ABSTRACT

Objective: Subglottic cysts are an important cause of pediatric airway obstruction, particularly in previously intubated infants. We describe the typical presentation and management of subglottic cysts, with historical recurrence rates between 12.5% and 71%, and our recommendations for treatment using the Bugbee fulgurating diathermy electrode as a technique to successfully manage subglottic cysts with a low recurrence rate.

Study Design: Case series and literature review.

Methods: A retrospective review was performed on a series of 18 consecutive patients treated for subglottic cysts with microaryngoscopy and cyst lysis by a single surgeon at a single tertiary care facility.

Results: The mean gestational age was 26.8 weeks with a male to female ratio of 3.5:1. All patients had a history of prior intubation, with an average of 1.7 prior intubations per patient. Biphasic stridor was the most common presenting symptom, occurring an average of 7.2 months after last extubation. At the time of diagnosis, patients were found to have a mean of 1.6 cysts, and the left lateral subglottic wall was the most common location of unilateral cysts. Sixty-one percent of patients had associated laryngeal pathology, with subglottic stenosis being the most common associated finding. In eight-nine percent of patients, cysts were ruptured with the Bugbee fulgurating diathermy electrode. Clinically significant cyst recurrence occurred in only 5.6% of patients, and no major complications were reported.

Conclusion: This review presents a treatment algorithm for patients with subglottic cysts that is safe, effective, and has a recurrence rate lower than previously reported.

INTRODUCTION

Pediatric subglottic cysts (SGC) are a rare but increasingly known cause of respiratory complications in neonates and children. SGC occur almost exclusively in premature infants with a history of endotracheal intubation. Cyst formation results from obstruction of native mucous glands due to subepithelial fibrosis and squamous metaplasia in the healing process.

The treatment of choice for symptomatic SGC is endoscopic marsupialization.3,7 There are no different reported options to achieve marsupialization, including cold steel microinstrumentation, CO2 laser, and laryngeal microdebrider.5,7,8 These methods have been shown to be the most effective from both a risk and recurrence perspective.1,4-6

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 abnormalities.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 18 consecutive patients with SGC and present a treatment algorithm that incorporates conventional endoscopic visualization and marsupialization and cyst lysis using the Bugbee electrode.

METHODS AND MATERIALS

We performed a retrospective review of a total of 18 patients who were treated at Batson Children’s Hospital at the University of Mississippi Medical Center for subglottic cysts between 2006 and 2012.

All patients were taken to the operating room for microaryngoscopy and bronchoscopy under general anesthesia. Patients were given inhalational anesthetics for induction of anesthesia. The airway was routinely exposed using a pediatric Miller intubating laryngoscope. The zero degree 4 mm or 2.7 mm Hopkins rod telescope was used to systematically examine the supraglottic, glottic, and subglottic larynx in detail while the patient’s respirations remained spontaneous. Anesthesia gases and oxygen were delivered through a cut-off laryngoscope positioned along the lateral orocavity. Photo documentation was obtained. The location and number of subglottic cysts was confirmed and documented.

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 of the patient. 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. Less commonly, cysts were lysed with the edge of the rigid bronchoscope as the bronchoscope entered or passed through the subglottic. This was generally only performed for smaller cysts. With patients that had numerous subglottic cysts, we also occasionally left smaller sessile, non-obstructing cysts unruptured in order to decrease potential scarring and cicatrization formation. After cyst rupture, patients were generally intubated with an appropriately-sized pediatric endotracheal tube and remained intubated overnight to compress the residual flaccid mucosa of the cyst wall to the cricoid lamina. Patients were placed on propofol pump inhibitor therapy to decrease the effect of reflux on denuded mucosa and other airway pathology.

Data was collected on patient characteristics, pathology, and results as described below.

RESULTS

Most patients (n = 14, 78%) were male. The most common presenting symptom was biphasic stridor. The mean gestational age at birth was 26.8 weeks gestation. The mean age at diagnosis was 38.4 weeks. All patients had a history of prior intubation. Patients presented with a mean of 1.7 prior intubations, 31.9 cumulative days of intubation, and a mean presentation time of 7.2 months after their last extubation.

Patients had a mean of 1.6 cysts, and the cysts were more commonly unilateral (n = 11, 61%) than bilateral (n = 7, 39%). Unilateral cyst location was variable and included left lateral wall (n = 5, 28%), right lateral wall (n = 2, 11%), posterior wall (n = 2, 11%), and anterior wall (n = 2, 11%).

Sixty-one percent (n = 11) of patients were diagnosed with associated laryngotracheal pathology, including subglottic stenosis (n = 9, 50%), which was the most common associated finding. In 89% (n = 16) of patients, cysts were ruptured with the Bugbee fulgurating diathermy electrode. The mean duration of follow-up was 95 weeks (0-310). No major or minor complications occurred.

Three out of 18 patients (17%) were found to have recurrence of SGC, but only one of these (5.6%) was deemed to be clinically significant. The other two patients with cyst recurrence developed only small, clinically insignificant, non-obstructing cysts that were asymptomatic. These were identified incidentally on regularly scheduled repeat endoscopic airway evaluation for other associated airway pathology. All three patients who developed any cyst recurrence had multiple and bilateral subglottic cysts at the time of initial presentation before any intervention.

DISCUSSION

Recurrence rates for SGC remain unsatisfactory with rates ranging from 12.5% to 71% of patients.1-8 In our series of eighteen patients, only one of eighteen (5.6%) of patients was found to have clinically significant cyst recurrence and required treatment. Although three of eighteen (17%) patients were found to have cysts on repeat endoscopy, two of these were symptomatic and would not have been identified if not for routine endoscopic surveillance of other airway pathology. It was difficult to determine if these were preexisting at the time of the original procedure and became more apparent on subsequent endoscopies. We believe the use of the Bugbee electrode diathermy technique is more effective at preventing recurrence than other previously described techniques because it facilitates coaptation of the subglottic mucosa to the cricoid cartilage.

The size of the pediatric airway often precludes the use of large or bulky instrumentation for lasers or other devices may be difficult. Advantages of the Bugbee flexible fulgurating electrode for treatment of SGC include the ability to pass the device through the bronchoscope during the course of normal endoscopic visualization and evaluation of the airway. This device provides a very small current that allows for targeted application of energy only to the cyst under endoscopic visualization. Low current diathermy minimizes trauma to normal tissues and also minimizes repeated instrumentation of the neonatal airway. The surgeon can 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 to other techniques.

Limitations include the retrospective nature of our analysis and the presence of other associated airway pathology that may confound the data. Finally, while our subset of patients is relatively large compared with other published series, SGC remain a rare entity overall and the number of patients is somewhat limited.

CONCLUSION

Subglottic cysts remain an important cause of stridor and airway obstruction in previously intubated infants and are frequently associated with other airway pathology. There are multiple techniques available to otolaryngologists for effective treatment of subglottic cysts. This review presents an algorithm for successful management of subglottic cysts that is safe, effective, and has a lower recurrence rate than previously reported techniques.

REFERENCES