OBJECTIVE: The study goal was to determine the impact of various surgical procedures for bilateral vocal cord paralysis in children by using established principles of meta-analysis.

STUDY DESIGN AND SETTING: We conducted a retrospective review of the literature in which a predetermined protocol was used to identify articles for meta-analysis. Six articles met inclusion criteria, and pertinent data were extracted.

RESULTS: Pooled data analysis demonstrated primary procedure-specific decannulation rates for external arytenoidopexy for 19 of 24 (79%), external arytenoidectomy for 14 of 19 (74%), CO₂ laser arytenoidectomy for 4 of 10 (40%), and costal cartilage graft procedures for 2 of 2 (100%). External arytenoid procedures are more efficacious than CO₂ ablative procedures ($P = 0.02$).

CONCLUSION: Meta-analysis of the existing literature reveals that external arytenoidopexy and external arytenoidectomy are equivalently effective procedures and that the two combined are significantly more effective than CO₂ ablative procedures.

SIGNIFICANCE: External procedures appear to be more effective as a first-line treatment in pediatric vocal cord paralysis, with arytenoidopexy with or without partial arytenoidectomy offering an attrac-
procedure involves the aim of decannulation on the one hand and vocal damage and aspiration on the other hand. Each technique chosen is performed with these risks and benefits in mind.

The initial and oldest treatment of bilateral VCP is a tracheostomy. Definitive surgical interventions are designed to overcome tracheostomy dependence. Multiple approaches have been championed, including a lateral cervical approach and a laryngofissure approach.13,14 External arytenoidectomies were first described in adults by Woodman,13 with subsequent application in children by Priest et al.15 Arytenoidopexy involves either a lateral cervical approach or a midline laryngofissure. In this procedure, lateralization sutures are passed through the vocal process and thyroid ala, thereby lateralizing the vocal cord without mucosal destruction.16 Endoscopic CO₂ laser arytenoidectomy was originally described by Ossoff et al.17,18 and involves ablation of the arytenoid to the level of the cricoid cartilage laterally. Posterior costal cartilage grafting represents a technique described by Cotton19 and Zalzal et al.20 for subglottic stenosis but may be applicable in selected cases of bilateral VCP. Laryngeal reinnervation procedures were pioneered by Tucker21 and involve implanting the ansa hypoglossi into the laryngeal musculature to restore abduction.

Most of the current literature addresses the management of bilateral VCP in adults. There have been attempts to correlate adult data with those in children, but there is a paucity of studies addressing the management of bilateral VCP specifically in children. Rosin et al.7 suggest that bilateral VCP in children should be considered an entirely different entity because of the disparity of etiologies and symptoms between adults and children with bilateral VCP.

Currently, there are no randomized controlled trials that document the increased efficacy of one method of surgical intervention over another. The current literature consists primarily of retrospective reviews of the experiences at individual centers. The results of these retrospective reviews are varied and have failed to produce uniform recommendations. The variability in results and experiences is likely secondary to low statistical power and individual training biases. We performed a systematic review of the published literature in an attempt to address these shortcomings. Although this report is not a traditional meta-analysis involving only randomized, double-blind, placebo-controlled studies,

<table>
<thead>
<tr>
<th>Study No.</th>
<th>First author</th>
<th>Year</th>
<th>Design</th>
<th>Patient age (y)</th>
<th>Treatment</th>
<th>Total No. of patients</th>
<th>Patients with preoperative tracheostomy</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Triglia</td>
<td>1996</td>
<td>Retrospective</td>
<td>Mean, 1.66 Range, 0.08-9</td>
<td>Arytenoidopexy, EL arytenoidectomy</td>
<td>15</td>
<td>12</td>
</tr>
<tr>
<td>2</td>
<td>De Guademer</td>
<td>1996</td>
<td>Retrospective</td>
<td>Mean, 3.8 Range, 1.4-14</td>
<td>Arytenoidectomy (laser), arytenoidopexy</td>
<td>7</td>
<td>2</td>
</tr>
<tr>
<td>3</td>
<td>Bower</td>
<td>1994</td>
<td>Retrospective</td>
<td>Mean, 7.3 Range, 1.5-18</td>
<td>Arytenoidectomy (EL), arytenoidectomy (laser), arytenoidectomy (Woodman)</td>
<td>30</td>
<td>29</td>
</tr>
<tr>
<td>4</td>
<td>Gray</td>
<td>1994</td>
<td>Retrospective</td>
<td>Mean, 4.1 Range, 1.9-7</td>
<td>Posterior glottic graft</td>
<td>3</td>
<td>2</td>
</tr>
<tr>
<td>5</td>
<td>Narcy</td>
<td>1995</td>
<td>Retrospective</td>
<td>NA</td>
<td>Arytenoidectomy, arytenoidectomy (laser)</td>
<td>7</td>
<td>4</td>
</tr>
<tr>
<td>6</td>
<td>Narcy</td>
<td>1990</td>
<td>Retrospective</td>
<td>Mean, 3.4 Range, 0.5-14</td>
<td>Arytenoidectomy, arytenoidectomy (Woodman)</td>
<td>16</td>
<td>6</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td><strong>78</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td><strong>55</strong></td>
<td></td>
</tr>
</tbody>
</table>

EL = External laryngofissure.
we used the principles governing meta-analysis to direct our review. Our aim was to use a systematic protocol to identify methodologically comparable retrospective studies that allowed statistical pooling of results to clarify the efficacy of the different surgical interventions for bilateral VCP. Our final objective was to provide an orderly review of the current literature to describe and report results of the various surgical interventions in a way that they might be directly compared.

**METHODS**

This review was undertaken using a systematic protocol based on the established principles of meta-analysis. Although randomized controlled trial data were not available for this project, we adhered to the principles governing meta-analysis. Namely, an a priori protocol was devised to conduct a systematic review of the literature with a strict focus to permit comparison of the outcomes of current surgical interventions for pediatric bilateral VCP.

The goal of our literature search was to identify relevant articles and to select those that could be statistically combined for comparison. Our literature search was conducted in systematic fashion using MEDLINE search databases, textbook chapters, and relevant review articles. After an initial review of textbooks and of the literature, selection criteria were formulated. The predetermined selection criteria to identify candidate articles were as follows. Each article had to (1) present a surgical intervention for bilateral VCP involving children under the age of 18, (2) discuss the management of pediatric bilateral VCP for children who previously underwent a tracheotomy, (3) clearly present the details of the preoperative, intraoperative, and postoperative course for each case, (4) be published after 1985, and (5) be an English-language article. Publication after 1985 was chosen as a criterion to reflect current practices as the use of flexible laryngoscopes for diagnosis and of the CO₂ laser became widespread. Articles that did not meet these criteria were excluded. A final exclusion criterion involved articles that referred to laryngeal reinnervation procedures as first-line treatment for pediatric bilateral VCP. Because reinnervation procedures remain controversial and outside the scope of conventional treatment alternatives for pediatric bilateral VCP, these articles were excluded from the study.

Source articles were initially obtained using a computerized MEDLINE literature search (1985 to 2000). The initial search involved creating a data set using “vocal cord paralysis” and associated medical subject heading (MeSH) terms as the subject combined with the MeSH term “surgery” as the subject. This yielded a database of 85 candidate articles, of which the titles and abstracts were read to determine the content of the article. If the information extracted from the title and abstract left any possibility of being a candidate article, the article was obtained and read to ensure that it met the predetermined inclusion criteria. Common exclusion criteria included primary focus of management of VCP in adults, nondefinitive surgical treatment (ie, tracheostomy only), unilateral VCP, and incomplete discussion of surgical details.

The references of the candidate articles were reviewed, and manual crosschecks of the references were performed. This elicited 4 additional articles that were not discovered on the initial literature search. To ensure that all applicable literature had been searched, manual reference crosschecks with current textbook chapters as well as additional variations of MEDLINE searches using the MeSH terms “pediatric,” “children,” “bilateral,” and “vocal cord paralysis” with the subheading “surgery” were performed. Of the articles reviewed, 6 met the predetermined inclusion criteria, and they compose the final data set.

Data extraction from the articles included general characteristics of the study such as design, surgical intervention, age at intervention, total number of patients, total number of operations, and preoperative tracheostomy dependence. In

### Table 2. Primary procedure decannulation rate

<table>
<thead>
<tr>
<th>Study No.</th>
<th>Operation</th>
<th>No. decannulated</th>
</tr>
</thead>
<tbody>
<tr>
<td>1, 5, 6</td>
<td>Arytenoidopexy</td>
<td>19/24 (79%)</td>
</tr>
<tr>
<td>2, 3, 6</td>
<td>External arytenoidectomy</td>
<td>14/19 (74%)</td>
</tr>
<tr>
<td>3</td>
<td>CO₂ laser arytenoidectomy</td>
<td>4/10 (40%)</td>
</tr>
<tr>
<td>4</td>
<td>Posterior cartilage graft</td>
<td>2/2 (100%)</td>
</tr>
</tbody>
</table>
addition, operative course, postoperative outcomes, and complications were extracted. Patients were grouped into subsets of preoperative tracheostomy dependence and nondependence.

Outcome criteria were defined by the end points presented in the articles. For patients in whom a preoperative tracheostomy was required, it was generally accepted that decannulation was the goal of treatment. This led to the calculation of operation-specific decannulation rates as a quantitative outcome measure that allowed numerical combination. We believed that a decannulation rate based on the outcome of a primary procedure alone allowed the data to be as homogeneous as possible. By specifically addressing the results of primary procedures alone, we believed that could eliminate the variability introduced by revision procedures. We believe that this numerical rate is an important tool and the strongest comparison in our meta-analysis.

Review of the pertinent articles revealed a subset of children without tracheostomies who underwent surgical procedures aimed at improving exertional dyspnea and other measures of health-related quality of life. Because these outcome measures were difficult to quantify for standard comparative purposes, this population was excluded from the study.

Data Analysis

Under the assumption of a power value of 0.9, an \( \alpha \) value of 0.05, and intergroup operation-specific decannulation rates that differed by 25%, an estimated sample size of 162 patients was calculated to be required. There are no accurate historic data on which to accurately define the intergroup differences. The data were then pooled, and discrete variables were recorded as primary outcome measures. After application of the strict inclusion and exclusion criteria, 55 children's records were suitable for data review. Statistical analysis was performed with \( \chi^2 \) analysis that compared the operation-specific decannulation rates between each procedure.

RESULTS

Studies included in the meta-analysis are summarized in Table 1. Studies were published between 1990 and 1996 in the otolaryngology literature. All studies were retrospective reviews of surgical outcomes in the treatment of bilateral VCP. Sample sizes ranged from 3 to 30 patients. All studies clearly described surgical techniques and outlined patient outcomes on an individual basis. The surgical techniques addressed included external arytenoidopexy,16,24,26,28 external arytenoidectomy,23,24,26 CO\(_2\) laser arytenoidectomy,23,24,26 and arytenoid separation with a posterior costal cartilage graft.25

As described earlier, results were calculated for all children who were tracheostomy dependent before intervention. We believe that our most meaningful data are derived in the calculation of a primary operation-specific decannulation rate. Although the actual number of children included in this study is fewer than the calculated sample size, the sample size calculations were made possible by a set of a priori hypotheses that lack support in the literature (the purpose of this report is to provide baseline information regarding surgical success rates for different operative procedures). The primary procedure decannulation data are presented in Table 2. A total of 55 patients were pooled. Results of \( \chi^2 \) analysis that compared the procedures is presented in Table 3. External arytenoidopexy and external arytenoidectomy were shown to be efficacious procedures, with a statistically similar decannulation rate of 19 of 24 (79%) and 14 of 19 (74%), respectively (\( P = 0.79 \)).
Because these 2 procedures are often performed in the same surgical setting, we elected to further substratify our results to look at external arytenoid procedures compared with endoscopic CO₂ procedures. This comparison demonstrated that external procedures have a significantly higher primary decannulation rate \( (P = 0.02) \). Both of the children who underwent posterior costal cartilage grafting were decannulated. Additional data calculated on this cohort included an overall operation-specific decannulation rate allowing for revision procedures (Table 4). These data correspond to the use of a particular procedure as a final procedure toward a goal of decannulation. The most notable change is the increased efficacy of CO₂ arytenoidectomy from 4 of 10 (40%) as a primary decannulation rate to 7 of 10 (70%) when multiple procedures are considered.

In addition, complications are reported by procedure for all patients (Table 5). The complications were dependent on accurate reporting by the authors and time of follow-up. Such reporting is subject to publication bias, and it is suspected that rates may be higher. The most common complication reported in most articles was dysphonia; unfortunately, it was a subjective complication and not documented according to a quantitative system.

**DISCUSSION**

Pediatric bilateral VCP is often considered a different entity than that in adults. Although patients with bilateral VCP have been known to undergo spontaneous recovery, it is generally agreed that approximately 50% of patients will require surgical intervention. Our study was designed to address the shortcomings of the current literature and to clarify the direction that surgical management of bilateral VCP is currently taking. The current literature is sparse and consists of essentially case reports of the experience at individual centers with particular interventions. We describe a method to compare individual experiences in a meaningful objective fashion through a primary procedure-specific decannulation rate. Although this is not a new concept, we believe that it allows the most objective comparisons possible.

Meta-analysis of the existing literature suggests that “open” procedures seem to be the most effective primary treatment for bilateral VCP in children. Both external arytenoidopexy and external arytenoidectomy demonstrated similar operation-specific decannulation rates. Both lateral and midline approaches have been described; endoscopic techniques may allow midline access without the need to violate the anterior commissure. In terms of interpretation of these results, in the face of equivalent results between arytenoidopexy and arytenoidectomy, our center is now performing a procedure that combines a partial arytenoidectomy (consisting of cutting the arytenoid just posterior to the vocal process) with a suture vocal cord lateralization as a primary operative procedure. We believe that these procedures represent effective, less-destructive, and potentially reversible procedures.

Minimally invasive or endoscopic CO₂ laser arytenoidectomies performed as a primary intervention do not appear to be as efficacious as “open” procedures. It is interesting to note that although only 4 of 10 (40%) patients were decannulated after the primary procedure, 7 of 10 (70%) patients underwent CO₂ laser arytenoidectomy as the final procedure that allowed decannulation. This alludes to a valuable role of such minimally invasive surgery as part of an initial attempt or as a revision surgery with less iatrogenic trauma as might be inflicted with an open procedure. The advantages of endoscopic intervention may justify the decreased efficacy compared with open procedures. It has been postulated that one reason for decreased efficacy of CO₂ laser ablation within the pediatric larynx is the smaller

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**Table 4. Overall decannulation rate allowing for revisions where procedure listed is final procedure**

<table>
<thead>
<tr>
<th>Study No.</th>
<th>Operation</th>
<th>No. Decannulated</th>
</tr>
</thead>
<tbody>
<tr>
<td>1, 5, 6</td>
<td>Arytenoidopexy</td>
<td>19/21 (90%)</td>
</tr>
<tr>
<td>2, 3, 6</td>
<td>External arytenoidectomy</td>
<td>15/19 (79%)</td>
</tr>
<tr>
<td>2, 3, 5</td>
<td>CO₂ laser arytenoidectomy</td>
<td>7/10 (70%)</td>
</tr>
<tr>
<td>4</td>
<td>Posterior cartilage graft</td>
<td>2/2 (100%)</td>
</tr>
<tr>
<td>3</td>
<td>Other airway procedure*</td>
<td>3/3 (100%)</td>
</tr>
</tbody>
</table>

*Refers to laryngotracheoplasty or cricoid split.*
scale of the anatomic landmarks and obstruction of the posterior glottis by a flaccid aryepiglottic fold.\textsuperscript{24} It may be that as the technique of CO\textsubscript{2} ablation evolves, novel strategies for providing postablative mucosal coverage will increase the overall success rate for this technique.

Posterior costal cartilage grafting for bilateral VCP is an attractive option, specifically for difficult revision cases. A current single-institution review is under way at Children’s Hospital Medical Center of Cincinnati, Ohio, to further elucidate the efficacy of this and other procedures for bilateral VCP. The primary fear with this procedure is the possibility of increased aspiration risk. We hope that the current study will address this issue in a quantifiable fashion.

The reporting of complications in articles that address bilateral VCP generally centers around a subjective description of dysphonia, aspiration, or dyspnea on exertion. In the articles presented, few patients underwent formal phonatory evaluation.\textsuperscript{23,25} There is a paucity of literature regarding the normal pediatric voice; literature regarding pathology of the pediatric voice is even more scarce. This is an area that needs much more work. Regarding other aspects of the reporting of complications, it is evident that complications reported on a subjective basis introduce reporting bias, in which different authors may have different thresholds for reporting. In a review in which most authors concentrated on a single type of intervention, the subjective nature makes the comparison of such results increasingly difficult. It is interesting that none of the included studies addressed an objective measure of aspiration and very few cases of clinical aspiration were reported. Most likely, there were cases of subclinical aspiration that went unreported, and long-term follow-up may demonstrate a higher incidence of aspiration symptoms. Dyspnea after decannulation was rarely reported in any of the series that we reviewed.

The effect of surgical intervention in patients who are not tracheotomy dependent is difficult to characterize. However, it is clear that surgical intervention can be used to improve the dyspnea on exertion that often affects patients with bilateral VCP.

Our study is based on established principles of a systematic literature review using a predetermined protocol. Although our study is not a conventional meta-analysis in that randomized controlled trials with clearly combinable quantitative end points are used, we believe that our operation-specific decannulation rate provides a valid means to combine and compare the outcomes of the different surgical interventions. By pooling the data of the current literature, an accurate comparison can be made with greater power than for independent article comparisons.

There are inherent weaknesses involved in the pooling of data as is done in meta-analyses. The most important opportunity for weakness is that meta-analysis relies on the strength of the individual articles reviewed. This deficiency is important in retrospective studies because the author often is already aware of the outcome. Thus, a significant opportunity for publication bias is introduced. Positive outcome studies are more likely to be published than are what may be pertinent negative studies. We believe that our stringent inclusion criteria allowed us to select scientifically sound articles for comparison. The issue of population heterogeneity is central to meta-analysis. In our meta-analysis, a potential weakness is the patient selection bias by individual authors. For example, Bower et al\textsuperscript{23} presented 29 of 30 patients who required preoperative tracheostomy, whereas in the 2 studies presented by Narcy,\textsuperscript{16,26} only 10 of 23 patients required a preoperative tracheostomy. We believe that we overcame this bias by selecting only those patients who required a preoperative tracheostomy for calculation of our primary quantitative result: the operation-specific decannulation rate.

### Table 5. Reported complications by procedure

<table>
<thead>
<tr>
<th>Study No.</th>
<th>Operation</th>
<th>Dysphonia</th>
<th>Aspiration</th>
<th>Dyspnea</th>
<th>Total No. of patients</th>
</tr>
</thead>
<tbody>
<tr>
<td>1, 2, 5, 6</td>
<td>Arytenoidopexy</td>
<td>9 (38%)</td>
<td>1 (4%)</td>
<td>2 (8%)</td>
<td>24</td>
</tr>
<tr>
<td>2, 3, 5, 6</td>
<td>All arytenoidectomy</td>
<td>4 (12%)</td>
<td>2 (6%)</td>
<td>1 (3%)</td>
<td>33</td>
</tr>
<tr>
<td>4</td>
<td>Posterior cartilage graft</td>
<td>2 (100%)</td>
<td>0 (0%)</td>
<td>0 (0%)</td>
<td>2</td>
</tr>
</tbody>
</table>


CONCLUSION

There are several different surgical alternatives designed to address the problem of pediatric bilateral VCP. Reviewing operation-specific decannulation rates is an important method to allow meaningful comparisons between these procedures. Meta-analysis of the existing literature reveals that external arytenoidopexy and external arytenoidectomy are equivalently effective procedures. Given this equivalency, external arytenoidopexy procedures with or without partial arytenoidectomy offer an attractive first-line surgical option. Based on limited available data, there seems to be a trend toward a higher decannulation rate in tracheotomized children with bilateral VCP who undergo these forms of open surgical correction. Posterior cartilage grafting may have a role in difficult revision cases. CO₂ laser ablation clearly has a decreased primary operation-specific decannulation rate; however, these procedures remain attractive, because they are endoscopic in nature and lack formal incision or violation of the pediatric anterior commissure. Moreover, its role as an adjunctive measure is clearly demonstrated.

REFERENCES