Endoscopic Management of Idiopathic Subglottic Stenosis in Twenty-Five Patients

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Introduction

While multiple causes exist for the development of subglottic stenosis, the mechanism responsible for idiopathic subglottic stenosis (ISS) remains unknown. Proposed etiologies include gastro-esophageal reflux (GERD) related exposure, telescoping of the first tracheal ring under the cricoid cartilage, a hormonal cause, and the possibility of an unidentified autoimmune process (1,3,6,8,12,18,19).

As ISS represents a rare disease of yet unknown etiology, the optimal management remains to be defined. The treatment options range from endoscopic management, which is less invasive but tends to show recurrence to open tracheal resection with anastomosis for those patients in whom endoscopic techniques are ineffective or have shown a high recurrence rate (5). Endoscopic management is an outpatient procedure, which shows limited voice alteration. In contrast, open resection requires a hospital stay, can be associated with changes in voice, and is reserved for centers with significant expertise due to the demanding nature of the procedure (11,15). However, the procedure results in complete removal of the inflamed and stenotic area (2,10).

The available literature shows limited reports for endoscopic management of ISS in more than twenty patients. This study sought to add to the current literature by reporting the experience at one institution, with a large number of patients during a relatively condensed time frame over which surgical technique has not varied. Specific areas of evaluation in this work include the presence of circumferential/cicatricial scarring which previously has indicated that endoscopic management would not be successful, as well as the goal for endoscopic management as the sole technique rather than a temporizing measure for open intervention. This study also sought to identify any characteristics related to interval of recurrence, which would help identify the patient population in which endoscopic intervention would be a favorable option.

Design

A retrospective chart review was completed to identify possible patients. Attributes including age at diagnosis, gender, smoking status and length of follow-up were obtained. Reported GERD symptoms and details of any medical management were acquired. A description of the grade, location, length, type and histopathology of the stenosis were attained where available. The number of endoscopic procedures, steroid use (oral/injected), Mitomycin C application and the longest interval between procedures were recorded. An assessment of whether any patient had undergone an open resection procedure including laryngotracheoplasty or larygotracheal resection was noted and if a tracheostomy had been placed if the patient was subsequently decannulated. The status of patient symptoms with regard to shortness of breath, dyspnea, hoarseness and cough were obtained to evaluate outcomes following intervention.

Endoscopic Management

Video bronchoscopy technique

Under monitored anesthesia care, topical nasal decongestant was applied, followed by topical anesthetic application to the supraglottis, glottis and subglottis. A video bronchoscope was introduced into the subglottis; characteristics of the stenosis were documented and laser safe precautions initiated. Laser radial incisions were made in the stenosis followed by balloon dilation.

MDL with flexible bronchoscopy

Following intubation the laryngoscope was introduced into the laryngea introitus and the patient was placed into suspension. Intermittent apneic anesthesia was utilized to allow intervention at the stenosis site. The video bronchoscope was then inserted allowing introduction of the laser for radial incision completion. Balloon dilation was then achieved and re-intubation completed.

Pharmaceutical adjuncts

At the discretion of the surgeon either Mitomycin C or Decadron were used in the following manner (16). Mitomycin C (1 mg per mL soaked cottonoid pledgets were placed over the area of intervention in the trachea. The pledgets were left in place for 4 minutes and then removed, after which cottonoid pledgets soaked in saline were used to wipe the area clear. Injection of Decadron (4 mg/mL) was carried out with 0.3-0.5 mL injected at the site.

Results

All patients in this study were female and the average age at diagnosis was 50.84 years with a range of 31-73 years. Tobacco use history indicated that 92% of patients never smoked and 8% reported being former smokers with a range of 5-10 pack years prior to cessation. Reflux symptoms were noted in 24%. The average grade was 2.15 by the Cotton-Myer grading system, with a range of 1- 3. The stenosis was described as circumferential in 36%. The average number of endoscopic procedures was 4.08 with a range of 1 – 14 and 48% of the group under went 2 or fewer. Twenty-eight percent received topical Mitomycin C application and 12% underwent Decadron injection; of the Mitomycin C group 12% underwent 2 or fewer procedures. One patient each underwent laryngotracheoplasty and laryngotracheal resection, representing 8% of the group. Tracheostomy tubes were placed at Jefferson in 16% and all were decannulated. Average follow-up for the patients totaled 3.6 years (1314.43 days) with a range of 0.2 – 10.22 years (72 – 3732 days).

Discussion

The current study showed a rate of open procedures of 2 patients or 8%, at the low end of the range of 5% to 16% in other similar studies. The rate of tracheostomy dependence is also the lowest of the group along with Giudice et al. at 0% (4,7,9,13,14,17,20).

Further comparison between the current study and the pooled data of the previously performed studies shows a similar average and range of age at diagnosis and percent of patients with reflux symptoms at 24% vs. 29% was noted. Patients in the current study showed an average grade stenosis of 2.15 by Cotton-Myer and a range of 1-3. The available literature showed wide variation in staging systems used and reporting of grade of stenosis, thus comparison data was not available. This study showed a lower average number at 4.08 and range of 1 – 14 endoscopic procedures. While wide variation in the use of intraoperative adjuncts in the outside studies made comparison impossible, our study showed 28% use of topical Mitomycin C and 12% use of Decadron injection. The average time interval between procedures was 1.86 years (680 days) with a range of 0.05 – 8.07 years (18-2,947 days). The average follow-up was approximately 3.6 years and 8 months less in the current study when compared to similar available literature (4,7,9,13,14,17,20). Symptomatically 56% of patients reported resolution of symptoms, 12% showed resolution then recurrence, 28% showed improvement with residual and 4% showed no change. No patients noted worsening of symptoms or sustained complications as a result of the endoscopic management employed.

The primary end point of the study was the longest interval between procedures and the multivariate model contains Mitomycin C application, circumferential stenosis and the age at diagnosis. The model indicates that if all other conditions are fixed, a patient given Mitomycin C shows 4.39 times longer duration between procedures. Certainly, a selection bias is possible in surgeon selection of patients who would benefit from application of this pharmacologic agent. Patients with circumferential stenosis showed 1.84 times longer duration between procedures. With regard to age, for patients showing circumferential stenosis, every 1 year increase in age at diagnosis would increase the longest interval by 1.07 times. For patients without circumferential stenosis, every 1-year increase in age at diagnosis would decrease the longest interval to 93% of the longest interval. During the follow-up period thus far 32% of patients underwent one procedure without recurrence. Due to the rare nature of this disease process and resulting low total number of patients included, overall the model explains 52% of total variance.

As ISS is an uncommon entity, the power of this study was not adequate to achieve statistically significant results regarding the impact of grade or type of stenosis on the number of endoscopic procedures, likelihood of having a tracheostomy tube placed or if grade and number of procedures was significant in symptomatic resolution. Further research is needed and will be completed based upon ongoing accrual.

Conclusion

ISS is a rare entity and this study adds to the available reported experience. Favorable outcomes were seen regarding symptom response, the number of endoscopic procedures needed and as in other studies, patients were encountered in this study that required only one endoscopic procedure for symptom control at the time of follow-up. Those who underwent an open procedure did so after thirteen and fifteen endoscopic procedures, which represent a much higher amount than the average of approximately four endoscopic procedures in the remaining population. The results obtained also showed an auspicious outcome for all patients regarding avoidance of a long-term tracheostomy tube with a low incidence of tracheostomy placement, mainly at the earlier stages of the study, and decannulation of all patients. Additionally, ISS patients with circumferential stenosis may still show success with endoscopic management.

With the positive results noted in this and prior studies in mind and as endoscopic intervention shows lower associated risks and is well tolerated, it is a rational choice as the initial surgical management and may be used in extended management for those who show continued response and improvement.

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