults has the following features: absence of past or present surrounding inflammation or malignancy, complete recovery after resection, and the presence of normal mucosa and absence of adherent lymph nodes[1,2,7]. We could confirm our case as a congenital TEF by the above mentioned features, even though it was found in a 70-year-old man.

The anatomy of TEF should be shown before surgery because the surgical approach depends on a correct evaluation of TEF. Conventional barium esophagography is considered to be the most sensitive test for diagnosing tracheoesophageal or bronchoesophageal fistula[9,10]. Other examinations that have been used but are less successful include esophagoscopy, bronchoscopy, and bronchography[9,10]. Three-dimensional (3D) displays of many organs and structures using multidetector-row CT have recently become a popular clinical examination tool with significant improvement which was made on quality of image due to a rapid progress in computer technology[11-13]. Multidetector-row CT esophagography correctly showed TEF in our case. Images of TEF could be easily made and provide crucial information for planning surgery. Fitoz et al report-
ed that 3D CT imaging are used to assist in the diagnosis of TEF\[14\].

Fitoz’s and our experience indicates CT esophagography might be clinically useful for the accurate diagnosis of tracheoesophageal fistula. CT esophagography without contrast medium is less invasive examination.

REFERENCES

1. Risher WH, Arensman RM, Ochsner JL. Congenital bronchoesophageal fistula. Ann Thorac Surg 1990; 49: 500-505
2. Azoulay D, Regnard JF, Magdeleinat P, Diamond T, Rojas-Miranda A, Levasseur P. Congenital respiratory-esophageal fistula in the adult. Report of nine cases and review of the literature. J Thorac Cardiovasc Surg 1992; 104: 381-384
3. Zach MS, Eber E. Adult outcome of congenital lower respiratory tract malformations. Thorax 2001; 56: 65-72
4. Braimbridge MV, Keith HI. OESOPHAGO-BRONCHIAL FISTULA IN THE ADULT. Thorax 1965; 20: 226-233
5. Acosta JL, Battersby JS. Congenital tracheoesophageal fistula in the adult. Ann Thorac Surg 1974; 17: 51-57
6. Demong CV, Grow JB, Heitzman GC. Congenital tracheoesophageal fistula without atresia of the esophagus. Am Surg 1959; 25: 156-162
7. Brunner A. Esophagobronchial fistulae. Munch Med Wochenschr 1961; 103: 2181-2184
8. Lansden FT, Falor WH. Congenital esophago respiratory fistula in the adult. J Thorac Cardiovasc Surg 1960; 39: 246-251
9. Hendry P, Crepeau A, Beatty D. Benign bronchoesophageal fistulas. J Thorac Cardiovasc Surg 1985; 90: 789-791
10. Kim JH, Park KH, Sung SW, Rho JR. Congenital bronchoesophageal fistulas in adult patients. Ann Thorac Surg 1995; 60: 151-155
11. Lou MW, Hu WD, Fan Y, Chen JH, E ZS, Yang GF. CT biliary cystoscopy of gallbladder polyps. World J Gastroenterol 2004; 10: 1204-1207
12. Nagata K, Endo S, Kudo SE, Kitanosono T, Kushihashi T. CT air-contrast enema as a preoperative examination for colorectal cancer. Dig Surg 2004; 21: 352-358
13. Ohtani H, Kawajiri H, Arimoto Y, Ohno K, Fujimoto Y, Oba H, Adachi K, Hirano M, Terakawa S, Tsubakimoto M. Efficacy of multislice computed tomography for gastroenteric and hepatic surgeries. World J Gastroenterol 2005; 11: 1532-1534
14. Fitoz S, Atasoy C, Yagmurlu A, Akyar S, Erden A, Dindar H. Three-dimensional CT of congenital esophageal atresia and distal tracheoesophageal fistula in neonates: preliminary results. AJR Am J Roentgenol 2000; 175: 1403-1407