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Post-operative bronchoscopy findings of esophageal atresia patients, initial phase of single-institute experience

Dina Hossam El Dine Hamed¹, Ayman Hussein², Basma Magdy^{2*} , Mohamed Elseoudi², Mohamed M. Elbarbary² and Ahmed M. K. Wishahy²

Abstract

Background: Esophageal atresia (EA) is not a rare congenital anomaly. It is usually accompanied by tracheoesophageal fistula (TEF). Respiratory morbidities are experienced in most of the survivors. Although it was believed that these symptoms tend to improve by age, permanent lung damage may continue to adulthood in some patients. We aimed to assess the respiratory tract in the follow-up period as an attempt to correlate possible findings with symptoms experienced which allowed us to pay more attention to the respiratory tract during the upcoming surgical interventions. Prospective observational study was conducted on patients presented to our institute, throughout 2018, with various degrees of refractory respiratory tract involvement, after having had their EA repaired inside or outside our institute. After exclusion of esophageal causes of these manifestations, bronchoscopy was performed to correlate the findings with the symptoms experienced by these patients. The study does not include patients repaired after 2018 as surgical modifications were performed and are still under study.

Results: Fifteen patients presented with various respiratory symptoms. Although recurrent chest infection is the commonest presentation, acute life-threatening events were reported in 5 patients. Blind-ended tracheal diverticulum was the most common finding observed in 80% of patients. About 76% of these patients were found to have tracheomalacia. Moreover, bronchoscopy was able to detect recurrent tracheoesophageal fistula in 4 patients. Vocal cords were affected in 13% of these patients.

Conclusions: Frequent respiratory symptoms in operated patients warrant careful long-term monitoring and appropriate management. Although bronchoscopy is considered a mandatory pre-repair procedure, it is a crucial tool in evaluating the survivors. It is pivotal to pay more attention to the respiratory tract to modify the surgical techniques to overcome these pitfalls.

Keywords: Esophageal atresia, Bronchoscopy, Tracheoesophageal fistula, Tracheal diverticulum

Background

Esophageal atresia (EA) has an estimated incidence of 1:5000 live birth and is usually accompanied by tracheoesophageal fistula (TEF). Recent surgical and neonatal intensive care amelioration have increased the success

rate to more than 90% [1]. The increased survival rate has unveiled the truth of the miscellaneous complications experienced in childhood till adolescence [2].

The respiratory morbidities of the survivors, which occupy a wide spectrum from mild to severe acute life-threatening events (ALTEs), are due either to intrinsic factors or to esophageal complication, respiratory tract involvement, or musculo-skeletal deformities. It was believed that these morbidities decrease in late

*Correspondence: basma.ibrahim@residents.kasralainy.edu.eg

² Department of Pediatric Surgery, Cairo University Childrens Hospital, Dr. Ali Ibrahim Street, El Mounira, Cairo, Egypt

Full list of author information is available at the end of the article

adolescence; however, permanent lung damage can persist to adulthood as bronchiectasis [2].

Bronchoscopy is considered a safe procedure that is able to elicit diagnostic information and therapeutic results. Although it is recommended to perform intraoperative bronchoscopy, it is not done in many institutes. Not only is intraoperative bronchoscopy useful in diagnosis and facilitating the procedure, but it also can somehow predict upcoming respiratory morbidities.

Bronchoscopy is an important diagnostic tool in survivors complaining of symptoms not responding to routine medications. The bronchoscopy is able to visualize the anatomy of the airway as well as to inspect for any recurrent or missed fistulas. Nevertheless, tracheomalacia and vocal cords can be thoroughly assessed [3].

Thus, in this study, we aimed to assess the findings of post-repair bronchoscopy in esophageal atresia patients and correlate with the symptoms to reach better management plans. That allowed us to pay more attention to the respiratory tract during the upcoming surgical interventions.

Methods

This study was a prospective observational study, conducted in Cairo University Childrens Hospital, after approval of the Ethical Committee, involving patients who presented with refractory respiratory symptoms, throughout 2018, after repair of EA/TEF.

Our study included patients who underwent elective repair either through open thoracotomy or thoracoscopic approach and presented with recurrent chest problems, which did not show any improvement on routine medical management. It is considered a level A recommendation by “The International Network of Esophageal Atresia, Respiratory Complications Working Group” to perform flexible bronchoscopy in these patients to evaluate tracheomalacia and in any patient with unexplained wheezes or exercise intolerance [4].

However, we excluded any patients with major cardiac anomalies that cause respiratory manifestations. Also, patients with persistent esophageal anastomotic stricture were detected by esophagogram and esophagoscopy and necessitated frequent esophageal dilation. Those with gastro-esophageal reflux (GER), diagnosed by contrast study, esophagoscopy, or Ph-metry and GER refractory to medical treatment, were also excluded. The primary aim of our study was to identify structural and functional deviations in the airway that could help in the management of these patients. Moreover, the results would aid the surgeons to refine the techniques to avoid certain fallacies.

Patients’ detailed history was retrieved as much as possible to include perinatal and peri-operative history.

We documented operative details regarding the operator, institute where the patient was admitted, method of repair either through thoracoscopic or open thoracotomy approach, duration of hospital stay, and any intra- or postoperative complications. Complaints and symptoms were meticulously assessed and investigated to exclude gastroesophageal causes.

Bronchoscopy was performed under inhalational sevoflurane for induction and maintaining anesthesia. We used Karl-Storz® bronchoscopes, flexible fiberoptic with sizes 2.7 and 3.7mm, depending on the age and weight of the patient. Although the flexible bronchoscopes are the preferred ones, we used a rigid bronchoscope 2.5 mm in only one patient, due to technical problems encountered while using the 2.7-mm flexible one.

The respiratory tract was screened for tracheomalacia, missed or recurrent TEF, tracheal diverticula or pouches, stenosis, or laryngeal cleft. Also, vocal cord mobility and integrity were assessed.

Both degree of tracheal collapse and abnormality in the shape of tracheal cartilage contribute to the tracheomalacia. Degree of collapse is either mild, moderate, or severe, if the collapse is 50–75%, 75–90%, and 90–100%, respectively. In tracheomalacia, the normal C-shaped cartilage is replaced by deformed U- or bow-shaped ones, and consequently, the posterior membrane will occupy more than the normal 20% ratio [5]. Fistula site was thoroughly inspected for tracheal diverticula, missed or recurrent tracheoesophageal fistula (rTEF).

Methylene blue test was performed by introducing Foley catheter in the upper esophagus and inflating the balloon in which diluted methylene blue is injected under pressure. If TEF is present, the methylene blue will be detected in the trachea.

Usually, bronchoscopy is performed as a day-case procedure, and patients are discharged after full recovery except if any instability develops.

Data was collected and coded in a master sheet and analyzed using Microsoft® Excel® 2013 (15.0.4420.1017) 32-bit software. Data was summarized using mean, standard deviation, median, minimum, and maximum in quantitative data and using frequency (count) and relative frequency (percentage) for categorical data. Descriptive data analysis and analytical analysis were done using *F* test two-sample for variances. *P* value was considered statistically significant if less than 0.05.

Results

The study involved 15 patients, 11 males and 4 females, who presented during the determined period and met the inclusion criteria. The ages of these patients ranged from 3 to 24 months with a median of 8 months and

Table 1 Main symptoms reported in the patients

Symptom	Number	Percentage
Recurrent infections	14	93
Cough	13	87
ALTEs	5	33
Ventilatory Support	4	27
Choking	4	27

ALTEs acute life-threatening events

weight ranging from 2 to 11 kg with median of 8 kg. All patients had gestational age of more than 36 weeks.

Out of these 15 patients, 7 had their fistula ligated and anastomosis completed through a thoracoscopic approach. The choice of open versus thoracoscopic approach depends on the expertise of the operating surgeon along with the anesthetist. The hospital stay ranged from 5 to 120 days with a mean stay of 32.5 days.

No intraoperative complications were experienced in any of these patients. Early complications such as leaks were detected in 2 patients (13%), while 4 patients (27%) had severe respiratory manifestations in the form of the recurrent need of ventilatory support. The leak in these patients was managed conservatively. The patients were kept on total parenteral nutrition, antibiotics, and regular chest care. The remaining patients were extubated easily after the operative procedures, had an uneventful early postoperative period, and were discharged without any early complications.

We documented the history of respiratory problems included in our sheet as shown in Table 1. Recurrent infection was the commonest symptom experienced while ALTEs were encountered by 5 patients.

Two guardians described ALTEs as blue spells, while 3 described generalized muscle tone associated with gag and inability to breathe. Four patients needed ventilatory support on frequent occasions, due to the development of severe pneumonia, two of them secondary to esophageal leak that developed postoperatively and two developed right upper lobe pneumonia and collapse secondary to aspiration.

During exhalation, the degree of tracheal collapse in at least one segment of the trachea was recorded in Table 2. Most of them, 13 patients (87%), had some sort of collapse in at least one segment of the trachea. Moreover, all of the patients had a lost C-shaped cartilage of the tracheal ring. However, none of these patients had preoperative bronchoscopy done to assess the changes in tracheomalacia. Bronchoscopy has become an indispensable preoperative procedure in our institute after studying these results.

Table 2 Patients with different degrees of trachea collapse

Degree of tracheal collapse	Number	Percentage
Absent	2	13
Mild	6	40
Moderate	3	20
Severe	4	27

Table 3 Findings at the site of the ligated fistula

Finding	Number	Percentage
Tracheal diverticulum	12	80
Stitch remnant	3	20
Positive methylene blue test	2	13

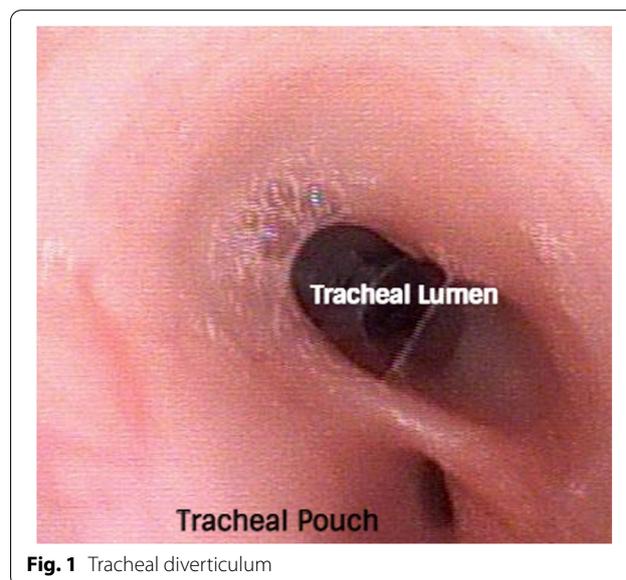


Fig. 1 Tracheal diverticulum

Site of the fistula was examined meticulously and the findings are shown in Table 3. Most of the patients with ligated fistula had developed diverticulum at the site of the primary fistula as demonstrated in Fig. 1. That finding was detected in 12/15 patients, 9/12 had the fistula ligated thoracoscopically but only 6 had the procedure completed thoracoscopically. Three patients had their fistula ligated and anastomosis performed through an open thoracoscopic approach. Also, bronchoscopy suggested rTEF by either stitch remnant or methylene blue test.

Tracheoesophageal fistula was recurrent in 6 patients in the current study, with age ranging from 20 days to 12 months with a median age of 9 months. The initial postoperative stay of these patients extended from 15 to 65

days with a median of 38.5 days. Anastomotic leak was not absolutely excluded in any of these patients. These patients presented with recurrent chest infections and about 4/6 required hospital readmissions. Cough was the obvious complaint in 5 patients. Choking fluids was prominent in half of them. Three of them experienced at least one attack of ALTEs.

Out of the 6 patients with recurrent tracheoesophageal fistula, 2 patients had their TEF ligated via open approach and 2 were ligated thoroscopically and esophageal anastomoses were performed through thoracotomy due to inconvenient operating field in these patients. Only one patient had the procedure completed thoroscopically and one had thoroscopic ligation of fistula then esophagostomy and gastrostomy due to a long gap between upper and lower esophageal pouches.

Although we had 6 patients diagnosed with recurrent TEF, bronchoscopy suggested its presence in 4 patients (67%). Stitch remnants, as Fig. 2, were detected in 3 patients. Methylene blue test to detect recurrent fistula was positive in only 2 patients.

There was a statistical significance between the diagnosis of rTEF and presence of stitch remnant with a *p*-value of 0.04396. However, *p* value was 0.24725, statistically insignificant, between the diagnosis of rTEF and positive methylene blue test.

Two rTEF were missed by bronchoscopy. One was diagnosed by esophagoscopy performed in a simultaneous session with bronchoscopy, and the other was diagnosed after re-exploration due to significant progressive stomach distention after positive pressure ventilation. The relationship between rTEF diagnosed by bronchoscopy out of the total rTEF is statistically significant with *p* value of 0.01099.

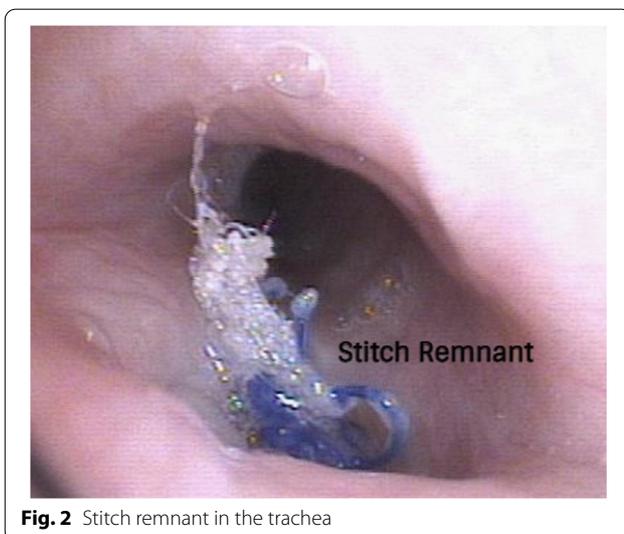


Fig. 2 Stitch remnant in the trachea

Inflamed vocal cords and swollen arytenoids were detected in 13 patients (87%) and weekly mobile vocal cords were visualized in 2 patients (13%). There was no statistical significance (*p* value= 1) between both associations, association between symptoms and vocal cords, as well as an association between symptoms and inflamed vocal cords.

Inflammatory signs in the form of edema, redness, abrasions, polyps and purulent secretions were found in 9 patients.

No complications were met during anesthesia. However, 3 patients (20%) developed bronchospasm, 2 of which were severe enough to require hospital admission. That bronchospasm developed after 4, 6, and 10 hours with an average of 6.7 hours.

Discussion

Great variation exists in the number of patients treated per year in single centers worldwide. Our institute is one of them with a high rate of admission of EA/TEF in the region. In our study, we aimed to use the bronchoscope to screen systematizing patients for congenital and acquired respiratory tract problems to find a correlation with those respiratory symptoms.

Bronchoscopic diagnosis of tracheomalacia is usually operator-dependent based on assessing the lumen collapse and cartilage shape. We diagnosed tracheomalacia in the majority (87%) of our patients, other studies reported its detection in up to two-third of the patients in preoperative bronchoscopy [6]. Out of our 15 patients, 2 symptomatizing patients (13%) had no tracheomalacia detected, However, in Thakkar et al.'s study, preoperative bronchoscopy revealed that about 65 % had tracheomalacia but only 25% developed symptoms [6]. That questions the number of survivors with asymptomatic tracheomalacia and emphasizes the importance of preoperative bronchoscopy.

In this work, a tracheal pouch was detected in 80% of presented patients, which is a much higher incidence than other studies [7]. We think the considerable difference may be attributed to the operative technique in which preservation of azygos vein impeded fistula ligation as near as possible to the trachea.

Tracheoesophageal fistula estimated recurrence rate is from 7 to 19% [7, 8]. The studies claim that recurrence occurs more in premature babies, however, in this study all patients were full term. Coran's 30-year review of 38 recurrent tracheoesophageal fistula involved anastomotic leak as a risk factor as it was encountered by 69% of patients who developed rTEF [8].

Bronchoscopy detected 67% (4/6) of rTEF, which was diagnosed by methylene blue test and suspected by the presence of stitch remnant. We think that ligation and

division of azygos vein intraoperatively might be a risk factor for recurrence due to adjoining suture lines [9].

Methylene blue test was positive in 2/6 patients, contradicting other studies that stated that this test has higher sensitivity [8]. Our patients who were not diagnosed by methylene blue test might be due to an esophageal opening that is not in the level of the tracheal one.

Half of the patients diagnosed with rTEF had stitch remnants, which had a statistical significance. No study reported stitch remnants as a suggestive mark for recurrence.

Vocal cords' involvement regarding mobility was detected in 13% of our patients, going in concordance with similar studies [10]. These patients were operated through open thoracotomy contradicting other studies that documented a higher rate of paralysis in thoracoscopic repair in comparison to open one [10].

Bronchospasm following bronchoscopy was reported in 14% of our patients, which matches the expected outcome in patients with asthma and bronchial hyperreactivity [11].

The low number of patients is considered a limiting factor to thoroughly correlate the findings with symptoms. Thus, our institute is heading to continue studying respiratory tract involvement and include further surgical modifications to overcome the mentioned problems.

Conclusions

As conclusion, frequent respiratory symptoms in operated patients warrant careful long-term monitoring and appropriate management. Thus, although bronchoscopy is considered a mandatory pre-repair procedure, it is a crucial tool in evaluating the survivors. It is pivotal to pay more attention to the respiratory tract to modify the surgical techniques to overcome these pitfalls.

Abbreviations

ALTEs: Acute life-threatening events; EA: Esophageal atresia; GER: Gastroesophageal reflux; rTEF: Recurrent tracheoesophageal fistula; TEF: Tracheoesophageal fistula.

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Authors' contributions

Dina Hossam El Dine Hamed interpreted and analyzed the data and drafted the work. Ayman Hussein Abdulsattar analyzed the data and revised the work critically. Basma Magdy collected the data and drafted the work. Mohamed Elseoudi collected the data and drafted the work. Mohamed M Elbarbary interpreted and analyzed the data and revised the work critically. Ahmed MK Wishahy designed the work and approved the final version. The author(s) read and approved the final manuscript.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

The study was approved by the Ethical Committee of Kasr Al-Ainy Medical School (I- 161016), Cairo University. Informed consent to participate was obtained from legal guardians.

Consent for publication

Not applicable.

Competing interests

The authors have no relevant financial or non-financial interests to declare.

Author details

¹Department of Pediatrics, Cairo University Childrens Hospital, Dr. Ali Ibrahim Street, El Mounira, Cairo, Egypt. ²Department of Pediatric Surgery, Cairo University Childrens Hospital, Dr. Ali Ibrahim Street, El Mounira, Cairo, Egypt.

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References

1. Rothenberg SS. Thoracoscopic repair of esophageal atresia and tracheo-esophageal fistula in neonates: evolution of a technique. *J Laparoendosc Adv Surg Tech Part A*. 2012;22(2):195–9. <https://doi.org/10.1089/lap.2011.0063>.
2. Sistonen SJ, Pakarinen MP, Rintala RJ. Long-term results of esophageal atresia: Helsinki experience and review of literature. *Pediatr Surg Int*. 2011;27(11):1141–9. <https://doi.org/10.1007/s00383-011-2980-7>.
3. Cartabuke RH, Lopez R, Thota PN. Long-term esophageal and respiratory outcomes in children with esophageal atresia and tracheoesophageal fistula. *Gastroenterol Rep*. 2016;4(4):310–4. <https://doi.org/10.1093/gastro/gov055>.
4. Koumbourlis AC, Belessis Y, Cataletto M, Cutrera R, DeBoer E, Kazachkov M, et al. Care recommendations for the respiratory complications of esophageal atresia-tracheoesophageal fistula. *Pediatr Pulmonol*. 2020;55(10):2713–29. <https://doi.org/10.1002/ppul.24982>.
5. Kamran A, Jennings RW. Tracheomalacia and Tracheobronchomalacia in Pediatrics: An Overview of Evaluation, Medical Management, and Surgical Treatment. *Front Pediatr*. 2019;7:512. <https://doi.org/10.3389/fped.2019.00512>.
6. Thakkar H, Upadhyaya M, Yardley IE. Bronchoscopy as a screening tool for symptomatic tracheomalacia in oesophageal atresia. *J Pediatr Surg*. 2018;53(2):227–9. <https://doi.org/10.1016/j.jpedsurg.2017.11.003>.
7. Porcaro F, Valfré L, Aufiero LR, Dall'Oglio L, De Angelis P, Villani A, et al. Respiratory problems in children with esophageal atresia and tracheoesophageal fistula. *Ital J Pediatr*. 2017;43(1):77. <https://doi.org/10.1186/s13052-017-0396-2>.
8. Coran AG. Redo esophageal surgery: the diagnosis and management of recurrent tracheoesophageal fistula. *Pediatr Surg Int*. 2013;29(10):995–9. <https://doi.org/10.1007/s00383-013-3395-4>.
9. Patkowski D, Rysiakiewicz K, Jaworski W, Zielinska M, Siejka G, Konsur K, et al. Thoracoscopic repair of tracheoesophageal fistula and esophageal atresia. *J Laparoendosc Adv Surg Tech A*. 2009;19(S1):s19–22. <https://doi.org/10.1089/lap.2008.0139.supp>.
10. Woo S, Lau S, Yoo E, Shaul D, Sydorak R. Thoracoscopic versus open repair of tracheoesophageal fistulas and rates of vocal cord paresis. *J Pediatr Surg*. 2015;50(12):2016–8. <https://doi.org/10.1016/j.jpedsurg.2015.08.021>.

11. Leiten EO, Martinsen EM, Bakke PS, Eagan TM, Grønseth R. Complications and discomfort of bronchoscopy: a systematic review. *Eur Clin Respir J.* 2016;3:33324. <https://doi.org/10.3402/ecrj.v3.33324>.

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