Heliyon 9 (2023) e15931

Contents lists available at ScienceDirect

Heliyon

journal homepage: www.cell.com/heliyon

Research article

# Effects of primary posterior tracheopexy in thoracoscopic repair of esophageal atresia

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ARTICLE INFO

*Keywords:*
Congenital esophageal atresia
Primary posterior tracheopexy
Thoracoscopy

ABSTRACT

*Background:* This study aimed to evaluate the effectiveness of primary posterior tracheopexy (PPT) in reducing ventilator dependence after repair of esophageal atresia (EA), and the risk of respiratory tract infections (RTI) requiring readmissions within one year.

*Methods:* This retrospective cohort study recruited patients with EA admitted to our hospital between June 2020 and December 2021.

*Results:* In the PPT group (n = 17), the time to extubation after surgery was 86.7 h for 12 patients, with one patient (8.3%) requiring repeated postoperation intubation; six-in-sixteen patients (37.5%) experience at least one RTI requiring hospitalization in one year. In the non-PPT group (n = 17), the time to extubation was 127.0 h for 14 patients, with six-in-fourteen patients (42.9%) requiring repeated intubation; twelve-in-seventeen patients (70.6%) experienced at least one RTI requiring hospitalization in one year.

*Conclusions:* Although the differences did not reach statistical significance due to limited number of participants, patients underwent PPT during EA repair had lower chance of repeated intubation and decreased risk of RTI requiring admissions within one year.

## 1. Introduction

Esophageal atresia with or without tracheoesophageal fistula (EA ± TEF) is a rare birth defect with an incidence rate of approximately 2.44 per 10,000 births [1]. With the advancement of surgical techniques and neonatal intensive care treatment, the survival rate of EA ± TEF has increased to over 90% at initial discharge. However, these patients have a high likelihood of readmissions after their initial discharge within one year, mainly caused by respiratory infections [2,3]. In the long-term follow-ups, 24.1%–44.2% of children with EA ± TEF suffer from recurrent respiratory tract infections [4,5]. Since the esophagus and respiratory tract both originate from the primitive foregut, children with EA ± TEF are often accompanied by tracheomalacia, with an increase of pars membrane form 1/3 to 1/2 or more [6,7].

In cases of severe tracheomalacia, posterior tracheopexy offers an effective surgical solution for improving clinical symptoms and reducing ventilator dependence [8]. Thoracoscopic posterior tracheopexy was firstly introduced into primary repair of EA ± TEF by the surgical team from Wilhelmina Children's Hospital reported in 2017 [9], which significantly decreased the rate of respiratory tract

*Abbreviations:* PPT, primary posterior tracheopexy; EA, esophageal atresia; TEF, tracheoesophageal fistula; RTI, respiratory tract infection.

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https://doi.org/10.1016/j.heliyon.2023.e15931
Received 6 March 2023; Received in revised form 25 April 2023; Accepted 27 April 2023
Available online 29 April 2023

infections (RTI) requiring antibiotics within one year [10]. However, there is still limited evidence supporting the effectiveness of primary posterior tracheopexy (PPT) during thoracoscopic repair of EA and more extensive studies with larger samples are needed.

The first case of PPT in a neonate with EA + TEF was performed in December 2017 and has been consistently implemented since June 2020 at our center. Therefore, the aims of this study were to evaluate whether PPT is effective in decreasing the incidence of RTI that require readmission within one year and reducing the need for ventilator after surgery during the initial hospitalization.

## 2. Materials and methods

This was a retrospective cohort study. All consecutive patients with EA ± TEF who were admitted to our hospital between June 1, 2020 and December 31, 2021 were recruited into this study. Patients whose Gross types were classified as A, B or E, whose ages on the day of surgery were greater than 28 days, or who deceased before 1 year old were further excluded from data analysis. Patients were followed up in the outpatient care at 1, 3, 6 and 12 months of age. Information on RTI requiring admissions were collected between October 2022 and December 2022 by pediatric surgeons who were not aware about the groupings of these patients, via telephone. All participants provided the Informed consent, and this study was approved by the review board of our hospital (HCHLL-2023-06).

Referring to the results of preoperative flexible bronchoscopy, the main eligible criteria for PPT in our study were defined as a "U" shaped collapse of the tracheal membrane during exhalation after ligation and transection of the distal tracheoesophageal fistula, assessed by a senior neonatal surgeon (Video 1). The patients were placed in an anterior left-lateral position. A 5 mm incision was made to insert the first trocar at the level of the right posterior axillary line near the subcircular angle. Thoracic pressure was maintained at 6–8 mmHg. Other two 3 mm trocars were inserted at the third and seventh intercostal of the right midaxillary line. After the tissue between the distal esophagus and trachea was carefully dissected, the degree of collapse of the posterior tracheal wall would be assessed. The posterior wall was then fixed to the anterior longitudinal spinal ligament with 2 or 3 absorbable sutures (Fig. 1A and B). The two ends of esophageal anastomosis were then performed with 5–0 absorbable sutures. Finally, saline solution was injected into the thorax to eliminate any leakage from the tracheal. After confirming no active bleeding, the trocars were removed and the gas was discharged. Chest drainage was placed and the wound was sutured with absorbable sutures.

Basic characteristics, including birthweight, gestational age, birthdate and combined malformations, were collected. The use of mechanical ventilator (MV) before and after surgery were recorded, along with perioperative data including age at surgery day, operation time, anastomotic leak, length of stay (LOS) and cost. The number of admissions for RTI within one year was calculated using both hospital records and reports from the patients' guardians via telephone. In addition, the number of balloon dilatation for esophageal stricture was also recorded. One patient was lost to follow-up in the PPT group.

Continuous data were presented as median and interquartile range (median [IQR]) and categorical data were presented as n (%). The Fisher's exact test or the Mann-Whitney *U* test was used to assess the differences between the PPT and non-PPT groups. Statistical significance was considered when a two-sided $p < 0.05$. All the analyses were performed by SPSS version 20.0 (IBM Corp., Armonk, NY).

## 3. Results

Fig. 2 shows the flow chart of our population. A total of 39 patients with EA ± TEF were admitted to our hospital between June 2020 and December 2021 and underwent repair surgery. After excluding two patients diagnosed as Gross type A, one patient aged over 28 days on the day of surgery, and two patients failed to thrive within one year (one underwent PPT, while the other did not), 34 patients were included in this study. Of these, 17 patients underwent primary posterior tracheopexy during EA repair. Thirteen patients in each group had pre-operation flexible bronchoscopy. In the PPT group, six patients (46.2%) didn't have obvious collapse of trachea or bronchus and the rest 9 patients collapsed 1/2 to over 90%. In the non-PPT group, ten patients (76.9%) didn't have obvious collapse of trachea or bronchus, one patient (7.7%) collapsed 1/2 to 3/4 and two patients (15.4%) collapsed 3/4 to 90% (Table 1).

Basic characteristics of the 34 participants are presented in Table 2. There were no obvious discrepancies between the PPT and non-PPT groups. The proportions of normal birthweight were 76.5% in the PPT group and 47.1% in the non-PPT group, respectively ($p = 0.157$). Half of participants had one or more VACTERL associated deformities. One patient was classified as Gross type D after the repair surgery and underwent PPT.

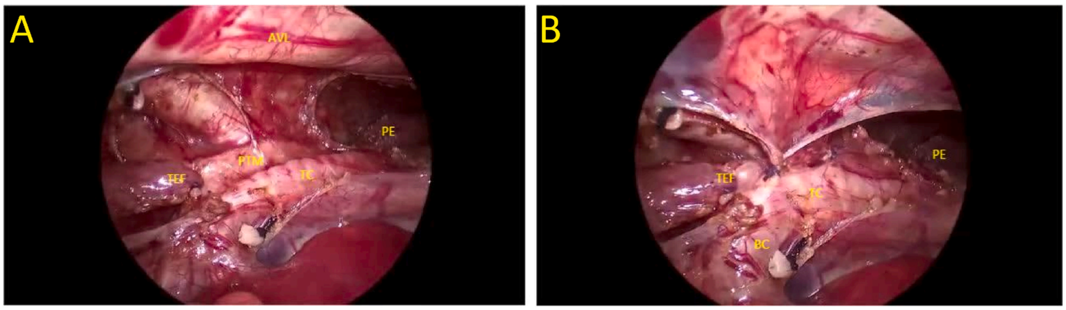


**Fig. 1.** Illustration of PPT during EA repair.

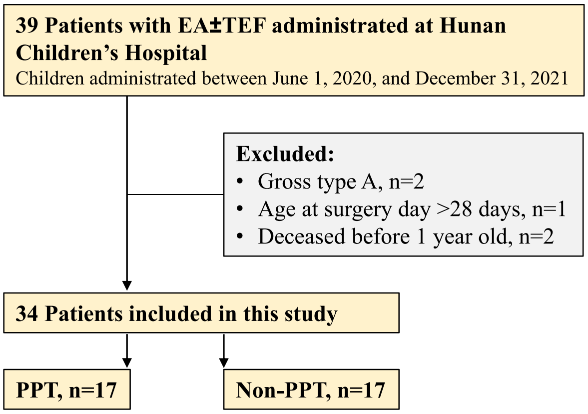


**Fig. 2.** Flowchart of the study population.

**Table 1**
Tracheomalacia evaluated by flexible bronchoscopy prior to EA repair.

| | Non-PPT | PPT |
|---|---|---|
| ≤50% | 10 (76.9) | 6 (46.2) |
| 50–75% | 1 (7.7) | 3 (23.1) |
| 75–90% | 2 (15.4) | 2 (15.4) |
| >90% | 0 | 2 (15.4) |

**Table 2**
Basic characteristics of all the participants.

| | All, n = 34 | Non-PPT, n = 17 | PPT, n = 17 | *p* |
|---|---|---|---|---|
| Sex, boy | 15 (44.1) | 8 (47.1) | 7 (41.2) | 0.73 |
| Age at admission, day | 1 [0, 1] | 1 [1,2] | 1 [0, 1] | 0.147 |
| Gestational age, week[a] | 38 [36, 39] | 37 [34, 39] | 39 [37, 39] | 0.208 |
| Gestational age group | | | | 0.438 |
| 32–36 weeks | 9 (26.5) | 6 (35.3) | 3 (17.6) | |
| ≥37 weeks | 25 (73.5) | 11 (64.7) | 14 (82.4) | |
| Birthweight, g | 2800 [2390, 3100] | 2800 [2300, 3150] | 2725 [2520, 2897] | 0.163 |
| Birthweight group | | | | 0.157 |
| <1500 g | 1 (2.9) | 1 (5.9) | 0 (0.0) | |
| 1500–2500 g | 12 (35.3) | 8 (47.1) | 4 (23.5) | |
| ≥2500 g | 21 (61.8) | 8 (47.1) | 13 (76.5) | |
| Associated anomalies | | | | |
| Cardiovascular* | 11 (32.4) | 5 (29.4) | 6 (35.3) | 0.714 |
| Gastrointestinal | 4 (11.8) | 1 (5.9) | 3 (17.6) | 0.601 |
| Genitourinary | 2 (5.9) | 1 (5.9) | 1 (5.9) | >0.99 |
| Skeletal | 6 (17.6) | 4 (23.5) | 2 (11.8) | 0.656 |
| VACTERL 2+ | 17 (50.0) | 9 (52.9) | 8 (47.1) | 0.732 |
| VACTERL 3+ | 3 (8.8) | 2 (11.8) | 1 (5.9) | 1.00 |
| Gross type | | | | >0.99 |
| C | 33 (97.1) | 17 (100.0) | 16 (94.1) | |
| D | 1 (2.9) | 0 (0.0) | 1 (5.9) | |

[a] n = 33; *Cardiovascular anomalies include atrial and ventral septal defects, tetralogy of Fallot.

The median age on the day of surgery day was 3 [3,5] days. Thirteen patients (38.2%) were intubated before the surgery, with a median duration of 63.9 [47.7, 78.5] hours. Two patients had open repair and 32 patients had thoracoscopic repair of EA. The median duration of the operation was 114 [96, 132] minutes, without differences between the two groups. Twenty-six patients (76.5%) were intubated after the surgery, with a median duration of 98.2 [40.5, 165.3] hours. Seven patients had repeated intubation after the surgery. The median LOS was 29 [19, 36] days and the cost was 73.5 [53.4, 90.2] thousand RMB. Patients in the PPT group had shorter duration of MV after surgery ($p = 0.231$) and only 1 patient (8.3%) had repeated MV after the surgery, which was lower than that in the non-PPT group, with a $p$ value around 0.05 (Table 3). No adverse complications of PPT were observed in the PPT group.

Out of the 34 patients, 11 were readmitted to our hospital within one year and a total of 25 readmissions were recorded. The leading two reasons for these readmissions were esophageal stricture (n = 13, 52.2%; median cost: 8.7 thousand RMB) and pneumonia (n = 6, 24%; median cost: 5.4 thousand RMB). Combining the self-reported RTI admitted to other hospitals, 12 of 17 participants (70.6%) in

**Table 3**
Clinical features of the participants in the two groups.

| | All | Non-PPT | PPT | p |
|---|---|---|---|---|
| Age at surgery, day | 3 [3,5] | 4 [3,5] | 3 [3,4] | 0.16 |
| Preoperation intubation | | | | 0.157 |
| No | 21 (61.8) | 8 (47.1) | 13 (76.5) | |
| Yes | 13 (38.2) | 9 (52.9) | 4 (23.5) | |
| Duration of preoperation intubation, h | 63.9 [47.7, 78.5] | 65.1 [57.3, 78.5] | 46.6 [38.3, 70.3] | 0.33 |
| Operation approach | | | | 0.485 |
| Open | 2 (5.9) | 2 (11.8) | 0 (0.0) | |
| Thoracoscopic | 32 (94.1) | 15 (88.2) | 17 (100.0) | |
| Operation time, min | 114 [96, 132] | 115 [95, 133] | 113 [105, 130] | 0.973 |
| Postoperation intubation | | | | 0.688 |
| No | 8 (23.5) | 3 (17.6) | 5 (29.4) | |
| Yes | 26 (76.5) | 14 (82.4) | 12 (70.6) | |
| Duration of postoperation intubation, h | 98.2 [40.5, 165.3] | 127.0 [53.9, 166.4] | 86.7 [34.8, 140.5] | 0.231 |
| Postoperation repeated intubation | | | | 0.085 |
| No | 19 (73.1) | 8 (57.1) | 11 (91.7) | |
| Yes | 7 (26.9) | 6 (42.9) | 1 (8.3) | |
| Anastomotic leakage | | | | 0.656 |
| No | 28 (82.4) | 15 (88.2) | 13 (76.5) | |
| Yes | 6 (17.6) | 2 (11.8) | 4 (23.5) | |
| Length of stay, day | 29 [19, 36] | 35 [19, 44] | 28 [20, 32] | 0.29 |
| Cost, thousand RMB | 73.5 [53.4, 90.2] | 74.4 [54.0, 100.1] | 67.3 [53.3, 79.7] | 0.413 |

the non-PPT group readmitted for respiratory reasons, which was higher than that in the PPT group (6 out of 16, 37.5%, $p = 0.056$). Four out of the 34 participants (11.8%) underwent balloon dilatation due to esophageal stricture, without differences between the two groups (Table 4).

## 4. Discussion

This study summarized the effectiveness of primary posterior tracheopexy during repair of EA in 34 patients. Although statistically nonsignificant, patients in the PPT group had remarkably shorter duration of intubation and lower rate of repeated MV after surgery. The occurrences of RTI requiring admissions in the first year of life were also lower in the PPT group than that in the non-PPT group.

Posterior tracheopexy was initially developed to prevent recurrent TEF [11] and has been proved to be a safe and effective treatment for severe tracheobronchomalacia, relieving clinical symptoms and degree of airway collapse [12–16]. Tytgat S et al. firstly reported the application of PPT during primary thoracoscopic EA repair [9]. In 2021, they summarized data from 64 patients, among whom 14 underwent PPT, and concluded that PPT could decrease the number of RTI in EA patients [10]. This meaningful outcome might be attributed to the effectiveness of PPT, or it could be a result of selection bias due to the comparison coming from different periods. Similar risk of selection bias may also exist in a study conducted by a surgery team in Japan [17]. Our data showed notably decreases in the duration of MV, rate of repeated intubation after surgery and the incidence of RTI in one year in the PPT group. The statistically nonsignificant results might be explained by small sample size. Dependence on MV was decided by physicians from the NICU at our hospital and the doctor who made the follow-up phone calls was unaware of the groupings of these patients, which meant low risk of measurement bias. The ages of these participants at follow-up were between 1 and 2.5 years old, which meant low risk of recall bias. After excluding the infant with very low birthweight in the non-PPT group, the clinical features of the participants in the two groups are provided in Supplementary Table 1. Patients underwent PPT had significantly lower rate of RTI within one year compared to those in the non-PPT group (37.5% vs. 75.0%, p = 0.033, Supplementary Table 2).

Currently there is no gold standard for the classification of TM severity in children and there is no consistent correlation between anatomical severity and clinical symptoms and signs [18]. Correspondingly, the eligible criteria for PPT were inconsistent across studies: moderate (1/3–2/3) to severe (over 2/3) collapse of trachea diagnosed by preoperative rigid bronchoscopy in Tytgat S's research [9]; coaptation in one or more areas of the trachea by bronchoscopy in combination with clinical symptoms in Shief H's study [14]; over half of the tracheal membrane collapsed during exhalation by bronchoscopy in Yasui A's research [17]. In our study, results of preoperative flexible bronchoscopy were referred and the eligibility criterion for PPT was the collapsibility of the tracheal posterior membrane judged by a senior neonatal surgeon. The results indicated that bronchoscopy had a good consistency with the collapsibility observed during operation, but a certain part of patients had increased flaccidity after dissection and transection of the distal fistula even though were mild in preoperative bronchoscopy. Further researches are needed to determine the criteria for primary posterior tracheopexy during repair of esophageal atresia.

There were also several limitations of our study. Firstly, the nonsignificant results might result from small sample size. A minimum of 36 patients in each group is needed to detect a difference of the incidence rate of RTI requiring readmission in one year from 70.6% to 37.5%, with a power of 80% and $\alpha = 0.05$. Secondly, not all participants underwent pre-surgery flexible bronchoscopy due to consistent MV or the guardians' refusal. There are three departments that can perform bronchoscopy at our hospital and two or more senior pediatricians were involved at each department. Assessment of changes in the lumen is subjective while there is a lack of consistent scoring system, which was the main reason why we did not consider bronchoscopy as the primary criterion for PPT. Our

**Table 4**
RTI requiring admissions in one year and esophageal dilatation.

| | All | Non-PPT | PPT | *p* |
|---|---|---|---|---|
| Readmission at our hospital in one year | | | | 0.714 |
| 0 | 23 (67.6) | 11 (64.7) | 12 (70.6) | |
| ≥1 | 11 (32.4) | 6 (35.3) | 5 (29.4) | |
| RTI admitted to our hospital in one year | | | | 0.199 |
| 0 | 27 (79.4) | 12 (70.6) | 15 (88.2) | |
| ≥1 | 7 (20.6) | 5 (29.4) | 2 (11.8) | |
| RTI requiring admissions in one year | | | | 0.056 |
| 0 | 15 (45.5) | 5 (29.4) | 10 (62.5) | |
| ≥1 | 18 (54.5) | 12 (70.6) | 6 (37.5) | |
| Esophageal dilatation | | | | >0.99 |
| 0 | 29 (87.9) | 15 (88.2) | 14 (87.5) | |
| ≥1 | 4 (12.1) | 2 (11.8) | 2 (12.5) | |

RTI, respiratory tract infection.

positive clinical outcomes of the PPT group suggest that flexibility of the tracheal membrane after dissection and ligation of the distal trachea-esophageal fistula, as judged by the senior neonatal surgeon, could be a good alternative criterion for PPT. Thirdly, patients in the PPT group tended to be full term with heavier birthweight and a lower rate of preoperative intubation, although nonsignificant. This suggested to a potential risk of selection bias by the personal preference of the senior neonatal surgeon. Randomized controlled studies are warranted.

In conclusion, the results of our study suggest that primary posterior tracheopexy during repair of EA can decrease the duration of MV, reduce the proportion of repeated intubation after surgery, and lower the incidence rate of readmissions for respiratory causes within one year, although the differences did not reach statistical significance due to limited number of participants. A large sample which requires multi-center collaboration, and randomized controlled trials are needed to verify the effectiveness and safety of primary posterior tracheopexy during thoracoscopic repair of esophageal atresia.

## Author contribution statement

Chonggao Zhou: Conceived and designed the experiments; Performed the experiments; Analyzed and interpreted the data; Wrote the paper.

Jie Dong: Conceived and designed the experiments; Analyzed and interpreted the data; Contributed reagents, materials, analysis tools or data; Wrote the paper.

Bo Li: Conceived and designed the experiments; Performed the experiments; Contributed reagents, materials, analysis tools or data.

Ming Li: performed the experiments; Contributed reagents, materials, analysis tools or data.

Chanjuan Zou, Yong Xiao, Guang Xu and Bixiang Li: Performed the experiments.

## Funding

Hunan Provincial Health Commission project (20230628536); Hunan Provincial Major Science & Technology Project of Collaborative Prevention & Treatment of Birth Defects (2019SK1015).

## Data availability statement

Data will be made available on request.

## Additional information

Supplementary content related to this article has been published online at [URL].

## Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

## Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.heliyon.2023.e15931.

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