

# Systematic review of surgery for persistent pediatric tracheocutaneous fistula (TCF)

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## ABSTRACT

**Title:** Systematic review of surgery for persistent pediatric tracheocutaneous fistula (TCF)

**Objective:** To compare the treatment outcomes & adverse event rates for primary closure compared to simple excision with healing by secondary intention for persistent TCF in children.

**Method:** Systematic review using PRISMA reporting standards. MEDLINE, EMBASE, Cochrane library, & "Reference" sections were used to identify articles. Inclusion criteria were case series or comparative studies of surgery for persistent TCF in children under age 18 years. Exclusion criteria were age 18 years or older, duplicate patient series, case series less than 2 patients, or case reports. Data were pooled using random effects meta-analysis to estimate incidence rates & comparative effectiveness.

**Results:** The initial data search identified 104 articles, 20 were included in full text review, 14 in the final data set after eligibility assessment by two independent reviewers. There were 413 patients treated with excision with primary closure and 233 patients treated with excision with closure by secondary intention. Pooled success rates were 95.7% with primary closure (95% CI, 93.1-97.4%) & 92.7% with secondary intention (95%, 88.4-95.4%). Subcutaneous emphysema or urgent airway problems were uncommon, occurring in 3.8 & 3.6% of patients, respectively. Five studies had data suitable for comparative meta-analysis, which showed no differences by closure technique for treatment success ( $p=0.480$ ), overall complications ( $p=0.551$ ), need for revision surgery ( $p=0.624$ ), or the incidence of subcutaneous emphysema or pneumothorax ( $p=0.512$ ), urgent airway problems ( $p=0.126$ ), wound infection ( $p=1.00$ ), or wound dehiscence or fistula ( $p=0.818$ ).

**Conclusion:** There are no differences in the rates of success or complications with surgical closure of TCF in children, whether performed by primary closure or by secondary intention. Both techniques have high success rates with a low incidence of serious complications.

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## INTRODUCTION

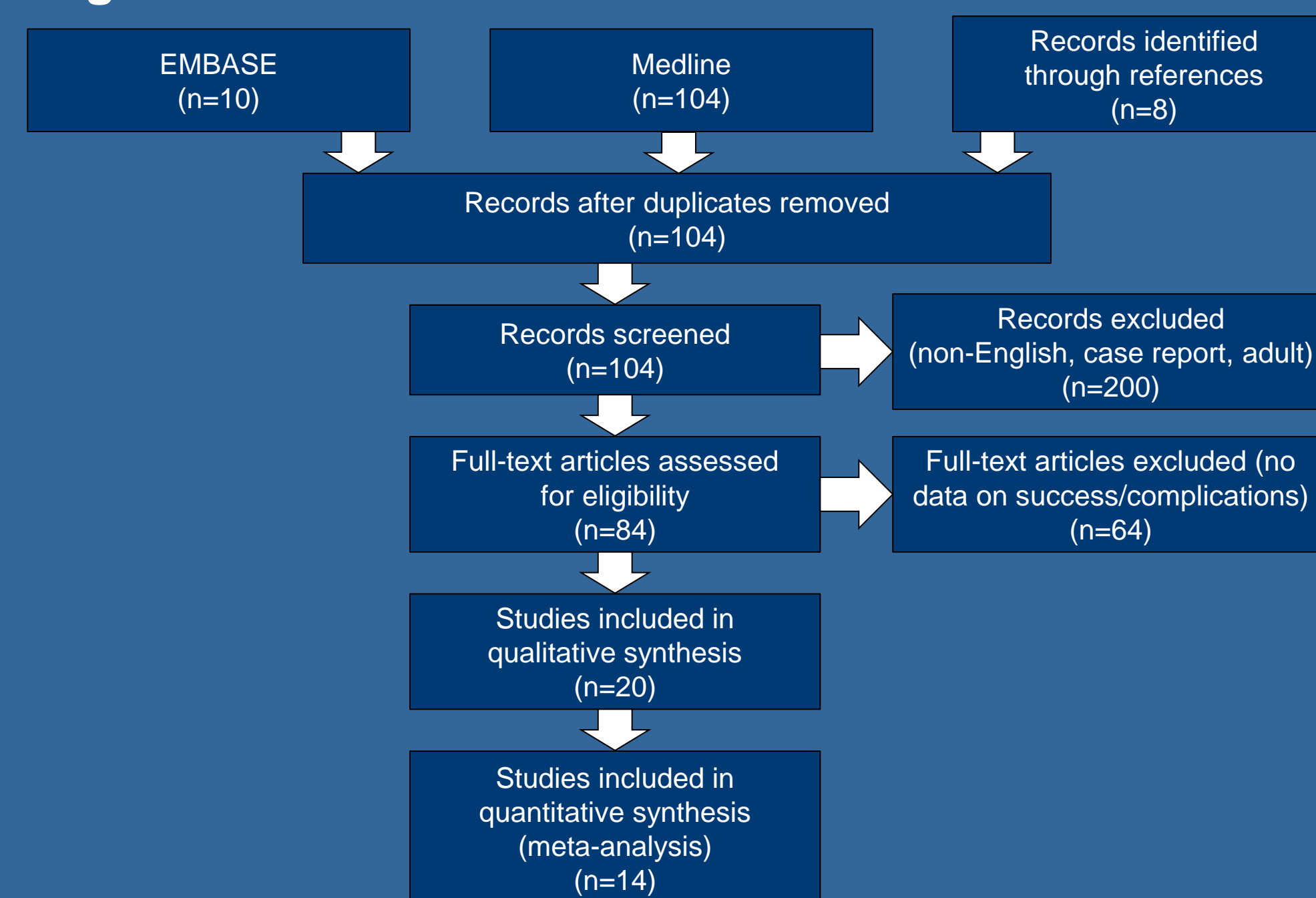
Tracheocutaneous fistula is a sequela seen in 1-3% of patients with previous tracheostomy. In majority of cases, the site of tracheostomy heals by secondary intention upon removal of the cannula.

The fistula can result in "persistent secretions with skin irritation, disturbed phonation, frequent infections, and poor cosmesis [1]." This may be an indication for closure and revision.

The most commonly performed surgical procedures to close the TCF are excision with primary closure in layers and excision with healing by secondary intention. Although majority of closures are successful without complication, there may be devastating complications after surgical closure for persistent fistulas. These range from infection and wound dehiscence, to subcutaneous emphysema, pneumomediastinum/pneumothorax, need for urgent intubation or recannulation.

This review seeks to determine which procedure, excision with primary closure or excision with secondary intention, is more successful and has less complications and adverse events.

Figure 1: PRISMA flowchart



## METHODS AND MATERIALS

An a priori protocol was established & was reported using PRISMA statement [2] (Fig 1). MEDLINE & EMBASE were used to access studies. Quality was assessed using MINORS Criteria [3] & random effects meta-analysis [4] was used for statistical analysis.

### Inclusion criteria:

Age < 18 years old; Data on surgical treatment of tracheocutaneous fistula & success and complications of surgical treatment of TCF

### Exclusion criteria:

Adults, duplicate patient series, case report

**Study characteristics:** year of publication, location, study design, inclusion criteria, exclusion criteria

**Population:** age range, gender, age of repair of TCF, age at tracheostomy, gestational age, length of cannulation, age at decannulation, time elapsed from decannulation to TCF repair, length of hospital stay

**Intervention:** surgical treatment of excision of TCF with primary closure with or without drain, surgical treatment of excision of TCF with closure by secondary intention

**Comparison:** comparing the different surgical treatment procedures

**Outcomes:** primary endpoint was "success" of closure reported by authors, rate of revision surgery, pneumothorax/emphysema, need for urgent airway; secondary endpoints were rate of complications such as infection, dehiscence, etc.

Table 1: Included Studies.

Publication	Year	Location	Cohort Study	# of Pts w/ Primary Closure	# of Pts w/ Secondary Closure	MINORS Score
Wine et al	2014	USA	Yes	30	22	20
Osborn et al	2013	USA	Yes	46	170	20
Priestly et al	2006	Australia	Yes	15	1	19
Stern et al	1998	Israel/USA	Yes	80	18	21
White et al	1988	Canada	Yes	6	9	14
Tasca et al	2010	UK	No	23	0	10
Schroeder et al	2008	USA	No	39	0	12
Geyer et al	2008	UK	No	100	0	12
Sautter et al	2005	USA	No	13	0	10
Eaton et al	2003	USA	No	0	13	11
Licameli et al	1997	USA	No	9	0	11
Haynes et al	1995	USA	No	12	0	10
Bressler et al	1994	USA	No	36	0	10
Goldsmith et al	1993	USA	No	4	0	11

## RESULTS

The pooled incidence for "success" was 95.7% (93.1-97.4) for primary closure and 92.7% (88.4%-95.4%) for closure by secondary intention, there was no statistical difference ( $p=0.480$ ).

For all complications, there was no difference ( $p=0.551$ ) between primary closure and secondary closure, 10% (6.3-17.0%) and 9.9% (6.6-14.5%).

The rates of revision surgery: 5.2% (2.5-10.3%) vs. 11.9 (6.9-19.9%), ( $p=0.624$ ); emphysema/pneumothorax: 3.8%(2.1-6.6%) vs. 3.8% (0.7-18.3%), ( $p=0.512$ ); and urgent airway: 3.5% (1.8-6.4%) vs. 3.6% (1.7-7.7%), ( $p=0.126$ ), were no different.

There was no difference between the rates of infection and dehiscence; 5.1% (3.0-8.3%) vs. 0.0% (-5.5%), ( $p=1.000$ ); 3.6% (2.0-6.4%) vs. 6.2% (3.7-10.3%), ( $p=0.818$ ) (Table 2, 3).

MINORS criteria to assess bias showed the range for non-comparative studies was 10-12, with a mean of 10.8. The range of comparative studies was 14-21, with a mean of 18.8.

Table 2: Comparison of outcomes and complications for primary closure vs. secondary intention

Outcome	N <sup>‡</sup>	Primary closure		Secondary intention		
		Pooled incidence, % (95% CI)*	I-sq <sup>†</sup>	N <sup>‡</sup>	Pooled incidence, % (95% CI)*	I <sup>‡</sup>
Success	13	95.7 (93.1 – 97.4)	0	5	92.7 (88.4 – 95.4)	0
Complications	13	10.5 (6.3 – 17.0)	47	5	9.9 (6.6 – 14.5)	0
Dehiscence or fistula	11	3.6 (2.0 – 6.4)	0	4	6.2 (3.7 – 10.3)	0
Infection	11	5.1 (3.0 – 8.3)	0	2	0 (0 – 5.5)	0
Revision surgery	11	5.2 (2.5 – 10.3)	38	4	11.9 (6.9 – 19.9)	10
Emphysema or pneumothorax	11	3.8 (2.1 – 6.6)	0	3	3.8 (0.7 – 18.3)	64
Urgent airway problem	11	3.5 (1.8 – 6.4)	21	3	3.6 (1.7 – 7.7)	20

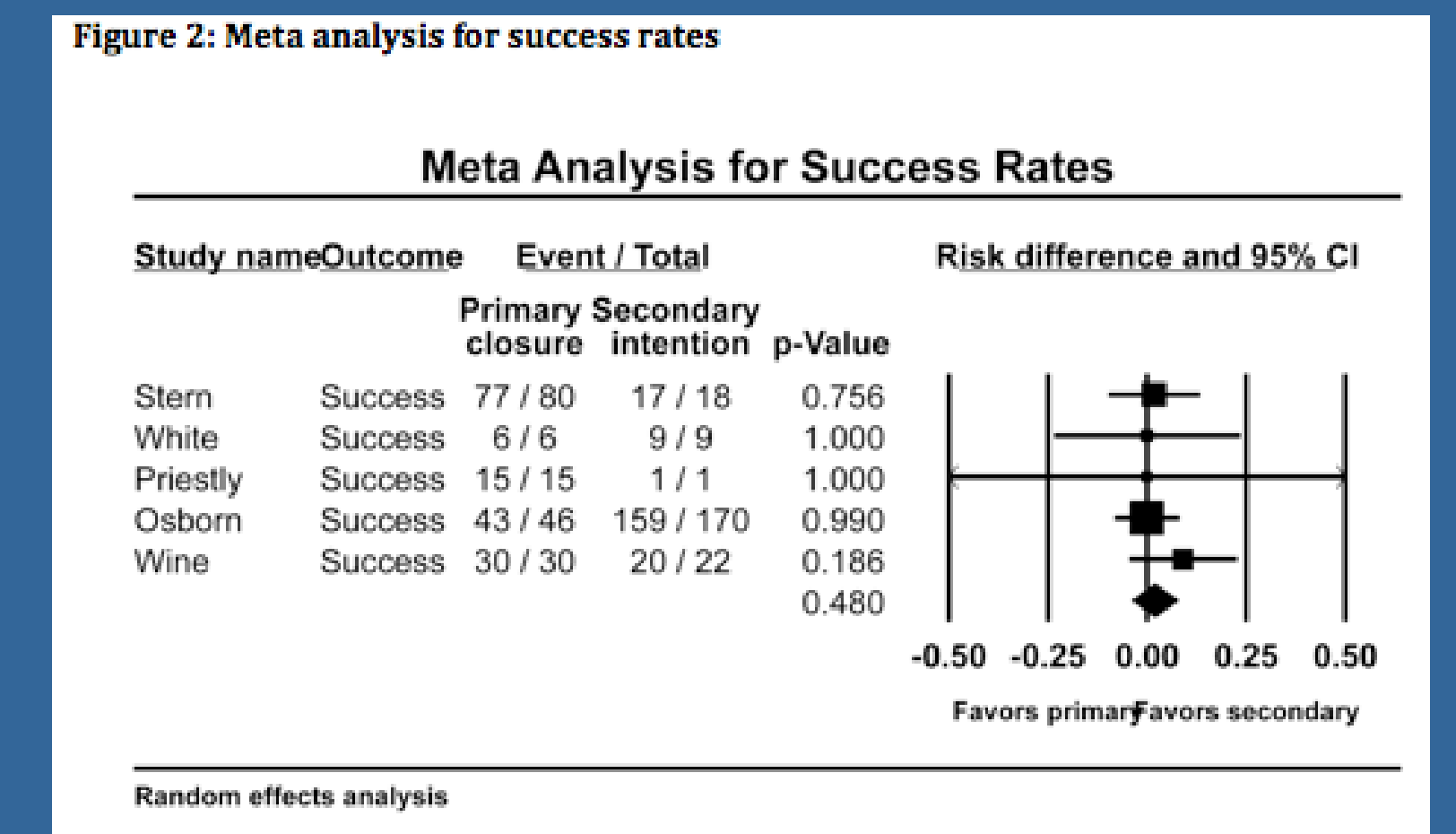
<sup>‡</sup>Number of studies contributing to the analysis

\*Random effects meta-analysis

<sup>†</sup>I-squared of 25% is low heterogeneity, 50% moderate heterogeneity, and 75% high heterogeneity

## REFERENCES

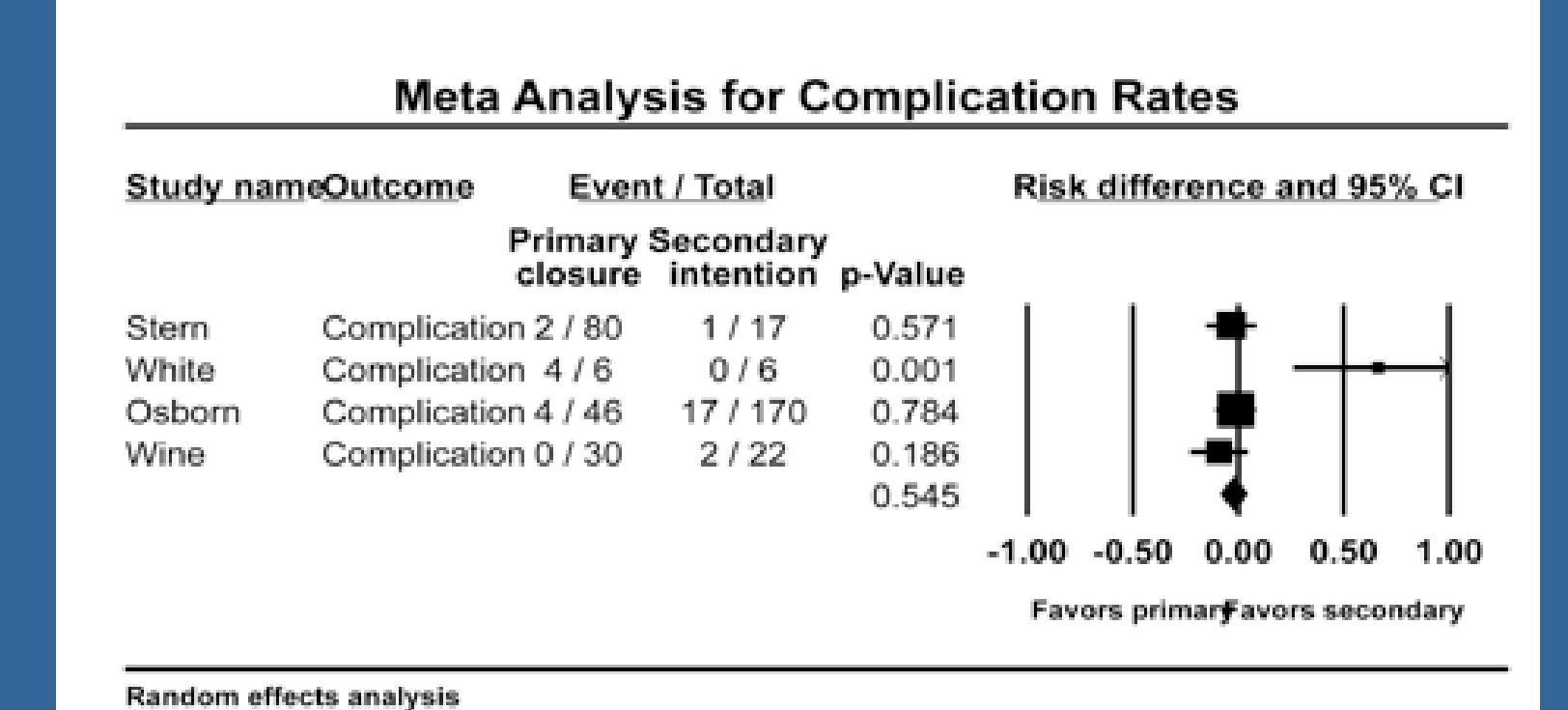
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## DISCUSSION

We did not find a difference between primary closure and secondary closure in urgent airway, emphysema/pneumothorax. We also did not find a difference between revision surgery rates. Using the random effects modeling, there were insufficient patients within the cohort studies to analyze revision surgery rates and urgent airway/emphysema complications. There were sufficient patients to look at "success" as well as all complications, and we found there to be no difference. We did not include an outcome of aesthetic appearance, nor could we extract data on specific reasons for revision surgery. We cannot conclude there is a difference in cosmesis or scarring from the two surgical procedures.

Figure 3: Meta-analysis for complication rates



## CONCLUSIONS

We have found, there is no difference between the two main surgical options for all complications, including "devastating complications," such as need for urgent airway or emphysema. Therefore, surgeon experience and patient preference will play a large role in which surgical option is performed.

Table 3: Meta-analysis of primary closure vs. secondary intention

Outcome	N <sup>‡</sup>	Rate difference, % (95% CI)*	P-value <sup>†</sup>	I-sq <sup>†</sup>
Success	5	2.1 (-3.6 – 7.7)	0.480	0
Complications	4	2.4 (-1.2 – 1.7)	0.551	75
Dehiscence or fistula	3	-0.1 (-5.5 – 4.3)	0.818	0
Infection	2	0 (-2.8 – 2.8)	1.000	0
Revision surgery	3	-2.4 (-7.6 – 2.8)	0.624	0
Emphysema or pneumothorax	2	-2.2 (-8.8 – 4.3)	0.512	31
Urgent airway problem	2	-2.8 (-6.5 – 0.1)	0.126	0

<sup>‡</sup>Number of studies contributing to the analysis

\*Absolute difference between groups; positive values favor primary closure for success rates; negative values favor primary closure for all other outcomes

<sup>†</sup>I-squared of 25% is low heterogeneity, 50% moderate heterogeneity, and 75% high heterogeneity