

# Thoracoscopic Repair of Esophageal Atresia and Tracheoesophageal Fistula

## A Multi-Institutional Analysis

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**Objectives:** For the past 60 years, successful repair of esophageal atresia (EA) and distal tracheoesophageal fistula (TEF) has been performed via a thoracotomy. However, a number of reports have described adverse musculoskeletal sequelae following thoracotomy in infants and young children. Until now, only a few scattered case reports have detailed an individual surgeon's success with thoracoscopic repair of EA/TEF. This multi-institutional review represents the largest experience describing the results with this approach.

**Methods:** A cohort of international pediatric surgeons from centers that perform advanced laparoscopic and thoracoscopic operations in infants and children retrospectively reviewed their data on primary thoracoscopic repair in 104 newborns with EA/TEF. Newborns with EA without a distal TEF or those with an isolated TEF without EA were excluded.

**Results:** In these 104 patients, the mean age at operation was 1.2 days ( $\pm 1.1$ ), the mean weight was 2.6 kg ( $\pm 0.5$ ), the mean operative time was 129.9 minutes ( $\pm 55.5$ ), the mean days of mechanical ventilation were 3.6 ( $\pm 5.8$ ), and the mean days of total hospitalization were 18.1 ( $\pm 18.6$ ). Twelve (11.5%) infants developed an early leak or stricture at the anastomosis and 33 (31.7%) required esophageal dilatation at least once. Five operations (4.8%) were converted to an open thoracotomy and one was staged due to a long gap between the 2 esophageal segments. Twenty-five newborns (24.0%) later required a laparoscopic fundoplication. A recurrent fistula between the esophagus and trachea developed in 2 infants (1.9%). A number of other operations were required in these patients, including imperforate anus repair in 10 patients (7 high, 3 low), aortopexy (7),

laparoscopic duodenal atresia repair (4), and various major cardiac operations (5). Three patients died, one related to the EA/TEF on the 20th postoperative day.

**Conclusions:** The thoracoscopic repair of EA/TEF represents a natural evolution in the operative correction of this complicated congenital anomaly and can safely be performed by experienced endoscopic surgeons. The results presented are comparable to previous reports of babies undergoing repair through a thoracotomy. Based on the associated musculoskeletal problems following thoracotomy, there will likely be long-term benefits for babies with this anomaly undergoing the thoracoscopic repair.

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In 1912, Jacobaeus first described the technique of thoracoscopy using a cystoscope to lyse pleural adhesions in patients with pulmonary tuberculosis.<sup>1</sup> This technique was widely used in the treatment of patients with pulmonary tuberculosis until effective antituberculous drug therapy became available in the 1950s. The modern era of thoracoscopy in children was introduced in a report in 1976 by Rodgers and Talbert describing 9 patients undergoing thoracoscopy for diagnostic purposes.<sup>2</sup> With the rapid progression and evolution of minimally invasive surgery in the 1990s, refinements in optical technology, instrumentation, and surgical skill has led to an explosion in the use of thoracoscopy for multiple purposes in children. To date, there have been only a few small reports of children undergoing thoracoscopy for the management of esophageal atresia (EA) and distal tracheoesophageal fistula (TEF).<sup>3–5</sup> Moreover, it is unclear if this technology can be used at multiple institutions with optimal results. Therefore, this report describes the use of thoracoscopy for repair of EA/TEF in newborn babies from 6 institutions around the world. By far, this is the largest reported experience using this approach for correction of this congenital anomaly.

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**METHODS**

A retrospective review of all patients undergoing thoracoscopic repair of EA/TEF in newborns was performed at 6 institutions around the world (Table 1). Data collected included the newborn's age and weight at the time of the operation, operative time, number of days of mechanical ventilation, duration of total hospitalization, the number of patients with leak and/or stricture formation, as well as the number of patients requiring esophageal dilation. In addition, other data included the number of operations that were staged or converted to a thoracotomy and the number of patients who required fundoplication, imperforate anus repair, aortopexy, duodenal atresia repair, and cardiac operations. Finally, other morbidity and mortality were also recorded.

**Technique**

All babies underwent tracheal intubation in this series. Moreover, all except 2 babies were placed in the 45° prone position to allow the lung to fall away from the posterior mediastinum. Three cannulas were used for access to the thoracic cavity in all but 5 patients. In these 5 patients, 4 ports were employed. This fourth port was used primarily for lung retraction. Carbon dioxide insufflation was used in all patients to effect lung collapse. The fistula was ligated using a variety of techniques. In 37 patients, the fistula was occluded with a suture ligature. In the remaining cases, a clip was placed on the fistula tract and the fistula was divided distal to it. The anastomosis was performed with either 4-0 or 5-0 suture. In 40 patients, PDS (polydioxanone, Ethicon, Inc., Somerville, NJ) was used and in another 46 patients, Vicryl (polyglactin, Ethicon, Inc., Somerville, NJ) suture was used. In 11 cases, silk was employed and "other" suture material was used in the remaining 7 patients. In 42 patients, the esophageal anastomosis was performed using extracorporeal knot-tying techniques and, in the remainder, intracorporeal suturing was used.

**RESULTS**

In this retrospective review, the mean age of the patients at the time of operation was 1.2 days ( $\pm 1.1$ ) and the

mean weight at operation was 2.6 kg ( $\pm 0.5$ ). The mean operative time for the entire series was 129.9 minutes ( $\pm 55.5$ ) and the mean days of mechanical ventilation were 3.6 ( $\pm 5.8$ ). The mean duration of total hospitalization was 18.1 days ( $\pm 18.6$ ). Twenty-one patients later required Nissen fundoplication and 4 underwent Thal fundoplication. Although relatively outdated, the data were further characterized according to the Waterston classification to allow comparison with previous studies utilizing thoracotomy. These data appear in Table 2.

In this series, only one newborn had a second fistula between the trachea and proximal esophagus. Two patients developed recurrent fistulae. In one of these, the recurring fistula was found at 3 months of age and repaired thoracoscopically. In the other patient, symptoms did not develop until the patient was 8 months of age. This patient underwent thoracotomy and successful repair at that time.

Six patients were found to have a right aortic arch. In 1 patient, the operation was converted from a thoracoscopic right procedure to an open left operation. In 3 patients, the conversion was from a thoracoscopic right operation to a thoracoscopic left operation. The right aortic arch was identified preoperatively and a left thoracoscopic operation was successfully performed in 2 patients.

One patient required a 2-stage operation for successful repair of a long gap esophageal atresia and fistula. At the first stage, an attempt was made to close the 2 esophageal segments. When this was not possible, the fistula was ligated thoracoscopically. At 3 months of age, a thoracotomy was used to anastomose the 2 esophageal segments. Even with this staged approach, 2 proximal esophageal myotomies were needed.

Five patients underwent conversion from a thoracoscopic procedure to an open thoracotomy. As mentioned previously, one patient with a right aortic arch (despite a negative echocardiogram to that effect) was converted to a left thoracotomy. Three patients underwent conversion because of intraoperative desaturations and a relatively long gap between the 2 esophageal segments. A 1.2-kg baby under-

**TABLE 1.** A Listing of Hospitals, Their Locations, and Authors Participating in This Study

Institution	Location	Authors
Children's Mercy Hospital	Kansas City, MO	Holcomb and Ostlie
Hospital for Infants and Children at Presbyterian-St. Lukes Medical Center	Denver, CO	Rothenberg
Wilhelmina Children's Hospital	Utrecht, The Netherlands	Bax and van der Zee
J.P. Garrahan National Children's Hospital	Buenos Aires, Argentina	Martinez-Ferro
Lucille Packard Children's Hospital	Palo Alto, CA	Albanese
Chinese University of Hong Kong	Hong Kong, China	Yeung

**TABLE 2.** A Description of the Results in the Series of 104 Babies Undergoing Thoracoscopic Repair of EA/TEF According to Their Preoperative Waterston Classification

	Waterston A* n = 62	Waterston B† n = 30	Waterston C‡ n = 12
Operation converted	2	2	1
Operation staged	1	—	—
Esophageal anastomotic leak	2	3	3
Stricture (on initial esophagram)	3	1	—
Patients needing only 1 dilation	7	5	—
Patients needing 2 dilations	9	1	2
Patients needing 3 dilations	—	3	1
Patients needing >3 dilations	3	2	—
Recurrent tracheoesophageal fistula	1	1	—
Fundoplication	19	6	1
Imperforate anus operations	4	4	2
Duodenal atresia repairs	—	2	2
Aortopexy	6	1	—
Death	1	—	2

\*Waterston A: >5.5 lb with no significant associated problems.

†Waterston B: 4–5.5 lb or higher weight with moderate pneumonia or congenital anomaly.

‡Waterston C: weight <4 lb or greater weight with severe pneumonia or congenital anomaly.

went conversion after placement of only one port because it was felt that the baby’s thoracic cavity was too small to perform the operation thoracoscopically.

A number of babies were found to have other significant anomalies (Table 3). Ten patients had anorectal atresia, with 7 having high anorectal atresia and 3 a low anomaly. Those with the high anomalies required colostomy followed by a pull-through at a later date. Four babies had duodenal atresia, all of whom underwent laparoscopic duodenal atresia repair. Seven patients underwent aortopexy, with 6 of them being performed thoracoscopically. Five patients required cardiac operations other than a ventricular/atrial septal defect repair.

Three patients died in this series. One Waterston A baby, who was recovering uneventfully, died at 7 months of age after the development of necrotizing enterocolitis, which was attributed to drinking herbal tea. Two Waterston C babies died. One baby died at day 10 of life from severe

congenital heart disease after successful thoracoscopic EA/TEF repair. A third baby was recovering uneventfully, but, because of desaturations related to congenital heart disease, required an emergency intubation in which the endotracheal tube was thrust through the fistula closure on the trachea. This baby died in the operating room at the time of attempted repair of this complication.

## DISCUSSION

In 1703, in his book *Anatomy of Humane Bodies Epitomized*, Thomas Gibson provides the first description of a baby with EA/TEF.<sup>6</sup> However, it would be several centuries later until any surgeon attempted primary repair of this congenital anomaly. In 1936, Thomas Lanman performed the first primary extrapleural anastomosis of this anomaly, but the child lived only 3 hours.<sup>7</sup> Three other attempts by Lanman, unfortunately, resulted in a similar outcome. On successive days in 1939, William Ladd and N. L. Leven performed gastrostomies on newborns with this anomaly. Both children lived to undergo subsequent extrapleural division of the fistula, cervical esophagostomy, and, much later, esophageal substitution.<sup>8,9</sup> As discussed in Gibson’s original report, until these 2 successes approximately 400 cases had been diagnosed and reported in the literature without successful repair.<sup>10</sup> In 1941, Cameron Haight ligated and divided the TEF in a baby and performed an end-to-end esophageal anastomosis through a left extrapleural approach.<sup>11</sup> The anastomosis leaked, but closed spontaneously. A gastrostomy was performed on the 10th postoperative day and the gastrostomy tube was removed on the 42nd day. A stricture developed at the anastomotic site that responded successfully to a single dilation 17 months after the operation. Although this patient was hospitalized for almost 18 months, she recovered uneventfully and is currently alive and well (Arnold Coran, personal correspondence, February 2005). After this successful outcome, other reports of successful primary repair were generated, and the care of babies with EA/TEF rapidly progressed.

Between 1945 and 1965, the focus centered on achieving successful repair in healthy babies with no associated anomalies and without severe pneumonia. In 1962, David Waterston reported his classification scheme in which babies with EA/TEF were placed in one of 3 groups based on their weight and the presence/absence of pneumonia or other congenital anomalies.<sup>12</sup> By 1965, with an increased understanding of infant physiology and advancements in anesthesia, a success rate of 80–90% was achievable for repair of this disorder in most Waterston A babies.<sup>13</sup> For the next 25 years, refinement in operative technique and attempts at improved survival with primary anastomosis in the low-birth-weight infant and the infant with coexisting morbidities (Waterston B and C) dominated the literature of this disorder.<sup>14,15</sup> Also, during these years, there were reports docu-

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**TABLE 3.** Associated Anomalies in the Group of 104 Patients Undergoing Thoracoscopic Repair of Esophageal Atresia and Fistula

Anomaly	No. Patients	Anomaly	No. Patients
Cardiac		Renal	
ASD/VSD	15	Horseshoe kidney	3
Right aortic arch	6	Unilateral agenesis	2
Tetralogy of Fallot	3	Crossed fused ectopia	1
Dextrocardia	3	VUR > Grade 3	1
PDA (ligation)	2	Duplex kidney	1
DORV	1	Ectopic kidney	1
Tricuspid atresia	1	Other	
Gastrointestinal		Vertebral anomalies	6
High imperforate anus	7	Radial aplasia	3
Duodenal atresia	4	Tethered cord	1
Low imperforate anus	3	Hydromyelia	1
Cloaca	1	Choanal atresia	1
Syndromes			
VACTERL (>2 anomalies)	10		
CHARGE	3		
Down	3		

ASD, atrial septal defect; CHARGE, Coloboma, Heart disease, Atresia choanae, Retarded growth and development, Genital anomalies, Ear anomalies; DORV, double outlet right ventricle; VACTERL, Vertebral, Anorectal, Cardiac, TracheoEsophageal, Renal, Limb; VSD, ventricular septal defect; VUR, vesicoureteral reflux.

menting the effectiveness of aortopexy for management of tracheal compression, the concept of staged repair in the very low–birth-weight and high-risk infants, and survival of low–birth-weight infants with gastrointestinal perforation.<sup>16–18</sup> Moreover, technical refinements in the operation were developed, including the concept of circular myotomy of the proximal esophageal pouch to aid in shortening the gap between the 2 esophageal segments.<sup>19</sup> With the rapid evolution of and increasing experience with thoracoscopy, it is only natural that this approach would be attempted for repair of this congenital anomaly.

In evaluating this current series, the patient characteristics are quite similar to other large reports in which open thoracotomy has been employed.<sup>20–24</sup> Three of these reports were published in the 1980s; one appeared in 1995, and another in 2001. Also, the incidence of postoperative complications is similar (Table 4). Five of these reviews, in which a number of the postoperative complications are well detailed, showed the incidence of anastomotic leak ranged from 10% to 21%. The incidence of stricture development averaged 18%, and the percentage of patients requiring fundoplication varied from 15% to 32.2%. In our thoracoscopic series, the leak rate (7.6%), stricture rate (3.8%), and rate of fundoplication (24.0%) favorably compares with these previous reports. Additionally, the rate of recurrent fistulization between the trachea and esophagus was between 2.2% and 12% in these open thoracotomy series,

whereas in this report, a recurrent fistula developed in 2 of 104 patients (1.9%). Even the most recent report had a 3.3% incidence of recurrent TEF.<sup>24</sup> The incidence of infants in this series requiring aortopexy for refractory symptoms due to tracheal compression (6.7%) compares favorably with 2 of these earlier reports.<sup>20,21</sup>

The association of gastroesophageal reflux in patients with EA/TEF is well documented. There is a recent trend toward more aggressive medical management resulting in the need for fewer funduplications.<sup>25</sup> Nevertheless, different surgeons have different thresholds for performing a fundoplication in these patients. Two recent reviews have documented a 23% incidence of fundoplication in 125 EA/TEF patients and a 100% fundoplication rate in 21 EA/TEF patients.<sup>26,27</sup> Of the 5 studies that were used for comparative purposes with this current thoracoscopic review, there was a 32.2% incidence of fundoplication in the most contemporary study.<sup>24</sup>

In evaluating the short-term results of this current series, although 33 patients (31.7%) required esophageal dilation, only 2 required revision of the anastomosis. These revisions were performed at the time of ligation of a recurrent TEF. The overall mortality was 2.8%, which also compares favorably with the other reports.<sup>20–24</sup> In fact, of the 3 patients in this current series who died, only one was related to a complication of the EA/TEF. It could easily be argued that

**TABLE 4.** An Attempt at Comparing this Thoracoscopic Series with Previous Reports Using Thoracotomy of Patients Undergoing Repair of Esophageal Atresia with Distal Tracheoesophageal Fistula (Gross Type C)

	Current Study	Engum, et al <sup>23</sup> (1971–93)	Spitz, Keily <sup>22</sup> (1980–84)	Randolph, et al <sup>24</sup> (1982–88)	Manning, et al <sup>21</sup> (1977–85)	Yanchar, et al <sup>25</sup> (1989–99)
Number of Patients	104	174	148*	39	63	90
Mean length of hospitalization (days)	18.1 (6–120)	NR	NR	NR	24 (9–174)	NR
Anastomotic leak	7.6%	NR	21%	10.2%	17%	16.6% <sup>†</sup>
Anastomotic stricture	3.8% <sup>‡</sup>	32.7% <sup>§</sup>	17.7%	33.3%	4.3% <sup>¶</sup>	17% <sup>  </sup>
Patients requiring at least 1 dilation	31.7%	32.7%	NR	33.3%	NR	17%
Anastomotic Revision	1.9%	0.9%	2.7%	5.1%	NR	NR
Fundoplication	24.0%	25.2%	18%	15.3%	16.9%	32.2%
Aortopexy	6.7%	NR	16%	NR	4.7%	NR
Mortality	0.9%	4.5%	14.8%	0%	3.1%	1.1%
Related EA/TEF	1.9%	(overall)	(overall)	7.6%	11.1%	3.2%
Unrelated EA/TEF	2.8%			7.6%	14.2%	3.3%
Recurrent fistula	1.9%	2.2%	12%	5.1%	6.4%	3.3%

\*87% are Gross Type C.

<sup>†</sup>Leak is defined as both a clinical and radiographic leak at the anastomosis.

<sup>‡</sup>Stricture in this paper is defined as requiring > 4 dilations.

<sup>§</sup>Stricture in this paper is defined as requiring > 2 dilations.

<sup>¶</sup>Stricture is defined as a significant narrowing on the initial esophagram.

<sup>||</sup>Stricture is defined as requiring one or more esophageal dilations > 6 wk after repair.

NR, not reported.

this complication was not related to the thoracoscopic approach.

If the operative results in this series are comparable to those achieved with the open operation, what may be the advantages of the thoracoscopic approach? The primary advantage lies in the potential for a reduction in the musculoskeletal sequelae that often develop following thoracotomy in the newborn. These complications have been fairly well detailed. In one report of 89 surviving patients who underwent right thoracotomy for repair of EA/TEF and who were available for follow-up longer than 3 years, 29 (32.5%) had significant musculoskeletal deformities.<sup>28</sup> Twenty-one of these 89 patients (23.6%) had a “winged” scapula, 18 (20.2%) had asymmetry of the thoracic wall secondary to atrophy of the serratus anterior muscle, and 16 (17.9%) had severe (>10%) thoracic scoliosis. Mammary maldevelopment was found in one of the 11 females and required reconstructive surgery. Similar results have been found in a number of other reports that have described musculoskeletal deformities secondary to thoracotomy in infants and young children.<sup>29–34</sup>

Spreading the ribs after entering the thoracic cavity may be one of the significant causes of postoperative pain following thoracotomy. In adults, chronic pain has been reported in more than 50% of patients undergoing thoracot-

omy.<sup>35,36</sup> This information has not been reported in children. Because of these known problems following thoracotomy, pediatric surgeons have tried to avoid these complications for over 50 years. When performing a thoracotomy, Sir Dennis Browne used a vertical skin incision, detached the serratus anterior muscle and then opened the intercostal space.<sup>37</sup> Soucy and colleagues described muscle sparing thoracotomy in children and many pediatric surgeons use this technique.<sup>38</sup> Bianchi et al advocated a transaxillary approach for thoracic exposure in neonates.<sup>39</sup> In an adult study, Khan and colleagues reported that a muscle-sparing thoracotomy preserves muscle strength but does not eliminate the chronic postoperative pain.<sup>40</sup> Thus, undesirable musculoskeletal morbidity following thoracotomy should be markedly lessened, if not eliminated, as a result of using the thoracoscopic approach.

Although the authors believe that the primary advantage of the thoracoscopic approach is a reduction in these musculoskeletal sequelae, other unanticipated benefits have been noted. One of these is superior visualization of the anatomy within the thoracic cage. This improved visualization is especially true for identification of the tracheo-esophageal fistula. Also, because the fistula is visualized perpendicular to its connection to the membranous trachea, the exact site for ligation is easily identifiable. Ligation of the fistula with 5-mm titanium clips was performed routinely in the

majority of operations in this series. Although one perceived disadvantage is migration of the clip through the wall of the fistula and the development of a recurrent fistula, only 2 patients developed this complication. This compares very favorably with other series of patients undergoing thoracotomy and suture closure or suture ligation of the fistula.

The transpleural route might be regarded as a disadvantage of the thoracoscopic approach. However, several reports have described patients undergoing a transpleural repair without a difference in mortality and morbidity when compared with the retropleural approach.<sup>41–43</sup>

When analyzing the results of this current series, an important caveat is that these operations were accomplished by surgeons who have been performing advanced laparoscopic and thoracoscopic procedures in infants and children for over 10 years. There is a certain amount of surgical expertise required to successfully complete this operation thoracoscopically. The authors feel that there is certainly a “learning curve” for this approach. Also, some patients are probably not good candidates for the thoracoscopic approach. The operation is difficult in babies less than 2 kg and in babies with significant lung disease, since it requires the ipsilateral lung to be compressed with the operative pneumothorax to achieve an adequate working space. Several of the authors used intracorporeal knot tying, but others preferred the extracorporeal technique. Although the extracorporeal knot tying may be simpler and may save operating time, some of the authors feel that intracorporeal tying is just as efficacious. Positioning of the baby in a three-quarters prone position is vitally important to allow the lung to fall away from the posterior mediastinum through gravity and positive pressure insufflation. Such positioning allows the surgeon to work behind the majority of the lung rather than over it and leads to very little, if any, prolonged damage to the lung. At the same time, continuous communication between the surgeon and anesthesiologist is imperative when performing these operations.

This phase I report shows that the thoracoscopic approach for repair of EA/TEF is safe and efficacious. A randomized trial is necessary to show that it is a better option than the open technique. The thoracoscopic approach is a further refinement in the pediatric surgeon's desire to achieve an optimal result in an infant or child with a minimally invasive approach. In addition, it is a natural evolution in the advancement of minimally invasive surgical and endoscopic techniques for management of congenital pediatric surgical diseases. It is through these advancements that improvements in patient care can be delivered to even the tiniest of patients.

## REFERENCES

- Jacobaeus HC. Über die möglichkeit die zystoskopie bei untersuchung se roser hohlungen anzuwenden. *Munch Med Wochenschr.* 1910;40:2090–2092.
- Rodgers BM, Talbert JL. Thoracoscopy for diagnosis of intrathoracic lesions in children. *J Pediatr Surg.* 1976;11:703–708.
- Lobe TE, Rothenberg S, Waldschmidt J, et al. Thoracoscopic repair of esophageal atresia in an infant: a surgical first. *Ped Endosurg Innov Techniqes.* 1999;3:141–148.
- Rothenberg SS. Thoracoscopic repair of tracheo-esophageal fistula in newborns. *J Pediatr Surg.* 2002;37:869–872.
- Bax NMA, van der Zee DC. Feasibility of thoracoscopic repair of esophageal atresia with distal fistula. *J Pediatr Surg.* 2002;37:192–196.
- Gibson T. *The Anatomy of Humane Bodies Epitomized*, 6th ed. London: Awnsham and Churchill; 1703.
- Lanman TH. Congenital atresia of the esophagus: a study of thirty-two cases. *Arch Surg.* 1940;41:1060–1083.
- Ladd WE. The surgical treatment of esophageal atresia and tracheo-esophageal fistulas. *New Eng J Med.* 1944;230:625–637.
- Leven NL. Congenital atresia of the esophagus with tracheo-esophageal fistula: report of fistulous communication and cervical esophagostomy. *J Thoracic Surg.* 1941;10:648–657.
- Ashcraft KW, Holder TM. The story of esophageal atresia and tracheo-esophageal fistula. *Surgery.* 1969;65:332–340.
- Haight C, Towsley H. Congenital atresia of the esophagus with tracheo-esophageal fistula: extrapleural ligation of fistula and end-to-end anastomosis of esophageal segments. *Surg Gynecol Obstet.* 1943;76:672–688.
- Waterston DJ, Bonham Carter RE, Aberdeen E. Oesophageal atresia: tracheo-esophageal fistula. *Lancet.* 1962;1:819–822.
- Koop CE, Hamilton JP. Atresia of the esophagus: increased survival with staged procedures in the poor-risk infant. *Ann Surg.* 1965;162:389–401.
- Pohlson EC, Schaller RT, Tapper D. Improved survival with primary anastomosis in the low birth weight neonate with esophageal atresia and tracheoesophageal fistula. *J Pediatr Surg.* 1988;23:418–421.
- Templeton JM Jr., Templeton JJ, Schnauffer L, et al. Management of esophageal atresia and tracheoesophageal fistula in the neonate with severe respiratory distress syndrome. *J Pediatr Surg.* 1985;20:394–397.
- Schwartz MZ, Filler RM. Tracheal compression as a cause of apnea following repair of tracheoesophageal fistula: treatment by aortopexy. *J Pediatr Surg.* 1980;15:842–848.
- Alexander F, Johanningman J, Martin LW. Staged repair improves outcome of high-risk premature infants with esophageal atresia and tracheoesophageal fistula. *J Pediatr Surg.* 1993;28:151–154.
- Holcomb GW III. Survival after gastrointestinal perforation from esophageal atresia and tracheoesophageal fistula. *J Pediatr Surg.* 1993;28:1532–1535.
- Lindahl H, Louhimo I. Livaditis myotomy in long-gap esophageal atresia. *J Pediatr Surg.* 1987;22:109–112.
- Manning PB, Morgan RA, Coran AG, et al. Fifty years' experience with esophageal atresia and tracheoesophageal fistula. *Ann Surg.* 1986;204:446–451.
- Spitz L, Kiely E, Brereton RJ. Esophageal atresia: five-year experience with 148 cases. *J Pediatr Surg.* 1987;22:103–108.
- Engum SA, Grosfeld JL, West KA, et al. Analysis of morbidity and mortality in 227 cases of esophageal atresia and/or tracheoesophageal fistula over two decades. *Arch Surg.* 1995;130:502–509.
- Randolph JG, Newman KD, Anderson KD. Current results in repair of esophageal atresia with tracheoesophageal fistula using physiologic status as a guide to therapy. *Ann Surg.* 1989;209:526–553.
- Yanchar NL, Gordon R, Cooper M, et al. Significance of the clinical course and early upper gastrointestinal studies in predicting complications associated with repair of esophageal atresia. *J Pediatr Surg.* 2001;36:815–822.
- Konkin DE, O'Hali WA, Webber EM, et al. Outcomes in esophageal atresia and tracheoesophageal fistula. *J Pediatr Surg.* 2003;38:1726–1729.
- Bergmeijer JH, Tibboel D, Hazebroek FW. Nissen fundoplication in the management of gastroesophageal reflux occurring after repair of esophageal atresia. *J Pediatr Surg.* 2000;35:573–576.
- Esposito C, Langer JC, Shaarschmidt K, et al. Laparoscopic antireflux procedures in the management of gastroesophageal reflux following esophageal atresia repair. *J Pediatr Gastroenterol Nutr.* 2005;40:349–351.
- Jaureguizar E, Vazquez J, Murcia J, et al. Morbid musculoskeletal

- sequelae of thoracotomy for tracheoesophageal fistula. *J Pediatr Surg*. 1985;20:511–514.
29. Freeman NV, Walkden J. Previously unreported shoulder deformity following right lateral thoracotomy for esophageal atresia. *J Pediatr Surg*. 1969;4:627–636.
  30. Durning RP, Scoles PV, Fox OD. Scoliosis after thoracotomy in tracheoesophageal fistula patients. *J Bone Joint Surg Am*. 1980;62:1156–1158.
  31. Cherup LL, Siewers RD, Futrell JW. Breast and pectoral muscle maldevelopment after anterolateral and posterolateral thoracotomies in children. *Ann Thorac Surg*. 1986;41:492–497.
  32. Westfelt JN, Nordwall A. Thoracotomy and scoliosis. *Spine*. 1991;16:1124–1125.
  33. Chetcuti P, Myers NA, Phelan PD, et al. Chest wall deformity in patients with repaired esophageal atresia. *J Pediatr Surg*. 1989;24:244–247.
  34. Emmel M, Ulbach P, Herse B, et al. Neurogenic lesions after posterolateral thoracotomy in young children. *Thorac Cardiovasc Surg*. 1996;44:86–91.
  35. Perttunen K, Tasmuth T, Kalso E. Chronic pain after thoracic surgery: a follow-up study. *Acta Anaesthesiol Scand*. 1999;43:563–567.
  36. Rogers ML, Duffy JP. Surgical aspects of chronic post-thoracotomy pain. *Eur J Cardiothorac Surg*. 2000;18:711–716.
  37. Browne D. Patent ductus arteriosus. *Proc R Soc Med*. 1952;45:719–722.
  38. Soucy P, Bass J, Evans M. The muscle sparing thoracotomy in infants and children. *J Pediatr Surg*. 1991;26:1232–1235.
  39. Bianchi A, Sowande O, Alizai NK, et al. Aesthetics and lateral thoracotomy in the neonate. *J Pediatr Surg*. 1998;33:1798–1800.
  40. Khan IH, McManus KG, McCraith A, et al. Muscle sparing thoracotomy: a biomechanical analysis confirms preservation of muscle strength but no improvement in wound discomfort. *Eur J Cardiothorac Surg*. 2000;18:656–661.
  41. Hicks LM, Masfield PB. Esophageal atresia and tracheoesophageal fistula. Review of thirteen years' experience. *J Thorac Cardiovasc Surg*. 1981;81:358–363.
  42. McKinnon LJ, Kosloske AM. Prediction and prevention of anastomotic complications of esophageal atresia and tracheoesophageal fistula. *J Pediatr Surg*. 1990;25:778–781.
  43. Bishop JP, Klein MD, Philippart AI, et al. Transpleural repair of esophageal atresia without a primary gastrostomy: 240 patients treated between 1951 and 1983. *J Pediatr Surg*. 1985;20:823–828.

## Discussions

DR. JAMES A. O'NEILL, JR. (NASHVILLE, TENNESSEE): As with many other procedures, we are in the process of evaluating whether minimally invasive approaches to standard operations bring added value in terms of outcomes and costs. In this excellent report of 104 carefully analyzed cases approached thoroscopically, the authors have proven without any doubt in my mind that operative management of this entity can be accomplished using minimally invasive techniques with reasonable morbidity and mortality. The techniques described in the manuscript are beautifully described and will be very helpful to those who have an interest in pursuing this further.

However, while the authors have proven that it can be done, the study is really a Phase I study, and the question still exists as to whether it is better than the traditional open approach which will require randomization. And there is some reason to question this issue.

The operative time of a little over 2 hours is slightly shorter than with reports about the open operation; time on a

ventilator postop of 3 and a half days is somewhat longer; and discharge at 18 days is similar. So where is the advantage?

It has been said that musculoskeletal sequelae are higher with open thoracotomy. Most experienced individuals have been able to avoid such sequelae for at least 10 to 15 years since modern techniques have been applied. It is also said that visualization of the fistula is better with the scope. But the real issue is whether you can do what you need to do when you get there. Therefore, I focused on the issue of post-op esophageal anastomotic complications.

The authors compared their results with 4 different series in the manuscript. But unfortunately, these reports range from 12 to 20 years in age and they are arguably not representative of contemporary outcomes. In a comparison of this series with our own recent contemporary results taking a sample of 63 cases, I will just give you some comparative figures of contemporary figures.

In our series we had 2 leaks, which is 3%, none requiring operation, as compared with 7.6% in the current series. 12% of our patients required dilatations as compared with 32% in the current series. There were no recurrent fistula in our series compared to 2% in the current one.

So contemporary results would provide data for better comparison in terms of this minimally invasive approach, at least in terms of anastomotic technique. And this once again begs randomized comparisons to see if this is better.

Would you comment on the following, Dr. Holcomb, since you are going to close the discussion: What is the threshold for success with this procedure, the learning curve, if you will? And did all the surgeons at institutions involved hit the standard? What are the limitations in terms of applicability to different gestational and size groups? Is CO<sub>2</sub> insufflation contraindicated in some patients and do you have a guideline such as resting PACO<sub>2</sub>?

This is an extremely attractive alternative to traditional open approaches, but is it precise enough in terms of the anastomosis and postop esophageal function, which makes me want randomized comparison even more because of the incidence of 25% of funduplications for reflux in your series and 8% in our own.

However, notwithstanding any of the questions I have posed, this is an outstanding study, and even a pioneering one, that I expect will bear fruit in the future with more experience, perhaps if the robot is used because of its greater accuracy, and randomized comparison is accumulated. I appreciated the fact that you presented it at this forum.

DR. GEORGE W. HOLCOMB, III (KANSAS CITY, MISSOURI): We would agree that this is a Phase I study and are not making any claim that it is a better approach. However, we do feel that it may have some advantages, primarily in the arena of musculoskeletal sequelae.

In the manuscript, we do compare it to 4 relatively old studies. These studies were published in the 80s. Unfortunately, these are the largest and most recent studies available in our literature. Also, we are pleased that our results are similar in many respects to the studies to which we were comparing our data.

You asked about size limitations. I believe that the patient who was 1.2 kilograms was probably was too small. I certainly think, in my own experience, that the baby should be 2 to 2.5 kilograms in weight for this approach. There are others in our group of authors who might feel that 2 or 1.5 kilograms may be a reasonable lower level of size. We all agree with the need for randomization. Perhaps we could start such a study among the centers that have participated in this retrospective review.

The robot is an attractive alternative. All of these operations were performed with 3-millimeter instruments. Currently the smallest instruments available for the robot are 5 millimeters. If the instrumentation can be miniaturized for the robot, I think that a robotic anastomosis might be applicable in this group of patients.

DR. JUDSON G. RANDOLPH (NASHVILLE, TENNESSEE): I think the very beautifully conducted study which encompassed 4 continents is remarkable in itself. The unbelievable mechanical ability that you all have demonstrated causes even those of us who operate on 2-kilogram babies to marvel. However, when you predicate a major reason for this operation on the complications of the incision, then a bit of history is necessary.

In 1939, before Ladd and Levin had their first successes, Dr. Tom Lanman of this Society and chairman of American Board of Surgery, wrote of 32 cases of esophageal atresia with fistula operated on without a single survivor. We don't have reports like that today. It was a very courageous report. But it pointed out what we needed to do. And one of the things he stressed was a retropleural approach.

Later as I learned pediatric surgery, we realized that there were problems with anesthesia and standing on that lung for a period of time, so speed became our hallmark. And we opened that chest from one side to the other between the ribs and got right in there and did what needed to be done swiftly and got out rather rapidly.

Most of the problems that we see today with the postop thoracotomy have been eliminated, as they were in Washington when our distinguished moderator began to develop an operation of very posterior thoracotomy, muscle sparing, and with a little sterile cotton swab pushed away the pleura and got into the retropleural space where the vision was and good, the operation could be carefully carried out. Dr. Anderson taught that to non-believers like myself and to our resident staff. While we wouldn't consider primacy in this because it was going on in other centers as well, the incision finally that

Dr. Anderson would use really was hardly more than what you all are making with your instruments and ports. I firmly believe that the complication of the chest wall is now a thing of the past. This marvelous technique which you have demonstrated so beautifully is to be studied, to be compared, but it is going to have to go some to beat the very tiny posterior incision.

Finally, I would just suggest to my colleague and friend Jim O'Neill, that in the talk he just gave us with some very important data, that he not speak about "separating the men from the boys" when we have such a distinguished lady moderator!

DR. GEORGE W. HOLCOMB, III (KANSAS CITY, MISSOURI): Dr. Randolph, I am not sure how I can respond to your comments, but I certainly appreciate your thoughts.

DR. JOHN E. FOKER (MINNEAPOLIS, MINNESOTA): I think there is no doubt that as the technical aspects advance, this approach will be used more and more for many of these repairs. Currently, however, there are several technical points worthy of discussion.

First, as has already been mentioned, are the late problems attributed to thoracotomies. The usual paper cited from 1985 describes very long incisions which are well beyond what is needed for an adequate repair. This paper serves as a straw man for favorable comparison.

I have found for a wide variety of difficult EA problems, a 3 centimeter or less posterior incision, which spares entirely the serratus anterior muscle, is more than adequate. The closure method also can greatly reduce the sequelae. Because there are few distractive forces on a thoracotomy incision, the pericostal closure need not bring the ribs together and a very normal intercostal spacing can be left behind. In addition, a small piece of thin silastic sheeting will ensure that rib fusion does not occur.

The second point concerns the esophageal repair. Because the quality of these repairs will be of long-term importance to these infants, at least 2 technical details may work against thoracoscopic techniques at the present. I believe finer sutures are better. And although they may eventually be used, the sutures illustrated, and in particular the needles, seem quite large. In my opinion, 6-0 and 7-0 sutures are preferable for newborns.

Finally, and perhaps more importantly, some degree of anastomotic tension will often be necessary. When longer gaps between the esophageal segments are present, I have found that by pre-placing the back row of sutures crossing them and then using increasing tension for several minutes, the ends will often come together and the individual sutures can be tied off tension. This maneuver, which is probably not practical thoracoscopically, is what I believe accounts for our uniform good operative results

and lack of leaks even for the very long gap infants. Otherwise, without this method, the first suture bears the brunt of the tension as it is tied.

Although the results of this series meet most older reported industry standards, the postoperative problems were greater than desirable. We have a more recent series – not large but more recent with far fewer problems. For now, in my opinion, one should not give up much on the quality of the anastomosis to avoid making a short incision.

But despite the points I raise, I commend you for advancing the technical aspects of the repair and I look forward to seeing it used more frequently and with increasingly good results in the future.

DR. GEORGE W. HOLCOMB, III (KANSAS CITY, MISSOURI): The needle that was being shown is a T-F needle, which is quite small. Obviously, with the magnification from the telescope, it can look large. We certainly wish there were smaller needles we could use as we could accomplish the operation more easily.

DR. JOHN E. FOKER (MINNEAPOLIS, MINNESOTA): There is no 5-0 needle that compares to a 7-0 needle.

DR. GEORGE W. HOLCOMB, III (KANSAS CITY, MISSOURI): That is probably true. We have traditionally used 4-0 and 5-0 suture, regardless of whether it is vicryl or PDS or silk, to perform these anastomoses. Various surgeons use different suture and different size needles, and we did not standardize that aspect of the operation as part of this report.

Finally, although I think all of us would agree that the most important part of this operation, however it is done, is to achieve a solid anastomosis, we feel that our results speak to the fact that we are able to achieve a good outcome using the thoracoscopic technique.

DR. MARSHALL Z. SCHWARTZ (PHILADELPHIA, PENNSYLVANIA): The previous discussants have been very thorough. I also agree that with this congenital anomaly it is all about the anastomosis and whether a thoracoscopic approach is as precise. These issues have been discussed. The question I have is: Why do you think that there was a much higher incidence of reflux? I can understand why there may be more strictures, but why was there a higher incidence of reflux and the need for funduplications?

DR. GEORGE W. HOLCOMB, III (KANSAS CITY, MISSOURI): In our manuscript, we compared our data to 4 large series from well-known institutions: From Dr. Grosfeld's institution in Indianapolis, from Drs. Spitz and Kyly in London, Dr. Randolph's group at Children's National Medical Center in Washington, and Dr. Peter Manning's paper from Ann Arbor with Dr. Coran. Our fundoplication rate was 24%, one of the others was 25%, and the other 2 were 18% and 15%. Thus, I am not sure that our rate of fundoplication is that high compared to these previous reports. Why we have a relatively higher rate, I can't answer. There are 6 individuals and all of us have different thresholds for when to perform a fundoplication in the patient who has previously undergone repair of esophageal atresia and fistula.

## AUTHOR QUERIES

### AUTHOR PLEASE ANSWER ALL QUERIES

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AQ1—Author: Spelled “Jacobeus” here but “Jacobaeus” in reference list. Which is correct?

AQ2—Author: Personal communications are cited in text. Subsequent references renumbered in both text and ref list.

AQ3—Author: Study of 89 patients mentioned, but numbers add up to only 84 (29, 21, 18, and 16).  
This OK?

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