Clinical Case Report

Medicine®

OPEN

Recurrent tracheoesophageal fistula in an adolescent without persistent symptoms

A case report

Jun-Hong Lin, MD[a], Li Deng, MD[a], Xing Li, MD[b,*]

Abstract

Rationale: Congenital esophageal atresia (EA) and tracheoesophageal fistula (TEF) ininfants have been treated well with surgery. Approximately 10% of children displayed recurrent fistula. In the present case, we reported recurrent TEF in an adolescent as a complication of EA/TEF in infancy.

Patient concerns: An infant was diagnosed with gross type C congenital EA and TEF and subsequentlyunderwent repair in early infancy, with division of the TEF and primary esophageal anastomosis. Postoperative esophageal strictures developed and were relieved by bougienage of the esophagus partially. Then, the child had normal growth with mild symptoms, mainly choking when drinking water. At 11 years of age, the child developed fever and cough, and massive bronchiectasis in lobus inferior pulmonis sinister was found.

Diagnosis: Recurrent tracheoesophageal fistula.

Interventions: Division of the TEF and esophageal replacement with gastric tube was performed as treatment

Outcomes: The child recovered well.

Lessons: Recurrent tracheoesophageal fistula aftercongenital EA and TEF could be diagnosed in adolescence. Massive bronchiectasis might develop without apparent symptoms.

Abbreviations: CT = computed tomography, EA = esophageal atresia, TEF = tracheoesophageal fistula.

Keywords: congenital esophageal atresia, esophageal atresia, tracheoesophageal fistula

1. Introduction

Congenital esophageal atresia (EA) and tracheoesophageal fistula (TEF) in infants have been treated well with surgery.[1] Approximately 10% of children with a history of EA and TEF display recurrent fistula.[2] Notably, the recurrent fistula in adolescents was very scarce, since its relevant symptoms are apparent. In the present case, we report a recurrent TEF in an adolescent as a complication of EA/TEF in infancy. The child displayed milder symptoms and normal growth, which delayed the diagnosis of this complication.

2. Case presentation

An infant was diagnosed with gross type C congenital EA and TEF and subsequently underwent repair in early infancy, with division of the TEF and primary esophageal anastomosis. Postoperative esophageal strictures developed and were relieved by bougienage of the esophagus partially. Then, the child had normal growth with occasional mild symptoms, mainly choking when drinking water. At 11 years of age, the child developed fever and cough that were not relieved after routine treatment. Subsequently, computed tomography (CT) scan illustrated massive bronchiectasis in the lobus inferior pulmonis sinister (Fig. 1A). In order to evaluate the bronchiectasis, bronchofiberoscopy was performed, and a TEF was found. Methylenum coeruleum stain and passage of barium confirmed TEF (Fig. 1B–D). Division of the TEF and esophageal replacement with gastric tube was performed as treatment, and the child recovered well. This study was approved by the Clinical Ethics Review Board at Guangzhou Women and Children's Medical Center. A written informed consent was obtained from the patient and the parents at the time of admission.

3. Discussion

The incidence of recurrent TEF was reported to be 5% to 14% after surgical repair of esophageal atresia.[2] It usually develops

Editor: N/A.

This study was supported by the National Clinical Key Specialty Project Foundation, 2015 National Health and Family Planning Commission Special Fund (201502025) and Natural Science Foundation of Guangdong (2016A030313302, 2016A030311046, and 2017A030313537), Guangzhou Science and Technology Plan (201709010030), and National Natural Science Foundation of China (31600710 and 81700645).

Take-away message: Recurrent TEF after congenital EA and TEF could be diagnosed in adolescence. Massive bronchiectasis might develop without apparent symptoms.

The authors have no conflicts of interest to disclose.

[a] Department of Respiration, Guangzhou Women and Children's Medical Center, Guangzhou Medical University, [b] Department of Medical Oncology, the Third Affiliated Hospital of Sun Yat-sen University, Guangzhou, People's Republic of China.

[*] Correspondence: Xing Li, Department of Medical Oncology, the Third Affiliated Hospital of Sun Yat-sen University, 600 Tianhe Road, Guangzhou 510630, People's Republic of China (e-mail: lixing9@mail.sysu.edu.cn).

Copyright © 2017 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the Creative Commons Attribution-NoDerivatives License 4.0, which allows for redistribution, commercial and non-commercial, as long as it is passed along unchanged and in whole, with credit to the author.

Medicine (2017) 96:46(e8668)

Received: 2 September 2017 / Received in final form: 24 October 2017 / Accepted: 26 October 2017

http://dx.doi.org/10.1097/MD.0000000000008668

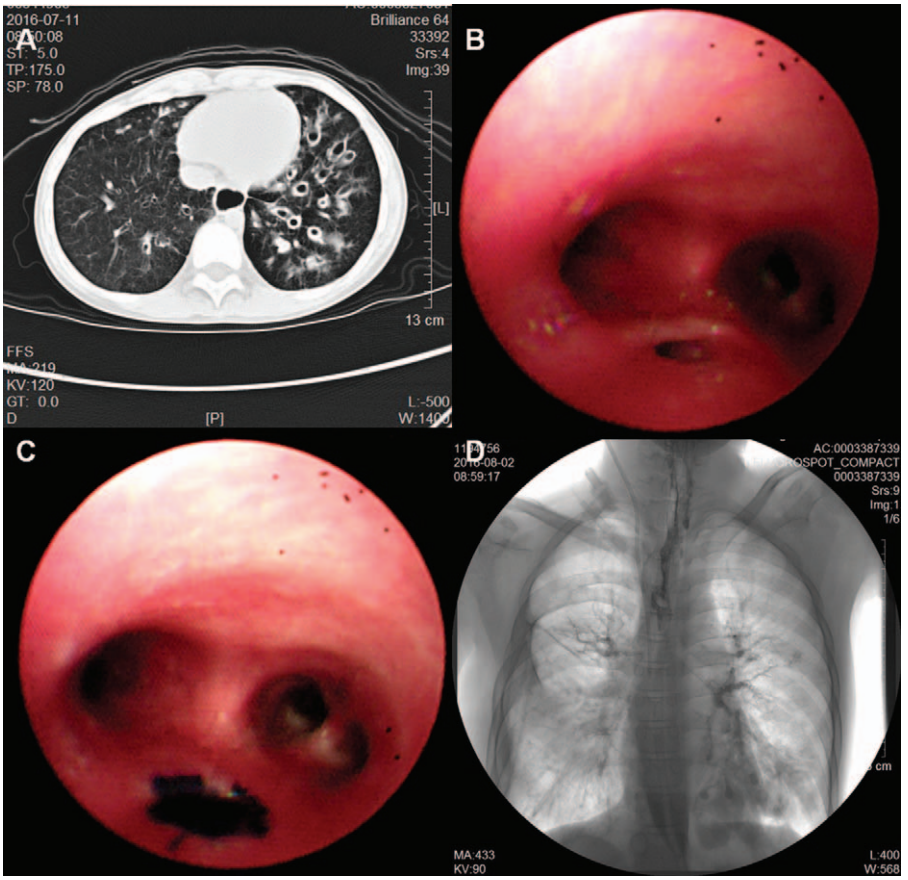

Figure 1. Images of recurrent tracheoesophageal fistula in an adolescent. Computed tomography (A), bronchofiberoscopy (B), methylenum coeruleum staining (C), and passage of barium (D) confirmed tracheoesophageal fistula.

within 18 months after the initial surgery with rare cases developing years later.[3] Currently, this complication is likely to decrease with the improvement of surgical instruments and imaging and surgical techniques. In the present case, the adolescent was diagnosed with recurrent TEF 11 years after the initial surgery. The recurrent fistula was not a second congenital fistula located in the pouch of the original fistula, which was suggested by bronchoscopy prior to the original TEF surgery. Recurrent fistula is more common after a primary esophageal repair under tension,[4] with ligation instead of complete division. Recurrent TEF may be suspected when a child presents with repeated lower respiratory tract infections with a history of EA. The adolescent in our study presented mild symptoms that delayed the diagnosis. However, the delay did not affect the growth of the adolescent. At 11 years of age, persistent symptoms of pneumonia and massive bronchiectasis lead to the performance of bronchofiberoscopy, which finally diagnosed the recurrent TEF.

This is a rare case of recurrent TEF, whose symptoms were very mild and did not influence the development of the adolescent. However, massive bronchiectasis developed in this adolescent. Thus, bronchofiberoscopy for children with history of EA might be necessary for early detection of recurrent TEF even years after surgery.

References

[1] Wang B, Tashiro J, Allan BJ, et al. A nationwide analysis of clinical outcomes among newborns with esophageal atresia and tracheoesophageal fistulas in the United States. J Surg Res 2014;190:604–12.
[2] Holland AJ, Fitzgerald DA. Oesophageal atresia and tracheo-oesophageal fistula: current management strategies and complications. Paediatr Respir Rev 2010;11:100–6.
[3] Bruch SW, Hirschl RB, Coran AG. The diagnosis and management of recurrent tracheoesophageal fistulas. J Pediatr Surg 2010;45:337–40.
[4] Kovesi T. Long-term respiratory complications of congenital esophageal atresia with or without tracheoesophageal fistula: an update. Dis Esophagus 2013;26:413–6.