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Eoghan E. Laffan · Alan Daneman · Sigmund H. Ein · Debbie Kerrigan · David E. Manson

Tracheoesophageal fistula without esophageal atresia: are pull-back tube esophagograms needed for diagnosis?

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Abstract Background: A pull-back tube esophagogram (PBTE) is widely accepted in the literature as the radiological investigation of choice for the diagnosis of tracheoesophageal fistula without esophageal atresia. However, PBTE is rarely performed in our institution, as we have been successful in confirming the presence of such fistulae with a contrast material swallow (CS). We hypothesized that PBTE is not the radiological investigation of choice for the diagnosis of the fistula in this condition. Objective: We sought to determine what proportion of patients with tracheoesophageal fistula without esophageal atresia can be diagnosed promptly by a CS and what the indications are for a PBTE. Materials and methods: We retrospectively analyzed the clinical and radiological findings in patients with tracheoesophageal fistula without esophageal atresia to determine whether the fistula was diagnosed with a CS or PBTE. Results: We identified 20 children (13 female and 7 male) with tracheoesophageal fistula without esophageal atresia. Their age at diagnosis ranged from 3 days to 168 months with a median of 9 days. The diagnosis was documented by CS in 12, PBTE in 7 and CT in 1. In three of the seven who had the fistula documented by PBTE, a previous CS had shown contrast material in the trachea, but no fistulous tract or aspiration was identified. Conclusion: We believe that CS should be the examination of choice in most patients suspected of having a tracheoesophageal fistula without esophageal atresia. A PBTE is indicated in patients who are intubated or are at significant risk of aspiration. Furthermore, a PBTE is also indicated where contrast

E. E. Laffan · A. Daneman (⋈) · D. Kerrigan · D. E. Manson Department of Diagnostic Imaging, Hospital for Sick Children, University of Toronto, 555 University Ave., Toronto M5G 1X8, Canada

e-mail: alan.daneman@sickkids.ca Tel.: +1-416-8136888

S. H. Ein Division of General Surgery, Hospital for Sick Children, University of Toronto, Toronto, Canada material is seen in the airway on CS and there is uncertainty whether this is due to aspiration or a fistula.

Keywords Esophagus · Fistula · Trachea · Children

Introduction

Congenital tracheoesophageal fistula (TEF) without esophageal atresia, often referred to as H-type TEF (H-TEF), accounts for only 5% of all TEFs [1, 2]. The fistula in these patients has an oblique orientation, passing forward and upward from its origin in the esophagus to its connection with the trachea. Because of this orientation, it should more accurately be referred to as an N-type TEF (N-TEF).

Difficulties in the diagnosis of these fistulae have been documented [1–15]. Failure to diagnose and treat the N-TEF early can lead to significant morbidity with recurrent chest infections, bronchiectasis and failure to thrive [2, 4, 13–18]. The first case was described in 1876 in the post-mortem examination of a young child, though nowadays it is rarely, if ever, fatal [13, 19–21].

Numerous diagnostic techniques have been described, but given the relatively low incidence of N-TEF, there are no randomized, controlled trials to support the use of one method over another [3, 8, 22–26]. It is stated in the literature and textbooks that an esophagogram performed by injecting contrast material down a tube with its tip in the esophagus, known as a pull-back tube esophagogram (PBTE), is the investigation of choice for the diagnosis of N-TEF [1, 3, 13, 15, 17, 27]. It has been documented in some reports that a routine contrast material swallow (CS) often has to be repeated and does not always provide the correct diagnosis [2, 3, 14, 16, 17]. Because of this, it has been suggested by some authors that all patients suspected of having N-TEF should undergo a PBTE [13, 15, 18, 27]. However, the data regarding this are anecdotal, with no large series proving whether the PBTE or a CS is more effective or safer in establishing the diagnosis.

Our purpose was to review all children with N-TEF in this institution to determine whether the diagnosis was

made by PBTE or CS or whether diagnosis required other modalities such as CT or endoscopy. We also sought to establish how many patients had a normal or negative CS and in whom the diagnosis of N-TEF was made by PBTE. Finally, we sought to establish a protocol for the investigation of N-TEF and to determine the indications for PBTE.

Materials and methods

After Research Ethics Board approval, we retrospectively analyzed all children with TEF without esophageal atresia in this institution from 1975 to June 2005. Patients were found via a word-search for the terms "H-type, tracheo, tracheoesophageal, fistula, H-TEF" and variations and combinations of the above in several databases: discharge diagnosis database, operating room diagnosis, pediatric radiology reports. Personal communications were also sent to current and retired surgical and radiological staff inquiring about cases.

This process revealed 24 patients with congenital TEF without esophageal atresia. Information about the imaging technique was available in only 20; these 20 were included in this study. In 17 of these patients, the static images were available for review by three radiologists (E.E.L., A.D., D.M.). In the other three children, seen between 1975 and 1985, only the reports of the imaging examinations were available. (Unfortunately, the full clinical and surgical records of the latter three were not available).

Prediagnostic symptoms and signs (cough, cyanosis/ apnea, choking or aspiration with feeds, failure to thrive, recurrent chest infections), age at diagnosis, number and type of studies performed (CS, PBTE, CT and endoscopy) were recorded. The prediagnosis radiographs were also reviewed for any opacification suggestive of pneumonia. The presence of gaseous bowel distension was also recorded. Comorbidities and associated syndromes were noted.

The choice of whether to perform a CS or PBTE in each patient was the decision of the radiologist responsible for the patient. During the study period, ten radiologists were responsible for the examinations. Water-soluble, low-osmolar contrast material was usually used, though barium was used in examinations performed during the earlier years of this review. Whether CS or PBTE is used, it is important to be sure there is good distension of the esophagus.

The CS examinations were performed with the patient lying horizontally on the fluoroscopy table in the right lateral decubitus position. The patients were fed contrast material from a bottle. The patients were allowed to suck from the bottle only until the fistula was adequately visualized to prevent too much contrast material from entering the trachea. The PBTE examinations were performed with the patient either lying horizontally on the fluoroscopy table in the right lateral decubitus position or lying prone on the footrest with the table upright. In both situations, a nasogastric feeding tube (no. 8 or 10) was

placed with its tip in the lower esophagus. Contrast material was slowly injected as the tube was slowly withdrawn until the tip was in the upper esophagus behind the level of the trachea. As with CS, the examination was stopped as soon as a fistula had been adequately visualized in order to prevent too much contrast material from entering the trachea.

Diagnosis of an N-TEF by CS or PBTE was made only if there was evidence of contrast opacification of a fistulous tract between the trachea and esophagus. The presence of contrast material in the trachea without visualization of the fistula itself was not considered diagnostic of N-TEF, and this prompted further imaging.

The vital signs of the sicker patients were monitored and the type of monitoring depended on the age of the patient and the presence of comorbidities (e.g., congenital heart disease).

Results

There were 20 children, 13 girls and 7 boys, with N-TEF for whom radiological information was available. Their median age at diagnosis was 9 days (range 3 days to 168 months), and 14 were diagnosed in their first month of life.

Among 14 of the 17 the neonates and young infants for whom adequate clinical records were available, the most common presenting complaint(s) was associated with feeding: cyanosis and apnea in six, choking and aspiration in six and coughing with feeds in two. However, several children had more than one of the above findings. Three older children presented late at 14, 36 and 168 months of age, respectively, and all had a longstanding clinical history. These older children all presented with recurrent lower respiratory tract infections, and one also presented with failure to thrive. However, on more detailed questioning there was also a history of choking with feeds in all three. Associated anomalies were present in five: VACTERL syndrome in two, CHARGE syndrome in one, and congenital heart disease in two. Two of these children had a right-side aortic arch.

Prior to the diagnosis of the fistula, imaging was available in only seven: a chest radiograph in six, an abdominal radiograph in two and a CT scan in one. Pulmonary parenchymal opacification was present in five and gaseous distension of bowel in five. One child was noted to have cardiomegaly and pulmonary edema caused by congenital heart disease.

Imaging examinations of the esophagus

In the 20 children, 24 imaging examinations of the esophagus were performed: 15 CS, 8 PBTE, and 1 CT. (The diagnosis was established by bronchoscopy in one neonate prior to a PBTE.) Only one imaging examination was performed in 16 children to document the presence of the fistula. The other four children had two imaging examinations, but the fistula was depicted correctly on the

first examination in one of these who had a CS.Of the 15 who had a CS as the first examination, 12 were positive for the presence of a fistula (Figs. 1, 2 and 3) and one of these was also shown by a subsequent PBTE. (The reason for performing a PBTE after a positive CS in the latter patient was not clearly defined in the chart.) In the other three patients who had CS, contrast material was noted in the trachea but no fistulous tract was identified. All three then had a PBTE that confirmed the presence of a fistula.

Four other children had a PBTE as the first imaging examination, and in all four the fistula was clearly demonstrated (Figs. 4, 5, 6 and 7). One of these was the neonate who had a fistula initially diagnosed on bronchoscopy, which was performed because of failure of extubation. A subsequent PBTE, performed while the patient was



Fig. 1 CS in a 3-day-old infant with N-TEF who presented with cyanosis during feeding. The infant was fed water-soluble contrast material from a bottle while lying horizontally in the right lateral decubitus position, and some contrast material is noted in the mouth. The *arrow* indicates the origin from the esophagus of the obliquely oriented fistula, which is at the C6/7 vertebral level. A small amount of contrast material has entered the trachea



Fig. 2 CS in a 9-day-old infant with N-TEF who presented with coughing. The infant was fed water-soluble contrast material from a bottle while lying horizontally in the right lateral decubitus position. The *arrow* shows the level of the fistula, which is at the C7 vertebral level. Some contrast material has entered the trachea and passed superiorly into the larynx. A nasogastric tube was present during this examination with its tip in the stomach. The presence of the tube did not prevent contrast material from passing through the fistula

still intubated, confirmed the presence of the fistula (Fig. 7). In this patient, the endotracheal tube obstructed the fistula almost completely while the patient was in the right lateral decubitus position. By changing the patient's position to the left lateral decubitus position, we were able to shift the tube off the orifice of the fistula and depict the fistula clearly. The last child, age 168 months, had a chest CT for investigation of possible bronchiectasis. An incidental TEF was documented, but the parents refused all further investigation. The fistula was considered to be congenital in nature. The patient was a recent immigrant to the country and it is not known whether other tests were carried out elsewhere.

Only one fistula was present in 19 children (Figs. 1, 2, 4, 5, 6, 7), and two fistulae were present in one child (Fig. 3). Thirteen fistulae were at or above the level of T1 (C5/6 two, C6/7 three, C7/T1 eight) and four were below T1 (T1/2 three, T2/3 one). The exact level of the fistula was not documented in the other three patients. All of the fistulae were oriented obliquely and extended forward and upward from the esophagus to the trachea (Figs. 1, 2, 3, 4,

Fig. 3 CS in a 5-day-old infant with two N-TEFs who presented with coughing during feeding. The patient was fed water-soluble contrast material from a bottle while lying horizontally in the right lateral decubitus position. The *arrows* show the levels in the esophagus where the two obliquely oriented N-TEFs are clearly depicted. The fistulae are at the C6 and C7 levels, respectively





5, 6, 7). They all, therefore, had an N configuration. An "esophageal nipple" sign was seen in 7 of the 17 contrast studies available for review (Fig. 7). The "nipple" is a small out-pouching of contrast material seen on the anterior aspect of the esophagus at the level of the esophageal ostium of the fistula. This was noted just prior to opacification of the fistula itself.

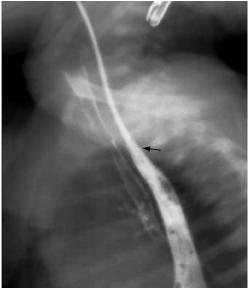
Five children required suctioning for respiratory complications during the examinations because of significant passage of contrast material through the fistulae into the lungs. This happened in two children during CS and in three during PBTE. In one patient, aspiration during CS caused significant distress with cyanosis and bradycardia, but the infant responded quickly to suctioning. The procedure was then abandoned and a positive PBTE was performed 2 days later.

Discussion

This study showed that CS can be used effectively for the diagnosis of TEF in those patients without esophageal atresia, as the fistula was accurately demonstrated in 80% (12/15) of the children in whom this technique was used. In all, the fistula was easily depicted on the first CS examination. PBTE was only required in the remaining 20% (3/15) as CS failed to depict the fistulous tract despite the fact that contrast material was noted in the trachea. Furthermore, CS compared favorably to PBTE when safety was considered. Airway suctioning to remove significant amounts of contrast material that entered the airway was required in only 13% (2/15) of the CS examinations, in contrast to 38% (3/8) of the PBTE examinations. The numbers are small, however, and one has to be prepared to suction the airway whichever technique is used.

Fig. 4 PBTE in a 5-day-old infant with N-TEF who presented with cyanosis. The examination was performed with the patient lying horizontally and somewhat obliquely in the right lateral decubitus position. The arrow shows the level of the obliquely oriented N-TEF clearly. The fistula was at the T1 vertebral level. Note the feeding tube, which has its tip below the level of the fistula. The amount of contrast material that has entered the trachea and larynx is greater than that seen in Figs. 1, 2 and 3





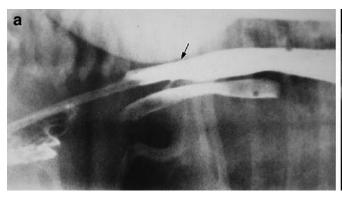
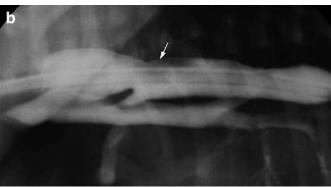


Fig. 5 PBTE in a 7-day-old infant (a) and a 3-month-old infant (b) with N-TEF. The examinations were done with the patients lying horizontally on the footrest of the fluoroscopy table in the prone position. In both images the *arrow* shows the level of the



obliquely oriented N-TEF, which is clearly depicted. The amount of contrast material that has entered the trachea is greater than that noted in Figs. 1, 2 and 3

This study had limitations: it was a retrospective analysis that could not compare the CS and PBTE in a controlled, randomized manner, and the decision whether to use a CS or a PBTE in each patient was made by the radiologist performing the examination in each instance. Despite this, based on our findings, we would recommend that the examination of choice for the evaluation of a patient for a possible N-TEF be a CS, as it is a simple, safe and accurate technique for documenting the presence of this entity.

Previous reports have suggested that the CS is not an effective technique for depicting N-TEF, that the technique often has to be repeated and, even then, does not always provide the diagnosis [2, 3, 14, 16, 28]. The alternative technique of the PBTE was described by Thomas and Chrispin [29] in 1969. Keats and Smith [30] suggested 4 years later that the tube esophagogram be performed with the immobilized infant lying prone or slightly Trendelen-



Fig. 6 Fluoroscopic spot view of the chest in a 4-day-old infant with an N-TEF who presented with cyanosis during feeding. The *arrow* shows the level in the esophagus where the obliquely oriented N-TEF is clearly depicted. The examination had been performed as a PBTE. Note the large amount of contrast material in the esophagus as well as in the airways extending out to the peripheral bronchioles

berg on the footrest of an upright fluoroscopy table. Since then a number of authors have described their experiences of infants with N-TEF who initially had a false-negative CS followed by a positive PBTE [14, 15, 18, 31].

The past difficulties of diagnosing N-TEF are emphasized by the variety of techniques described by various authors, including bronchoscopy and esophagoscopy [3, 4, 14, 16, 20, 22, 23, 28, 31] and even CT [24, 32]. Sonography has recently been successfully used by Gassner and Geley [25] to depict N-TEF in two of three patients [25]. Air bubbles were seen to cross the mediastinum from trachea to esophagus in one and ascend the esophagus in two; this is also known as the "atomizer" sign [33]. It is not known whether this sign can also be seen in reflux. The lack of radiation is an advantage, but sonography is operator-dependent and its effective use in larger numbers of patients in several institutions is required before this technique could be considered routine.

Of the above techniques, we prefer the CS as it is easier to perform with the patient lying horizontally in the right lateral decubitus position and requires no esophageal tube placement or injection of contrast material. The PBTE is technically more difficult than a CS and appropriate monitoring, restraining and suctioning of the infant is more difficult in the prone position on the footrest. Therefore, we believe that CS should be the examination of choice in most patients suspected of having an N-TEF. However, PBTE is indicated in patients who are intubated and if there are associated abnormalities that significantly increase the risk of aspiration. PBTE can be valuable in certain patients when contrast material is seen in the airway on CS and the radiologist is uncertain as to whether it is because of aspiration or an N-TEF. This situation occurred in three of the patients in this series.

There remains the question as to whether one should perform a PBTE in the face of a normal CS. We seldom do this, as we believe that PBTE is seldom required in such a situation as we have no record in our institution in which an N-TEF has been depicted by PBTE in a patient in whom a previous CS had been completely normal. Nevertheless, there are a few published reports of such cases [16, 18, 31]. This does not necessarily reflect a technical superiority of a

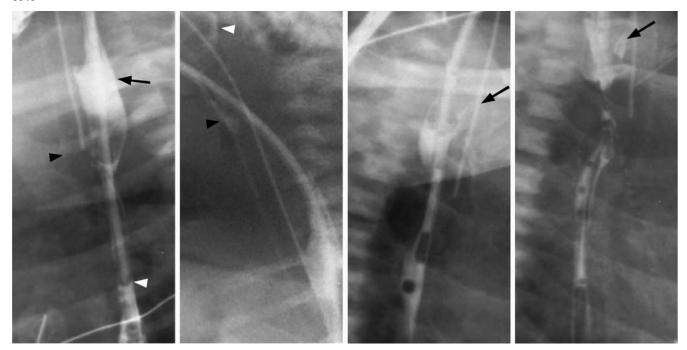


Fig. 7 PBTE in a 3-week-old infant with craniofacial anomalies and respiratory distress with an N-TEF. Images **a**—**d** were obtained with the infant lying horizontally in various obliquities on the fluoroscopic table. Images **a** and **b** were obtained with the patient in the right lateral decubitus position. In **a** the *arrow* shows the level in the esophagus where there is an anterior out-pouching, or nipple, suggesting the presence of an N-TEF. The *black arrowhead* shows the position of the tip of the endotracheal tube, and the *white arrowhead* indicates the tip of the esophageal tube noted in the lower esophagus. Note that despite the fact that the tube is in the lower esophagus, contrast material has passed quite far proximally

in the esophagus. **b** Later during the study contrast material was seen in the pharynx (*white arrowhead*) and in the larynx (*black arrowhead*). From these images it was uncertain whether the contrast material in the larynx had entered via aspiration from above or through an N-TEF. **c**, **d** The patient was then placed in a left lateral decubitus position, and more contrast material was injected into the esophagus. In these two images the *arrow* indicates a greater amount of contrast material entering the N-TEF and passing into the larynx. These images illustrate that when an endotracheal tube is in place it can prevent the contrast material from entering the fistula and airway when the patient is in one position but not in another

PBTE compared to a CS. The symptoms and signs of an N-TEF, while often present since birth, can be intermittent, and it is possible that depiction of the fistula on imaging might also be achievable in some patients only intermittently. This is thought to a result of intermittent occlusion of the N-TEF by redundant esophageal mucosa, which can act like a valve, or by muscle contraction or spasm of the muscular layers of the esophagus, which might also block off the fistula during swallowing [2, 4, 13, 28].

The decision whether to do a PBTE in the face of a completely normal CS depends on the clinical findings and the strength of the clinical suspicion for the presence of an N-TEF. The combination of coughing, cyanosis, aspiration pneumonia and abdominal distension (which may be severe) have been considered useful findings [16, 34]. In our patients we found the most useful clinical clues were the "three Cs" of cough, cyanosis (or apnea) and choking (or aspiration) during feeds. Although the three older children in this study came to medical attention primarily with recurrent lower respiratory tract infections, further review of their history showed they had evidence of the three Cs as well.

Independent of which technique is used, meticulous attention to technique is required, and one should attempt to produce good esophageal distension with contrast material. However, with CS the infant should not be fed

too rapidly and with PBTE the contrast material should not be injected too rapidly or forcefully in order to avoid flooding the airway with contrast material, either via the fistula or by aspiration. Particular attention should be paid to visualizing the upper trachea and larynx during fluoroscopy. This is important not only because the sites of most of the fistulae we encountered were high, but also so that contrast material seen in the upper airway because of aspiration is not confused with contrast that has passed through a fistula. Although rare, the possibility of two fistulous tracts should always be considered [11]. Attention should also be paid to the presence of the esophageal nipple sign, as this might be seen just prior to opacification of the fistulous tract and can be present in those patients in whom the tract itself is not opacified.

In our experience contrast material injected down a tube with its tip in the esophagus very often initially passes proximally up the esophagus rather than distally. This is an extremely important practical point, as it is important to know how far up the esophagus to pull the tube while slowly injecting the contrast material. The fistulae are usually high, and if the tube tip is pulled all the way up to that level the injected contrast material can pass proximally into the pharynx, making aspiration more likely. It is better to leave the tube tip slightly lower than the expected level of the fistula.

It should also be stressed that the presence of an endotracheal tube can obstruct the orifice of the fistula, preventing contrast material from passing from the esophagus through the fistula into the trachea. This indeed occurred in one of our patients who was intubated. By turning this patient to the contralateral decubitus position we were able to alter the tube's position slightly and demonstrate the N-TEF (Fig. 7).

Conclusion

We believe that CS should be the examination of choice in most patients suspected of having an N-TEF. However, PBTE is indicated in those patients who are intubated or who have associated abnormalities that significantly increase the risk of aspiration.

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