**Endoscopic Balloon Dilation of Pediatric Subglottic Stenosis: A Meta-Analysis**

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**ABSTRACT**

Objective: Systematically review the currently available evidence regarding the use of Endoscopic balloon dilation (EBD) to manage pediatric subglottic stenosis that could preclude the need for tracheostomy and/or laryngo-tracheal reconstruction (LTR).

Data Sources: MEDLINE, EMBASE, and the Cochrane databases

Review Methods: A systematic review of pediatric EBD was performed and then reported in compliance with PRISMA principles. Inclusion criteria consisted of sample size of five or greater, pediatric patients, and primary EBD without adjuvant procedures. Meta-analysis was performed with random effects modeling and pooled data regression.

Results: Seven studies were included in the final dataset with 150 total subjects. All studies were case series (level 4 evidence). The mean sample size was 20 subjects (range=5–44) and the grand mean age was 2.2 years (range=2–60 months). The estimate of the overall treatment success was 65.3% (k=6 studies, 95% CI=60.1-70.6%, p<0.001, Q test for heterogeneity=3.98, p=0.552, I squared=0%). Follow-up was inconsistently reported but averaged 4.6 months (range=0.25–12.5 months). Only one study reported significant complications (one death, two tracheal lacerations). Pooled data multivariate regression indicated increasing Cotton-Myers grade was associated with decreased odds of success (OR=0.198, 95% CI=0.045-0.870, p=0.032).

Conclusions: EBD is successful in the majority of patients over short-term follow-up. The reported complication rates are low. Increasing severity of SGS increases odds of treatment failure.

**INTRODUCTION**

Endoscopic balloon dilation (EBD) of pediatric subglottic stenosis has the potential to provide a simple and non-invasive treatment that could effectively manage many cases of pediatric SGS. However, the outcomes of pediatric EBD, the determination of which patients are best suited for the procedure, and its limitations and complications are not well known.

The primary aim of this study was to systematically identify all relevant published data regarding the use of primary EBD alone for the management of pediatric subglottic stenosis, critically evaluate the success of the technique, clarify which patients are the best candidates for the procedure, and estimate the complication rate for the procedure (to include the need for a repeat dilation procedure).

**METHODS AND MATERIALS**

The MEDLINE, EMBASE, and Cochrane databases were systematically searched in January 2013 using multiple search terms with the aid of a biomedical librarian. Inclusion criteria consisted of (1) sample size of 5 or greater (2) use of EBD for pediatric patients (0-18 years) (3) use of EBD as the primary treatment of pediatric subglottic stenosis to avoid more definitive airway management to include tracheostomy and/or laryngo-tracheal reconstruction (LTR).

The primary outcome measure was treatment success (%) defined as the avoidance of more invasive procedures such as tracheostomy and/or laryngo-tracheal reconstruction (LTR). Secondary outcome measures of need for revision EBD and complications were also recorded. Effect modification by age and the severity of subglottic stenosis as measured by the Cotton-Myers grade was also assessed. The data were extracted by two reviewers independently with a third reviewer used to settle any discrepancies if needed.

Random effects modeling (standard error flowchart (PRISMA format)) was used to calculate summary effect measures. The I² statistic was used to assess between-study heterogeneity. Data were pooled to perform logistic regression to assess the effects of age, number of dilations, and Cotton-Myers grade. Possible publication bias was assessed using graphical funnel plot analysis. Duval and Tweedie “Trim and Fill” methods were then used to estimate the effects of possible publication bias.

**RESULTS**

Literature search results are shown in Figure 1. The final study group included seven studies. Only one study reported complications which included atelectasis (3 patients), tracheitis (2 patients), pneumomediastinum (asymptomatic, 1 patient), tracheal laceration (2 patients), and death (1 patient, from tracheal laceration).

The summary estimate of primary treatment success with EBD of pediatric SGS was 65.3% (k=6 studies, 95% CI=60.1-70.6%, p<0.001, I squared=0%). Use of EBD as a secondary therapy following either tracheotomy or LTR was very similar at 61.2% (k=3 studies, 95% CI=44.5-78.0%, p<0.001, I squared=88%).

Publication bias was assessed with graphical funnel plot analysis. Visual inspection suggests a possibility of publication bias with an absence of studies reporting lower success rates of EBD. The Duval and Tweedie nonparametric “trim and fill” method of “filled” summary estimate was 64.2%, 3 studies “filled”, 95% CI = 59.1-69.2%) suggested the statistical effects of publication bias may be negligible.

Graphical evaluation of the trends of the pooled raw data did suggest that a lower Cotton-Myers Stage had a higher treatment success rate. Multivariate logistic regression showed increasing Cotton-Myers grade was associated with decreased odds of treatment success (OR=0.198, 95% CI=0.045-0.870, p=0.032).

**DISCUSSION**

EBD as a primary treatment to prevent the need for tracheostomy and/or LTR was successful in approximately two thirds of patients over follow-up of approximately 4 months. The use of EBD as a secondary treatment after tracheotomy and/or LTR was also reported to be successful in approximately two thirds of patients. Complications were rarely reported but were potentially severe including one death due to tracheal laceration. Subgroup analysis suggested that increasing severity of subglottic stenosis was associated with increasing odds of treatment failure.

There are several important limitations to consider in the interpretation of the results of this study. As with any meta-analysis this study is limited by the heterogeneity of the evidence table (Table 1) shows, there were several methodological differences between the studies. An additional weakness is that all the included studies were case series. Case series do not contain a control group and can be prone to selection bias and confounding. Nonetheless, EBD is unquestionably simpler and less invasive than tracheostomy and LTR to which it might be compared. As a result, any measurable success of EBD can still be considered important and useful.

Lastly, funnel plot analysis indicated the possibility of publication bias was present. Specifically, there were no published studies that reported a low success rate with EBD. Combining this with the finding that only one study reported severe complications with EBD raises reasonable concern that the negative effects of EBD have not yet been reported. This is a legitimate concern that will need to be addressed with future studies.

**CONCLUSIONS**

The use of endoscopic balloon dilation as a primary treatment of pediatric subglottic stenosis is successful in the majority of patients with short-term follow-up. Reported complications are rare but can be severe and life threatening. Increasing severity of subglottic stenosis may be associated with increasing odds of treatment failure. Age does not appear to be predictive of treatment outcomes.

**REFERENCES**