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Tracheocutaneous fistula closure in children

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ABSTRACT

Objective: Systematic review of surgical techniques for tracheocutaneous fistula closure in children and successful closure or development of adverse events.
Data sources: PubMed, EMBASE, Web of Science, and Cochrane Library.
Review methods: A medical librarian was utilized to perform a systematic review.
Results: Fourteen studies were identified. Eight studies reported outcomes for primary closure alone; one discussed results for secondary closure alone; and five included a combination of children who underwent either primary and secondary closure alone. No difference between surgical techniques was identified for effectiveness of closure (RR = 1.03, 95% CI: 0.97 to 1.10) and major (RR = 1.68, 95% CI: 0.56 to 5.05) or minor complications (RR = 1.28, 95% CI: 0.50 to 3.27). No mortalities were identified.
Conclusions: Both primary and secondary closure techniques for persistent tracheocutaneous fistulas in children are effective and associated with acceptable rates of complications. Given the available evidence, neither approach appears to be superior. Considerations for decision making in surgical approach may include family and social considerations, facility resource utilization, and cost differential. Further investigation may be directed at improving patient selection.

1. Introduction

Tracheocutaneous fistula formation following decannulation in children with a previous tracheotomy is a well recognized issue, affecting approximately one-fourth of children with tracheotomies in place for over a year [1]. With a shift towards tracheotomies for chronic diseases such as prolonged respiratory failure and congenital malformation from that of acute respiratory tract infections, the tracheocutaneous fistula has become a more common entity [2–4]. Mucocutaneous overgrowth and persistent squamous epithelialization preventing closure of the artificial lumen at the stoma site is typically described [5]. The persistence of a fistula can be burdensome for both patient and family alike, including issues with poor hygiene, aspiration, and cosmesis [6]. Although decannulation typically results in complete closure, the rate of a persistent tracheocutaneous fistula has been reported to range from 6.2% to 37.1% in children [7–10].

The surgical management of tracheocutaneous fistulas has been debated in the literature. Primary closure includes excision of the fistulous tract and multilayered closure versus a secondary closure, which allows healing by secondary intention following tract excision [11,12]. Proponents of secondary closure believe that secondary closure decreases the likelihood of serious complications such as subcutaneous emphysema and pneumomediastinum, while other studies have verified the safety profile of primary closure and support the immediate resolution the procedure provides [7,13]. Despite relatively strong opinions regarding both surgical techniques, a systematic review of the literature has yet to be performed. We performed a systematic review examining tracheocutaneous fistula repair in children, utilizing either primary or secondary closure techniques, and an assessment of successful closure or development of adverse events.

2. Methods

Using a medical librarian's services, a query of the PubMed, EMBASE, Web of Science, and Cochrane databases for studies

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examining the results of tracheocutaneous fistula closure in children (aged 0–17 years) was undertaken (Appendix 1). Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines and checklist were utilized. Titles and abstracts were reviewed by two independent reviewers for appropriate inclusion criteria including: 1) original study reporting individual or aggregated patient outcomes; 2) children with a tracheocutaneous fistula resulting from a prior tracheotomy; 3) studies reporting primary closure defined as excision of tracheocutaneous fistula tract or fistulectomy with layered primary closure; 4) studies reporting secondary closure defined as excision of tracheocutaneous fistula tract or fistulectomy with closure by secondary intention; 5) closure technique utilizing intervention in the operative theater; and 6) results of tracheocutaneous fistula closure were reported, either successful closure or adverse events.

Studies published from all available years in English were considered. Exclusion criteria included: 1) case reports containing 5 subjects or less; 2) case series that included adult patients (aged > 18 years); and 3) results reported in non-human subjects. Also, studies that did not obviously fulfill inclusion criteria, including review articles, basic science studies, as well as unpublished abstracts and expert opinion were also excluded. The remaining articles underwent full-length review and data was extracted for individual-level analysis (Fig. 1). References were reviewed for further potentially applicable studies. No contact with the authors was performed given the retrospective nature of the reports. Quality assessment was also performed for each of the included studies reporting adverse events according to the tool developed by Chou and Helfand for Agency for Healthcare Research and Quality (AHRQ) systematic review [14] (Appendix 2).

There were 5 studies that reported outcomes for both primary and secondary closures and appropriate for meta-analysis. Analysis was then performed to examine for any association between successful closure outcomes, major, and minor complication rates between primary closure and secondary intention techniques.

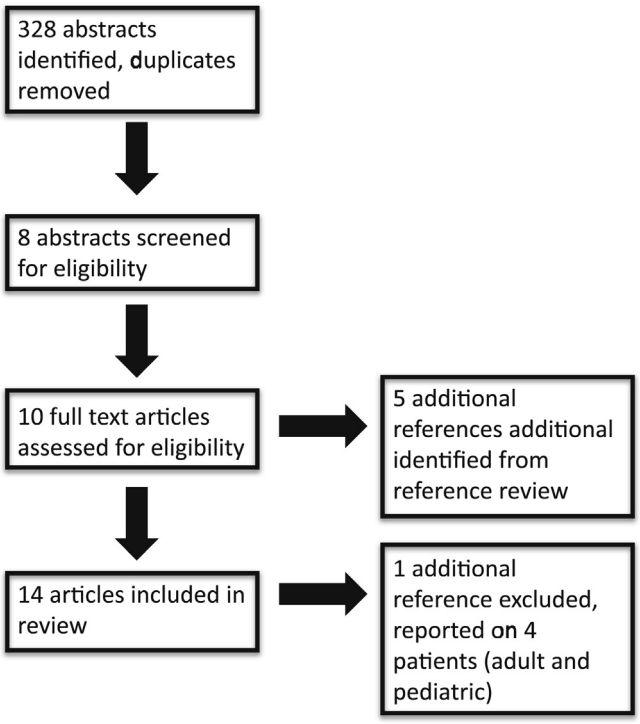

Fig. 1. Flow diagram for study inclusion.

Successful closure was defined as adequate cosmesis postoperatively and closure without undergoing a secondary procedure. Only operation specific complications were included. Major complications were defined as mortalities, subcutaneous emphysema requiring intervention, re-intubation or respiratory compromise requiring replacement of tracheotomy tube, or any other incident that required further surgical intervention. Minor complications included postoperative, superficial wound infections managed with oral antibiotics, airway granuloma, minor bleeding air leak, supplemental oxygen requirements without the need for invasive or non-invasive ventilator support, stridor or increased work of breathing, or other wound issues that were managed conservatively.

For these studies, a meta-analysis using the Mantel-Haenszel method for calculating effect estimates was conducted. Fixed effect models and pooled estimates of the relative risk (RR) were used. Heterogeneity across studies was evaluated with the Q statistic, which uses a chi-square test to assess heterogeneity. The degree of heterogeneity was reported using the I^2 statistic (Higgins) [15,16]. A sensitivity analysis excluding the largest study was conducted to assure that the findings were not unduly influenced by a single study. Due to the relatively small number of studies, a funnel plot was not able to be used to assess publication bias.

All statistical analysis was conducted in R version 3.1.0 (The R Foundation for Statistical Computing, 2014). Confidence intervals were reported at 95% unless otherwise specified.

3. Results

Three hundred and twenty-eight titles and abstracts were reviewed after applying our literature search strategy and removing duplicates. Fourteen articles, including other applicable studies identified by reviewing article references, were identified as appropriate for inclusion and underwent full length review (Table 1). Eight studies reported outcomes for primary closure alone; one discussed results for secondary closure alone; and five included a combination of children who underwent either primary and secondary closure alone (Tables 2 and 3). All were observational and retrospective case series and contained no comparison or control groups. One study did not report any successful closure outcomes or adverse events. No mortalities were identified. Overall reported rates of successful closure ranged between 85 and 100%.

Analysis of the 5 studies which contained both primary and secondary closure outcomes reported revealed no significant association between closure type and successful closure, RR = 1.03 [0.97 to 1.10] (Fig. 2). There was minimal heterogeneity, $\tau^2 = 0$, between-study variance (Q = 1.55, P < 0.8185). The percentage of variability attributable to heterogeneity was 0% ($I^2 = 0\%$). No significant association was found between closure type and major complications, RR = 1.68 [0.56 to 5.05] (Fig. 3). There was minimal heterogeneity, $\tau^2 = 0.3476$, between-study variance (Q = 4.63, P < 0.3271). The percentage of variability attributable to heterogeneity was 13.7% ($I^2 = 13.7\%$). The analysis for minor complications only included 3 studies, as 2 of the studies had 0 observed minor complications in both the primary and secondary closure groups. No significant association was found between closure type and minor complications, RR = 1.28 [0.50 to 3.27] (Fig. 4). There was minimal heterogeneity, $\tau^2 = 0$, between-study variance (Q = 0.82, P < 0.6626). The percentage of variability attributable to heterogeneity was 0% ($I^2 = 0\%$).

There were no obvious outliers, as all results were consistent. All 95% CI of RR included 1, indicating non-significant findings. We did analyze the data after removing the largest study to examine if it had undue influence (Osborn n = 216); the results for all outcomes were unchanged. Thus, the sensitivity analysis supports the use of all five studies in the meta-analysis.