Unilateral repair of tracheoesophageal fistula with dilatation of incomplete subglottic stenosis

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Abstract
A case of TEF along with subglottic stenosis in a patient as a result of prolong intubation. We performed left unilateral tracheal repair to avoid injury to left recurrent laryngeal nerve followed by endoscopic balloon repair of stenosis. Due to critical condition of the patient and COVID-19 infection, she expired.

KEYWORDS
laryngeal, stenosis, tracheal, tracheoesophageal fistula

1 | INTRODUCTION

Tracheoesophageal fistula (TEF) is a rare complication that can be due to tumor extension from the mediastinum or tracheoesophageal injury as a result of endotracheal intubation,1,2 bronchoscopy,3 and endoscopy.4 It is characterized with high risk of respiratory failure therefore requires urgent management.5,6 Nonoperative mortality rate is 80% in TEF patients.7 Commonly, surgical practice includes closure of the membranous trachea in one layer and closure of the esophagus in membrane and muscle layer.8 Muscular or pericardial or pleural flap is placed between the trachea and esophagus to minimize structural interposition and recurrence of TEF.9,10

2 | CASE PRESENTATION

On December 18, 2019, a 25-year-old girl was taken to hospital by Glasgow Coma Scale (GCS) 3 after suicidal hanging. After primary cardiopulmonary resuscitation and intubation, the patient was transferred to the intensive care unit in a deep coma. Brain CT-Scan of the patient was normal and echocardiogram and forensic medical counseling was provided. The patient under gastric tube, follicular catheter and a central venous catheter. Due to prolonged intubation, the patient underwent tracheostomy after 1 week, in which she developed right lung pneumonia, recurrent fever, and secondary pleurisy due to aspiration of the gastric tube. At this stage, after the fluid was removed by gavage, tracheoesophageal fistula was identified. 1 week later, due to bleeding around the tracheostomy tube, the anticoagulant therapy was discontinued (Table 1).

After 16 days of hospitalization in the ICU, a PEG (percutaneous endoscopic gastrostomy) was placed for feeding. At the time of hospitalization, troponin was initially elevated to 5 ng/mL, which was reduced to 0.2 ng/mL with treatment. On January 25, 2020, the patient underwent endoscopy and was diagnosed with esophageal fistula. She had positive blood culture, the central venous line was replaced, and due to a right pneumonothorax, the chest tube was placed on February 2. After treatment and stabilization, the patient underwent thoracic surgery, in which the gastric tube was removed due to increased risk of fistula and bronchoscopy was performed. In the initial bronchoscopy, the incomplete stenosis of the subglottic and complete tracheal ring was identified with TEF. Finally, after jejunostomy feeding, left unilateral repair of TEF surgery was performed (Tables 2 and 3).
During this surgery, the patient was placed in a supine position with hyperextension of the neck and abduction of the arms. For tracheostomy, with a vertical incision from the top to bottom of the trachea to the inlet thorax, the muscles in the midline were released, and the trachea was cut near the second ring, and the tube was placed into the third ring of the trachea released. Then, from the lower edge, we opened trachea at 1 cm anterior level and inserted the spiral tube No. 7 into the distal end of the trachea, which was then connected to tube 8 for the induction of anesthesia (Figure 1).

In the exploration, there was no stenosis and fibrosis of the trachea, but there was incomplete anterior subglottic stenosis and approximately 1 cm long TEF in the posterior wall of the membranous part to the esophagus at the anterior surface. To prevent trauma to the left recurrent laryngeal nerve, the dissection was performed from the left side of the trachea to its posterior, without damaging the large arteries, the site of attachment of the esophageal fistula to the posterior elastic wall of the membrane and trachea. The anterior surface of the esophagus and fistula site was then repaired in two layers, the mucosal layer with the Vircyl 0000 thread and the muscle wall with the polypropylene 000. After repairing the esophagus, the sternocleidomastoid muscle was released from the upper edge and passed through the middle of the sternothyroid muscle in the lower edge, placed on the esophagus and fixed. Then, the membrane of the trachea, which is made of loose connective tissue and is located between the esophagus and the trachea, was repaired with Vircyl 0000 sutures.

Subglottic stenosis was removed with endoscopic balloon dilation and scar was resected using CO$_2$ laser along with transoral exposure of the tracheal scar. After removing the distal tracheal tube, the proximal tracheal tube was placed at the bottom of the repair site of the posterior wall of the trachea with low pressure, and the anterior wall was repaired with polypropylene 000, and the wall was closed by hemobag (Figure 2).

### Table 1
The results of laboratory test

| Date (MM-DD-YYYY) | Troponin (Normal range up to 0.06) | CKMB (Normal range 6-25) | CRP (Normal range 24-190) | INR |
|-------------------|-----------------------------------|--------------------------|---------------------------|-----|
| 18-12-2019        | 0.2                               | 13                       | 62                        |     |
| 20-12-2019        | 1.24                              |                          |                           | 2.2*|

*With using Venlafaxine and ASA.

### Table 2
The result of Glasgow coma scale

| Date (MM-DD-YYYY) | Glasgow coma scale |
|-------------------|--------------------|
| 18-12-2019        | 3                  |
| 24-12-2019        | 5                  |
| 10-1-2020         | 10-11              |
| 15-1-2020         | 8                  |
| 4-2-2020          | 6-7                |

*The patient was placed on a T-piece.

### Table 3
Different stage of blood received

| Date (MM-DD-YYYY) | Receive blood |
|-------------------|---------------|
| 2-1-2020          | 30-1-2020     |
| One unit          | One unit      |

*Figure 1* Gradual dilatation of incomplete subglottic stenosis with gradual increase in the pressure of the tracheal tube cuff

*Figure 2* Extension tube input from the chip distal
On the fourth day after surgery, the patient was placed on a T-Piece with 95% \( \text{O}_2 \) saturation. Unfortunately, due to the concurrence of the COVID-19 in the patient and 48 hours after positive coronavirus test, the patient expired (Table 4).

### 3 | DISCUSSION

In this case, TEF was presented between the posterior elastic membrane wall of the trachea and the serous esophagus and the third ring of the trachea below the tracheostomy site,\(^{11,12}\) without the occurrence of fistula stenosis in the trachea with subglottic stenosis.\(^{13,14}\) Exploration revealed that by resecting left side of the trachea and attaching it without the need for the resection of membranous layer, the muscle tissue was separated from the esophagus and mucosal and muscle layer was sutured.\(^{15,16}\) In cases where there is a history of surgery on one side, it is preferable to dissect the trachea from the esophagus from the anterior side of the neck to reduce the risk of injury to the recurrent laryngeal nerve.\(^{17,18}\)

### 4 | CONCLUSION

By one-sided resection of the trachea, the duration of surgery can be reduced. The possibility of bilateral damage to the vocal cords is eliminated due to the unilateral dissection of the neck area.

With a more limited dissection of the posterior space of the upper mediastinum, the risk of infection and mediastinitis decreases along with probability of partial tracheal necrosis and the occurrence of tracheal fibrosis.

### CONFLICT OF INTEREST

The authors deny any conflict of interest in any terms or by any means during the study.

### AUTHOR CONTRIBUTIONS

SMR: conceptualized and designed the study, drafted the initial manuscript, and reviewed and revised the manuscript.

PS: Designed the data collection instruments, collected data, carried out the initial analyses, and reviewed and revised the manuscript. HRT: Coordinated and supervised data collection, and critically reviewed the manuscript for important intellectual content.

### HUMAN AND ANIMAL RIGHTS

No animals were used in this research. All human research procedures followed were in accordance with the ethical standards of the committee responsible for human experimentation (institutional and national), and with the Helsinki Declaration of 1975, as revised in 2013.

### CONSENT FOR PUBLICATION

Informed consent was obtained from each participant.

### DATA AVAILABILITY STATEMENT

All relevant data and materials are provided with in manuscript.

| Date       | PH   | \( \text{PaCO}_2 \) | \( \text{PaO}_2 \) | T  | H\( \text{CO}_3 \) | Sat\( \text{O}_2 \) | Acute respiratory distress syndrome (ARDS) |
|------------|------|-----------------|----------------|----|---------------|-------------|-------------------------------------------|
| 18-12-2019 | 7.43 | 37.8            | 38.8           | 24.9 |               |             |                                           |
| 21-12-2019 | 7.46 | 35.7            | 44.2           | 24.9 |               |             | No                                        |
| 4-1-2020   | 7.42 | 33.6            | 43.4           | 21.3 |               |             |                                           |
| 28-1-2020  | 7.38 | 42              | 103            | 24.8 |               |             |                                           |
| 29-1-2020  | 7.35 | 56              | 26             | 30.9 |               |             |                                           |
| 31-1-2020  | 7.34 | 38              | 59             | 20.5 |               |             |                                           |
| 6-2-2020   |      |                 |                |      | 95% Yes       |             |                                           |
| 18-2-2020  | 7.38 | 59              | 37             | 34.9 | 97% No        |             |                                           |
| 19-2-2020  |      |                 |                |      | 93% No        |             |                                           |
| 20-2-2020  |      |                 |                |      | 98%           |             |                                           |
| 21-2-2020  |      |                 |                |      |               |             |                                           |
| 22-2-2020  |      |                 |                |      | 94%           |             |                                           |
| 23-2-2020  |      |                 |                |      | 100%          |             |                                           |
| 24-2-2020  |      |                 |                |      | 99%           |             |                                           |

### TABLE 4

Vital signs after surgery until death
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How to cite this article: Marzban-Rad S, Sattari P, Taheri HR. Unilateral repair of tracheoesophageal fistula with dilatation of incomplete subglottic stenosis. Clin Case Rep. 2021;9:2036–2039. https://doi.org/10.1002/ccr3.3937