ABSTRACT

Objective: To describe the clinical profile of patients with laryngotracheal stenosis over a 7-year period and discuss strategies for its prevention.

Methods:

Design: Retrospective Case Series
Setting: Tertiary Government Hospital
Participants: Thirteen (13) patients with laryngotracheal stenosis confirmed by laryngoscopy and/or bronchoscopy.

Results: Twenty-one patients were evaluated for laryngotracheal stenosis from January 2008 to June 2015, but only 13 with complete data were included in this study. Of the 13 patients, nine (69.2%) belonged to the pediatric age group. Ten (77%) were males and three (23%) were females. Laryngotracheal stenosis following endotracheal tube (ET) intubation was seen in 11 (84.6%) while 2 had thyroid masses and no history of prior ET intubation. Presenting symptoms or reasons for referral were wheezing (n=4), stridor (n=4), failure to decannulate the tracheostomy tube (n=3), and dyspnea (n=2). Duration of ET intubation was four to 60 days. The highest frequency of ET reintubation was 5 times. Among those intubated, stenosis was glottic in one, subglottic in five and tracheal in five patients. Three had Cotton-Myer grade I stenosis, two had grade II, three had grade III and three had grade IV stenosis. Those with thyroid masses had tracheal stenosis.

Conclusion: Strategies for prevention of laryngotracheal stenosis should include routine airway endoscopy for patients with longstanding neck masses and for those with prolonged ET intubation, for whom the option of early prophylactic tracheostomy is worth considering. Otherwise, immediate post-extubation endoscopy may facilitate documentation and appropriate intervention.

Keywords: acquired laryngeal stenosis; tracheal stenosis; endoscopy; intubation, intratracheal; tracheostomy

Laryngotracheal stenosis is a challenging airway problem, most often from sequelae of local tumors, infection, trauma and intubation. Laryngotracheal stenosis is an umbrella term, encompassing luminal compromise of the larynx, subglottis and trachea. Injury is initiated by ischemic necrosis of the mucosa, leading to ulceration of cartilage, inflammation with granulation and fibrous contraction. Although numerous studies describe various treatment modalities, there is no standard approach to laryngotracheal stenosis. Successful treatment...
despite various options remains a challenge to the otolaryngologist, and prevention is still the best way to overcome laryngotracheal stenosis. Understanding the etiologies of stenosis and avoidance of instigating factors may help prevent its development. To the best of our knowledge, no local data has been published on the profile of patients with laryngotracheal stenosis. An initial survey may thus be used to explore prevention strategies. This study describes the clinical profile of patients with laryngotracheal stenosis over a 7-year period and discusses strategies for its prevention.

METHODS

With institutional review board approval, a retrospective chart review of patients with laryngotracheal stenosis who were seen at a tertiary government hospital from January 2008 to June 2015 was conducted. Patients with incomplete data were excluded. Diagnosis of laryngotracheal stenosis was confirmed by laryngoscopy and/or bronchoscopy. Age, sex, causal factors for stenosis, presentation of stenosis, history of endotracheal tube (ET) intubation (including indication for intubation, size of ET tube used, duration of intubation, frequency of re-intubation, interval from latest extubation until stenosis diagnosis), and grading of stenosis based on Cotton-Myer classification and location of stenosis were recorded.

RESULTS

A total of 21 patients were evaluated for laryngotracheal stenosis from January 2008 to June 2015, but only 13 patients with complete data were included in this study. Table 1 shows the age, sex, causal factors of stenosis, presentation of stenosis, history of ET intubation, Table 1. Clinical profile of patients with laryngotracheal stenosis from 2008-2015

<table>
<thead>
<tr>
<th>Age</th>
<th>Sex</th>
<th>Probable cause of stenosis</th>
<th>Presentation of stenosis</th>
<th>ET size used</th>
<th>Total Duration of ET intubation (days)</th>
<th>Frequency of ET re-intubation</th>
<th>Interval from latest extubation to stenosis diagnosis (days)</th>
<th>Stenosis Grade</th>
<th>Stenosis Location</th>
</tr>
</thead>
<tbody>
<tr>
<td>24 days</td>
<td>M</td>
<td>Intubation due to pneumonia</td>
<td>Stridor</td>
<td>3.0</td>
<td>5</td>
<td>2</td>
<td>2</td>
<td>I</td>
<td>Subglottis</td>
</tr>
<tr>
<td>52 days</td>
<td>M</td>
<td>Intubation due to pneumonia, failure to thrive</td>
<td>Stridor</td>
<td>3.0</td>
<td>33</td>
<td>5</td>
<td>29</td>
<td>III</td>
<td>Subglottis</td>
</tr>
<tr>
<td>22 mos</td>
<td>M</td>
<td>Intubation due to congenital diaphragmatic hernia</td>
<td>Stridor</td>
<td>4.5</td>
<td>4</td>
<td>2</td>
<td>5</td>
<td>II</td>
<td>Trachea</td>
</tr>
<tr>
<td>3 yrs</td>
<td>M</td>
<td>Intubation due to pneumonia</td>
<td>Stridor</td>
<td>4.5</td>
<td>60</td>
<td>1</td>
<td>4</td>
<td>I</td>
<td>Subglottis</td>
</tr>
<tr>
<td>6 yrs</td>
<td>M</td>
<td>Intubation due to blunt abdominal trauma due to vehicular accident</td>
<td>Wheeze</td>
<td>5.5</td>
<td>15</td>
<td>1</td>
<td>3</td>
<td>III</td>
<td>Subglottis</td>
</tr>
<tr>
<td>8 yrs</td>
<td>M</td>
<td>Intubation due to pneumonia</td>
<td>Failure to decanulate</td>
<td>4.0</td>
<td>42 Converted to tracheostomy</td>
<td>2</td>
<td>378</td>
<td>II</td>
<td>Subglottis</td>
</tr>
<tr>
<td>12 yrs</td>
<td>F</td>
<td>Intubation due to Acute gastroenteritis, hypokalemia</td>
<td>Wheeze</td>
<td>5.5</td>
<td>22</td>
<td>3</td>
<td>3</td>
<td>I</td>
<td>Glottis</td>
</tr>
<tr>
<td>14 yrs</td>
<td>M</td>
<td>Multinodular nontoxic goiter, compression</td>
<td>Wheezing</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>I</td>
<td>Trachea</td>
</tr>
<tr>
<td>16 yrs</td>
<td>M</td>
<td>Intubation due to craniectomy due to vehicular accident</td>
<td>Dyspnea</td>
<td>6.0</td>
<td>9</td>
<td>1</td>
<td>7</td>
<td>IV</td>
<td>Trachea</td>
</tr>
<tr>
<td>32 yrs</td>
<td>M</td>
<td>Intubation due to cardiopulmonary arrest</td>
<td>Failure to decanulate</td>
<td>6.0</td>
<td>23 Converted to tracheostomy</td>
<td>1</td>
<td>217</td>
<td>IV</td>
<td>Trachea</td>
</tr>
<tr>
<td>35 yrs</td>
<td>F</td>
<td>Intubation due to ectopic pregnancy</td>
<td>Failure to decanulate</td>
<td>6.0</td>
<td>18 Converted to tracheostomy</td>
<td>1</td>
<td>128</td>
<td>IV</td>
<td>Trachea</td>
</tr>
<tr>
<td>41 yrs</td>
<td>F</td>
<td>Multinodular nontoxic goiter, compression</td>
<td>Wheezing</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>III</td>
<td>Trachea</td>
</tr>
<tr>
<td>63 yrs</td>
<td>M</td>
<td>Intubation due to altered sensorium, due to vehicular accident</td>
<td>Dyspnea</td>
<td>6.0</td>
<td>28</td>
<td>1</td>
<td>68</td>
<td>III</td>
<td>Trachea</td>
</tr>
</tbody>
</table>

Table 1. Clinical profile of patients with laryngotracheal stenosis from 2008-2015
grading and location of stenosis. Of the 13 patients, nine (69.2%) belonged to the pediatric age group. Ten (77%) were males and three (23%) were females. Laryngotracheal stenosis following ET intubation was seen in 11 (84.6%) of the patients. The indications for ET intubation were diverse. Two patients had thyroid masses with no history of prior ET intubation. Presentations of stenosis were wheezing (n=4), stridor (n=4), failure to decannulate (n=3), and dyspnea (n=2). All 11 patients who had a history of ET intubation had endotracheal tubes compatible with the ideal size for age. The duration of ET intubation ranged from 4 to 60 days. Five patients had a history of being intubated more than once. The highest frequency of ET re-intubation was five times. Among the eight patients who did not undergo conversion to tracheostomy, stenosis diagnosis was established 2 to 68 days after ET extubation. Three patients were diagnosed very late (378, 216, and 128 days respectively) when they were referred due to failure of tracheostomy tube decannulation. Among those intubated, three had grade I stenosis, two had grade II, three had grade III and three had grade IV stenosis. The stenosis location was glottic in one patient, subglottic in five patients and tracheal in five patients. Those with thyroid masses had tracheal stenosis.

**DISCUSSION**

Laryngotracheal stenosis is a term implying the presence of airway compromise resulting from healing by secondary intention resulting in submucosal fibrosis and scar contraction involving the larynx, trachea or both. Probable causes of laryngotracheal stenosis in this study were ET intubation and neck mass compression.

An iatrogenic cause of laryngotracheal stenosis is usually ET intubation. Nearly 10% of intubated patients subsequently develop laryngotracheal stenosis1 with a 0-2% incidence of subglottic stenosis2 and 6% to 21% incidence of tracheal stenosis.3 In our study, stenosis following ET intubation was seen in 11 patients. The cuff of the ET tube can cause ischemic necrosis leading to stenosis in the trachea and/or subglottic region.4 Tracheal stenosis was seen in adults and adolescents which may be due to the cuffed ET usually used in this age-group. The tube itself can cause pressure injury to the glottis that can result in severe commissural scarring and necrosis especially if ET tube was excessively wide. However, appropriateness of ET tube size was probably not a factor causing stenosis in our study since all patients had appropriate ET tube sizes. There is no consensus regarding the limits for safe periods of intubation. Longer intubation periods correlated with broader and deeper ulcers, injury in the subglottis and posterior glottis. In neonates, perichondrium of the cartilage was exposed when intubation was longer than 8 days.5 In adults, orotracheal intubation for more than 96 hours has been associated with permanent damage.6 Stenosis was seen in 2% of patients who were intubated between three to five days while stenosis increased to 5% in patients intubated between six to ten days.4 In our study, the duration of ET intubation associated with stenosis was 4 to 60 days. Based on this limited experience, the authors suggest that patients may be evaluated as early as 4 days after ET intubation for possible stenosis. Tracheostomy may be contemplated based on the clinical features on endoscopy, particularly in those unlikely to be extubated early.5 The highest frequency of intubation was five times in a patient. Stenosis was seen even in patients who had been intubated only once, including those with Grade IV stenosis. This may suggest that intubated patients should be evaluated regardless of the times intubated.

In our study, diagnosis of stenosis was made 2 to 68 days after ET extubation. Delays in diagnosis may be due to symptoms not manifesting immediately or due to symptoms that presented immediately but were treated as asthma or pneumonia before eventual referral to our Larynx Clinic.

The diagnosis may be missed if the patient exhibited airway symptoms and was immediately re-intubated without stenosis evaluation. In this study, the diagnosis was as late as 378 days after ET extubation for patients who were referred after failure of decannulation. Wittekamp et al. found that post-extubation laryngeal edema developing symptoms within 30 minutes after extubation was the cause of 15% of all reintubations. Laryngeal edema, once diagnosed, can be prevented by giving corticosteroids. By preventing laryngeal edema, reintubations and in turn stenosis may be prevented. All these reasons for delayed diagnosis were related to failure to evaluate patients immediately post-extubation. A study by Smith et al reports that fiber-optic laryngoscopy may be performed in the first hours after extubation, with fewer complications.10 Flexible endoscopy is rapidly performed and provides accurate conditions of the larynx that may be useful for the diagnosis of lesions due to intubation. This would be less aggressive compared to direct laryngoscopy under general anesthesia.10

The Cotton-Myer system, originally developed for congenital subglottic stenosis, is currently the generally used classification in evaluating laryngotracheal stenosis severity.11 Grade I is 0 to 50% obstruction of the lumen, Grade II 51-70%, Grade III 71-99% and Grade IV no detectable lumen.12 Among our patients, Grade I, II and IV stenosis were mostly seen but the exact etiopathology cannot be determined. Figure 1 shows examples of the grading of stenosis from our study subjects.

The combination of factors such as age, infection, duration and multiple extubations-reintubations may have led to stenosis.
Pneumonia, an indication for intubation in four of our patients, could have predisposed the previously injured laryngotracheal framework to bacterial contamination. Age could be contributory. Subglottic stenosis was seen in 11.38% of intubated children. Majority of laryngotracheal stenosis in this study was in the pediatric group where subglottic stenosis was common. The subglottis is the most vulnerable area of the pediatric airways because it is the only site surrounded by a complete cartilage ring, the narrowest anatomic site, and the site most exposed to intubation trauma.

Our study could not account for other factors that may have been potential risk factors for injury such as ET cuff inflation, inadequate sedation and less cooperation on the part of the patient, technique of ET intubation, number of attempts and difficulty scale, movement of the tube from ventilator motion or manual suctioning and gastric reflux.

Tracheal injury was seen in patients with thyroid masses. This may be due to the anatomic location of the thyroid gland causing extrinsic compression and compromising the patency of the trachea. In our study, two patients had benign goiter measuring 3 and 4 cm, presenting 3 and 2 years, respectively. Consequent tracheal stenosis was diagnosed warranting prophylactic tracheostomy.

Tracheal stenosis is often misdiagnosed as asthma. Respiratory distress develops in patients with more than 50% narrowing of the tracheal lumen. In our study, three patients were treated as having asthma. Two of them presented with wheezing and neck masses and upon routine laryngoscopy for the neck mass were suspected to have tracheal stenosis. Laryngoscopy and bronchoscopy confirmed the diagnosis. Another patient presented with stridor after extubation. Since there was no resolution of stridor after 28 days of bronchodilator, referral to our Larynx Clinic was made which confirmed subglottic stenosis. History of prolonged ET intubation with respiratory symptoms unresponsive to bronchodilators should be an index of suspicion for stenosis.

The limitation of the study is the sparse number of cases that have been well documented. In 7 years, there were only 13 cases confirmed to have laryngotracheal stenosis with complete records, out of only 21 patients referred for evaluation. Deficiency of referrals due to lack of awareness of possibility of stenosis, failure to evaluate patients post-extubation, missed and misdiagnosis of patients may explain the small number of reported patients.
This study observed the varied frequency of intubation and intervals from latest extubation to stenosis diagnosis. The exact time when stenosis developed cannot be identified. Future research can be geared toward elucidation of stenosis evolution among the general population of intubated patients. Correlation of frequency and duration of intubation to the grading of stenosis can also be explored. Best time of intervention can then be determined.

Awareness of potential endotracheal intubation-related injuries is critical for early diagnosis and appropriate treatment. A systematic laryngeal examination after extubation decreases the chances of morbid sequelae by initiating appropriate measures. Strategies for prevention of laryngotracheal stenosis should include routine airway endoscopy for patients with longstanding neck masses and for those with prolonged ET intubation, for whom the option of early prophylactic tracheostomy is worth considering. Otherwise, immediate post-extubation endoscopy may facilitate documentation and appropriate intervention.

REFERENCES