Pediatric subglottic cysts are a rare but increasingly known cause of respiratory complications in neonates and children. Pediatric subglottic cysts are associated with prematurity, and while congenital cysts have been reported in patients without prior intubation, subglottic cysts occur almost exclusively in premature infants with a history of endotracheal intubation. Cyst formation occurs during healing of the subglottis after traumatic mucosal injury and results from obstruction of native mucous glands secondary to subepithelial fibrosis.²

An infant with a history of prematurity and intubation who presents with biphasic stridor should raise clinical suspicion for the presence of a subglottic cyst.³-⁴ Other symptoms associated with subglottic cysts may include apnea, recurrent croup, and feeding problems. Some infants are also inappropriately treated for pulmonary disease because of their clinical presentation of wheezing. Because these signs and symptoms are nonspecific, other pathologic conditions, including subglottic stenosis, laryngomalacia, vocal cord paralysis, hemangiomas, lymphangiomas, and foreign bodies, must be ruled out.⁵ Flexible laryngoscopy is a useful adjunctive procedure and may allow diagnosis in the office setting.⁶ While the diagnosis of a subglottic cyst is often suggested by history, physical examination, and imaging, the gold standard for diagnosis of subglottic cysts remains microlaryngoscopy and bronchoscopy.

The current treatment of choice for symptomatic subglottic cysts is endoscopic marsupialization.⁶ The reported techniques for marsupialization include cold steel microinstrumentation, various laser treatments, and the laryngeal microdebrider.³-⁴,⁷,⁸ These methods have been shown to be the most effective from both a risk and recurrence perspective.²-⁵,⁹ If the cyst is smaller and more fragile, rupture with the rigid bronchoscope is sometimes performed, but this method has not been proven to be as effective as marsupialization owing to an increased risk of recurrence.¹⁰

The Bugbee fulgurating electrode is an important tool used in urologic surgery and can also be effective in the treatment of various upper aerodigestive tract disorders.¹¹,¹² This electrode is a sterile, flexible device with a small diameter that delivers radiofrequency energy to soft tissue via a monopolar electrode tip. We describe treatment of 16 consecutive patients with subglottic cysts and present a treatment method that incorporates conventional endoscopic visualization techniques with marsupialization and cyst lysis using the Bugbee electrode.

**Methods**

Following the University of Mississippi Medical Center’s institutional review board review and approval, we performed a ret-
A prospective review of 16 patients who were treated at Batson Children's Hospital at the University of Mississippi Medical Center for subglottic cysts between 2006 and 2012. The patients were almost exclusively referred for stridor, which initiated an airway evaluation by the pediatric otolaryngology service.

If not intubated at the time of consultation, patients underwent bedside flexible fiberoptic laryngoscopy. All patients were then taken to the operating room for microlaryngoscopy and bronchoscopy following induction of general anesthesia. Patients were given inhalational anesthetics for induction of anesthesia, and the airway was exposed using a pediatric Miller intubating laryngoscope. The zero-degree Hopkins rod telescope was used to systematically examine the supraglottic, glottic, and subglottic larynx in detail while the patient’s respirations remained spontaneous. The location and number of subglottic cysts was confirmed and documented, as shown in the Figure.

The appropriate rigid pediatric bronchoscope was then attached to the Hopkins rod telescope and inserted to the level of the true vocal cords, at which point the anesthesia circuit was connected to the bronchoscope for ventilation. The Bugbee fulgurating electrode was then threaded through the suction port of the bronchoscope, and the cyst wall was blanched and perforated with the Bugbee electrode. Most commonly, purulent or mucoid-appearing fluid was expressed from the cyst. The Bugbee electrode was occasionally used to pierce larger cysts in several locations. Once the cyst was adequately decompressed, bronchoscopy was performed. In 1 patient, smaller cysts were lysed with the edge of the rigid bronchoscope as the bronchoscope passed through the subglottis. For patients who had numerous subglottic cysts, we occasionally left smaller sessile, nonobstructing cysts unruptured to decrease potential scarring and cicatrix formation, as recommended by Albright and Magit.9 After cyst rupture, patients were generally intubated with an appropriately sized pediatric endotracheal tube and remained intubated overnight. A dose of corticosteroids was given to all patients intraoperatively and repeated prior to extubation for all patients who remained intubated overnight. Patients who were not already receiving proton pump inhibitor (PPI) therapy were placed on PPI therapy indefinitely to decrease the effect of reflux on denuded mucosa and other airway pathologic conditions.

Data were collected on each patient’s gestational age at birth, sex, age at diagnosis, presenting symptoms, number of intubations, length of each intubation, total days intubated, interval from extubation to presentation, number of subglottic cysts, location of subglottic cysts, method of cyst lysis, days intubated postoperatively, other comorbid airway pathologic condition, follow-up duration, cyst recurrence, and location of cyst recurrence.

Results

Table 1 and Table 2 summarize the key portions of the data. Most patients (n = 13 [81%]) were male. The most common presenting symptom was biphasic stridor, followed by recurrent croup. The mean gestational age at birth was 26.9 weeks’ gestation, and patients were almost exclusively preterm neonates (n = 15 [94%]). The mean age at diagnosis was 38.9 weeks after birth. All patients had at least 1 prior intubation. Patients presented with a mean of 1.8 prior intubations, 29.8 cumulative days of intubation, and a mean presentation time of 7.3 months after their last extubation.

At the time of diagnosis, patients were found to have a mean of 1.6 cysts, and the cysts were more commonly unilateral (n = 10 [63%]) than bilateral (n = 6 [38%]). Unilateral cyst location was variable and included left lateral wall (n = 5 [31%]), right lateral wall (n = 1 [6.3%]), posterior wall (n = 2 [13%]), and anterior wall (n = 2 [13%]).

Nine patients (56%) were diagnosed as having an associated laryngotracheal pathologic condition, including subglottic stenosis (n = 8 [50%]), which was the most common asso-
Associated finding. Other associated pathologic conditions included a supraglottic hamartoma identified in 1 patient (6%). In all 16 patients, cysts were ruptured with the Bugbee fulgurating diathermy electrode.

One patient died of bronchopulmonary dysplasia prior to hospital discharge. For the 15 patients who did not die secondarily to other comorbid medical conditions before any outpatient follow-up, the mean duration of follow-up was 86 weeks (range, 0-310 weeks). This includes 3 patients who never returned for their first follow-up appointment after hospital discharge and were lost to follow-up. No major or minor complications occurred.

Of the 16 patients, 3 (19%) were found to have any recurrence of subglottic cysts, but only 1 of these patients (6%) was symptomatic and required repeated cyst lysis. The other 2 patients with cyst recurrence developed only small, asymptomatic, nonobstructing cysts that did not require treatment. All 3 patients who developed any cyst recurrence had multiple and bilateral subglottic cysts at the time of initial presentation before any intervention.

**Discussion**

Despite increasing surgical options for treating subglottic cysts, the recurrence rates remain unsatisfactory, with rates ranging from 12.5% to 71.0% of patients.3,7 Watson et al3 reported a 50% recurrence rate for patients treated with microinstru-
mentation, while Agada et al found a recurrence rate of 29%. In the largest case series to date, Lim et al used a combination of microinstruments and carbon dioxide laser in a series of 55 patients and reported a 43% recurrence rate with follow-up bronchoscopy between 3 and 6 months. In a series of 9 patients, Smith et al reported no recurrence after 6 months to 2.5 years while using a carbon dioxide laser and the Bugbee electrode, but several of these patients were treated with multiple procedures for cyst lysis. In another recent series, Ransom et al documented success using a laryngeal microdebrider, reporting only 1 symptomatic recurrence among 8 patients (12.5%). Also, mitomycin C therapy after surgical intervention has recently been shown to have an advantage on recurrence rates by potentially reducing scarring. In our series of 16 patients, only 1 (6%) was found to have symptomatic cyst recurrence and required treatment.

There are several properties of the Bugbee fulgurating electrode that make it effective at preventing recurrence. This device provides a very small current that allows for targeted application of energy only to the cyst under endoscopic visualization, facilitating coaptation of the subglottic mucosa to the cricoid cartilage. Low current diathermy minimizes trauma to normal tissues and also minimizes repeated instrumentation of the neonatal airway. The size of the pediatric airway often precludes the use of large or bulky instrumentation, and exposure for lasers or other devices may be difficult. This technique allows the surgeon to leave the bronchoscope and endoscope in place, continuously visualizing and controlling the airway, while threading the electrode into place. This helps to reduce further trauma and scarring of the airway and may also help to decrease operative time compared with other techniques.

Patients were generally left intubated postoperatively for short-term airway protection. After cyst rupture, the endotracheal tube was left in place to compress the residual flaccid mucosa of the cyst wall to the cricoid lamina and prevent immediate return of mucoid cystic fluid. Three patients were deemed safe for extubation immediately postoperatively because of minimal redundant cyst mucosa. We believe that the benefits of short-term airway protection with an appropriately sized endotracheal tube inserted in an atraumatic fashion outweigh any risk of further subglottic trauma.

With respect to cyst location, previous studies have reported a predominance of left-sided subglottic cysts, possibly because of iatrogenic mucosal injury, since most clinicians performing intubation are right handed. In our series, the most common location of unilateral cysts was again the left lateral subglottic wall, with 31% of our patients presenting with unilateral left-sided cysts.

The mean gestational age at birth for our population was 26.9 weeks, which correlates well with other previously reported ranges of 27 to 28 weeks. Of our patients, 15 (94%) were preterm neonates, and previously reported rates of prematurity in populations of patients with subglottic cysts range from 93% to 100%. The mean duration of intubation for our population was 29.8 cumulative days. Previous studies have reported ranges from 10 to 33 days. The number of intubations remains a key aspect of cyst formation because mucosal trauma is thought to be the etiology of cyst formation. The patients in our study underwent a mean of 1.8 intubations per patient and were diagnosed at a mean of 7.3 months after the last extubation. Halimi et al previously reported a mean of 1.9 intubations per patient and an interval from extubation to diagnosis of 8.2 months in their review of 17 patients with subglottic cysts.

Limitations of the present study include the retrospective nature of our analysis and the presence of other associated airway pathologic conditions, which may confound the data. While some studies have reported recurrence rates after scheduled repeated endoscopy for all patients, others have reported recurrence rates based on symptomatic patients. We did not perform repeated endoscopy on asymptomatic patients because we believe this would subject patients to unnecessary risk. Of the 3 patients with recurrent cysts, 2 were asymptomatic and would not have been recognized without airway surveillance of other associated pathologic conditions. It was also difficult to determine if these were present at the time of the original procedure and became more apparent on subsequent endoscopies. One of these 2 patients did not undergo any airway intervention for cyst recurrence and remained asymptomatic. The second patient was tracheostomy dependent secondary to grade 3 subglottic stenosis at the time of diagnosis of cyst recurrence and eventually underwent laryngotraceoeploplasty. It is unclear if this patient would have developed symptomatic recurrence if severe subglottic stenosis was not also present. Finally, while our population is relatively large compared with other published series, subglottic cysts remain a rare entity overall, and the number of patients is somewhat limited.

Conclusions

Subglottic cysts remain an important cause of stridor and airway obstruction in previously intubated infants and are frequently associated with other airway pathologic conditions. There are multiple techniques available to otolaryngologists for effective treatment of subglottic cysts. This review presents a method for successful management of subglottic cysts that is safe and effective and has a lower symptomatic recurrence rate than previously reported techniques.
Analysis and interpretation of data: Richardson, Reed.
Drafting of the manuscript: Richardson, Winford.
Critical revision of the manuscript for important intellectual content: Richardson, Norris, Reed.
Statistical analysis: Richardson, Winford.
Administrative, technical, or material support: Norris.
Study supervision: Norris, Reed.
Conflict of Interest Disclosures: None reported.

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