



Recurrent tracheobronchitis caused by H-type tracheoesophageal fistula: A case report

Abbas Dabaghzadeh¹, Sara Sadr², Sepideh Yekkekar², Hamed Jafarpour^{2*}

¹ Assistant of Pediatrics, Pediatrics Department of Bou'ali Sina Hospital, Faculty of Medicine, Mazandaran University of Medical Sciences, Sari, Iran

² Medical Student, Student Research Committee, Faculty of Medicine, Mazandaran University of Medical Sciences, Sari, Iran.

Tracheoesophageal fistula is a connection between the posterior wall of the trachea and the anterior wall of esophagus, without atresia. This is a rare condition presentation in 4-5% of all congenital esophageal anomalies. In contrast to other forms of tracheoesophageal fistula, the H-type is not diagnosed in early infancy and the diagnosis is usually delayed. A 7-month-old boy presented with a chief complaint of a consistent cough and wheezing with an onset of a month prior to the visit. A cough was productive and persistent but no vomiting, cyanosis or problems with feeding were reported. In surgery the fistula was completely extracted and both the esophagus and the trachea were repaired and the incision site was sutured. There were no significant post-op complications, and a barium meal performed one week post-operation showed no connection between the esophagus and the trachea. Tracheoesophageal fistula is rare condition presenting with variety of symptoms. As in this case the diagnosis is often delayed for months or even until adulthood.

Keywords: Tracheoesophageal fistula, Tracheobronchitis, H-type fistula

How to cite this article: Dabaghzadeh A., Sadr S., Yekkekar S., Jafarpour H., Recurrent tracheobronchitis caused by H-type tracheoesophageal fistula: A case report. Tabari Biomed Stu Res J. 2019;1(2):38-40

Introduction

An H-type tracheoesophageal fistula (TEF) is an oblique connection between the posterior wall of the trachea and the anterior wall of the esophagus, without a presentation of atresia. This rare condition represents 4-5% of all congenital esophageal anomalies [1, 2]. The etiology of this condition is not fully known but an incomplete separation of the trachea and esophagus in the early stages of embryogenic development is thought to be responsible for this anomaly. In contrast to other forms of tracheoesophageal fistula, the H-type is not diagnosed in early infancy; even though the anomaly is in place since before birth, diagnosis is usually delayed [3, 4].

Case presentation

The patient is a 7-month-old boy, presented with a chief complaint of a consistent cough and wheezing with an onset of a month prior to the visit. A cough was productive and persistent but no vomiting, cyanosis or problems with feeding were reported. On auscultation, an inspiration/expiration generalized wheeze was detected. He was first admitted to our medical center for an allergy and hyper-responsive airway work-up.

After an allergy works up, a rigid bronchoscopy was performed with a suspicion of a partial obstruction caused by a foreign object, which resulted in a diagnosis of tracheobronchitis. The exudates were removed by suction, but no foreign object was detected. The patient then underwent a gastrointestinal and reflux investigation which did not point to a pathology. The chest x-ray was also reported normal.

A spiral CT-Scan with contrast was performed that showed a cervical tracheal stenosis and a tract

* Corresponding author: **Hamed Jafarpour**, Medical Student, Student Research Committee, Faculty of Medicine, Mazandaran University of Medical Sciences, Sari, Iran. Email: hamed.jafarpour7@gmail.com

between the esophagus and the trachea, which suggested a TEF. To confirm the diagnosis, an esophagography was performed which demonstrated an oblique fistula at the level of the second or third thoracic intervertebral space

Surgical technique

The patient went under general anesthesia and endotracheal intubation. The patient's head was positioned in hyperextension and a vertical incision in the medial border of the right sternocleidomastoid muscle was made. Then, the muscular layer, platysma muscle, and the inferior thyroid artery were moved aside and a good view of the cervical esophagus and the fistula was obtained. Then the fistula was completely extracted and both the esophagus and the trachea were repaired and the incision site was sutured

There were no significant post-operation complications, and a barium meal performed one-week post-op showed no connection between the esophagus and the trachea

Discussion

A congenital tracheoesophageal fistula, without a present atresia known as an h-type TEF, is, in fact, an oblique connection between the esophagus and the trachea; which is an N-like opening from the cephalad part of the posterior wall of the trachea to the caudal part of the anterior wall of the esophagus.

With an incidence of one in every 5000-8000 live births, this anomaly represents 4% of all esophageal anomalies [5].

The classic triad of sudden paroxysmal severe a cough, choking (caused by food entering the trachea) and abdominal distention (caused by air entering the stomach through the esophagus and recurrent aspiration pneumonia), is used for diagnosis of a fistula; although several signs and symptoms are needed to build a suspicion of an H-type fistula but in our case respiratory symptoms were prominent and a chest x-ray was performed with consideration of the possibility of a foreign body obstruction, although no abnormalities were seen. In further investigations, using a rigid bronchoscopy, tracheobronchitis was confirmed and without any sign of a foreign body, the exudates were suctioned. Our patient management was in accordance with recommendations given by Crabbe *et al.* [6].

Many different radiologic and endoscopic modalities are used to confirm the diagnosis of an H-type fistula, although success is majorly depended

on the imaging technique and aiming angles, which makes making a diagnosis difficult and in many cases such as ours, a diagnosis is not made until later in life, when prominent symptoms are presented [1, 7].

In the presented case a gastrointestinal workup was done and in a spiral CT-Scan with contrast, a stenosis in the cervical trachea and a tract between the esophagus and the trachea were seen; which suggests a TEF as the underlying pathology. To confirm our diagnosis and esophagography was performed which showed an oblique fistula at the level of the second or fourth cervical intervertebral space.

Closure of the fistula is the treatment goal for which the best approach is surgical. In multiple studies, it has been shown that a cervical surgical approach is preferable than a thoracic approach [1]. In the presented case the surgical procedure was performed using a thoracic right side posterolateral approach; similar to a case reported by Karnak *et al.* [8]. In the procedure, the attempt was not to tear the pleura. A retro-pleural dissection was performed and the procedure was done in the posterior mediastinum. The fistula was closed and extracted.

Conclusion

Tracheoesophageal fistula is rare condition presenting with variety of symptoms. Unfortunately, the diagnosis is often delayed for months or even until adulthood. The diagnosis maybe difficult due to the small size and the intermittency of esophageal content into trachea. Early diagnosis of this condition can decrease associated morbidities and result in better prognosis so this condition must be kept in mind for newborns who are experiencing recurrent pulmonary infections.

Conflicts of interest

The authors declare that there is no conflict of interest regarding the publication of this paper.

Acknowledgment

The authors would like to express their thanks to the Department of Pediatrics, Clinical Research Development Unit of Bou'Ali Sina Hospital and Student Research Committee of Mazandaran University of Medical Sciences to support this research

References

1. Zani A, Jamal L, Cobellis G, Wolinska JM, Fung S, Propst EJ, et al. Long-term outcomes following H-type tracheoesophageal fistula repair in infants. *Pediatr Surg Int.* 2017;33(2):187-90.
2. Tröbs R, Finke W, Bahr M, Roll C, Nissen M, Vahdad M, et al. Isolated tracheoesophageal fistula versus esophageal atresia—Early morbidity and short-term outcome. A single institution series. *International Journal of Pediatric Otorhinolaryngology.* 2017;94:104-11.
3. Amat F, Heraud MC, Scheye T, Canavese M, Labbe A. Flexible bronchoscopic cannulation of an isolated H-type tracheoesophageal fistula in a newborn. *J Pediatr Surg.* 2012;47(10): 9-10.
4. Boybeyi O, Kose M, Ersoz DD, Haliloglu M, Karnak I, Senocak ME. Achalasia-like findings in a case with delayed diagnosis of H-type tracheoesophageal fistula. *Pediatr Surg Int.* 2008;24(8):965-9.
5. Antao B, Soccorso G, Bateman N, Shawis R. H-type tracheoesophageal fistula with type III laryngotracheoesophageal cleft. *Eur Arch Otorhinolaryngol.* 2007;264(11):1373-6.
6. Crabbe D. Isolated tracheo-oesophageal fistula. *Paediatric respiratory reviews.* 2003;4(1):74-8.
7. Aziz GA, Schier F. Thoracoscopic ligation of a tracheoesophageal H-type fistula in a newborn. *J Pediatr Surg.* 2005;40(6):e35-6.
8. Karnak Ib, Şenocak ME, Hiçsönmez A, Büyükpamukçu N. The diagnosis and treatment of H-type tracheoesophageal fistula. *Journal of pediatric surgery.* 1997;32(12):1670-4.