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Management of congenital isolated H-type tracheoesophageal fistula: A case report

Azalia Avila; Nina D Kosciuszek; Jacob Campbell; Katherine Kavanagh; Christine Finck*

***Corresponding Author: Christine Finck**
292 Washington Street, Hartford, CT 06106, USA.
Tel: 860-545-9634; Email: cfinck@connecticutchildrens.org

Abstract

Introduction: Isolated H Type Tracheoesophageal Fistulas (H-TEF) with esophageal continuity are rare and diagnosis requires a high index of suspicion. In addition, the incidence of vocal cord paralysis following repair of an H-type Tracheoesophageal Fistula (TEF) is relatively high compared to other types of TEF, primarily due to the proximity of the recurrent laryngeal nerves to the fistula tract.

Case presentation: A 4-month-old female born at 41 weeks 1 day gestation with an otherwise uneventful perinatal course presented to our institution due to failure to thrive with stridor and gurgling when feeding. We performed a contrast esophagography with pullback technique and demonstrated an H-type cervical tracheoesophageal fistula. Preoperative evaluation of the vocal cords demonstrated equal movement bilaterally. Preoperative planning with bronchoscopy identified the fistula and a ureteral catheter was fed across the fistulous tract. In addition, the light from the bronchoscope at the level of the fistula was marked externally on the neck to guide the location of the incision. Ligation of the fistula was performed via a cervical approach and postoperative evaluation of the vocal cords revealed symmetric movement.

Conclusion: Precise preoperative localization with the bronchoscope light externally marking the site of this fistula is useful in guiding location of the incision. Bronchoscopic placement of a catheter across the fistula aids in minimizing excessive dissection, thus reducing risk to the recurrent laryngeal nerve.

Keywords: H-TEF: H-type tracheoesophageal fistula; Bronchoscopy; Recurrent laryngeal nerve; Vocal cord paralysis; Case report.

Abbreviations: H-TEF: H-Type Tracheoesophageal Fistula; TEF: Tracheoesophageal Fistula.

Introduction

Tracheoesophageal Fistulas (TEF) are congenital abnormalities of the foregut characterized by

an aberrant communication between the esophagus and trachea with subtypes further classified by the presence of esophageal atresia [1]. Symptoms consistent with TEFs typically appear soon after birth and may include respiratory distress, cyanosis, coughing, choking during feeds, and inability to intubate the esophagus with an orogastric or nasogastric tube when esophageal atresia is present H-Type TEF (H-TEF) comprise approximately 4% of all TEF anomalies (1 in 50,000-80,000) [2]. This subtype is unique as it occurs without the presence of an esophageal atresia. The esophagus remains in continuity; therefore, feeding may be initially tolerated which may lead to a delay in diagnosis. These neonates are at an increased risk for recurrent pulmonary infections, cyanosis, and failure to thrive secondary to ongoing aspiration. Diagnosis of H-type TEF requires a high index of suspicion, as well as dedicated testing with esophagography and/or bronchoscopy to determine the location and provide subsequent surgical ligation [3]. In addition, the incidence of vocal cord paralysis following repair of an H-TEF is high at around 20-50% compared to other types of TEF repair primarily due to the proximity of the recurrent laryngeal nerves to the fistula tract [4,5]. Therefore, preoperative planning and use of a multi-disciplinary team is essential to minimize this risk. We present a case presentation where a multi-disciplinary team and the use of transillumination from a bronchoscope is useful in surgical planning. In addition, placement of a catheter via a bronchoscope aid in fistula tract dissection while reducing the risk of recurrent laryngeal nerve injury. This manuscript was prepared following the CARE guidelines (https://www.care-statement.org).

Case Presentation

A 4-month-old female born at 41 weeks and 1 day, with a birth weight of 6 pounds and 11 ounces, presented to the hospital due to failure to thrive, intermittent abdominal distension, and “bubbling noises” with feeds. There were no cyanotic spells or choking reported by the parents. In addition, no pertinent patient or family history was noted. Initial differential diagnosis was most consistent with oropharyngeal dysphagia given gurgling with feeds. This prompted further imaging studies with a modified barium swallowed that raised suspicion for a TEF. Dedicated imaging with an esophagogram utilizing a “pullback” technique with a Nasogastric Tube (NGT) was performed which allowed for complete identification of a cervical H-TEF [6,7]. In this technique, an NGT was inserted, and a small amount of injected contrast confirmed the presence of a TEF at the level of the cervical esophagus, just above the thoracic inlet with communication to the trachea. The distention of the intrathoracic esophagus with contrast below the level of the fistula demonstrated no additional fistulas with contrast passing into the stomach without obstruction. There was no evidence of esophageal stricture (Figure 1). Preoperative evaluation of the vocal cords with an awake flexible laryngoscopy was performed by Otolaryngology and demonstrated equal true vocal cord movement bilaterally on phonation and inspiration.

Operative management

The patient was brought to the operating room and placed in the supine position on the operating room table. After general anesthesia was administered, direct laryngoscopy and bronchoscopy were performed to identify the fistula and evaluate the vocal cords. Typically, rigid bronchoscopy is optimal for evaluation and enables placement of a ureteral catheter across the fistula site. Prior to incision, bronchoscopy showed intact and symmetric vocal cords and identified the tracheoesophageal fistula in

the proximal trachea, right of midline and posteriorly (Figure 2A). Following identification, the patient's neck was marked with a surgical marking pen at the level of the fistula using transillumination from the bronchoscope (Figure 2C).

The fistula tract was then cannulated with a 3 French ureteral catheter under direct vision (Figure 2B). Next, the patient was intubated with a 3.5 cuffed endotracheal tube under direct vision and the endotracheal tube, as well as our ureteral catheter, were secured.

A shoulder roll was placed, and the patient's head was rotated towards the left side. The neck and chest were prepped and draped in standard sterile fashion. A 3 cm transverse incision was made along a natural crease of the right neck at the level previously marked. The platysma was divided, and dissection was carried down to the level of the sternocleidomastoid muscle. This muscle was preserved and retracted laterally. Next, the trachea was identified medially and the carotid sheath laterally. Care was taken to preserve the recurrent laryngeal nerve. The dissection was then carried posteriorly until the esophagus was encountered. The ureteral catheter was then utilized to identify the fistula, which was then encircled with a vessel loop (Figure 3). Absorbable stay sutures were placed on both ends of the fistula with two on the tracheal side and two on the esophageal side. The ureteral catheter was then removed, and the fistula was divided sharply. The trachea was then repaired transversely with absorbable sutures in a running fashion. The esophagus was then repaired longitudinally as a single layer of multiple interrupted absorbable sutures. A Valsalva maneuver with saline placed in the surgical field did not reveal any leak. A fibrin sealant was applied in the neck. A portion of the sternocleidomastoid muscle was mobilized to buttress the repair, and the neck incision was then closed in multiple layers. The patient tolerated the procedure well and was successfully extubated. A repeat laryngoscopy at the end of the case immediately following deep extubation revealed both vocal cords in their expected normal anatomic abducted position. Normal abduction and adduction of the vocal cords was observed bilaterally. Both vocal cords moved smoothly without any issues. Patient tolerated this procedure well and was brought to recovery without any issues.

Follow Up and Outcome

This patient's postoperative course was unremarkable with no postoperative complications. Following surgery, she was tolerating tube feeds via a nasogastric tube and underwent an esophagram on postoperative day 5. The esophagram did not show any evidence of a residual fistula connection between the trachea and esophagus. The patient was subsequentially transitioned to full oral feeds, and the nasogastric tube was removed. The patient was then discharged home on full oral feeds on postoperative day 6.

Three weeks following surgery, the patient was evaluated in clinic and was progressing well at home. Parents did not notice any recurrent episodes of choking or "bubbling noises" with feeds. She continued to tolerate fortified formula and was rapidly gaining weight. Her parents did note occasional hiccups that were self-resolving. Her breathing was not noisy, and parents denied any stridulous breathing. Patient is planned for a 2 month follow up with a goal of introducing puree food at that time.

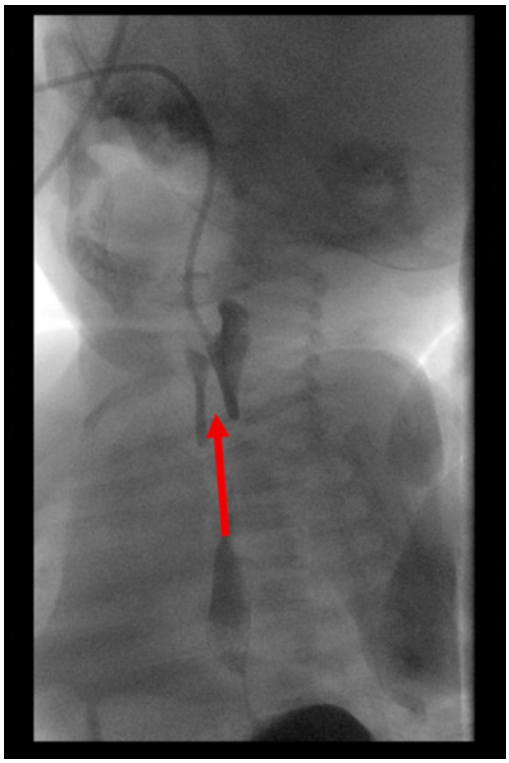

Figure 1: Esophagogram performed via a nasogastric "pullback" method demonstrating a cervical H-type tracheoesophageal fistula (red arrow).

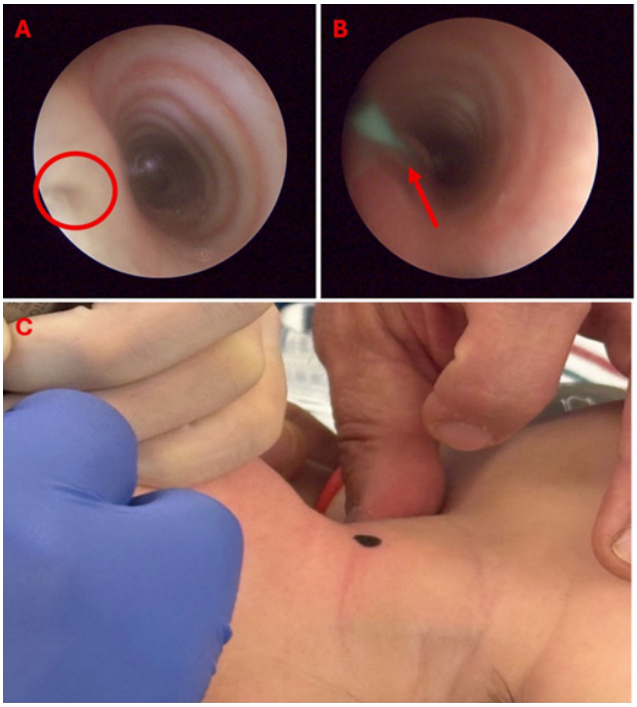

Figure 2: (A & B) Intraoperative bronchoscopic image illustrating a posteriorly located tracheoesophageal fistula (Red Circle in Panel A) and intubated with a catheter (Red Arrow, Panel B) **(C)** The level of the fistula marked using transillumination from the bronchoscope.

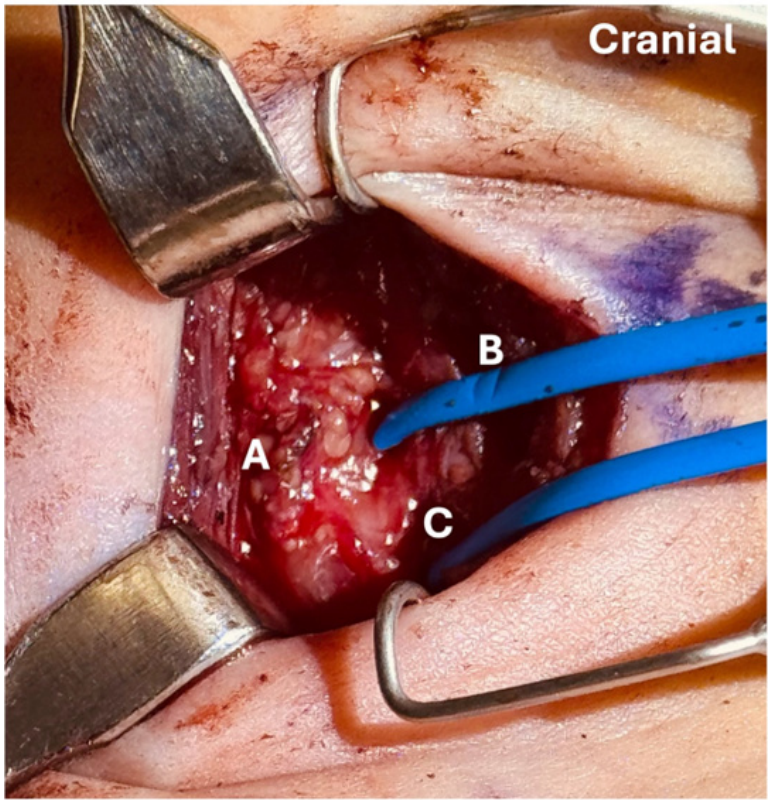

Figure 3: Image of esophagus **(A)**, trachea **(B)** and tracheoesophageal fistula **(C)** after neck exploration and dissection. A blue vessel loop surrounded the fistula.

Discussion

Herein, we present a 4-month-old female born at 41 weeks and 1 day with a relatively uncomplicated perinatal period except for failure to thrive, intermittent abdominal distension, and "bubbling noises" with feeds who was admitted to the gastroenterology service for further care and workup. A modified barium swallow raised concerns for a tracheoesophageal fistula for which she underwent an esophagogram with a "pullback" technique via a nasogastric tube. This study demonstrated a cervical H-TEF, prompting surgical consultation and repair. With guidance from otolaryngology, utilization of transillumination during bronchoscopy enabled easy identification of the fistula relative to the patient's neck externally, allowing for precise operative planning. Placement of a trans-fistula catheter assisted with intraoperative identification of the fistula, minimizing extensive dissection which could inadvertently increase the risk of injury to the recurrent laryngeal nerve. Lastly, our case highlights the benefits of a multidisciplinary team with expertise in TEF leading to a prompt diagnosis and treatment.

The diagnosis of H-TEF may not be clinically evident in an otherwise healthy neonate, as we have seen in this case [8]. Feeding intolerance, cyanosis, and recurrent pulmonary infections are symptoms of a H-TEF and should raise suspicion and prompt workup. It is imperative to evaluate all patients with TEFs for VACTERL anomalies, which often occur in conjunction. The diagnosis of this patient did not occur until she was 4 months old due to the subtlety of her symptoms, normal birth history, and absence of other VACTERL anomalies. In addition, H-TEFs are extremely rare with an incidence of 1 in 100,000 live births or roughly 4%; therefore, it is important for pediatricians to be cognizant of the subtle signs and symptoms that present with H-TEFs [9]. Thus, this pathology's rarity may have also contributed to the delay in diagnosis for this patient.

A video esophagography is often the preferred diagnostic tool to identify H-TEFs as fistulas can be missed with a standard contrast esophagogram in up to 50% of cases [8]. Additionally, utilizing a NGT with a "pullback" technique as the contrast is being administered may be more precise, with some studies revealing multiple fistulas with this method [3,6]. It is important that the diagnostic studies are performed by a radiologist that is familiar with these anomalies as there is a risk of aspiration [7].

Bronchoscopy can also be utilized to identify the fistula intraoperatively. A catheter advanced into the fistula tract eases intraoperative identification of the fistula. Surgical approach is performed either through a cervical or thoracic incision. Most H-TEFs are high (between C7 and T2); therefore, a cervical approach is often performed. However, lower lesions may benefit from a thoracic approach as reported by Toczewski et al., who illustrate a thoracoscopic approach [10]. When using a cervical approach, a right neck incision is more widely used given the more lateral location of the recurrent laryngeal nerve [3]. Additionally, the fistula more commonly angles towards the right, making its identification easier from a right sided cervical approach. However, some surgeons opt to perform a left sided cervical approach as the esophagus lies slightly to the left of midline [11]. In our patient, we cannulated the fistula using a ureteral catheter via bronchoscopy. We elected to approach this fistula via a right cervical incision, which provided us with proper exposure of the esophagus, identification of the recurrent laryngeal nerve, and access to the fistula in its expected location.

As previously mentioned, H-type fistulas can either be high or low in the neck with some instances of the fistula being in the chest [10]. We used transillumination to assist us in determining the optimal location for a right neck incision, which allowed for a more direct dissection down to the fistula (Figure 2C). This method of localizing the fistula has been reported since 2005 and has been shown to make repairs straightforward which is a strength of this technique [12]. During bronchoscopy, a ureteral catheter or guide wire can be passed through the fistula while the light from the bronchoscope can be directed through the fistula and out to the neck tissue and skin. This allows for better understanding of the fistula's location in relation to the patient's body from an outside perspective. However, limitations of this technique are poor depth perception and variable operator experience. If the fistula is deep in the neck, light from the scope may be faint making it difficult to identify the exact location of the fistula. In addition, the technique is operator dependent and can be challenging for someone with limited experience.

Post operative fistula recurrence after repair is uncommon, ranging between 0–14% of patients who have had H type TEF repairs and usually occurs in the early post operative period [11]. The most common post operative complication is recurrent laryngeal nerve dysfunction occurring in 15 -50% of patients. This incidence is believed to be related to the location of the fistula in the neck necessitating more extensive dissection along the course of the vagus nerve and laryngeal nerves [13-15]. Although some centers describe the use of intraoperative nerve monitoring to delineate the recurrent laryngeal nerve, this can be difficult in the small airways of neonates [16]. Additionally, endoscopic intervention to close off the fistulous tract can also resolve TEFs with minimal risk to the recurrent laryngeal nerves. A caveat to this approach is a higher rate of TEF recurrence compared to a transcervical repair. One systematic review reported an overall success rate of 60% in patients who underwent an endoscopic intervention for congenital TEF [17]. While another study demonstrated a 96% success rate in a transcervical approach [18]. Therefore, this highlights that open surgery remains superior to endoscopic interventions which may be due to the fact that endoscopic approaches are only applicable to neonates who have long, narrow fistulas.

Using a stepwise approach that identifies the fistula tract and minimizes dissection is imperative in reducing the risk of nerve injury. In our case, we utilized transillumination to mark the tract as well as a catheter to help with intraoperative identification. In addition, presurgical and post-surgical evaluation of the vocal cords can further establish if the nerve has been injured. This technique was successfully used in our patient.

Conclusion

H-type tracheoesophageal fistulas may pose a diagnostic and therapeutic challenge to clinicians. A high index of suspicion is imperative for early diagnosis with definitive treatment being surgical ligation of the fistula. Locating the fistula prior to surgical exploration facilitates, proper identification and successful ligation and minimizes the risk of recurrent laryngeal nerve injury.

Declarations

CRediT authorship contribution statement

All authors attest that they meet the current ICMJE criteria for Authorship; **Azalia Avila:** Writing – review & editing, Writing – original draft, Validation, Methodology, Investigation, Formal analysis, Data curation. **Nina Kosciuszek:** Writing – review & editing, Validation, Methodology, Formal analysis. **Jacob Campbell:** Review & editing, Validation, Supervision, Resources, Investigation, Data curation, Conceptualization. **Katherine Kavanagh:** Review & editing, Validation, Supervision, Resources, Investigation, Data curation. **Christine Finck:** Writing – review & editing, Validation, Resources, Investigation, Data curation, Conceptualization.

Statements

Informed consent was obtained from the patient's guardian. All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of generative AI Use

During the preparation of this work, we used Deep L translator in order to improve the readability and language of the manuscript. After using this tool/service, we reviewed and edited the content as needed and take full responsibility for the content of the published article.

Declaration of competing interests and funding

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper. No funding was received for this case report.

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Authors Information: Azalia Avila[1]; Nina D Kosciuszek[1]; Jacob Campbell[1,2]; Katherine Kavanagh[2,3]; Christine Finck[1,2,4]*

[1]Connecticut Children's Medical Center, Division of Pediatric General and Thoracic Surgery, Hartford, CT, USA.

[2]University of Connecticut School of Medicine, Department of Pediatrics, Farmington, CT, USA.

[3]Connecticut Children's Medical Center, Department of Otolaryngology, Hartford, CT, USA.

[4]Connecticut Children's Medical Center, Fetal Care Program, Hartford, CT, USA.

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