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# Laryngotracheobronchoscopy prior to esophageal atresia and tracheoesophageal fistula repair—its use and importance

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ABSTRACT

*Background:* Pure esophageal atresia (EA) and esophageal atresia with tracheoesophageal fistula (EA-TEF) are commonly associated with various anomalies. Associated anomalies, especially those of upper airways may alter the management strategies. This study was designed to find out the role of preoperative laryngotracheobronchoscopy (LTB) just prior to the standard surgical procedure.
*Study design:* This was a retrospective study. The data of all the newborn babies (n = 88) with a provisional diagnosis of EA or EA-TEF with preoperative rigid LTB, using 2.5/3.0/3.5 F rigid bronchoscope were analyzed. This additional procedure entailed documenting the abnormalities, endoscopic lavage and noting the site of the fistula. The fistula was cannulated by 3.0 F ureteric catheter just prior to the standard surgical procedure. Management strategies were changed as per the additional findings.
*Results:* Out of 88 patients, 77 had EA-TEF while 11 had pure EA. LTB was performed in all of them. Additional findings in bronchoscopy were noted in 18 (20.46%) babies. These additional findings were: fistula at unusual site in 12, laryngotracheal cleft in 2 and vallecular cyst in 1 neonate. The diagnosis of pure EA turned out to be EA-TEF in 3 cases. Unusual fistula sites were carinal/subcarinal in 4/12 (33.33%), upper pouch fistula in 1/12 (8.33%), double fistula in 2/12 (16.67%) and fistula from main bronchus in 5/12 (41.67%) cases.
*Conclusions:* LTB performed just prior to the definitive surgical procedure in EA and EA-TEF would diagnose, document and may aid in the surgical management strategies.



Pure esophageal atresia (EA) and esophageal atresia with tracheoesophageal fistula (EA-TEF) are common anomalies of the foregut. Its association with the anomalies of other organ systems is well known. The incidence of EA-TEF is approximately 1 in 2500 to 4500 live births [1]. The diagnosis of EA and EA-TEF is straightforward and a careful clinical confirmation by a red rubber catheter has been the standard practice. Often there are other associated anomalies, especially those of the upper airways which may need careful planning and surgical management. Thus a thorough screening of the patient for associated anomalies with confirmation of the same and documenting the exact site of the fistula (thus preempting any surprising intraoperative findings) may help during the definitive surgical procedure. This study was designed to elucidate the role of preoperative laryngotracheobronchoscopy (LTB) just prior to the standard surgical procedure in newborn babies with EA/EA-TEF.

## 1. Study design

This is a retrospective study.

## 2. Methods

All the consecutive newborn babies (n = 88) admitted between January 2005 and September 2012 with the provisional clinical diagnosis of EA/EA-TEF under a single pediatric surgery consultant (MS) formed the basis of the present analysis. Just prior to the definitive surgical procedure, these babies were subjected to rigid LTB using a 2.5/3/3.5 F rigid bronchoscope (ENDOWORLD® 120-E Rigid Universal bronchoscopes, KARL STORZ GmbH & Co. KG, Tuttlingen, Germany). Bronchoscopy was done by the senior author with extensive experience in performing pediatric bronchoscopy. After the delineation of the airway anatomy, the findings were noted and if a tracheoesophageal fistula was present it was cannulated using a 3.0 F ureteric catheter (GS-7018 open tip, Romson's, Nunhai, Agra, India) [Fig. 1]. A 5 F infant feeding tube was used in case of large fistulae where the stomach needed to be decompressed. After bronchoscopy, the baby was repositioned to left lateral position. This was followed by thoracotomy for the definitive repair. The advantage of pre-operative LTB was that the surgical plan could be refined and modified accordingly. In laryngotracheal cleft a planned repair was done at a later date. In vallecular cyst marsupialization was done. Subglottic stenosis was managed by an otorhinolaryngologist at a later date by laser incision under tracheostomy cover. During LTB, bronchial secretions were also collected and sent for bacterial culture

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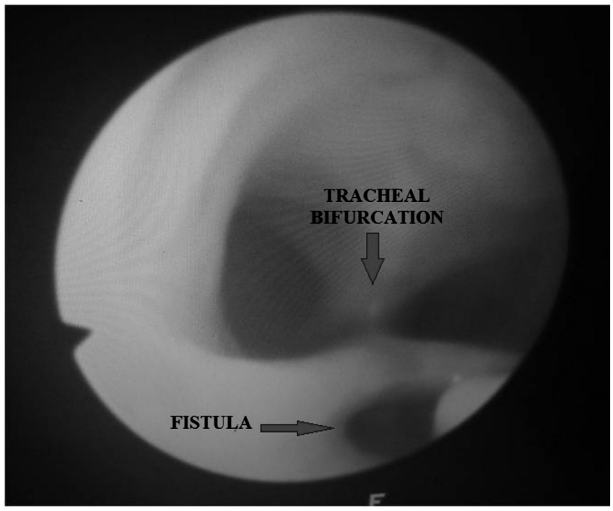


**Fig. 1.** Laryngotracheobronchoscopy showing tracheoesophageal fistula cannulated with ureteric catheter.

and antibiotic sensitivity. On observing the growth of any pathogen, antibiotics were changed and added accordingly.

## 3. Results

There were a total of 88 newborn babies which formed the study group. Based on the clinical examination, passage of red rubber catheter and X ray abdomen, there were 77 (87.5%) cases of EA-TEF and 11 (12.5%) cases of pure EA. LTB was performed in all of them. LTB findings were the same as the clinical diagnosis in 70 (79.54%) patients while additional findings were seen in 18 (20.46%).

The additional findings noted on LTB were as shown in Table 1. An "unusual" fistulous opening was seen in 12/18 (67%) cases. Laryngotracheal cleft was discovered in 2/18 (11%) cases. One of these two cases was a type 1 cleft while the other one was a type 2 cleft. These cases were managed appropriately at a later date. Vallecular cyst was found in 1 (5%) case. In 3 (17%) cases with a preoperative diagnosis of pure esophageal atresia, laryngotracheobronchoscopy revealed a tracheoesophageal fistula (Table 1). In this subset of the cases, LTB offered the greatest benefit because these cases were then managed by primary repair instead of diversion. Out of the 12 cases with "unusual" fistula, carinal/subcarinal fistula were present in 4/12 (33.33%) and upper pouch fistula in 1/12 (8.33%) cases. In 2/12 (16.67%) cases, both upper and lower pouch fistulas were seen. Five of the fistulae were seen to arise from the main bronchus (41.67%). Three of these were from the right main bronchus and the rest from the left main bronchus (Table 2). Besides these additional findings subglottic stenosis was noted in 3 cases. In one case with the clinical suspicion of choanal atresia, a nasopharyngoscopy was added and the diagnosis was confirmed and managed accordingly. Large fistulas in two sick babies were occluded by a Fogarty embolectomy catheter. These children were managed at a later date when they became stable and the respiratory morbidity had decreased significantly. In five cases a trans-fistula stent was used for gastric decompression. Stenting also helped in the identification of the fistula especially in those with a long gap and distal communications. There were 13 such cases. The final diagnosis as per Ladd and Gross classification [2] post-LTB was shown in Table 3.

## 4. Discussion

In the present study, LTB was performed just prior to the definitive surgical procedure in newborn babies with EA and EA-TEF. It was found that the potential benefits were confirmation of the diagnosis, documentation of the exact site of the fistula and diagnosis of any associated airway abnormalities. EA/EA-TEF cases routinely undergo surgery in the emergency operation theater at our institute and all the cases of the present study were posted for surgical procedure soon after presentation and preliminary resuscitation (if required). The additional procedure (LTB) of the present study did not delay the initiation of the definitive surgical procedure.

**Table 1**
Details of additional findings (n = 18) noted by laryngotracheobronchoscopy.

| Abnormal findings | Number (%) |
|---|---|
| Unusual fistula | 12 (66.67%) |
| Laryngotracheal cleft | 2 (11.11%) |
| Vallecular cyst | 1 (5.55%) |
| Pure EA turned out to be EATEF | 3 (16.67%) |

Note. The percentages have been calculated from 18 ie, total number of cases with additional findings.

Esophageal atresia is a relatively common anomaly. A large number of studies have been published regarding the classification, associated anomalies and management of this congenital abnormality. Esophageal atresia can either be pure EA or associated with a tracheoesophageal fistula. Ladd and Gross classified esophageal atresia into 5 types [2] (from A to E) depending upon the type of atresia and the location of the fistula. A literature search revealed differing incidences of the various types of fistulae. As per the Ladd and Gross classification, upper pouch fistula (type B) is seen in 5% cases, lower pouch fistula (type C) in 84%, both pouch fistula (type D) in 1%, no fistula i.e. pure esophageal atresia (type A) in 6% and fistula without esophageal atresia (type E) in 4% cases [2]. In a study by Atzori et al [3] it was found that lower pouch fistula (type C) was seen in 82.2% cases, while pure esophageal atresia without fistula (type A) was seen in 9.7% and upper pouch fistula (type B) was seen in 4.9%. In the present study, lower pouch fistula (Type C) was present in 74/88 (84.09%), pure EA (type A) in 8/88 (9.09%), double fistula (type D) was present in 2/88 (2.27%) and upper pouch fistula (type B) was present in 1/88 (1.13%) (Table 3).

Regarding the exact anatomical location of the fistula in EA-TEF, Benjamin [4] described the first experience of tracheobronchoscopy in newborn babies with EA. Filston et al [5], first described the technique of endoscopic insertion of a Fogarty balloon catheter for temporary occlusion of TEF to facilitate positive-pressure ventilation of non-compliant premature lungs before surgical closure of the fistula. Laryngotracheobronchoscopy may also be useful in detecting associated malformations of the respiratory tree. In his extensive review, Usui et al [6] described various kinds of fatal deformities of the upper respiratory tract in patients with EA and TEF: tracheal agenesis, laryngeal atresia, congenital tracheal stenosis, and pulmonary agenesis. The same series showed an overall 47% incidence of associated tracheobronchial anomalies, frequently on the right side, such as ectopic tracheal bronchus and trifurcated trachea.

In the present study, LTB revealed additional findings in 18 out of 88 (20.45%) newborn babies. The most commonly noted findings involved the variable location of the opening of the tracheoesophageal fistula. There were 12 cases where unusual fistula sites were noted on

**Table 2**
Details of unusual fistulas sites seen on laryngotracheobronchoscopy (n = 12).

| Additional findings | Number (%*) |
|---|---|
| Carinal/subcarinal fistula | 4 (33.33%) |
| Upper pouch fistula | 1 (8.33%) |
| Double fistula | 2 (16.67%) |
| Fistula from bronchus | 5 (41.67%) |

Note the percentage have been calculated from 12 i.e. those where unusual fistula sites were noted.

**Table 3**
Details of different type of anomalies according to the Ladd and Gross Classification (n = 88).

| Type of Anomaly | Number (%) |
|---|---|
| Type A (Pure esophageal atresia) | 8 (9.09%) |
| Type B (Upper pouch fistula) | 1 (1.13%) |
| Type C (Lower pouch fistula) | 74 (84.09%) |
| Type D (Double fistula) | 2 (2.27%) |
| Type E (Fistula without atresia) | 0 (0%) |

laryngotracheobronchoscopy. In clinically suspected pure EA (n = 11), LTB could demonstrate fistula in 3 cases converting the diagnosis from pure EA to EA-TEF. This subset of the cases (pure EA) benefited most from the LTB. Upper pouch fistula was present in 1/3 (33.33%) and in remaining 2/3 (66.67%) a lower pouch fistula was present. These fistulae were cannulated with 3 F ureteric catheters to help during the definitive surgical repair. Thus a thoracotomy and primary repair were done in these cases instead of diversion/delayed primary repair. This prevented the otherwise disastrous consequences of missed tracheoesophageal communications.

Vallecular cyst was documented as an additional finding in one case. This would have otherwise led to serious air way problems in the postoperative period. LTB helped in diagnosing this condition and the cyst was marsupialized prior to the TEF repair. A laryngotracheal cleft was found in two cases. It was type 1 in one child which did not require any additional intervention while in the other case it was type 2. The latter case was managed subsequently. These cases would have resulted in difficulties in the immediate postoperative period. Emergent LTB during the immediate post-operative period in cases of missed airway issue carry a risk of disrupting the anastomosis. A preoperative LTB would rule out such conditions without jeopardizing the anastomosis.

Atzori et al [3] in their study reported additional findings in 5/62 (8%) cases. An upper pouch fistula was seen in 3 cases and the H type fistula was seen in 2 cases. These cases were thus approached cervically and subsequently diverted. According to them tracheobronchoscopy helped in a total of 15/62 (24%) cases. These included 5 cases with additional findings and 10 cases of long gap TEF where dissection was facilitated by cannulation of the fistula and primary repair was possible. In the present study additional findings were seen in 18/88 (20.46%) cases. Preoperative cannulation of the fistula helps in easy identification of fistula during thoracotomy especially when the fistula is from the left main bronchus. Of 88 newborn babies, 5 (5.68%) were very sick for the primary repair, a LTB followed by cannulation of the stomach was done in these cases. This helped us to keep the stomach decompressed till a definitive primary repair was done.

Thus in the present study, additional findings were seen in 18/88 cases (Table 1). Stenting also helped in the identification of the fistula especially in those with a long gap and distal fistula.

There were 3 other cases where a subglottic stenosis was found on preoperative laryngotracheobronchoscopy. These cases were managed subsequently by laser incision in the otorhinolaryngology department.

Thus the potential advantages of preoperative LTB are enumerated below:

1. It confirms and documents the airway anatomy.
2. It documents the true incidence of other airway anomalies.
3. It delineates the exact site of fistula.
4. LTB cannulation offers decompression of the stomach till definitive repair is undertaken in sick newborn babies who require preoperative ventilation.
5. LTB and occlusion of fistula with a Fogarty embolectomy catheter helps in preoperative ventilation till definitive repair can be undertaken in sick newborn babies.
6. It offers an opportunity to modify treatment plans in cases where additional findings are observed.
7. Bronchoscopic lavage during LTB may be useful for changing the antibiotics based on the culture and sensitivity reports when required.
8. The diagnosis of conditions like subglottic stenosis, tracheobronchomalacia, etc. has prognostic and therapeutic implications.
9. The identification of fistula from left man bronchus and carinal/subcarinal fistula during the primary repair for EA-TEF may be difficult. LTB and cannulation offer easier identification.

Laryngotracheobronchoscopy in neonates needs good technical expertise. Complications of LTB neonates have been well described: oxygen desaturation, laryngospasm, bronchospasm, coughing and bleeding. Life-threatening complications such as pneumothorax have been also reported during the procedure in children [7–9]. None of the patients in our series had any of these complications.

## 5. Conclusion

LTB performed just prior to the definitive surgical procedure in EA and EA-TEF can help in the diagnosis and documentation of additional associated respiratory anomalies and thus aid in the surgical management strategies.

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