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Incidence of Operative Endoscopy Findings in Recurrent Croup

Noel Jabbour, MD1,2, Noah P. Parker, MD1,2, Marsha Finkelstein, MS1, Timothy A. Lander, MD1,2, and James D. Sidman, MD1,2

Abstract

Objective. Develop an evidence-based model for predicting operative endoscopy findings in patients with recurrent croup.

Study Design. Case series with chart review.


Subjects and Methods. Retrospective chart review was performed on 124 patients who received consultation for recurrent croup between 2000 and 2009. Direct laryngoscopy and bronchoscopy findings were categorized as normal, mildly abnormal (incidental findings or grade I subglottic stenosis), moderately abnormal (grade II subglottic stenosis), or severely abnormal (grade III-IV subglottic stenosis).

Results. Of 124 consultations for recurrent croup, 81 patients (average age 3.5 years) underwent operative endoscopy. Normal examinations occurred in 33 of 81 (41%). Abnormal findings were encountered with the following frequency: mildly abnormal, 40 of 81 (49%); moderately abnormal, 6 of 81 (7.5%); and severely abnormal, 2 of 81 (2.5%). Relative risk (RR) of either moderately abnormal or severely abnormal findings was increased for patients who had a history of previous intubation (RR = 9.8; P = .002), prematurity (RR = 6.4; P = .01), or inpatient consultation (RR = 5.3; P = .028). The rate of moderately or severely abnormal findings in patients without the risk factors of intubation and age younger than 1 year was 0 of 48 (0%; confidence interval, 0%-7.4%). Mild abnormalities in this group were encountered in 27 of 48 (56%).

Conclusion. Mild airway abnormalities are common in children with recurrent croup and cannot be ruled out based on history. In the absence the risk factors of previous intubation, age younger than 1 year, or inpatient consultation, the incidence of a significantly abnormal finding is quite low. A predictive model based on this evidence is discussed.

Keywords
recurrent croup, laryngotracheobronchitis, bronchoscopy, laryngoscopy

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Croup is a common clinical syndrome of infancy and childhood marked by acute onset of barking cough, stridor, and varying degrees of hoarseness and respiratory distress.1 It may affect approximately 15% of children at some point in childhood.2 Large population studies have shown that 5% to 6.4% of children suffer from recurring episodes of this illness, which is commonly referred to as recurrent croup.3,4

It is important to note that recurrent croup is not a diagnosis; rather, it is a recurring syndrome that should alert physicians to assess for nonviral etiologies.4 Recurrent croup has been shown to be commonly associated with gastroesophageal reflux, asthma, allergy, eosinophilic esophagitis, or extrinsic compression or intrinsic narrowing of the trachea or subglottis.5-7 Recurrent croup may be manifested by recurrent episodes of acute laryngotracheobronchitis or by spasmodic croup, which involves recurrent croup-like symptoms often in the absence of a fever or a viral prodrome.

Recurrent croup is a common cause for referral to an otolaryngologist. Significant controversy and variance in practice exists with regard to the diagnostic workup of these patients. The decision of whether to undergo diagnostic laryngoscopy and bronchoscopy should be made with an understanding of the likelihood of detecting an underlying condition. However, few studies have correlated risk factors noted at the time of consultation with direct laryngoscopy and bronchoscopy findings, and applicability of these studies has been limited because of small sample sizes and limited statistical analysis.4,7,8,10,11

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Our aim is to identify risk factors that correlate with a higher likelihood of discovering a significant abnormality during diagnostic laryngoscopy and bronchoscopy. A secondary aim is to identify risk factors or combinations of risk factors, which, when absent, have a high negative predictive value for operative endoscopy. These data could enable otolaryngologists and patient families to make more informed decisions on whether to proceed with diagnostic laryngoscopy and bronchoscopy in the setting of recurrent croup.

Methods

Institutional review board (IRB) approval was obtained from the Children’s Hospitals and Clinics of Minnesota IRB to retrospectively review the charts of patients who had been referred to a pediatric otolaryngology practice for croup between July 2000 and September 2009. Charts of patients with recurrent croup were selected for review. Only patients who had undergone direct laryngoscopy and bronchoscopy at our institution were included for review.

Information tabulated for each patient included age, gender, prematurity status, past medical history, frequency of croup episodes, previous intubation status, whether the initial consultation was performed in the inpatient or outpatient setting, age at the time of endoscopy, and operative findings. Degree of subglottic stenosis was graded according to the Meyer-Cotton grading system. Endoscopic findings were categorized as follows:

- **Normal**—no abnormalities noted.
- **Mildly abnormal**—grade I subglottic stenosis or other minor abnormality not requiring surgical intervention or further monitoring with operative endoscopy.
- **Moderately abnormal**—grade II subglottic stenosis or other non-life-threatening abnormality requiring surgical intervention or further operative endoscopy for monitoring.
- **Severely abnormal**—grade III or IV subglottic stenosis or other life-threatening abnormality requiring surgical intervention and repeat operative endoscopy for monitoring.

Moderately and severely abnormal findings were grouped together for statistical analysis as both required further diagnostic or surgical intervention.

The effect of preprocedure risk factors on the incidence of moderately or severely abnormal findings was evaluated by calculating relative risk, confidence intervals, and 2-tailed \( P \) values by Fisher exact test. Risk factors analyzed were history of previous intubation, history of prematurity, location of consultation (inpatient vs outpatient), presence of significant comorbidities, age younger than 1 year, and age younger than 3 years. Comparisons of the absence of each risk factor to the rate of mildly abnormal findings or moderately or severely abnormal findings are reported with 95% confidence intervals based on the standard error for proportions.

Using a classification algorithm, CHi-squared Automatic Interaction Detector (CHAID), available in SPSS AnswerTree (SPSS, Inc, an IBM Company, Chicago, Illinois), a decision tree was generated classifying patients into the 2 groups: (1) NL/Mild (normal or mildly abnormal findings) and (2) Mod/Sev (moderately or severely abnormal findings). All of the risk factor variables were entered into the analysis for consideration. Because of the limited number of cases, a learning set was not designated. The results represent all of the available cases.

Multivariate analysis was performed using logistic regression to calculate the adjusted odds ratios for risk factors that met statistical significance using univariate analysis but were not discriminating factors in the discriminant analysis model.

Results

Of the 124 patients who met criteria for recurrent croup, 81 underwent direct laryngoscopy and bronchoscopy. There were 59 boys and 22 girls. Age ranged from 2 months to 10.5 years (mean age 41 months; median age 32 months).

A summary of operative endoscopy findings for these 81 patients is shown in Table 1. Normal examinations occurred in 33 of 81 (41%). Some form of abnormality was noted in 48 of 81 (59%), although the majority of these, 40 of 48, were considered mildly abnormal. Thus, a normal or mildly abnormal finding was noted in 73 of 81 (90%). Grade I subglottic stenosis was noted in 17 of 40 mildly abnormal findings. Eighteen of the 40 mildly abnormal findings were found outside the subglottis.

### Table 1. Summary of Direct Laryngoscopy and Bronchoscopy

<table>
<thead>
<tr>
<th>Finding (n = 81)</th>
<th>No. (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Normal</td>
<td>33 (40.7)</td>
</tr>
<tr>
<td>Mildly abnormal</td>
<td>40 (49.4)</td>
</tr>
<tr>
<td>Grade I SGS</td>
<td>17*</td>
</tr>
<tr>
<td>TVF nodules</td>
<td>7</td>
</tr>
<tr>
<td>Signs of LPR</td>
<td>4</td>
</tr>
<tr>
<td>Adenoid hypertrophy</td>
<td>1</td>
</tr>
<tr>
<td>Mild asymmetry of TVF motion</td>
<td>1</td>
</tr>
<tr>
<td>Small subglottic cyst</td>
<td>1</td>
</tr>
<tr>
<td>Small vallecular cyst</td>
<td>1</td>
</tr>
<tr>
<td>TVF edema</td>
<td>1</td>
</tr>
<tr>
<td>Moderately abnormal</td>
<td>6 (7.4)</td>
</tr>
<tr>
<td>Grade II SGS</td>
<td>4</td>
</tr>
<tr>
<td>Large subglottic cysts</td>
<td>1</td>
</tr>
<tr>
<td>Subglottic hemangioma</td>
<td>1</td>
</tr>
<tr>
<td>Severely abnormal</td>
<td>2 (2.5)</td>
</tr>
<tr>
<td>Grade III SGS</td>
<td>1</td>
</tr>
<tr>
<td>Severe tracheobronchomalacia</td>
<td>1</td>
</tr>
</tbody>
</table>

Abbreviations: LPR, laryngopharyngeal reflux; SGS, subglottic stenosis; TVF, true vocal fold.

*One patient was described as having laryngomalacia and grade I SGS and is categorized twice.
The incidence of significant findings (moderately abnormal or severely abnormal) by risk factor is shown in Table 2. Relative risk (RR) of moderately/severely abnormal findings to normal/mildly abnormal findings was significant for patients who had a history of previous intubation (RR = 9.8; \(P = .002\)), prematurity (RR = 6.4; \(P = .01\)), inpatient consultation (RR = 5.3; \(P = .028\)), or age younger than 1 year (RR = 6.3; \(P = .009\)). No statistically significant increased risk was noted for the risk factors of comorbid condition (RR = 2.4; \(P = .19\)) and age younger than 3 years (RR = 2.8; \(P = .267\)).

Comparison of the absence of specific risk factors to the presence of significantly abnormal endoscopic findings is shown in Table 3. The rate of moderately or severely abnormal findings in patients without the risk factors of intubation or age younger than 1 year was 0 of 48 (0%; confidence interval [CI], 0%-7.4%). However, mild abnormalities in this group were encountered in 27 of 48 (56%). Similarly, the rate of moderately or severely abnormal findings in patients without the risk factors of intubation or prematurity was 1 of 57 (1.8%; CI, 0%-9.4%); mild abnormalities in this group were encountered in 29 of 57 (51%). For patients whose only risk factor was intubation, significantly abnormal findings were noted in 1 of 7 (14%; CI, 0.4%-58%).

The classification tree revealed 3 discriminating risk factors: intubation, age younger than 1 year, and inpatient consultation. That is, within this study, these risk factors alone could be used to stratify patients into 2 groups: those who did and those who did not have moderately or severely abnormal findings.

### Discussion

In this study, we found that only 65% of patients who presented for a consultation of recurrent croup underwent diagnostic laryngoscopy and bronchoscopy. Although diagnostic endoscopy was routinely offered, the recommendation was tempered for some patients with less concerning histories and for patients who lacked perceived risk factors for significant abnormalities (eg, previous intubation). This was done without statistical evidence to support such decisions.

Indeed, for the 81 patients who underwent diagnostic laryngoscopy and bronchoscopy, the likelihood of finding any abnormality was moderately high; 48 of 81 patients (59%) were found to have some abnormality. This rate is within the wide range of rates for abnormal findings previously reported, which range from 25% to 100%.7,8,11 This range is widened by the high rate of subglottic stenosis reported by Hoa et al10 (98%) and Kwong et al11 (100%) from the same institution.

One difficulty in comparing previous reports is that a dichotomous classification of normal vs abnormal is often used. This obscures the true continuum of abnormalities that exists, ranging from minor abnormalities that may or may not be contributing to the syndrome of recurrent croup to severe, life-threatening abnormalities. For this reason, we have found it helpful to further categorize the degree of abnormality into mild, moderate, and severe. In our analysis, it also seems reasonable to group these endoscopic findings into those that do not require surgical intervention or further operative endoscopy (normal or mildly abnormal) and those that do (moderately abnormal and severely abnormal).

Although the likelihood of finding a minor abnormality was relatively high in these 81 patients, the likelihood of finding a moderately or severely abnormal finding was low, 8 of 81 (9.9%). If we assume that the 43 patients who did not undergo bronchoscopy would have been even less likely to have significantly abnormal findings, this percentage may be
even lower when considering the rate for all patients presenting for consultation. Because of this low rate, one should expect to have a normal or only mildly abnormal finding in more than 90% of patients. Indeed, when the data presented by Chun et al.\(^8\) are analyzed using the classification scheme presented in this article, 28 of 30 (93%) had a normal or mildly abnormal finding. This high rate of normal or only mildly abnormal findings can be distressing to primary care providers and otolaryngologists who are conscious about cost containment and minimizing patient risks for “unnecessary” procedures. However, decisions to defer or forgo diagnostic laryngoscopy and bronchoscopy should not be done without a thorough understanding of the risk factors that are correlated with a high incidence of significantly abnormal findings.

In this study, intubation was the risk factor most correlated with moderately or severely abnormal findings, with a relative risk of 9.8. In the discriminant analysis tree, a history of intubation was the most discriminating risk factor. This is not surprising since the most common etiology of acquired subglottic stenosis is transglottic endotracheal intubation. Importantly, as shown in Table 3, the presence of this risk factor, even in the absence of all other risk factors, yielded a moderately or severely abnormal finding in 1 of 7 patients. Given the small size of this subgroup, the 95% confidence intervals for the incidence of a significantly abnormal finding range from 0.4% to 58%. We thus can find no evidence in this study or in other previously published studies to support the practice of forgoing diagnostic laryngoscopy and bronchoscopy in patients with a history of intubation.

Baker\(^\text{13}\) argued that any patient whose croup symptoms were severe enough to warrant hospitalization and intubation merits operative endoscopy. This study further suggests that any patient whose recurrent croup symptoms are severe enough to warrant inpatient otolaryngology consultation deserves a diagnostic laryngoscopy and bronchoscopy. The relative risk of finding a moderately or severely abnormal finding in patients whose initial consult was performed in the inpatient setting was 5.3 (\(P = .028\)). In addition, inpatient consultation was a discriminating risk factor in the discriminant analysis model.

Documented of whether a patient had ever been admitted for croup was not present consistently in the text of every consult note; therefore, the presence of an inpatient otolaryngology consultation note was deemed to be a more reliable method to decipher which patients had been hospitalized for recurrent croup. We acknowledge that the location of consultation depends heavily on referral patterns of individual practices and institutions. We thus present this risk factor of “inpatient consultation” only insomuch as it is a corollary to “inpatient admission” for recurrent croup. It is reasonable to assume that patients admitted for croup may have a severity of disease that is greater than that of their counterparts who have not been admitted for recurrent croup. It is not surprising, therefore, that patients with recurrent croup who were seen during an inpatient admission for croup, in our study, had a higher relative risk of moderately or severely abnormal findings.

Age younger than 3 years was found by Chun et al.\(^8\) to be a group with a higher likelihood of abnormalities, but in this study, no significant increase in moderately or severely abnormal findings was noted for this age group (\(RR = 2.8; P = .267\)). In this study, age younger than 1 year was a risk factor for having a significant abnormality (\(RR = 6.3\)), and this was statistically significant (\(P = .009\)). Age younger than 1 year was also a discriminating factor in the discriminant analysis model.

A history of prematurity seemed to be correlated with a higher likelihood of a significant abnormality (\(RR = 6.4\)). However, logistic regression analysis revealed no statistically significant predictive value, and prematurity was not a discriminating risk factor in the discriminant analysis model. Similarly, the presence of a significant comorbidity was not correlated with an increase in the likelihood of a moderately or severely abnormal finding (\(RR = 2.8; P = .267\)).

For the 81 patients who underwent direct laryngoscopy and bronchoscopy, the absence of any of these 3 risk factors—intubation, inpatient consultation, and age younger than 1 year—was correlated with a very low likelihood of a significantly abnormal finding. Actually, the absence of 2 of these risk factors alone, intubation and age younger than 1 year, yielded a 0% rate of moderately or severely abnormal findings. It is important to note that because the size of this subgroup is only 48 patients, the CI ranges from 0% to 7.4%. Further studies would be needed with larger sample sizes to assess if this rate is truly closer to 0% as was found in this study.

It is also notable that even in this subgroup, the rate of finding a minor abnormality was 27 of 48 (56%). Although these

### Table 3. Incidence of Abnormal Findings with Absence of Risk Factors

<table>
<thead>
<tr>
<th>Absence of the Following Risk Factors</th>
<th>(n = 81)</th>
<th>Mild Abnormality</th>
<th>Moderate/Severe Abnormality</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td>No. (%)</td>
<td>95% CI, %</td>
</tr>
<tr>
<td>Prematurity and comorbidity</td>
<td>48</td>
<td>29 (60)</td>
<td>45-74</td>
</tr>
<tr>
<td>Prematurity and age &lt;1 y</td>
<td>57</td>
<td>32 (56)</td>
<td>42-69</td>
</tr>
<tr>
<td>Intubation and premorbidity</td>
<td>57</td>
<td>29 (51)</td>
<td>37-64</td>
</tr>
<tr>
<td>Intubation and age &lt;1 y</td>
<td>48</td>
<td>27 (56)</td>
<td>41-71</td>
</tr>
<tr>
<td>Prematurity, comorbidity, and age &lt;1 y but positive for intubation</td>
<td>7</td>
<td>4 (57)</td>
<td>18-90</td>
</tr>
</tbody>
</table>
minor abnormalities do not require further surgical intervention or diagnostic bronchoscopy, there may be value to the patient, family, primary care provider, and other physicians to know this information. Some authors have argued that such findings may help to better direct treatment of asthma, gastroesophageal reflux, or eosinophilic esophagitis.

Based on the data from this study, an evidence-based model was developed to assess which patients should undergo diagnostic laryngoscopy and bronchoscopy. Of note, the classification tree model identified intubation and age younger than 1 year as sufficient primary discriminating factors. That is, no other questions are needed to separate patients with normal or mildly abnormal findings from those with significantly abnormal findings. This is because 48 of the 73 patients with normal or mildly abnormal findings had an absence of these 2 risk factors. The use of inpatient consultation in this model is only as a secondary discriminator for patients who were not intubated but whose age was younger than 1 year. We acknowledge that because the size of this subgroup was only 14, a conservative approach would include inpatient consultation as another primary discriminating risk factor. Using these discriminating factors as decision tree nodes, a recommendation can be made that patients with any of the risk factors of intubation, age younger than 1 year, or inpatient consultation, should undergo diagnostic laryngoscopy and bronchoscopy. Patients without these risk factors should be counseled that the likelihood of a mildly abnormal finding may be approximately 50% to 60%, but the rate of a significantly abnormal finding is likely below 7.5% and may indeed approach 0%. The value of finding a mild abnormality should also be explained to the family. On the basis of this evidence, a family will be able to make an informed decision about proceeding with or deferring direct laryngoscopy.

It should be noted that this model is based on retrospective data correlating patient risk factors with operative endoscopy findings. Additional testing of this model on a separate retrospective data set or on a prospective cohort would further strengthen its predictive value.

A limitation of this study is that the small number of patients with significantly abnormal findings (8/81) decreases the power of multivariate analysis and increases the uncertainty of discriminant analysis. In the future, a multicenter retrospective study could provide a larger sample size to strengthen both multivariate analysis and discriminant analysis. Indeed, in an era of increased interest in health care cost reduction, such an analysis may be of interest to payers and physicians.

**Conclusion**

Recurrent croup is a common cause for referral to an otolaryngologist. Decisions on whether to proceed with diagnostic laryngoscopy and bronchoscopy should be made with a thorough understanding of the risk factors that correlate with significant airway abnormalities.

A high index of suspicion for significant abnormalities should exist for patients with risk factors of previous intubation, age younger than 1 year, and inpatient consultation. Mild airway abnormalities are common in children with recurrent croup and cannot be ruled out based on history alone. However, for patients without a history of previous intubation, age younger than 1 year, or inpatient consultation, the likelihood of finding a significant abnormality is quite low (0%-7.5%). This information may be useful to referring physicians and to otolaryngologists who are counseling patients and their families about the utility of diagnostic laryngoscopy and bronchoscopy.

**Author Contributions**

Noel Jabbour, conception, design, acquisition of data, analysis of results, drafting of article, and final approval; Noah P. Parker, acquisition of data, writing, and final approval; Marsha Finkelstein, analysis of data, writing, and final approval; Timothy A. Lander, conception and design, critical review, and final approval; James D. Sidman, conception and design, interpretation of data, writing, and final approval.

**Disclosures**

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References