Tracheocutaneous Fistula Closure in the Pediatric Population: Should Secondary Closure Be the Standard of Care?

Alexander J. Osborn, MD, PhD¹, Alessandro de Alarcon, MD, MPH¹,², Catherine K. Hart, MD¹,², Robin T. Cotton, MD¹,², and Michael J. Rutter, MBChB, FRACS¹,²

Sponsorships or competing interests that may be relevant to content are disclosed at the end of this article.

Abstract

Objective. Tracheocutaneous fistula (TCF) closure is achieved by excision followed either by primary closure or healing by secondary intention. Although primary closure provides immediate resolution of the fistula, it is associated with more severe potential complications. Healing by secondary intention minimizes these potential complications; however, it is inconvenient for the patient and may be more likely to require revision surgery. We have had 2 life-threatening complications after primary closure, and as a result, we largely changed our practice pattern. We compared complication and success rates of the 2 methods since this change to determine its ramifications.

Study Design. Historical cohort study.

Setting. Academic tertiary care pediatric otolaryngology practice.

Subjects and Methods. Two hundred sixteen patients who underwent TCF closure between January 2004 and August 2012.

Results. Forty-six (21.3%) fistulae were addressed by primary closure, and 170 (78.7%) were addressed by secondary intention. The complication and revision rates were not significantly different between the 2 methods (8.7% vs 10% and 8.7% vs 14.7%, respectively).

Conclusion. In our study, we did not see any statistical differences between the 2 methods studied but could not exclude clinically important differences that may have favored one method over the other. Although our comparative results were inconclusive, we have adopted secondary closure as standard practice for management of pediatric TCF. Individual surgeons and patients may use the data presented to help guide decisions concerning which procedure is most appropriate.

Keywords
pediatric, tracheotomy, tracheocutaneous fistula, tracheocutaneous fistula closure, airway complication

Received May 28, 2013; revised July 12, 2013; accepted July 18, 2013.

TracheotomY in children is a commonly performed surgical procedure. Rates of tracheotomy in those admitted to pediatric or neonatal intensive care units (PICU/NICU) are approximately 1% to 2%.¹² Given best estimates of PICU and NICU admissions, this implies at least 2500 to 4500 tracheotomies are performed annually on children in the United States.³ Short-term complications of tracheotomy are relatively rare, and the most common long-term complication is peristomal granulation tissue.⁴,⁵ Serious complications or adverse events associated with tracheotomy are rare.

In most cases, decannulation results in the tracheostomy closing spontaneously; however, the rate of a persistent tracheocutaneous fistula (TCF) ranges from 6.2% to 37.1% in children.⁴,⁶,⁷ In children, the development of TCF appears to be related to duration of tracheostomy and age at tracheotomy.⁷ In adults, additional risk factors such as radiation, previous tracheotomy, and obesity also predispose to developing a fistula.⁸ These factors are likely to also predispose to TCF formation in children but have not been identified in studies, presumably because of their relative rarity in the pediatric population.

TCF closure is achieved by excision of the fistulous tract with multilayered primary closure or excision of the tract with healing by secondary intention.⁹,¹⁰ Primary closure often results in better cosmetic outcome and is easier for the patient to maintain in the immediate postoperative period;

¹Division of Otolaryngology–Head and Neck Surgery, Cincinnati Children’s Hospital Medical Center, Cincinnati, Ohio, USA
²Aerodigestive, Esophageal, and Sleep Center, Cincinnati Children’s Hospital Medical Center, Cincinnati, Ohio, USA

This article was accepted for presentation at the 2013 AAO-HNSF Annual Meeting and OTO EXPO⁹⁰; September 29–October 3, 2013; Vancouver, BC, Canada.

Corresponding Author:
Alexander J. Osborn, MD, PhD, Division of Otolaryngology–Head and Neck Surgery, Cincinnati Children’s Hospital Medical Center, 3333 Burnet Ave ML 2018, Cincinnati OH 45229, USA.
Email: alexander.osborn2@cchmc.org
Table 1. Characteristics of children who underwent TCF closure.

<table>
<thead>
<tr>
<th>Characteristics</th>
<th>Total</th>
<th>Primary Closure</th>
<th>Secondary Intention</th>
</tr>
</thead>
<tbody>
<tr>
<td>Patients</td>
<td>216</td>
<td>46</td>
<td>170</td>
</tr>
<tr>
<td>Male</td>
<td>120</td>
<td>26</td>
<td>94</td>
</tr>
<tr>
<td>Female</td>
<td>96</td>
<td>20</td>
<td>76</td>
</tr>
<tr>
<td>Median (IQR) age at tracheotomy, mo</td>
<td>3.07 (1.7-4)</td>
<td>3.77 (2.7-2)</td>
<td>3.07 (1.7-3)</td>
</tr>
<tr>
<td>Median time (IQR) with tracheotomy, mo</td>
<td>37.8 (23.1-62)</td>
<td>23.9 (15.4-51.2)*</td>
<td>40.5 (27.4-63.7)*</td>
</tr>
<tr>
<td>Median (IQR) age at decannulation, mo</td>
<td>49.8 (30.4-81.7)</td>
<td>39.1 (23.3-79.3)</td>
<td>50 (32.8-81.5)</td>
</tr>
<tr>
<td>Median (IQR) time between decannulation and TCF closure, mo</td>
<td>6.6 (4.1-11.1)</td>
<td>6.3 (3.8-11.5)</td>
<td>6.7 (4.1-11.1)</td>
</tr>
<tr>
<td>Median (IQR) age at TCF closure, mo</td>
<td>60.3 (41.2-88.4)</td>
<td>50.1 (32.7-88.3)</td>
<td>60.8 (44.9-87.8)</td>
</tr>
</tbody>
</table>

Abbreviations: IQR, interquartile range; TCF, tracheocutaneous fistula.

*The primary closure and secondary intention groups are statistically significantly different with respect to this variable (P = .004).

however, the risk of serious complications such as subcutaneous emphysema, pneumomediastinum, pneumothorax, and respiratory distress appear higher with this method.19

Many variations of primary closure have been developed and claim to reduce risks while maintaining cosmetic benefit and convenience; however, large and long-term studies of such methods are absent.11-16 We have previously compared the 2 methods of TCF closure at our institution and found no significant differences in risks or efficacy between primary closure and secondary intention.9 Shortly after this study was published, we experienced 2 serious complications using the primary closure method. This prompted a gradual cultural shift in our Division toward the secondary intention method. Our aim is to evaluate the efficacy and complication rates of the 2 methods during this transition and to use these data to determine the consequences of such a shift and if there were drawbacks to using the method of secondary intention.

Methods

Approval for the study was obtained from the Cincinnati Children’s Hospital Institutional Review Board (protocol 2012-2128). Our surgical records between January 2004 and August 2012 were queried for CPT codes 31820 (surgical closure tracheostomy or fistula; without plastic repair) and 31825 (surgical closure tracheostomy or fistula; with plastic repair), and procedures were verified by examination of the operative reports. The inpatient and outpatient charts of these patients were then reviewed for the following information: gender, date of birth, date of tracheotomy, date of decannulation, date of fistula closure, method of closure, postoperative complications, need for revision, and additional postoperative procedures other than surveillance. The timing of TCF closure as well as the decision of which procedure was performed was left to the discretion of the operating surgeon. Children who underwent primary closure had part of the tract imbricated and oversewn, multilayered closure of the overlying tissue, and a rubber band or Penrose drain. Children who underwent closure by secondary intention had the tract excised and a small tracheotomy tube placed within the wound to serve as an escape valve should the patient cough or buck during emergence. The tube was removed once the child was fully awake in the recovery area. All children were observed overnight regardless of which method of closure was performed. Parametric and nonparametric statistical tests were used for our analysis as appropriate, depending on whether the variables examined fell into a normal distribution or not.

Results

We reviewed 216 children who underwent TCF closure by either primary closure or secondary intention, and key characteristics are listed in Table 1. When comparing the 2 groups, there were no significant differences in gender distribution, age at tracheotomy, age at decannulation, age at TCF closure, or time between decannulation and TCF closure (P > .05 in all cases, Mann-Whitney test). There was a significant difference in duration of cannulation, with the children undergoing primary closure having on the whole a shorter time with the tracheotomy tube in place (P = .004).

Complication rates between the 2 methods were similar (8.7% primary vs 10% secondary), and the odds ratio was not significant (P = .79; Table 2). The most frequent complication was desaturations requiring supplemental oxygen. Two children in the secondary intention group went back to the operating room for bronchoscopy due to episodes of hypoxia. Both of these children, as well as 1 with stridor and 1 with subcutaneous air, had their tracheotomy tubes replaced within the first 24 hours after surgery. In all instances, this tracheotomy tube was removed within 2 weeks. In the instances of subcutaneous emphysema, air was limited to the neck and did not result in pneumothorax or pneumomediastinum. Both cases resolved spontaneously. No one in the primary closure group required such treatment.

Fourteen children, 3/46 (6.5%) in the primary closure group and 11/170 (6.5%) in the secondary intention group, required revision surgery for persistent TCF. No child required a second revision, and there were no complications associated with revisions. One child in the primary closure group and 14 in the secondary intention group required additional surgery as a result of the tracheotomy but not to close persistent TCF. The most common additional
Table 2. Complications and revision surgery in the 2 cohorts.

<table>
<thead>
<tr>
<th></th>
<th>Primary Closure</th>
<th>Secondary Intention</th>
<th>Odds Ratio (Primary:Secondary)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Total complications</td>
<td>4/46 (8.7%)</td>
<td>17/170 (10%)</td>
<td>0.86 (95% CI, 0.27-2.68)</td>
</tr>
<tr>
<td>Return to OR</td>
<td>0/4 (0%)</td>
<td>2/17 (11.7%)</td>
<td>0.69 (95% CI, 0.028-17.12)</td>
</tr>
<tr>
<td>Wound exploration/tracheotomy tube replaced</td>
<td>0/4 (0%)</td>
<td>4/17 (23.5%)</td>
<td>0.33 (95% CI, 0.15-7.48)</td>
</tr>
<tr>
<td>Revision surgery</td>
<td>4/46 (8.7%)</td>
<td>25/170 (14.7%)</td>
<td>0.55 (95% CI, 0.18-1.68)</td>
</tr>
</tbody>
</table>

Abbreviations: CI, confidence interval; OR, operating room.

*Complications included desaturation, O₂ requirement (2), stridor and increased work of breathing (1), and stitch abscess (1).

Revision methods included cautery (2), and excision with primary closure (1); other revision surgery included endoscopic removal of granuloma (1).

Revision methods included excision with primary closure (5), excision with secondary intention (5), and cautery (1); other revision surgery included scar revision (11), endoscopic removal of granuloma (2), and tracheoplasty with graft to alleviate stenosis (1).

procedures were scar revision (6.5% of all secondary intention patients) and removal of airway granulation. Again, odds ratios demonstrated no significant differences between the 2 groups ($P = .29$).

Discussion

We reviewed our experience with TCF closure, and the current series and our previously published experience are the only 2 published reports that directly compare large numbers of patients who have undergone TCF closure by primary closure or secondary intention. The demographics and timing of surgery for our series are similar to those of other published reports. We did notice a significant difference in duration of cannulation between the 2 groups. This is likely a correlation rather than true causation, and duration of cannulation did not influence the approach the surgeon took to TCF closure. If anything, this may reflect the fact that the surgeons in our practice who preferred primary closure also had a predilection toward earlier decannulation or were managing airway problems that were eligible for decannulation earlier.

A number of studies have been published regarding TCF closure (Table 3). Looking at the studies chronologically, one can see there has been a shift over time toward advocating for primary closure. Early studies, as well as several case reports, advocate the method of secondary intention for TCF closure to avoid serious complications such as subcutaneous emphysema resulting in respiratory distress, pneumomediastinum, pneumothorax, or death. Subsequently, advocates of primary closure, most notably from centers in Chicago, Cincinnati, and London, have published large series generally highlighting the safety of the technique.

Two caveats are worth mentioning when considering these series. The first is that despite generally advocating for primary closure, the authors highlight the need to wisely select patients who undergo this procedure. Children with predilection toward or currently active upper respiratory infections, pulmonary issues, cough, and large defects ($\geq 5$ mm) would be considered poor candidates for primary closure and at higher risk of serious complications that can arise from this method. The second caveat is that despite following these guidelines for careful patient selection, each series had at least 1 serious complication. Schroeder et al described 1 instance of cervical subcutaneous emphysema in which the child developed a cough 1 week postoperation requiring the wound be opened. Geyer et al described 4 instances of subcutaneous air developing, 1 of which required an emergency return to the operating room. Finally, Stern et al at our own institution described 1 instance of respiratory distress after primary closure requiring recannulation. Even though the rates of serious complications are low in each of these large series, and none resulted in death or negative long-term sequelae, clearly the potential for catastrophic events is present even with careful patient selection.

This potential is evidenced by the fact that in the period immediately following the publication of our original series, we had 2 major complications after primary closure of TCF. One patient developed massive subcutaneous emphysema, pneumothorax, and respiratory distress having been discharged after 24-hour observation. The fact that she was not in the hospital transformed this event from a serious complication to a near-death event. The second child developed bilateral pneumothorax and respiratory distress in the postanesthesia care unit requiring bilateral tube thoracostomy. Finally, we were involved in caring for a child, the primary TCF closure for whom was done elsewhere, who ultimately died after developing subcutaneous air and respiratory distress, which progressed to respiratory failure. These 3 events have set in motion a gradual cultural shift in our department away from primary closure of TCF.

We took advantage of this period of time during which most physicians in our group had changed from primary to secondary closure to undertake the current study and determine the ramifications of this shift. Success rates for the 2 procedures are the same in our series. The rate of scar revision for the secondary intention group is 6.5%. In addition, the rates of complication of the 2 methods in our series were similar, and no serious complications were encountered in either group during this study period, underscoring the rarity of serious complications from this procedure regardless of method. In light of equal efficacy, the only
drawback of the secondary intention method appears to be a relatively low rate of scar revision. We feel that the low but not insignificant rate of scar revision is an acceptable trade-off for the potential, albeit unproven, benefit of avoiding life-threatening complications that can arise from primary closure of TCF. Accordingly, since these results came to light, our departmental policy has been to use the secondary intention method for all TCF closures.

The results of this study, those of the previous study from our institution, as well as any study in which the rates of an index event are low must be interpreted with caution. Low complication rates between the 2 groups were not statistically different; however, the wide confidence intervals illustrate 2 points. First, despite our advocating for secondary closure, our data do not really answer the question of which method is truly safer. Based on Table 2, secondary intention may have as little as one-fourth or as high as 2½ times as many complications as primary closure. Second, they suggest the study is underpowered. Complication rates in our study are similar to those of other published series, and assuming a complication rate of 10%, the sample size of the study would have to be 566 to detect a 2-fold difference in rates of complication ($\alpha = .05, \beta = .2$). A smaller difference would require an even larger sample set. Given that 566 patients is a larger cohort than all of the previously published studies on the topic combined, further certainty in this arena may require multi-institutional collaboration, meta-analysis, or systematic review in order to accrue enough patients in each cohort.

Although the rate of scar revision is low, it is not inconsequential. The rate of scar revision likely underestimates the rate of parental or patient dissatisfaction with cosmesis after healing by secondary intention, and we did not consistently gather data regarding this subjective variable in the course of caring for these patients. Both the differences in cosmetic outcome between the methods and the fact that insurers are sometimes reticent in covering scar revision should be discussed preoperatively. It is worth testing on a large scale whether partial closure methods might improve cosmesis.

Recently, Wine et al (personal communication, May 2013) made an argument for TCF closure by secondary intention based on resource utilization. They similarly found no difference in rates of success or complication between

<table>
<thead>
<tr>
<th>Study</th>
<th>Year</th>
<th>n, Method</th>
<th>Comp.</th>
<th>Revisions</th>
<th>Recommendation</th>
</tr>
</thead>
<tbody>
<tr>
<td>Khaja et al (adult)</td>
<td>2011</td>
<td>13, Loose primary closure</td>
<td>0 (0%)</td>
<td>1 (7.7%)</td>
<td>Loose primary closure</td>
</tr>
<tr>
<td>Tasca and Clarke (adult)</td>
<td>2010</td>
<td>23, Primary closure with or without drain</td>
<td>4 (17.4%)</td>
<td>0 (0%)</td>
<td>Primary closure</td>
</tr>
<tr>
<td>Geyer et al (pediatric)</td>
<td>2008</td>
<td>100, Primary closure with or without drain</td>
<td>11 (11%)</td>
<td>Not mentioned</td>
<td>Primary closure except for large defects</td>
</tr>
<tr>
<td>Schroeder et al (pediatric)</td>
<td>2008</td>
<td>39, Primary closure with drain</td>
<td>5 (12.8%)</td>
<td>0 (0%)</td>
<td>Primary closure with drain but patient selection is important</td>
</tr>
<tr>
<td>Priestley and Berkowitz (pediatric)</td>
<td>2006</td>
<td>15, Primary closure without drain</td>
<td>3 (20%)</td>
<td>0 (0%)</td>
<td>Primary closure with drain but patient selection is important</td>
</tr>
<tr>
<td>Sautter et al (pediatric)</td>
<td>2006</td>
<td>13, Primary closure without drain</td>
<td>3 (23%)</td>
<td>0 (0%)</td>
<td>Primary closure without drain</td>
</tr>
<tr>
<td>Eaton et al (pediatric)</td>
<td>2003</td>
<td>13, Cautery and secondary intention</td>
<td>0 (0%)</td>
<td>1 (7.7%)</td>
<td>Cautery and secondary intention</td>
</tr>
<tr>
<td>Stern et al (pediatric)</td>
<td>1999</td>
<td>80, Primary closure with drain</td>
<td>1 (1%)</td>
<td>3 (3%)</td>
<td>Both equally safe and efficacious, but patient selection is important</td>
</tr>
<tr>
<td>Drezner and Cantrell (adult)</td>
<td>1998</td>
<td>5, Primary closure</td>
<td>2 (18%)</td>
<td>Not mentioned</td>
<td>Secondary intention especially for large tracheal defects</td>
</tr>
<tr>
<td>Licamelli et al (pediatric)</td>
<td>1997</td>
<td>8, De-epithelialization and primary closure</td>
<td>0 (0%)</td>
<td>1 (13%)</td>
<td>8 De-epithelialization and primary closure</td>
</tr>
<tr>
<td>Bressler et al (pediatric)</td>
<td>1994</td>
<td>36, Primary closure with drain</td>
<td>4 (11.1%)</td>
<td>0 (0%)</td>
<td>Primary closure</td>
</tr>
<tr>
<td>White and Smitheringale (pediatric)</td>
<td>1989</td>
<td>6, Primary closure</td>
<td>4 (26.7%)</td>
<td>Not mentioned</td>
<td>Secondary intention</td>
</tr>
</tbody>
</table>
the groups and demonstrated that procedure times were approximately one-third as long in the secondary intention group. Hospital stays were also significantly shorter in those children who underwent TCF closure by secondary intention compared with primary closure. This was primarily because children who underwent primary closure spent 1 night in the ICU while those who underwent closure by secondary intention were often discharged on the day of surgery. Our practice is to observe both groups of children overnight, and thus at our institution, benefit with regard to resource utilization would be limited to that gained by decreased operative times, and this gain would vary among institutions depending on postoperative monitoring protocols.

Our series is unusual in that 2 of our patients who underwent closure of TCF by secondary intention did experience subcutaneous emphysema, a complication that is canonically eliminated by using this technique; however, subcutaneous air is a known complication of tracheotomy itself, which essentially entails leaving an open wound. The cases were self-resolving, but one was worrisome enough to prompt the managing physician to replace the tracheotomy tube temporarily. Retrospective review of the charts did not reveal any predisposing factors or explanation for these events, and the episodes highlight the fact that air can dissect into the tissue planes of the neck even with the secondary intention method of TCF closure. This highlights the fact that perhaps overnight observation and close monitoring are more important than choice of closure method. The development of subcutaneous air in these cases was surprising, but to us supported our decision to leave the wound open. If subcutaneous air can develop with either method, our preference is to have an open wound since closure might force air into the mediastinum and thorax, as was seen in the anecdotal index cases that initially encouraged us to change our practice pattern. Although not proven in this or any other series due to a lack of statistical power, the less severe nature of subcutaneous air in the setting of an open wound as compared with a closed wound seems intuitively obvious. The fact that opening the wound and replacing the tracheotomy tube would be the first line of treatment for subcutaneous emphysema after fistula excision and primary closure supports this intuition.

With standard tracheotomy techniques, the rate of TCF varies widely and ranges from approximately 5% to 40%. The “starplasty” technique, advocated by Koltai and colleagues for its lower rate of accidental decannulation, lower risk of pneumothorax, and more rapid stoma maturation, does seem to result in a higher rate of TCF. Although we do not use the starplasty technique in our institution, we do mature the stoma using 3 or 4 stitches at the corners of the tracheostomy incision. This method is similar to that described by Simons and colleagues and that has been demonstrated by this group not to lead to an increased rate of TCF. Thus, the large number of children seen with TCF in our center is more likely a reflection of referral patterns and the high number of complex airways requiring tracheotomy at our center rather than a higher rate of TCF secondary to stoma maturation.

Conclusion
Primary and secondary means of TCF closure appear equally effective. Complication rates appear similar between the 2 methods, given the caveat that the study was underpowered. Although we did not find any life-threatening complications of primary TCF closure within our study period, we present 3 such events prior to our study period and list examples documented in the literature. Even careful patient selection does not seem to safeguard against these potentially catastrophic events. Given equivalent rates of efficacy, we feel that the potential risks of primary closure outweigh the benefit of avoiding scar revision, and we therefore have adopted a uniform approach of TCF closure by secondary intention. The information presented in the current study should guide the surgeon, patient, and family in assessing risk and in making the decision as to which procedure is appropriate.

Acknowledgment
The authors wish to thank Cynthia Fitton, ANP for her assistance in retrieving the medical records of those patients treated prior to the study period.

Author Contributions
Alexander J. Osborn, concept and design, acquisition of data, analysis and interpretation of data, drafting the article, final approval; Alessandro de Alarcón, concept and design, revision for intellectual content, final approval; Catherine K. Hart, concept and design, revision for intellectual content, final approval; Robin T. Cotton, concept and design, revision for intellectual content, final approval; Michael J. Rutter, concept and design, revision for intellectual content, final approval.

Disclosures
Competing interests: Michael J. Rutter is a former member of the Scientific Advisory Board of Acclarent and is an unpaid consultant for Boston Medical Products, Hood Laboratories, Bryan Medical, and Karl Storz. He receives trivial royalties from Gyrus/Olympus. These relationships do not influence the interpretation or presentation of findings in this article as none of the products from any of these companies are represented or promoted therein.

Sponsorships: None.

Funding source: None.

References


