Novel Technique: Noninvasive Ventilation Support Flexible Endoscopy for Preoperative Manage Neonates of Esophageal Atresia with Tracheoesophageal Fistula and Respiratory Distress

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Abstract

Introduction Pre-operative management of neonates with esophageal atresia and tracheoesophageal fistula (EA/TEF) requiring positive pressure ventilation (PPV) support is clinically challenging. This study evaluates the safety, feasibility and value of flexible endoscopy with noninvasive ventilation and sustained pharyngeal inflation (FE-NIV-SPI) in diagnosis and placing a naso-tracheo-fistula-gastric (NTFG) tube before surgery. Methods A retrospective study conducted from 2017 to 2020 in neonates with Type-C EA/TEF and respiratory distress, where FE-NIV-SPI performed with NTFG tube placement before surgery. Results Five neonates were collected, one with duodenal atresia and one with transposition of great artery. At FE-NIV-SPI, median body weight was 2,399 g and mean age was 15.2 hours. Four neonates yielded severe (>80% collapsed) tracheomalacia. With this FE-NIV, all tracheal, fistulas and esophageal lumens could clearly assess and manage. All fistulas were less than 8mm proximal to carina with mean orifice width of 5 mm. All NTFG tubes placed successfully after confirmed the EA/TEF. Three neonates had co-intubated with nasal endotracheal tube and 2 neonates had received nasal prongs PPV. Mean procedural time of FE-NIV was 13.6±4.5 minutes. All neonates received gastric decompression and feeding via NTFG tubes for mean of 11.4±18.2 days and had stable pre-surgical courses. No adverse associated complication noted. Conclusion FE-NIV-SPI technique enables safe and accurate measurement of EA/TEF anatomy and placing NTFG tube. It could avert emergent gastrostomy, aid gastric decompression, feeding, and ETT intubation, improve PPV, provide pre-surgical stabilization and identify the fistula location during the surgical correction.

Title pages

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Key words:

Esophageal atresia, Flexible bronchoscopy, Noninvasive ventilation, Sustained pharyngeal inflation, Tracheoesophageal fistula

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Abbreviated title:

Pre-Op flexible scopy with NIV manage EA/TEF neonates

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Introduction

Preoperative management of neonates with esophageal atresia and tracheoesophageal fistula (EA/TEF) remains challenging and requires multidisciplinary support including neonatologist, pulmonologist, surgeon and anesthetist. EA/TEF may frequently complicate with respiratory distress due to coexisted prematurity,[1-3]tracheomalacia (TM),[4,5] gastric distension or aspiration pneumonitis. However, respiratory support with positive pressure ventilation (PPV) may be ineffective with large fistulas due to ventilation

shunting through the fistula. This shunting may lead severe gastric distention that consequently restricts lung expansion, impaired pulmonary function and overall clinical deterioration. The main goal of preoperative management of EA/TEF is to stabilize the general status of the patient so that definite surgical correction can be carried out under best possible conditions. In the published literatures, the use balloon of Fogarty catheter to occlude the fistula[6] or umbilical catheter through the fistula for gastric decompression[7] has been suggested to accomplish such means.

"Pharyngeal oxygen with optional nose-close and abdomen-compression (PhO_2-NC-AC)" is a novel model of noninvasive ventilation (NIV).[8-10] Its clinical application and operation are simple without using any artificial devices such as Ambu-bag, face or nasal mask, laryngeal mask airway, endotracheal tube (ETT), or mechanical ventilator. This approach has already been demonstrated to provide adequate oxygenation and PPV during flexible endoscopy (FE) interventions in pediatric patients, even those in severe asphyxiated status. In this NIV, a prolonged duration of nose-close, sustained pharyngeal inflation (SPI), can create positive pressure and make expansion in the laryngopharyngeal space.[11]

Here, we present our experience in preoperative management of neonates with EA/TEF and moderated respiratory distress where FE with this NIV and SPI (FE-NIV-SPI) facilitated safe, accurate examination and placement of naso-tracheo-fistula-gastric (NTFG) tube.

Patients and Methods

In this case series, retrospective review of neonates with Type-C EA/TEF and compromised respiratory status who underwent preoperative FE-NIV-SPI for anatomic measurements and subsequent placement of NTFG tube. Between dates of June 2017 to May 2020, five neonates were identified from two tertiary referral medical centers in Taiwan, Children's Hospital of Taichung China Medical University and Department of Pediatric of Taipei Veterans General Hospital. Data collection included demographics, comorbidities, gestational age, procedural details, and relevant findings. Both hospital ethical committees approved the study. This study was approved by the Institutional Review Board (IRB) of Taipei Veterans General Hospital (IRB No. 2018-08-001CC).

FE-NIV technique

All neonates underwent FE-NIV-SPI in bedside of neonatal intensive care unit with continuous vital signs monitoring. Intravenous procedural sedation was recommended with midazolam (0.1-0.3 mg/kg) and topical anesthesia of 2% lidocaine solution 1-2 ml/kg, and preserved spontaneous ventilation as possible. Details of this FE-NIV-SPI approach have reported previously[8-11] and describes in below.

NIV-SPI

A small nasopharyngeal (suction) catheter (8 Fr.) was inserted via one (right) nostril to a length of 0.5 cm less than the measured distance from nose tip to ipsilateral tragus which ensured the tip positioned in the oropharynx. Then, a heated humidified continuous oxygen flow was provided through the catheter at rate of 1.0 L/kg/min. So that oxygen could fill the entire upper airway space (Figure 1), which bearing similarities to "high flow nasal cannula". Endoscopist's index finger of right hand hooked under the neonate's chin while both thumb and mid-finger placed around ala nasi in a pinching position as well as holding the endoscope at nose. Then, following maneuvers were executed optionally to deliver PPV:

(1) Inspiration phase was accomplished by nose(mouth)-closure with pinching thumb, index and mid-finger of right hand. Cricoid pressure might also be applied concurrently with ipsilateral little finger. While the SPI with duration of 2 to 5 seconds might also provide. (2) Expiration phase was started passively with the release of the nose-closure, which could be further facilitated with simultaneous mild compression of umbilical region and release of cricoid-pressure. Above steps were performed optionally at the rate of 5–10 cycles per minute.

Flexible Endoscopy (FE)

Flexible endoscope with an out-diameter of 2.6 mm and working length of 30 cm (ENF-V3, Olympus) was introduced via patient's nose (left) and advanced to aeroesophageal tracks for inspection. Nose-closure maneuver with SPI duration of 2-5 seconds was performed to gradually expand the associated lumens in order to facilitate a complete and dynamic evaluation. FE assessed the structures in following order: pharynx, larynx, trachea, TEF, bilateral bronchi, lower esophagus, stomach, then withdrew back to pharynx and went to upper esophagus. Commonly associated anatomic anomalies such as TM, fistula and upper esophageal pouch were clearly identified. The fistula location and opening width were estimated with FE. The distance from the upper edge of the carina to the lower edge of fistula was measured using the FE tip technique.

Results

Five neonates with Gross type-C EA/TEF with compromised respiration were enrolled. Table 1 shows the summaries of their clinical characteristics and associated management. Four neonates had body weights of less than 2,500 g and two neonates had gestation age less than 37 weeks. All of them developed respiratory distress immediately after birth and had received nasal bi-prongs PPV supports before the FE. In four inborn neonates, these FE interventions were performed within six-hours of age, while Case 5 was referred from another medical center and the FE performed on her third day of life (DOL).

After diagnosis and measurements were confirmed with FE-NIV-SPI, NTFG tube was immediately placed as early therapeutic intervention in all neonates. In three neonates, FE-NIV-SPI also aided in nasal ETT intubations, with the presence of the NTFG tube, by safely guiding the ETT tip passed over the fistula opening and appropriately positioned just above the carina. Both NTFG tube and ETT were well visualized as wedging into the vocal cords. The mean procedural duration of the whole FE-NIV, including both diagnostic and therapeutic, was 13.6±4.5 minutes.

Afterward, all neonates received regular gastric feeding and intermittent gastric decompression via the NTFG tubes. Their vital signs and respiratory statuses were stable prior to surgical correction of EA/TEF, except for Case 4, where the patient required urgent cardiac surgery due to associated cardiovascular comorbidity. During correction surgery of EA/TEF, after confirming the fistula sites in all patients, surgeons removed the NTFG tubes just before ligation of the fistula. The mean duration of NTFG tube placement was 11.4±18.2 days. All neonates survived and had follow-up for more than 18 months. There were no complications associated with the FE-NIV-SPI and NTFG tube placement including air leakage, airway bleeding, laceration or stenosis. Three neonates (Case 1, 3 and 4) received 2 to 4 courses of laser therapy and balloon dilatation due to mid-esophageal stenosis at the anastomosis sites.

Case presentation (Case 4)

A full-term male neonate, birth weight 2,390 g with prenatal polyhydramnios and echocardiographic diagnosis of transposition of great artery, was born by spontaneous delivery. Respiratory distress soon developed after birth and he underwent nasal ETT (3.0 mm inner diameter) intubation with mechanical ventilation settings: fraction of inspiratory oxygen 50%, PEEP 6 cmH_2O, PIP 20 cmH_2O and rate of 20 per minute. Unfortunately, he gradually developed obvious stomach distention and failed multiple insertion of gastric tube, therefore presence of EA/TEF malformation was suspected.

Diagnostic FE-NIV-SPI was performed at age of 5 hours old with ETT extubation. A folded esophageal tube was seen at hypopharynx (Figure 2) with severe (>95% collapsed) TM in mid-tracheal which occluding scope view of the distal trachea. After applying 2 to 3 seconds of SPI, the collapsed portion gradually opened and a large fistula with width of 6 mm (Figure 3) was revealed at the posterior wall, 8 mm proximal to the carina level, while estimated the length of TM about 15 mm. The scope then advanced through the fistula, down into the lower esophagus and found stomach cavity, no obvious structure of sphincter over gastroesophageal junction was noted. After completing tracheal, fistula and low esophageal assessment, the scope was drawn back to pharynx and inserted into the inlet of esophagus. With aid of SPI (2 to 5 seconds), a gradual dilated esophageal blind pouch of about 2 cm length was disclosed. Therefore, FE-NIV-SPI technique could assist the accurate diagnosis of EA/TEF and associated anatomic relationships.

After confirming the diagnosis, therapeutic FE-NIV-SPI was carried out. An 8 Fr. suction catheter was selected as the NTFG tube with appropriate insertion depth to the stomach marked. With aid of Magill forceps, the catheter was inserted nasally, through the vocal cords and into the tracheal lumen (similar to nasal ETT intubation). The scope (ENF-V3, 2.6 mm) then advanced through the nose and wedged into the tracheal lumen (Figure 4) alongside the NTFG tube. Under FE guidance and SPI (2 to 3 seconds), endoscopist manipulated the tube into the wide-open fistula orifice smoothly, and propelled it forward into the gastric cavity. The endoscope itself could also advance into the low esophagus, alongside the tube, and confirmed the tube's tip located inside the gastric cavity.

After placement of NTFG tube, a nasal ETT with internal diameter of 3.0mm was wedged alongside of the NTFG tube through the vocal cords with aid of Magill forceps (Figure 5). Then, a slim flexible endoscope (OD 1.8 mm, LF-P, Olympus) was cannulated through the ETT lumen and assisted in guiding the tip of ETT passed over the TEF till it reached just above the level of carina. Finally, the nasal ETT was fixated and connected to mechanical ventilator while the NTFG tube was used for feeding and gastric decompression.

Cardiac surgery was performed on the 10^{th} DOL with a complicated course. On 44^{th} DOL, cardiac function finally stabilized and a primary one-stage repair of the EA/TEF was carried out. During operation, the NTFG tube assisted in identifying the fistula location before its removal. Postoperative clinical course was uneventful, except for the esophageal stenosis, where he received two courses of laser therapy to cut off the fibrotic scare and four courses of balloon dilatations. These therapeutic FE all performed under the FE-NIV approach.[13,16] At the time of this article writing, he is two years old with excellent respiratory function and oral feeding status.

Discussion

The main goals of EA/TEF management are timely recognition of underlying malformation and reduction of associated complications. To our knowledge this is the first study concerning preoperative management of neonates with EA/TEF and moderate respiratory distress; where we utilized the novel FE-NIV-SPI approach to make accurate diagnosis, insert NTFG tube and subsequent management to stabilize condition before surgical intervention.

Shunting of ventilation through the fistula presents a major challenge in managing neonates of EA/TEF with respiratory distress requiring PPV support. Placing the ETT tip distal to the fistula is the principle key to minimize ventilation loss through fistula. However, majority of the TEFs reported are located within 1.0 cm from carina or even at the level of carina, making this distal placement of ETT difficult.[17] Routine preoperative endoscopy in neonates with EA/TEF remains inconsistent, but several studies highlighted its usefulness in locating the fistula and associated airway anomalies for deciding operating approach.[6,7,18–21]Previous literatures described using Fogarty catheter[6]or a semi-rigid umbilical catheter[7] either to directly balloon-occlude the fistula or prevent gastric distention, respectively, for improving lung ventilation. This case series demonstrated that the FE-NIV offered a practical and effective benefits in both pre- and post-operative clinical management.

This FE-NIV-SPI technique provides several advantages especially in high-risk neonates. 1) In our previous study[11] of same setting, the SPI with duration of 0 to 5 seconds could create well correlated positive pressure levels of 4.1±3.3 to 65.5±18.5 cmH_2O in laryngopharyngeal space. This transient, dynamic and controllable positive pressure is safe and enough to expand the collapsed aeroesophageal lumens in case of EA/TEF. 2) Oxygen insufflation through the nasopharyngeal catheter can directly flush upper airway dead space, as the effects of "apneic oxygenation",[22-26] which may significantly prolong onset of desaturation. 3) Both assist inspiration and expiration can be optionally achieved by this simple maneuver to provide enough oxygenation and ventilation. 4) No instruments such as mask, ETT, ventilation bag or mechanical ventilator are needed, hence eliminate preparation time and provide an unobstructed and unimpeded viewing field for FE assessment and manipulation. 5) With improved PPV and visual field, that facilitates accurate FE assessment of aeroesophageal tracks such as length of TM, orifice diameter of TEF, distance between TEF and carina, depth of the blind esophageal pouch; and consequent interventions such as precise insertion of NTFG

tube, ETT intubation, laser and balloon measurement and even stent implantation in these compromised neonates.

The NTFG tube placement has the following advantages. 1) Early occlusion of the fistula prevents PPV loss that eliminate distension of stomach or intestine, as the Case 1 with duodenal atresia (DA), and allows for gastric feeding. 2) It facilitates effective lung ventilation, even in neonates who require ETT PPV. 3) It offers stabilizing option that can avert the need for emergent gastrostomy. 4) It preserves intact peritoneal cavity and provides future benefits for intra-abdomen procedures. 5) During surgical repair of EA/TEF, NTFG tube decompression declines risk of surgical field contamination by gastric contents. 6) NTFG tube can also act as a guide for locating the fistula during operation. For patients similar to Case 1, staged repair consisting of gastrostomy and fistula ligation followed by DA repair one week later had been reported.[27] This approach could allow for a less hurried and well-prepared simultaneous surgical correction of both EA/TEF and DA lesions in one session. Nevertheless, cannulation of NTFG tube or concurrent ETT intubation require skillful technique of FE-NIV, especially when performed in small neonates with multiple comorbidities. Further large studies are required for validation and widespread application.

Conclusion

Using this FE-NIV-SPI approach for preoperative assessment and NTFG tube placement can safely delineate aeroesophageal malformations and effectively stabilize through gastric feeding and ETT intubation to improve ventilation in neonates with EA/TEF and moderate respiratory distress. This approach may also improve diagnostic accuracy of FE and allows for simultaneous surgical correction of EA/TEF and associated anomalies.

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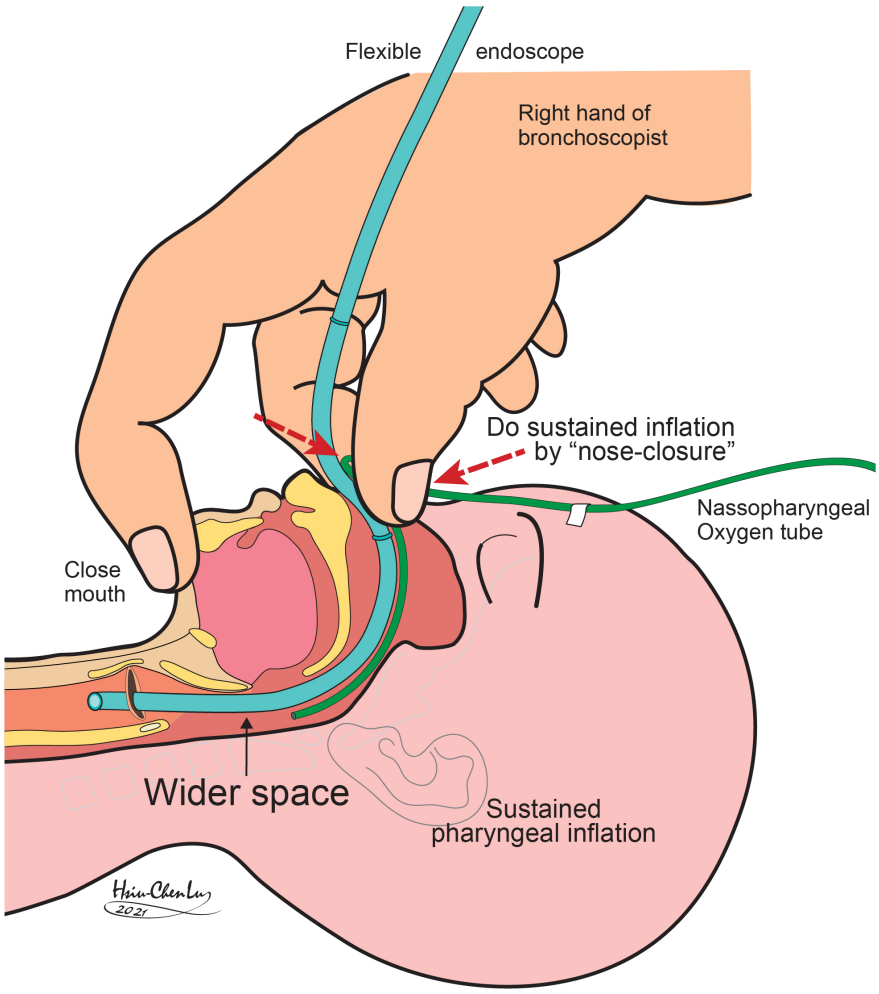

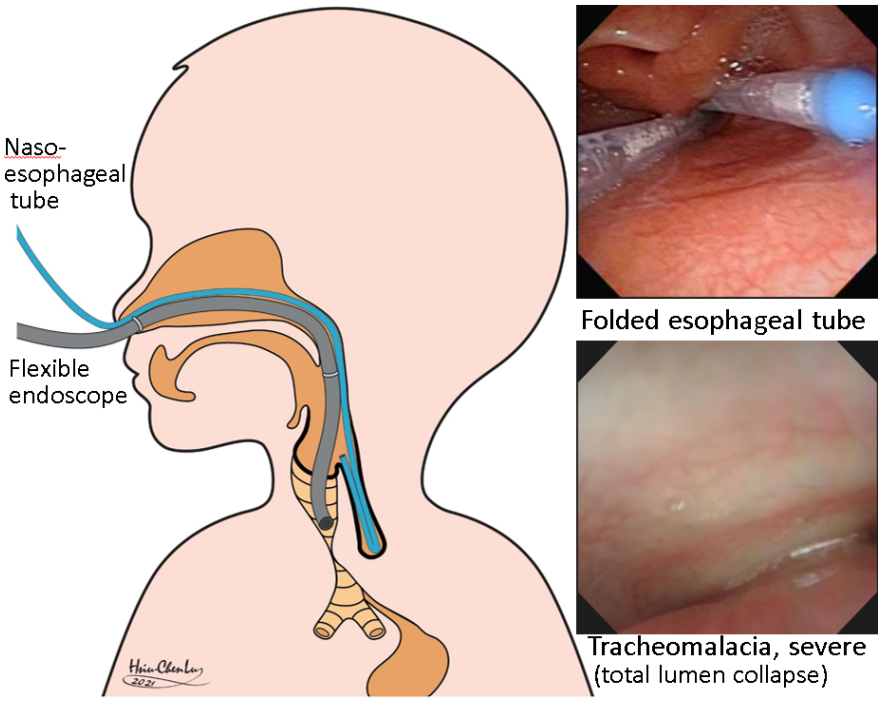

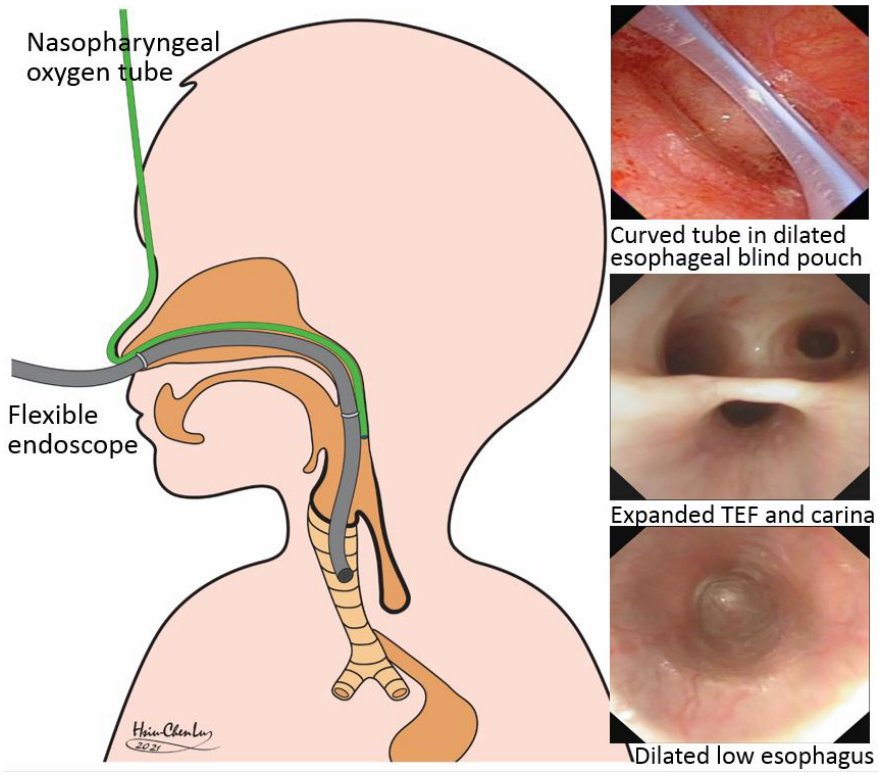

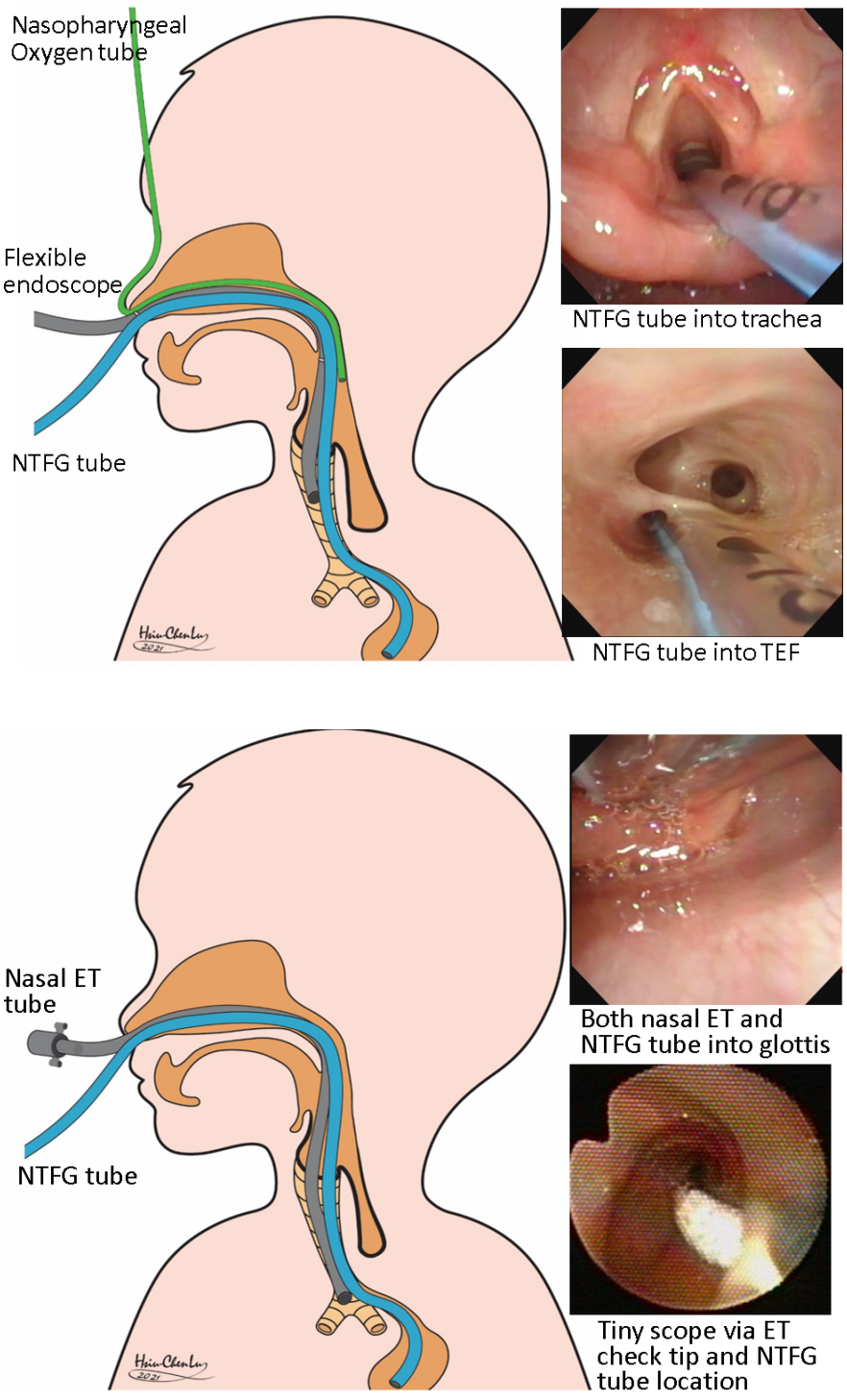

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