

**CASE REPORT****Gastroenterology**

# Sedation-free transnasal esophagoscopy to evaluate and monitor esophageal diseases in children with esophageal atresia-tracheoesophageal fistula

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**Abstract**

Patients with esophageal atresia and tracheoesophageal fistula (EA-TEF) are at increased risk of conditions including gastroesophageal reflux, peptic esophagitis, gastric metaplasia, anastomotic strictures, eosinophilic esophagitis, and dysphagia. Patients with TEF-EA may need serial endoscopy in their lifetime given the known short- and long-term GI complications. There has been increased interest in pediatric unsedated transnasal endoscopy (TNE) as an endoscopic alternative as it is lower cost, has shorter recovery time, and eliminates potential risks associated with anesthesia. We report on the use of TNE with EA-TEF in four patients: One patient had gastroesophageal reflux disease, one patient had eosinophilic esophagitis and TNE was used for surveillance in two patients. Use of TNE allowed for close endoscopic monitoring and changes in medication management. The third and fourth patients underwent TNE as part of routine EA-TEF screening which is recommended by societal guidelines (Krishnan et al, *J Pediatr Gastroenterol Nutr.* 2016;63(5):550-570). Unsedated TNE is an alternative endoscopic approach in the management of patients with EA-TEF.

**KEYWORDS**

EA-TEF, pediatrics, unsedated endoscopy

## 1 | INTRODUCTION

Esophageal atresia (EA) is one of the most common congenital malformations, occurring in one in 2400–4500 births worldwide.<sup>1</sup> Most EA patients have concomitant tracheoesophageal fistula (TEF).<sup>2</sup> In 2016, the European Society for Paediatric Gastroenterology Hepatology and Nutrition (ESPGHAN) and the North American Society for Pediatric Gastroenterology, Hepatology and Nutrition (NASPGHAN) developed joint societal guidelines for the evaluation and treatment of children with EA-TEF. These patients are at increased

risk of conditions including gastroesophageal reflux (GER), peptic esophagitis, Barrett's esophagus/gastric metaplasia, anastomotic strictures, EoE, esophageal dysmotility, and dysphagia.<sup>3</sup> The joint guidelines recommend routine surveillance endoscopy after stopping proton pump inhibitor (PPI) therapy, before the age of 10 years, and at the time of transition to adulthood.<sup>3</sup> There has been increased interest in pediatrics in the use of in-office, virtual reality (VR) distraction and dissociation assisted, unsedated transnasal esophagoscopy (TNE) to monitor upper gastrointestinal tract conditions, such as eosinophilic esophagitis (EoE),

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which has been proven to be safe, well-tolerated and cost-efficient.<sup>4,5</sup> We report on the use of sedation-free, VR assisted, TNE in four children with EA-TEF. These 4 patients were all evaluated in a multidisciplinary Aerodigestive clinic and did not need other procedures under anesthesia; therefore, they were offered the option of unседated TNE. All patients underwent TNE as previously described.<sup>4,5</sup> Briefly, patients were asked not to eat or drink for 2 h before TNE. All patients utilized VR goggles for distraction. They all received six sprays of 4% aerosolized lidocaine intranasally and one spray orally for topical anesthesia. They underwent TNE in an outpatient clinic room while sitting upright in a chair designed for outpatient laryngoscopy procedures and all tolerated the procedure with no adverse events.

## 2 | CASE SERIES

### 2.1 | Patient 1

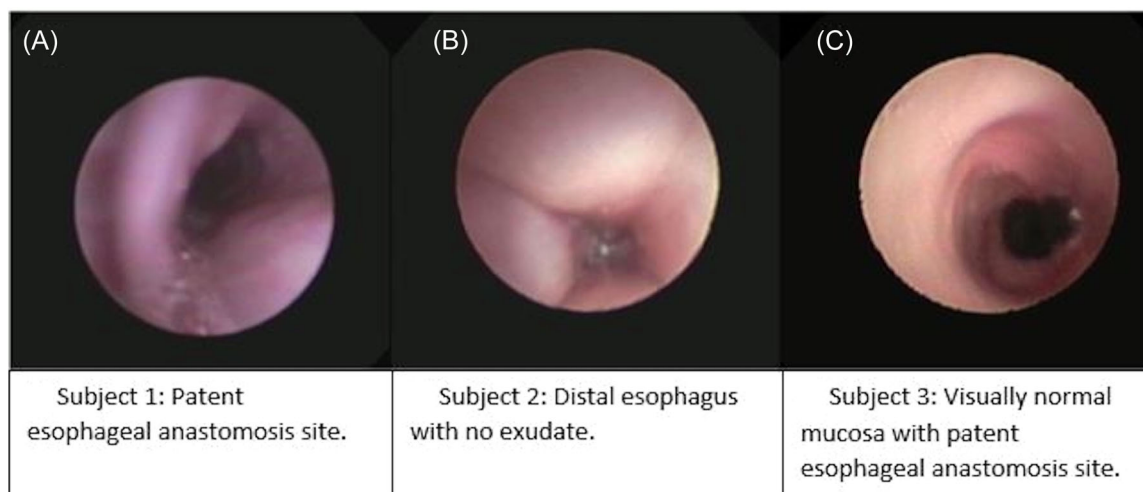
Nine-year-old female with EA-TEF status post-repair who presented to the Aerodigestive Multidisciplinary Clinic with symptoms of dysphagia and regurgitation after eating. She underwent a fluoroscopic upper gastrointestinal series with no esophageal stricture. She subsequently underwent an esophagogastroduodenoscopy (EGD) under anesthesia that showed distal esophagitis with 10 eosinophils per high power field (eos/hpf) secondary to reflux. Proton Pump Inhibitor (PPI) was increased to twice daily and she had improvement in symptoms. Six months later, she underwent TNE which showed the distal esophagitis had resolved and a patent anastomosis site (Figure 1A). The PPI was decreased to daily dosing.

### 2.2 | Patient 2

Four-year-old male with a history of long gap EA (status post Foker repair) in infancy, who subsequently was diagnosed with EoE at 15 months of age. He was started on swallowed topical steroids (TCS) and PPI without improvement in his EoE or reflux. An esophagram showed no esophageal stricture. He had previously undergone two EGDs under anesthesia and family now wanted to pursue TNE. While on PPI twice daily, he underwent TNE at the age of 7 years that was visually normal, but with 60 eos/hpf in the proximal esophagus and 160 eos/hpf in the distal esophagus. His treatment was modified, and he was started on a regimen of PPI, H2 blocker, TCS and removal of dairy with follow up EGD showing a patent anastomosis site, but with exudate and furrowing consistent with active EoE. His swallowed topical steroid was increased to twice daily and a subsequent TNE was visually normal (Figure 1B) with improved histology (1 and 5 eos/hpf in proximal and distal esophagus, respectively).

### 2.3 | Patient 3

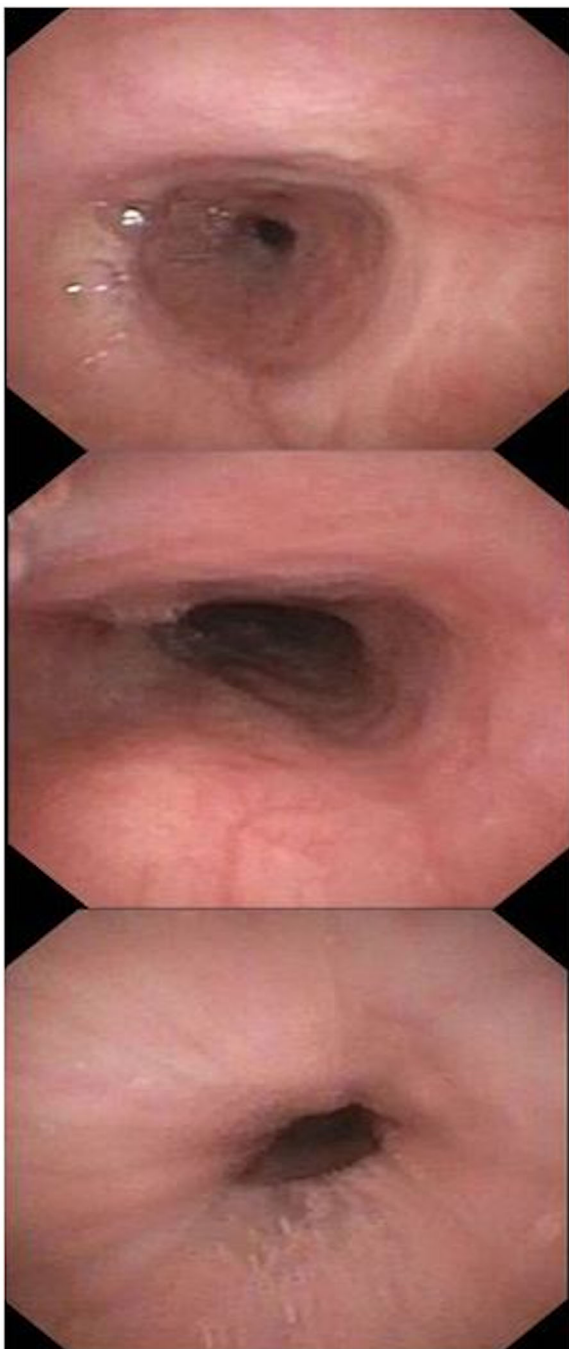
Five-year-old female with EA-TEF status post-repair was seen in the Aerodigestive Multidisciplinary Clinic and underwent EGD under anesthesia that was visually and histologically normal (Figure 1C). She continued to be followed in clinic with occasional regurgitation symptoms. At the age of 10, she underwent TNE for routine EA-TEF monitoring, per the NASPGHAN guidelines that showed a patent anastomosis site and normal biopsies without evidence of esophagitis or EoE.



**FIGURE 1** (A) Subject 1: Patent esophageal anastomosis site. (B) Subject 2: Distal Esophagus with no exudate. (C) Subject 3: Visually normal mucosa with patent esophageal anastomosis site.

## 2.4 | Patient 4

A 10-year-old otherwise healthy child with a history EA-TEF reported worsening dysphagia and had stopped acid suppression 2 months prior. Due to previous history of multiple fluoroscopic studies, patient and family expressed desire to consider non-radiographic evaluation to avoid radiation. Patient was offered TNE and underwent sedation-free TNE with biopsies.



**FIGURE 2** Top photo: esophageal anastomosis, middle photo: mid-esophagus, bottom photo: lower esophageal sphincter of subject 4.

Biopsies revealed mild esophagitis with <15 eos per hpf in both the distal and proximal esophagus. There was no visual evidence of stricture noted. (Figure 2) Given these findings, decision was made to continue off acid suppression and monitor symptoms clinically.

## 3 | DISCUSSION

Here we describe the novel use of sedation-free TNE in 4 pediatric patients with EA-TEF as a feasible and safe alternative to sedated oral flexible endoscopy under anesthesia. Unsedated pediatric TNE has been well described to monitor EoE, however, this case series is the first report of its use in a series pediatric patients with EA-TEF.<sup>5,6</sup>

Patients with EA-TEF are at risk for short- and long-term GI disease sequelae throughout their life and into adulthood.<sup>7</sup> Joint guidelines by ESPGHAN-NASPGHAN currently recommend routine surveillance endoscopy in asymptomatic children at certain points (after stopping PPI, before the age of 10 years, and at the time of transition to adulthood) and symptomatic children may need to undergo endoscopy more often.<sup>3</sup> Many patients with EA-TEF may have comorbid respiratory problems such as tracheomalacia or bronchiectasis which may put them at higher risk for general anesthesia.<sup>8</sup> Given that up to 60% of adverse events related to pediatric sedated endoscopy are from anesthesia/sedation complications, sedation-free TNE provides a safe option for patients who require endoscopic monitoring.<sup>9</sup> Furthermore, several studies have shown patient and parent satisfaction with pediatric unsedated TNE.<sup>4,10</sup> The use of VR in an immersive environment for distraction and dissociation may have a positive impact on the high procedural success rate in children, a population that typically might not tolerate unsedated endoscopy as well as adults. VR has been shown to reduce pain and anxiety and improve satisfaction in children undergoing painful medical procedures such as intravenous line placements and burn dressing changes.<sup>11</sup> TNE has several advantages compared to endoscopy under anesthesia. These include cost-efficiency, decreased risks that could be associated anesthesia, and less time away from work/school.<sup>4-6</sup> Nguyen et al described that TNE charges were over 50% lower when compared to EGD under anesthesia at a single center while Sabe et al showed that the mean visit duration for TNE was much lower (36 min) compared to sedated endoscopy (162 min).<sup>6</sup>

In conclusion, this case series highlights the use of pediatric unsedated TNE as an alternative and reliable method of evaluating symptomatic individuals or patients in need of endoscopic evaluation with EA-TEF. Furthermore, it provides a less costly endoscopic option and decreases the endoscopic risks associated

with anesthesia in a patient population who may have comorbid respiratory disease. Patient selection is helpful when performing a successful TNE. With EA-TEF, it is often important to rule out significant anastomotic strictures with fluoroscopic imaging, however, as described in Patient 4, TNE could be used to rule out significant stricture in selected patients who are hesitant to undergo fluoroscopic evaluation due to radiation. TNE, however, has not been shown to be effective to evaluate underlying fibrosis or dysmotility that could also be missed on fluoroscopic or endoscopic studies. As described here, unsedated TNE is well tolerated and is an alternative endoscopic approach for patients with EA-TEF. It can be used to evaluate and monitor the long term esophageal sequelae of the condition as well as for routine surveillance as recommended by the ESPGHAN-NASPGHAN guidelines.<sup>3</sup>

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Institutional Review Board approval was obtained for retrospective chart review used in this case series. The authors have no funding to report.

### CONFLICT OF INTEREST STATEMENT

Nathalie Nguyen has served as a consultant for Regeneron/Sanofi and EvoEndo, Inc. Joel A. Friedlander is the Chief Medical Officer and Board Member of EvoEndo, Inc. He is the co-inventor/coinventor on several patents and patents pending related to endoscopic and virtual reality technologies. Clint Smith is an employee of EvoEndo, Inc. The authors declare no conflicts of interest.

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