Radiology Case Reports 18 (2023) 3672–3674

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Congenital esophageal stenosis caused by tracheobronchial remnants: A case report ☆

Soung Hee Kim, MD

Department of Radiology, Sanggye Paik Hospital, Inje University College of Medicine, 1342 Dongil-ro, Nowon-gu, Seoul 01757, Korea

ARTICLE INFO

Article history:
Received 12 April 2023
Revised 18 June 2023
Accepted 23 July 2023

Keywords:
Congenital esophageal stenosis
Congenital esophageal stricture
Tracheobronchial remnants
Esophagography

ABSTRACT

Congenital esophageal stenosis caused by tracheobronchial remnants is a rare anomaly in infants and children that results in dysphagia and recurrent pneumonia. An accurate diagnosis is needed because this anomaly does not respond well to instrumental dilatation, such as bougienage and balloon dilatation, and requires surgical resection. Herein, I report a case of congenital esophageal stenosis caused by tracheobronchial remnants with characteristic imaging findings.

Introduction

Congenital esophageal stenosis caused by tracheobronchial remnants is a rare anomaly in infants and children [1–4]. Such patients usually present with dysphagia and recurrent vomiting leading to aspiration pneumonia, particularly after ingestion of solid food [5]. This case report describes the characteristic radiological and pathological findings of congenital esophageal stenosis caused by tracheobronchial remnants.

Case report

A 20-month-old boy was admitted to our hospital with a fever and cough. He also had a history of recurrent vomiting since 6 months of age, which worsened after eating solid foods. He was delivered full-term without complications. Aside from the recurrent vomiting, there were no reported developmental abnormalities.

Diagnostics were performed to determine the cause of the persisting vomiting. A 24-hour pH monitoring test, using an intraesophageal catheter, was conducted to assess for gastroesophageal reflux; there was no gastroesophageal reflux detected. Esophagoscopy was performed to identify obstruction; however, the rigid esophagoscope could not pass through the lower esophagus. Esophagitis was not observed in the visible region.

As a follow-up, a barium esophagogram was performed to evaluate the lower esophagus. Esophagography revealed an abrupt narrowing of the distal esophagus with marked proximal dilatation (bird-beak appearance) (Fig. 1A) and tracheal aspiration of the contrast media. Small linear barium collections projecting perpendicularly from the stenotic esophagus were also observed (Fig. 1B). Surgical resection was performed

☆ Competing Interests: The authors declare that they have no competing interests.

E-mail address: sounheerad@gmail.com

https://doi.org/10.1016/j.radcr.2023.07.063

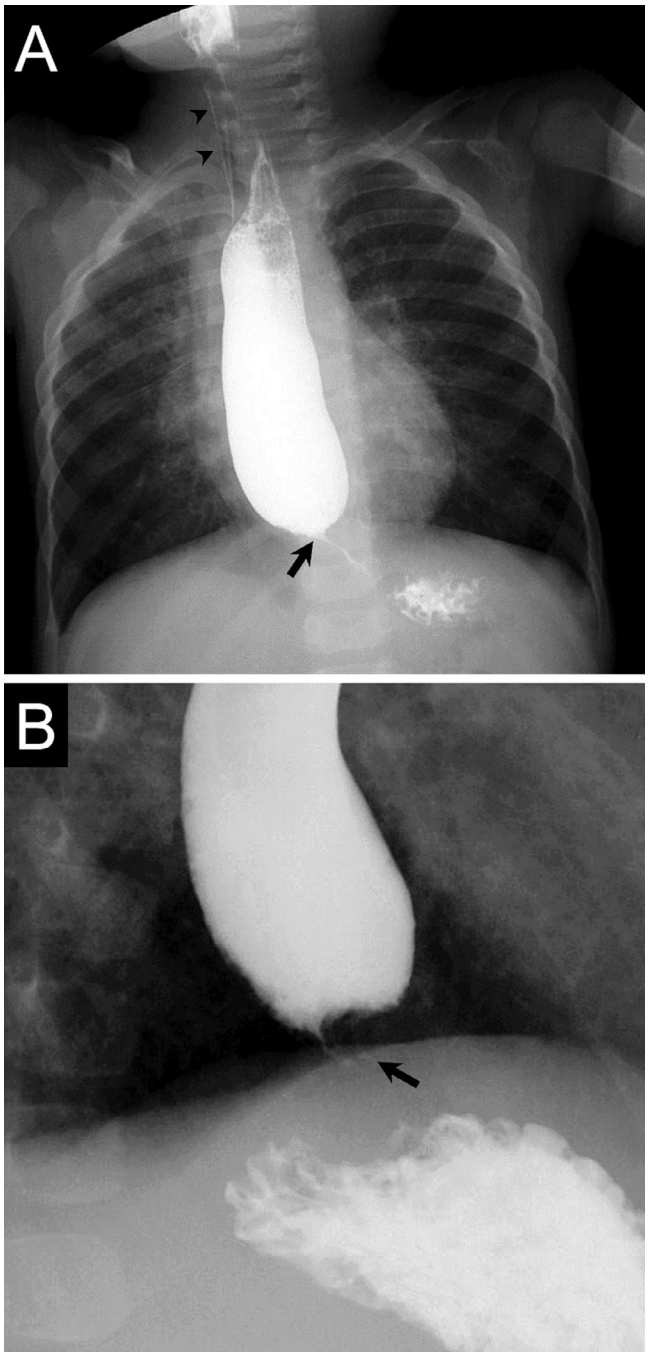

Fig. 1 – Barium esophagogram. (A) Esophagography shows luminal narrowing in the distal esophagus (arrow) with marked proximal dilatation. A small amount of contrast media is aspirated (arrow heads) to the trachea. The perihilar and infrahilar infiltrations of both lungs suggest aspiration pneumonia. (B) Left posterior oblique view shows multiple small linear projections (arrow) of barium extending intramurally from the stenotic esophagus.

without instrumental dilatation. During surgery, a hard ring-shaped stenosis approximately 1 cm in length was noted in the distal esophagus. Segmental resection of the lower esophagus was performed using end-to-end anastomosis and fundoplication. Pathological examination of the resected specimen revealed respiratory epithelia with cartilage and mucous glands in the esophageal musculature (Fig. 2). Postoperatively, the patient had no vomiting, dysphagia, or pneumonia during the 9-month follow-up period.

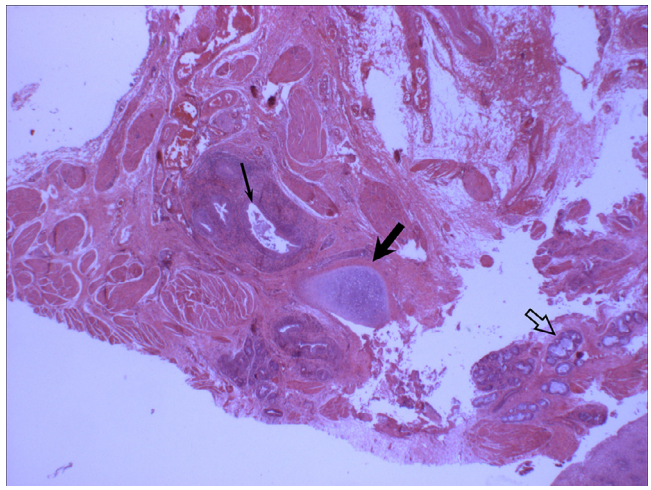

Fig. 2 – Histopathology. (A) photomicrograph of the histopathological specimen shows respiratory epithelia (thin arrow) with cartilage (thick arrow) and mucous glands (empty arrow) in the esophageal wall (Hematoxylin-eosin stain, x 40).

Discussion

Congenital esophageal stenosis is categorized into 3 histological types: membranous web, fibromuscular thickening, and tracheobronchial remnants. Membranous webs can be cured by dilatation; however, the other 2 types are usually refractory to dilatation and require surgical treatment [1–4]. Treatment varies depending on type; therefore, it is important to distinguish between them. A membranous web is usually located in the mid-esophagus or at the junction of the middle and lower thirds. Esophagography shows a linear filling defect perpendicular to the esophageal wall. Fibromuscular thickening is 1-4 cm in length, has tapered stenosis with an hourglass configuration, and is located at the junction of the middle and lower thirds of the esophagus. Tracheobronchial remnants are found in the lower third, within 3 cm of the gastric cardia, and often show high-grade obstruction [4].

Congenital esophageal stenosis caused by tracheobronchial remnants is a rare anomaly in infants and children [1–4], refractory to instrumental dilatation and often diagnosed after surgery [3,4]. It is caused by a failure of separation of the embryonic respiratory tract from the primitive foregut and results in the sequestration of tracheobronchial precursor cells in the esophageal wall [1–5]. The clinical features of congenital esophageal stenosis caused by tracheobronchial remnants vary depending on the degree of stenosis and include dysphagia and recurrent vomiting, which may lead to aspiration pneumonia, particularly after the ingestion of solid food [3]. Stenosis occurs in the distal esophagus, in contrast

to fibromuscular thickening, in which stenosis occurs in the middle third of the esophagus [2–4]. Pathologically, the resected specimen shows respiratory epithelia with cartilage and mucous glands in the esophageal wall. Unlike achalasia or other fibrous strictures, esophageal stenosis caused by tracheobronchial remnants is not responsive to instrumental dilatation, such as bougienage or balloon dilatation [1–4]. Surgery, such as resection of the stenotic segment with end-to-end anastomosis or circular myectomy, is recommended [1,3,6,7]. Maeda et al. [6] reported that congenital esophageal stenosis caused by tracheoesophageal remnants could be successfully managed with circular myectomy without resection.

Zhao et al. [3] classified the esophagographic findings of tracheobronchial remnants into 3 types: type I, distal esophageal stenosis with either tapered narrowing (Ia) or abrupt narrowing (Ib); type II, flask-shaped shadow; and type III, type I or II with a linear projection of barium at the level of stenosis. They explained that the imaging findings varied depending on whether the stenotic segment was completely or incompletely surrounded by cartilage and how far away the stenosis was from the gastroesophageal junction. Type I is a nonspecific finding that may be observed in patients with stenosis of other causes, such as secondary to the ingestion of caustic agents, peptic esophagitis, or achalasia. Type III is the least common but shows characteristic radiological findings, such as multiple linear barium collections that extend perpendicularly from the stenotic esophagus, that differentiate it from stenoses of other causes [3,5]. In type III, sinus tracts or ducts of the bronchial glands connecting with the esophageal lumen are unique pathological findings, which could explain the linear projection of barium seen in esophagography.

Although it is difficult to diagnose esophageal stenosis caused by tracheobronchial remnants, it is important to carefully check for its characteristic esophagography findings to avoid unnecessary instrumental dilatation. Esophageal stenosis caused by tracheobronchial remnants should be considered when small linear projections of barium extending intramurally from the stenotic esophagus are observed in esophagography in young children or infants.

Patient consent

Consent for publication has been obtained.

REFERENCES

[1] Amae S, Nio M, Kamiyama T, Ishii T, Yoshida S, Hayashi Y, et al. Clinical characteristics and management of congenital esophageal stenosis: a report on 14 cases. J Pediatr Surg 2003;38:565–70. doi:10.1053/jpsu.2003.50123.
[2] Gao Z, Wang L, Liu H, Zhang X. Congenital esophageal stenosis caused by tracheobronchial remnants: a case report. J Int Med Res 2022;50:1–7. doi:10.1177/03000605221132704.
[3] Zhao LL, Hsieh WS, Hsu WM. Congenital esophageal stenosis owing to ectopic tracheobronchial remnants. J Pediatr Surg 2004;39:1183–7. doi:10.1016/j.jpedsurg.2004.04.039.
[4] Ramesh JC, Ramanujam TM, Jayaram G. Congenital esophageal stenosis: report of three cases, literature review, and a proposed classification. Pediatr Surg Int 2001;17:188–92. doi:10.1007/s003830000458.
[5] Anderson LS, Shackelford GD, Mancilla-Jimenez R, McAlister WH. Cartilaginous esophageal ring: a cause of esophageal stenosis in infants and children. Radiology 1973;108:665–6. doi:10.1148/108.3.665.
[6] Maeda K, Hisamatsu C, Hasegawa T, Tanaka H, Okita Y. Circular myectomy for the treatment of congenital esophageal stenosis owing to tracheobronchial remnant. J Pediatr Surg 2004;39:1765–8. doi:10.1016/j.jpedsurg.2004.08.016.
[7] Olguner M, Ozdemir T, Akgür FM, Aktuğ T. Congenital esophageal stenosis owing to tracheobronchial remnants: a case report. J Pediatr Surg 1997;32:1485–7. doi:10.1016/s0022-3468(97)90570-4.