An anesthetic for the adult patient with congenital tracheoesophageal fistula: A case report

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A 20-year-old male with a history of recurrent pneumonia was diagnosed as having an N-type tracheoesophageal fistula. A general anesthetic was planned to facilitate the repair of the tracheoesophageal fistula using a left anterior cervical approach. Intraoperatively, the surgeons were unable to identify the defect after surgical exposure. To facilitate location of the tracheoesophageal fistula, a flexible pediatric fiberoptic bronchoscope was passed through an elbow adapter connected to the endotracheal tube. The scope was then visually passed via the trachea through the defect into the esophagus. The surgeons were able to palpate the fistula with the bronchoscope passed through the defect. Ease of identification allowed the tracheoesophageal fistula to be quickly repaired. At the completion of the surgery, the patient was extubated, and recovery was uneventful. The patient was discharged 48 hours postoperatively.

Key words: Tracheoesophageal fistula in the adult, congenital tracheoesophageal fistula, flexible fiberoptic pediatric bronchoscope.

Introduction
The occurrence of esophageal atresia with a tracheoesophageal (TE) fistula in the pediatric patient is rare, with an incidence of 1 in 3,000 live births to an incidence of an H-type fistula without esophageal atresia of about 1 in 100,000 live births. The diagnosis of esophageal atresia is commonly made close to the time of delivery, but it may be delayed until attempts at feeding are made. However, the diagnosis of a TE fistula can be delayed until adolescence in some cases or, in rare instances, until adulthood.

Since 1929, approximately 20 cases of congenital TE fistula have been reported in the adult. All reported cases have been of the H-type without esophageal atresia. The H-type TE fistula is the most compatible with long-term survival and generally the most difficult to diagnose.

A review of literature reveals that numerous cases of acquired TE fistula have been reported. These cases cite causes trauma, inflammatory processes caused by prolonged intubation or tracheostomy, caustic ingestion, esophagoscopy, and laser treatment for esophageal stricture, and they differ from the congenital cases. In acquired TE fistula diagnosis may be easier due to the size and location of the defect as well as a precipitating event, such as prolonged intubation. Conversely, cases of congenital TE fistula in an adult seem to be more difficult to diagnose due to the subtleness of symptoms and variations in the anatomy of the defect.

The reported cases of congenital TE fistula in adults have all been characterized by difficulty in diagnosing the defect. The most probable cause of
this difficulty appears to be the intermittent symptomatology that all patients describe. On initial presentation, all patients describe occasional respiratory difficulties as their primary reason for seeking care. The respiratory difficulties described by patients include sudden coughing after the ingestion of fluids, hemoptysis, choking spells, frequent coughing, productive cough, and fever as well as chest pain on inspiration. Interestingly, only after lengthy and in-depth questioning do patients reveal chronic respiratory problems associated with dysphagia as problematic.

The most probable cause of the intermittent symptomatology is an “N” configuration of an “H”-type defect. In the adult, the “N”-type configuration of the TE fistula is present because of differential growth of the esophagus and the trachea. In the newborn, the defect is recognized as being the H-type defect, but if it goes untreated differential growth of the trachea and esophagus causes the esophageal attachment to be pulled more caudal, while the tracheal attachment remains more cephalad (Figure 1).

Another anatomical difference that causes difficulty in diagnosis is the presence of a small tissue flap on either the esophageal or tracheal opening of the defect. The presence of such a flap has been described in two cases, in which the tissue flap was thought to act as a valve, intermittently preventing the passage of air and contrast material, even under pressure, from the esophagus to the trachea. These anatomical differences may help explain the intermittent and positional symptoms described by the patients.

A unique factor associated with the congenital TE fistula in the adult is the anatomical location of the defect. Of the cases described in the literature, 70% of the defects occur at or above the level of the second thoracic (T-2) vertebra. This may help explain the viability of adult patients, as higher level defects appear to be associated with fewer and less severe symptoms. This would also have an impact on anesthetic and surgical techniques. With 70% of the defects occurring at or above T-2, repair of most defects could be accomplished via a cervical or transverse supraclavicular versus a posterolateral thoracic approach.

Another factor in the patient who presents with a TE fistula is the association of other congenital anomalies. The newborn with TE fistula will present with major congenital anomalies in other organ systems 30-50% of the time. The association of such anomalies has been reported to occur at a rate of 27% in patients with the H-type defect and at a rate of approximately 50% with the other TE fistulas.

Anomalies associated with a TE fistula are numerous and include cardiac defects such as atrial septal defect, patent ductus arteriosus, and left ventricular anomalies. Other defects include anorectal agenesis, renal hypoplasia, and multiple vertebral anomalies. One reported case described a 15-year-old patient with a TE fistula who presented with numerous other congenital anomalies including an omphalocele, a clubfoot, hypospadias, a horseshoe kidney, a supernumerary rib, and osseous abnormalities of the thoracolumbar spine.

**Case summary**

A healthy, 80-kg, 20-year-old male presented after follow-up for chronic pulmonary problems including a vague history of chronic cough, occasional wheezing, and a history of recurrent pneumonia. The patient was evaluated as an outpatient for chronic cough and a low-grade fever. A consul-
tation with pulmonology services revealed the patient had a history of recurrent pneumonia, the last episode of which was 2 years before this visit. A chest radiograph revealed questionable infiltrates. Primary radiographic studies, a chest radiograph, and a barium swallow revealed "normal" appearing anatomy with the exception of the branch of the right upper lobe, which occurred at the distal trachea rather than at the right mainstem bronchus (Figure 2). Because of the patient's history of chronic symptoms and the presence of questionable infiltrates, a bronchoscopy was performed that resulted in identification of the fistula.

A barium swallow was again performed; however, this time the patient's position was changed throughout the procedure. The Trendelenberg position resulted in confirmation of the TE fistula with contrast medium (Figure 3).

A diagnosis of tracheoesophageal fistula was made, and the patient was referred to thoracic surgery and anesthesia service for surgical evaluation. The patient interview revealed a history of pulmonary symptoms with no "abnormal" findings. The patient stated that he had no difficulty in swallowing. However, further inquiry revealed that the patient had always considered swallowing, coughing, and swallowing again as "normal." The patient also revealed that nocturnal coughing associated with a "sour stomach" occurred regularly.

The patient was kept on nothing by mouth status after midnight, except for a gastric preparation, which included oral ranitidine 150 mg the night before and another 150 mg orally with 10 mg of metoclopramide by mouth the morning of surgery. He was counseled for general anesthesia with endotracheal intubation and also for an anterior cervical and a possible thoracic approach to repair his TE fistula.

Induction of anesthesia was performed with 20 μg of sufentanil followed by 10 mg of etomidate administered intravenously after the patient had been preoxygenated for 5 minutes. Endotracheal intubation was facilitated with 10 mg of vecuronium. The patient remained apneic after the administration of the induction agent until the endotracheal intubation was performed.

Cricoid pressure was administered on induction. This technique was employed although we realize that we could not assure protection of the patient's airway from gastric contents because the location of the defect could have been distal to the area of cricoid pressure. Endotracheal intubation was easily performed with the endotracheal tube cuff placed distal to the defect and above the carina. This was confirmed with sustained positive pressure insufflation of the lungs without gastric distension. Maintenance of anesthesia was carried out with an air, oxygen, and isoflurane mixture, as well as titrated doses of sufentanil.
Muscle relaxation was maintained with vecuronium and monitored with a nerve stimulator. Monitors that were used throughout the case included electrocardiographic and oxyhemoglobin saturation monitors, an end-tidal carbon dioxide monitor, a noninvasive blood pressure monitor, an in-line inspiratory oxygen analyzer, and a combination esophageal-temperature stethoscope.

An anterior cervical approach was used. An hour after incision, the surgeons noted difficulty in identifying the abnormality, either visually or by palpation. Because of the difficulty of surgical identification, the anesthetist recommended that a flexible pediatric fiberoptic bronchoscope be passed through the endotracheal tube and into the defect, which would enable the surgeons to palpate the defect through the bronchoscope. A flexible, pediatric fiberoptic bronchoscope was passed into the endotracheal tube through a fiberoptic bronchoscope swivel adapter. The adapter with seal allowed for continuous positive pressure ventilation. The fiberoptic bronchoscope passed easily through the endotracheal tube, but the defect was not visualized.

After withdrawing the endotracheal tube 1-2 cm, the defect was seen distal to the tip of the endotracheal tube. The tip of the flexible bronchoscope was passed through the defect into the esophagus, facilitating palpation and identification of the defect by the surgeons (Figure 4), who then passed suture ligatures around both ends of the TE fistula before the flexible fiberoptic bronchoscope was removed. Manual positive pressure ventilation was uninterrupted throughout this procedure.

After completion of the surgical procedure, muscle relaxation was reversed with neostigmine 2.5 mg and glycopyrrolate 0.5 mg intravenously. Oropharyngeal suctioning was performed, and after demonstrating appropriate muscle strength by sustained head lift, the patient was extubated and transferred to the postanesthesia recovery room. The patient showed no signs of difficulty postoperatively and was discharged 2 days after surgery. He was seen 2 weeks after surgery by the surgeon, at which time the patient stated he no longer had dysphagia or multiple swallowing episodes. The patient also noted that the nocturnal coughing episodes had stopped. No postoperative studies were carried out.

Discussion
This case does not appear to be unique in presentation, based on a review of other cases reported; in fact, there are many similarities.

As stated earlier, other congenital defects are associated with patients who have a TE fistula, although fewer of those associated anomalies were noted in the previously undiagnosed adult. In our case, it was observed that the patient had an abnormal tracheobronchial tree; no other anomalies were noted.

Another similarity between our case and others reported was the level of the TE fistula. About 70% of the previously reported cases had defects at or above the level of the second thoracic vertebra, a fact that was true in our case as well, because the defect was only 2 cm below the level of the vocal cords.

The configuration of the defect was also similar to other reported cases. In our case, the patient demonstrated an N-type configuration of the TE fistula. As in other reported cases, this may help to explain the patient’s intermittent symptoms and those elicited by abrupt position changes.

Our case allowed us to employ a technique that has not been described previously. When the surgeons had difficulty recognizing the defect, the anesthetists were able to assist them by passing a flexible pediatric fiberoptic bronchoscope through the endotracheal tube and into the defect to locate the fistula.

Although this technique seemed to be unique, a similar method has been found in the literature. A previous case describes the use of a vascular wire, which was passed through a TE fistula using a bronchoscope before induction of anesthesia. Once the wire was in place, the patient was brought to the operating room, where induction of anesthesia was carried out. The defect was rapidly located due to the placement of the vascular wire. The
wire was palpated through the surgical incision, and the fistula was easily repaired.

A fiberoptic bronchoscope was used in two other cases of TE fistula. In the first case, the scope was passed through the defect via the endotracheal tube to assist with the percutaneous placement of a Fogarty embolectomy catheter and gastrostomy tube. Once it was in place in the distal esophagus, the catheter allowed for positive pressure ventilation and optimum oxyhemoglobin saturation. The repair was performed without difficulty in the pediatric patient, and no respiratory embarrassment was noted.

In the second case, a fiberoptic esophagoscopy was performed to occlude a fistula that resulted from the repair of an esophageal stricture caused by a mediastinal carcinoma. A balloon catheter was placed at the site of the defect in an attempt to prevent the movement of gastric contents into the pulmonary system. This procedure was performed as a temporary measure to prevent further respiratory compromise, but it proved ineffective.

We note here the importance of a good interview technique. Our patient, as has been noted with previous patients, could not describe his symptoms as anything but normal. Since our patient had experienced these same symptoms throughout his life, he believed that his method of multiple swallowing was normal. Therefore, it was more appropriate to let the patient describe his symptoms rather than have him answer direct questions.

This patient had sought treatment for respiratory symptoms in the past. In fact, he had been diagnosed and treated for the symptoms of pneumonia on more than one occasion. We recognize that the patient’s complaints of coughing and wheezing accompanied by a history of recurrent pneumonia were compatible with asthma or reactive airway disease. However, the previous questioning and examination did not take into account the possibility of gastrointestinal involvement in the patient’s symptoms. We believe that if a more detailed history that included the patient’s description of his symptoms had been taken, the recurrence of pneumonia might have been suspected or diagnosed as a congenital TE fistula at an earlier time.

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