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Original Article

Bronchoscopic Localization of Tracheoesophageal Fistula in Newborns with Esophageal Atresia: Intubate Above or Below the Fistula?

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ABSTRACT

Purpose: In neonates with suspected type C esophageal atresia and tracheoesophageal fistula (EA/TEF) who require preoperative intubation, some texts advocate for attempted "deep" or distal-to-fistula intubation. However, this can lead to gastric distension and ventilatory compromise if a distal fistula is accidently intubated. This study examines the distribution of tracheoesophageal fistula locations in neonates with type C EA/TEF as determined by intraoperative bronchoscopy.

Methods: This was a single-center retrospective review of neonates with suspected type C EA/TEF who underwent primary repair with intraoperative bronchoscopy between 2010 and 2020. Data were collected on demographics and fistula location during bronchoscopic evaluation. Fistula location was categorized as amenable to blind deep intubation (>1.5 cm above carina) or not amenable to blind deep intubation intubation (≤1.5 cm above carina or carinal).

Results: Sixty-nine neonates underwent primary repair of Type C EA/TEF with intraoperative bronchoscopy during the study period. Three patients did not have documented fistula locations and were excluded (n = 66). In total, 49 (74 %) of patients were found to have fistulas located ≤1.5 cm from the carina that were not amenable to blind deep intubation. Only 17 patients (26 %) had fistulas >1.5 cm above carina potentially amenable to blind deep intubation.

Conclusions: Most neonates with suspected type C esophageal atresia and tracheoesophageal fistula have distal tracheal and carinal fistulas that are not amenable to blind deep intubation.

Level of Evidence: Level III.

1. Introduction

Esophageal atresia (EA) is a rare congenital anomaly characterized by discontinuity of the upper gastrointestinal tract, often accompanied by a tracheoesophageal fistula (TEF) [1]. At a reported incidence of 1 in 3000 to 1 in 4500 live births, EA/TEF is one of the most common gastrointestinal birth defects worldwide and one of the most difficult conditions pediatric surgeons manage in the modern era [1,2]. The most common EA/TEF abnormality, Gross type C, describes a proximal EA with a distal TEF, representing a challenge in anesthetic and airway management.

In neonates with suspected type C EA/TEF, endotracheal intubation and positive pressure ventilation should be avoided, if possible, prior to surgical correction of the fistula due to the risk of accidental intubation of the fistula and difficulty with ventilation secondary to preferential airflow into the stomach via the TEF with resulting gastric distension [3]. If a newborn requires intubation without bronchoscopic guidance, some neonatology and anesthesia texts advocate for attempted "deep" intubation with the hopes of bypassing the fistula, such that the endotracheal tube is positioned just above the level of the carina and distal to the suspected fistula location [3–5]. Because this is done without visual guidance and direct confirmation of the TEF location, this strategy may be misguided, as one is simply guessing where the fistula could be; if the fistula is mistakenly intubated due to its distal location, deep intubation may lead to gastric distension, reflux, ventilatory compromise, and ultimately, death [4,6].

Reported incidence of distal tracheal and carinal fistulas have been inconsistent in the literature, ranging from 9 to 55 % [7–11]. This study aims to examine the distribution of TEF location in

Abbreviations: EA, esophageal fistula; TEF, tracheoesophageal fistula.

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neonates with type C EA/TEF as determined by intraoperative bronchoscopy. We hypothesized that the majority of neonates with suspected type C EA/TEF have distal fistulas that are not amenable to blind "deep" distal-to-fistula intubation.

2. Methods

This study was approved and waiver for consent was obtained from the Boston Children's Hospital Institutional Review Board (IRB-P00004344).

2.1. Study population & data source

This was a single-center retrospective review of consecutive neonates with suspected Type C EA/TEF who underwent primary repair with intraoperative bronchoscopy at a free-standing tertiary pediatric hospital between 2010 and 2020. Data were collected on patient demographics, intraoperative details, and fistula characteristics during intraoperative bronchoscopic evaluation.

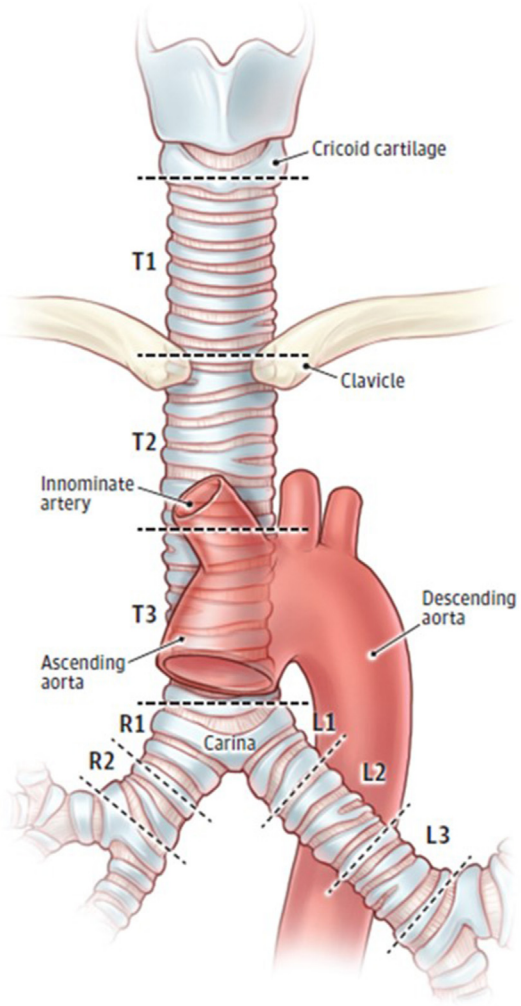

Fig. 1. The upper airway and trachea are subdivided into standardized T1/T2/T3 anatomical regions. T1, or the cervical trachea, is defined as the upper third of the trachea from the cricoid cartilage to the clavicles. T2 is the middle third of the trachea from the clavicles to the takeoff of the innominate artery (visualized bronchoscopically as an anterior tracheal impression), and T3 is the lower third of the trachea from the innominate artery takeoff to the carina (From Choi et al., 2019 in JAMA Otolaryngology, with permission under licensing agreement).

2.2. Outcome measurements & study variables

A rigid bronchoscopy was routinely performed in the OR prior to each EA/TEF repair by the operating surgeon. Whenever possible, this was performed as a combined airway evaluation with the Otolaryngology team to assess for clefts and subglottic/sublaryngeal pathology. Details on fistula location were obtained from surgeon-dictated reports. Fistula location was either quantified as estimated centimeters from the carina, or by standardized T1/T2/T3 anatomical regions (Fig. 1). T1, or the cervical trachea, is defined as the upper third of the trachea from the cricoid cartilage to the clavicles, T2 as the middle third of the trachea from the clavicles to the takeoff of the innominate artery (visualized bronchoscopically as an anterior tracheal impression), and T3 as the lower third of the trachea from the innominate artery takeoff to the carina [12]. Reports utilizing T1/T2/T3 nomenclature were recategorized into estimated centimeters, with T1 and T2 defined as >2 cm from carina. Fistulas reported as T3, which encompasses all fistulas ≤2 cm, were defined as 2 cm from carina − and therefore amenable to blind intubation as per our cutoff of >1.5 cm. Carinal and TEFs located ≤1.5 cm from carina (Fig. 2A-B) were considered not amenable to blind deep intubation distal to fistula, while TEFs located >1.5 cm from carina were considered amenable to blind deep intubation (Fig. 2C-D).

Descriptive statistics are provided as counts (percentages) for categorical variables, and as mean (standard deviation) for continuous variables. All statistics were calculated using SPSS (IBM Corp. Released 2021. IBM SPSS Statistics for Windows, Version 28.0. Armonk, NY: IBM Corp).

3. Results

Sixty-nine neonates underwent primary repair of type C EA/TEF during the study period. All patients underwent intraoperative bronchoscopy. Of these, 3 patients did not have clearly documented fistula locations and were excluded from analysis (n = 66). The majority (58 %) of neonates were male, and born at term gestation (71 %). Of premature infants (29 %, n = 19), 12 were born at 34–36 weeks gestation and 7 were born at 30–33 weeks gestation; no infants were born prior to 29 weeks gestation. On average, infants underwent EA/TEF repair 2 days after birth (SD = 1.2). The average weight at time of EA/TEF repair was 2636 g (SD = 623). Eight infants (12 %) required preoperative intubation for an average of 1.4 days (SD = 0.7). Premature infants were not significantly more likely to require preoperative intubation (p = 0.16).

In total, 49 (74 %) of patients were found to have fistulas located ≤1.5 cm from the carina that were not amenable to blind deep intubation. Of these, 25 patients (38 %) had carinal fistulas and 24 (36 %) had fistulas approximately ≤1 cm from carina. Only 17 patients (26 %) had fistulas >1.5 cm above carina potentially amenable to blind deep intubation. Of these, eight patients (12 %) had fistulas approximately 2 cm from carina and nine (14 %) had fistulas documented as >2 cm from carina (Fig. 3).

4. Discussion

Despite advances in perioperative management of EA/TEF, airway management in neonates with suspected type C EA/TEF remains challenging [4]. Positive pressure ventilation in patients with distal TEF can lead to gastric distension and respiratory compromise [3]. We questioned the safety and appropriateness of a classic teaching within the neonatal critical care and anesthesia literature that advocates for "deep" or distal-to-fistula intubation to bypass the TEF, prevent gastric insufflation and facilitate ventilation [3,5,6]. Our study results show that the majority of neonates with

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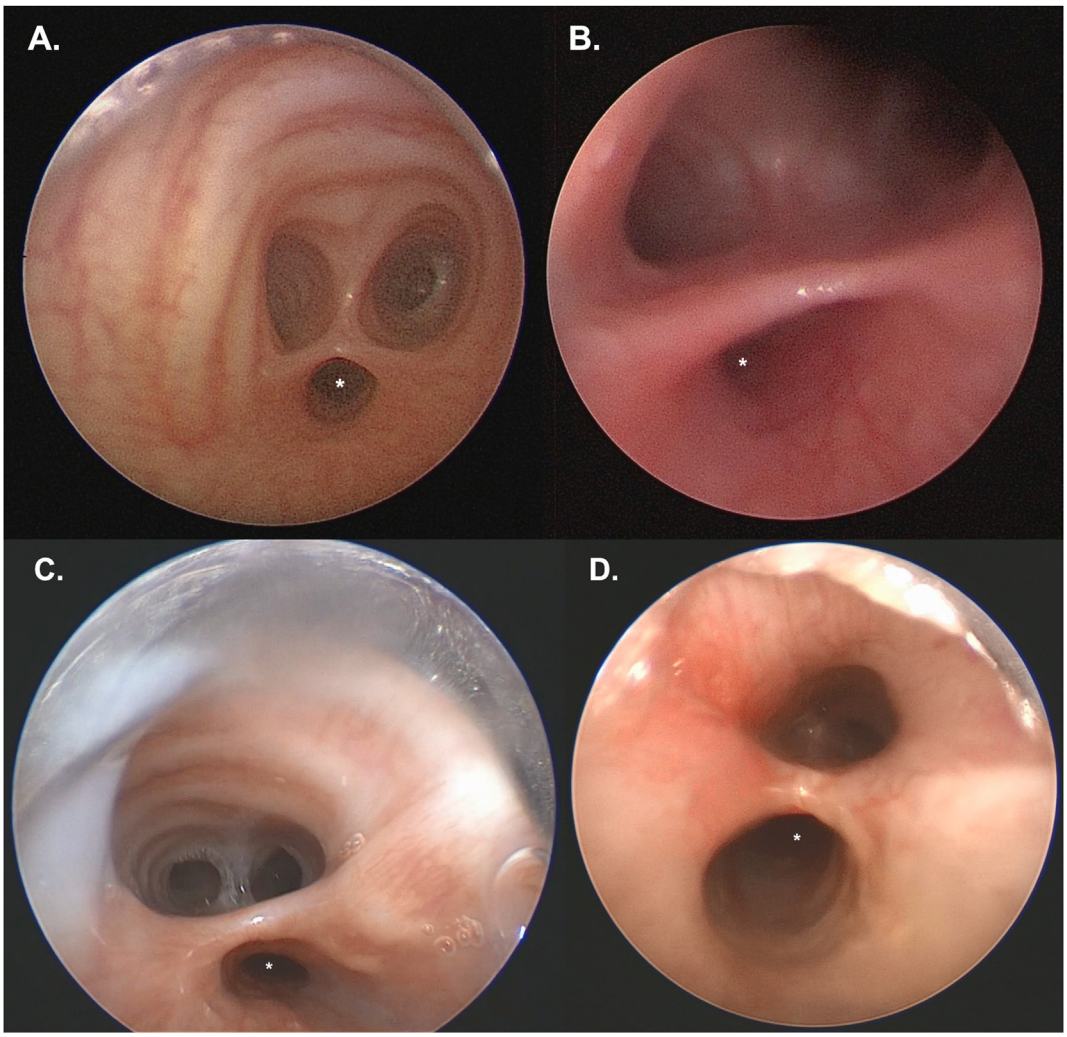

Fig. 2. Tracheoesophageal fistulas in neonates with type C EA/TEF during intraoperative bronchoscopy evaluation (fistulas depicted by *). Carinal fistulas (2A) and distal fistulas located approximately 1 cm from carina (2B) are not amenable to blind deep intubation. Fistulas located approximately 2 cm above carina (2C) and high fistulas >2 cm from carina (2D) were categorized as amenable to blind deep intubation in our study, although other factors such as size of fistula opening, may play an additional role in the risk of accidental fistula intubation during a blind intubation attempt.

suspected type C EA/TEF have distal TEFs located ≤1 cm from the carina (Fig. 2A-B). Hence, most type C EA/TEF children are not amenable to a "deep" endotracheal intubation strategy. Instead, blind attempts (not bronchoscopically guided) at deep intubation in this cohort of patients may carry increased risk of accidental fistula intubation.

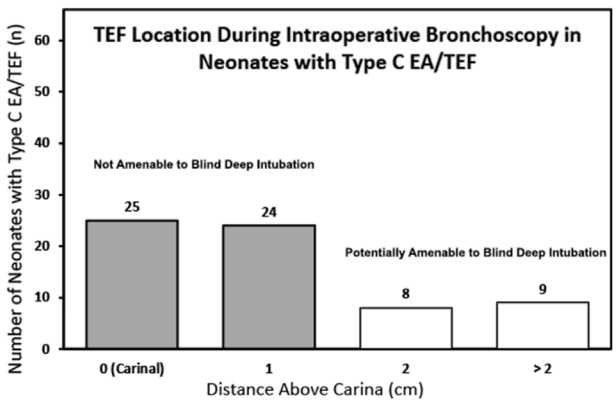

Fig. 3. The majority of patients in our cohort had fistulas ≤1.5 cm from the carina that were not amenable to blind deep intubation. Only 17 patients had fistulas >1.5 cm above carina that were considered amenable to blind deep intubation.

The dangers of accidental fistula intubation in neonates with EA/TEF have been previously reported in the literature. A case report by Alabbad et al. warns against the feasibility of blind deep intubation by highlighting an intraoperative mortality caused by the unknown passage of an endotracheal tube into a distal TEF located 1.5 cm above the carina in a neonate with type C EA/TEF [4]. Similarly, Hwang et al. reported a case of incidental intraoperative fistula intubation into a TEF located 1.02 cm above the carina [6]. After removal of the initial endotracheal tube, a second attempt at intubation was performed using video laryngoscopy. The fistula was again intubated despite direct visualization, but appropriate endotracheal tube positioning was ultimately achieved with careful repositioning. Such interventions are possible during controlled intraoperative settings, but may not be feasible during emergency airway control with attempted blind intubation prior to surgical correction.

The incidence of distal and carinal TEFs in neonates with type C EA/TEF have been previously examined in the literature, though reports vary widely from 9 to 55 % [7–11]. The earliest report and

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highest incidence of carinal fistula localization was published in 1958, when Roberts et al. reported a case series of 33 infants with type C EA/TEF, 18 of whom had carinal fistulas (54.5 %) [8]. Subsequent series looking at similar data varied in their reported incidence, ranging from a 9.3 % incidence of carinal fistulas reported by Holzki et al. to 45.2 % reported by Atzori et al. [7,11]. Of all published studies, only Holzki et al. reported on the presence of non-carinal TEF located <1 cm above the carina (21.1 % in his cohort of 161 patients) in 1992 [7], and our data represents the only modern study to also report on this distinction. In our cohort, we found a 37.9 % incidence of carinal fistulas, which was within the range of carinal fistula incidence previously reported in the literature. Importantly, we also found an equally high percentage of distal TEF located ≤1 cm above carina at 36.4 %. Because even TEF located as high as 1.5 cm from carina have been shown in the literature to carry the risk of accidental fistula intubation with deep intubation [4], the high incidence of TEF located within 1 cm above carina has significant clinical implications. In aggregate, neonates with fistulas at the carina and at ≤1 cm above carina comprise the majority of our cohort and also have anatomy that is not feasibly amenable to deep intubation.

Our categorization of carinal and fistulas ≤1.5 cm from carina (Fig. 2A-B) as not amenable to blind deep intubation was estimated from the aforementioned cases of incidental distal fistula intubations [4,6], as well as a review of the endotracheal tubes used in our patient cohort intraoperatively. The majority of our patients used a size 3.0 cuffed endotracheal tube. The length of the tube beyond the inflated cuff in a 3.0 endotracheal tube measures approximately 1.5 cm. With the cuff inflated, positive pressure ventilation would preferentially cause airflow to be directed distal to the cuff, and therefore, partially into the TEF. It is important to note, however, that emergency intubations in many NICUs would use an uncuffed endotracheal tube. Though an inflated cuff would not direct airflow into a TEF, the risk of incidental intubation into a distal fistula remains, as highlighted by the published cases of accidental fistula intubations located 1.5 cm and 1.02 cm from carina [4,6]. To date, no data exists on the incidence of accidental fistula intubation during emergency intubation in neonates with type C EA/TEF.

Our study has several limitations. Firstly, our measurements of fistula distance are estimated from intraoperative bronchoscopy. Despite our center's experience with a dedicated esophageal and airway team who performs a high volume of EA/TEF repairs, with routine preoperative rigid bronchoscopy, measurements made during intraoperative bronchoscopy are generally visual estimates, without the use of fluoroscopy or other direct methods to measure distance. Secondly, from an anatomical perspective, a 1.5 cm measurement in the airway of an 800 g neonate does not translate linearly to that of a 3.8 kg baby. A standardized T1/T2/T3 nomenclature (Fig. 1) may be more anatomically accurate in babies of different sizes [12]. However, as the majority of our bronchoscopy reports utilized a centimeter-based estimation of distance from carina, we believe the centimeter measurement is more accurate in representing our data. For the patients where only the T1/T2/T3 nomenclature was used in their bronchoscopy reporting, we converted these conservatively to centimeter distance estimates. As a result, all patients with T1/T2/T3 fistulas were categorized as ≥2 cm − and therefore, amenable to distal intubation by our definition of ≥1.5 cm from carina. As a consequence, our results may underpredict the incidence of distal TEF ≤1.5 cm from carina and an even greater percentage of patients may have a TEF that is not amenable to blind deep intubation.

Our study focuses on distal fistula location as an important risk factor for incidental TEF intubation, but does not account for other factors that may also similarly increase risk, such as the size of the fistula opening or fistula orientation along the trachea. In theory, a wide-mouthed proximal fistula may also carry the risk of fistula intubation during a blind intubation attempt. For example, Fig. 2D depicts a wide-mouthed proximal fistula (denoted by *) whose lumen appears larger than the true trachea, whereas Fig. 2C depicts a fistula (denoted by *) located approximately 2 cm from carina whose lumen is smaller than the true trachea. In our study, both of these fistulas would be classified as "amenable to deep intubation". In reality, however, the risk of accidental fistula intubation is relatively high in a neonate with anatomy depicted in Fig. 2D who has a wide-mouthed proximal fistula. While it is impossible to accurately measure the size of the fistula opening in every infant during bronchoscopic evaluation, the presence of these and other potential risk factors for fistula intubation underscore the importance of avoiding blind intubation in a neonate with suspected type C EA/TEF whenever possible. In the event that intubation is required for a rapidly decompensating infant, it is important to recognize the need for expeditious surgical control of the TEF. Even in a more controlled intraoperative setting, it is important to obtain direct visualization of TEF along the trachea with preoperative bronchoscopy. Whenever possible, the use of a Fogarty balloon to occlude the fistula lumen (Fig. 4) has been shown to provide adequate temporary control of the TEF until it can be repaired surgically [13]. Video-guided endotracheal tube placement under direct visualization further provides the ability to place the endotracheal tube in an ideal location proximal to the carina but distal to the fistula, which is extremely difficult to achieve during blind intubation in an emergent setting. Other strategies to lessen the chance of inadvertently intubating a high fistula blindly include rotating the bevel of the ETT posteriorly, such that the tube may preferentially slide into the trachea instead of the fistula, even if not attempting to intubate past the fistula.

In the setting of ventilatory difficulties (e.g. intermittent lack of end-tidal CO2, low tidal volumes, large system/circuit leak, distended abdomen) in a neonate with unrepaired EA/TEF type C, we suggest the following elements to consider. First, confirmation of ETT within the airway via direct laryngoscopy, auscultation of bilateral breath sounds, and/or X-ray. Next, attempt to suction out secretions from the ETT. Ensure adequate function of the naso-esophageal tube to suction secretions from the upper esophageal pouch. Finally, perhaps most importantly, is to pull-back the ETT roughly 1−2 cm, depending on the intubation depth, in order to

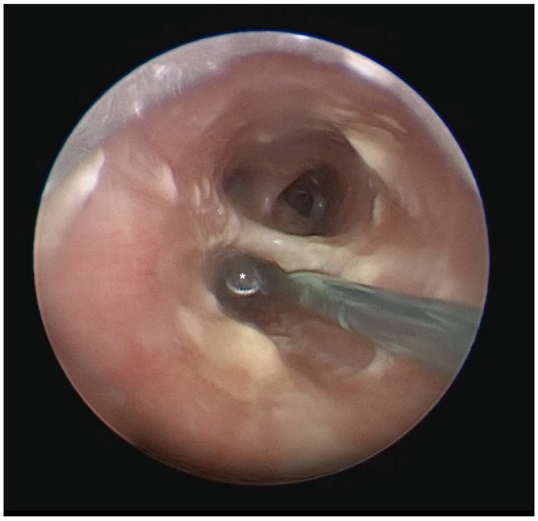

Fig. 4. A Fogarty balloon (depicted by *) can be used to occlude the fistula lumen and provide temporary control of the tracheoesophageal fistula until definitive surgical repair.

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come out of the TEF in case of accidental TEF intubation. If the patient's tracheal anatomy is unknown at time of intubation, it is safer to place the ETT shallowly to avoid accidental fistula intubation rather than attempt to place the ETT blindly past the fistula. In our center, cuffed endotracheal tubes are preferentially used, and if preoperative intubation is required, our goal is to intubate just deep enough to have the cuff past the vocal cords. When placing the ETT shallowly, the airway must be securely fastened to avoid inadvertent extubation and ETT dislodgement. Once stabilized, flexible bronchoscopy via the ETT can confirm adequate ETT position. Importantly, expeditious plans to surgically address the TEF in the operating room should be made.

In conclusion, despite the classic teaching of “deep” distal-to-fistula intubation in neonates with suspected type C EA/TEF, most neonates in our study had fistulas arising from the distal trachea located at the carina or $\leq$1.5 cm above the carina. Thus, the majority of such patients are not amenable to distal-to-fistula intubation without bronchoscopic guidance due to increased risk of accidental fistula intubation. Hence, we do not recommend blind deep intubation in this cohort of neonates.

Previous Communication

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Conflicts of interest

The authors have no sources of financial support or conflicts of interest that are relevant to this work.

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