Open Excision of Subglottic Hemangiomas to Avoid Tracheostomy

Shyan Vijayasekaran, FRACS; David R. White, MD; Benjamin E. J. Hartley, FRCS(ORL); Michael J. Rutter, FRACS; Ravindhra G. Elluru, MD, PhD; Robin T. Cotton, MD

Objective: To assess the efficacy of open excision as an alternative to tracheostomy in the management of subglottic hemangioma.

Design: A retrospective review of patients undergoing open surgical excision of subglottic hemangiomas over a 10-year period.

Setting: A tertiary pediatric center.

Patients: The study included 22 children ranging in age from 2 to 42 months (median age, 5 months) who underwent open excision of subglottic hemangioma over a 10-year period.

Results: Twenty-one patients were treated with single-stage procedures, with postoperative endotracheal intubation for an average of 5 days. One patient who had a preexisting tracheostomy was treated with a 2-stage procedure and underwent decannulation 2 months after excision. Seven other patients were tracheostomy dependent at the time of excision and underwent decannulation at the time of the procedure. Cartilage grafts were inserted in 10 patients. There were no problems with subglottic stenosis. Twenty-one patients reported good voice and no airway symptoms after a mean follow-up period of 42 months. Five patients had significant postoperative problems. Three patients required further endoscopic procedures for removal of granulation tissue, and 1 patient, who remains minimally symptomatic, developed an anterior glottic web. One patient required a 6-month course of steroids after surgery to treat residual glottic hemangioma.

Conclusions: Open surgical excision of subglottic hemangiomas can be performed as a single procedure, avoiding a tracheostomy, when modern surgical techniques developed for laryngotracheal reconstruction are incorporated. This approach can avoid repeated endoscopic procedures, prolonged treatment with corticosteroids, and years spent with a tracheostomy waiting for spontaneous involution of the hemangioma.

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N EONATES WITH SUBGLOT- tic hemangiomas usually have no stridor at birth. A small, red macule may be visible in the subglottis. The hemangioma then enters a proliferative phase and progressively enlarges, obstructing the airway and causing biphasic stridor at an average age of 5 weeks. The proliferative phase often lasts through the first year of life. Untreated, the lesion then enters a stationary phase, during which it grows no further. At a later stage, resolution occurs, with spontaneous regression of the lesion.

A wide range of treatment modalities have been reported. The aim of the majority of treatments is to reduce the size of the lesion and therefore to maintain an adequate airway until the lesion spontaneously regresses. However, a significant number of the larger lesions do not respond sufficiently to local treatments, resulting in the placement of a tracheostomy to bypass the obstructed portion of the airway. This problem has led to the consideration of open excision using a surgical approach that was developed for single-stage laryngotracheal reconstruction as a treatment option for subglottic hemangioma.

Surgical excision of subglottic hemangiomas was first described in 1949. Subglottic surgery at that time necessitated a tracheostomy. It was argued that if a tracheostomy was going to be performed for an obstructive lesion that was going to spontaneously resolve, it would be better instead to perform a tracheostomy alone and wait for resolution, thereby avoiding the risk of subglottic stenosis. In 1949, the development of subglottic stenosis in a child was a serious complication, often resulting in lifelong tracheostomy. The prin-
principle cause of subglottic stenosis in the early part of this century was inappropriately high tracheostomy (placed at or just inferior to the cricoid cartilage), which was usually performed for inflammatory airway disease such as diphtheria. Surgeons were discouraged from splitting the cricoid. The Rethi procedure,5 which was used to treat subglottic stenosis in adults, was not described until 1956 and was not used in children until 1971.6 In 1972, Fearon and Cotton7 described the use of cartilage grafts to treat subglottic stenosis in children. In modern practice, subglottic stenosis is surgically correctable. By using single-stage laryngotraceal reconstruction techniques, surgeons can reliably operate on the pediatric subglottic airway, without the need for a tracheostomy. The use of cartilage grafts, particularly the readily available thyroid alar graft, has given surgeons the option to expand the airway caliber in cases in which subglottic stenosis is thought to be likely to develop.

Successful excision of subglottic hemangiomas, avoiding the use of a tracheostomy, has been described.8,9 While there are still surgeons who advocate performing a tracheotomy and waiting for spontaneous resolution of the hemangioma, this process may take years. The mortality and morbidity of pediatric tracheostomy are well documented. Increasingly, we are becoming aware of the effect on the parents and the difficulties of raising a child with tracheostomy. We are now able to offer surgical excision of subglottic hemangioma as a single-stage procedure, providing an alternative to tracheostomy. In this series, we report our experience with open excision for the treatment of subglottic hemangioma.

METHODS

After obtaining approval from the institutional review board, we reviewed the medical records of 21 children who underwent open excision of subglottic hemangiomas. The variables that were analyzed included age, sex, presenting symptoms, age at operation, previous treatment, existing tracheostomy, hemangioma site, adverse effects of previous treatment, operation date, graft used, complications, length of intubation, follow-up, and symptoms.

The surgical approach is similar to that used for single-stage laryngotracheal reconstruction.2,3 Initially, laryngoscopy is performed to confirm the exact position and size of the hemangioma (Figure 1). The patient is then intubated with an appropriate-sized endotracheal tube. The lesion is compressible, and endotracheal intubation can usually be achieved. The patient is positioned on the operating table with the neck in extension. A horizontal skin incision is made at the level of the cricoid cartilage. Superior and inferior subplatysmal flaps are elevated. The strap muscles are separated in the midline to expose the thyroid cartilage, cricoid cartilage, and trachea. If necessary, the thyroid isthmus is divided in the midline for access to the trachea. A temporary intraoperative tracheotomy is formed, and a cut oral Ring-Adair-Elwyn (RAE) tube is placed in the tracheostomy to ventilate the patient. The oral endotracheal tube is withdrawn. The cricoid cartilage is split vertically in the midline and held open with retraction sutures, providing exposure of the subglottic hemangioma (Figure 2). Local anesthetic with epinephrine is infiltrated around the hemangioma to reduce bleeding. The mucosa is incised over the hemangioma, and the hemangioma is dissected out in a submucosal plane. The subglottic mucosa is preserved and draped over the cricoid lamina. In our experience, it is difficult to establish a plane of dissection between the lesion and the mucosa of the subglottis in patients who have undergone extensive laser surgery in the past. In some cases, a laryngofissure is performed for access. After excision of the hemangioma, the patient is nasally intubated with an endotracheal tube that is half a size smaller than the age-appropriate tube. If the cricoid cartilage approximates loosely around the tube, then the anterior cricoid split is closed with 4-0 prolene sutures. If a simple closure will be under tension, a small thyroid alar graft is harvested from the superior border of
the thyroid cartilage and sutured into the midline anterior cricoid split. The neck is closed in the usual manner with a Penrose drain in situ.

## RESULTS

Twenty-two patients (11 girls and 11 boys) underwent open excision of subglottic hemangiomas between January 1995 and December 2004 (Table). They ranged in age from 2 to 42 months (mean age, 19.1 months). The presenting symptom of the hemangioma was stridor in all cases. Six patients were referred with tracheostomies in situ. One patient was referred because of multiple complications resulting from steroid therapy after 10 previous laser procedures. This patient failed to tolerate a steroid taper and hence required placement of a tracheostomy before reconstruction. Twenty patients underwent single-stage procedures with no postoperative tracheostomy, and 1 patient who had an existing tracheostomy tube was treated with a double-stage procedure. In the latter case, decannulation was achieved 6 weeks after the hemangioma had been excised. One patient, who initially underwent a single-stage procedure, required placement of a tracheostomy for 11 days to manage postoperative wound dehiscence.

Cartilage grafts were positioned in 15 patients. Ten patients had anterior thyroid alar grafts placed, and 3 patients had anterior costal cartilage grafts placed. Two patients had posterior costal cartilage grafts positioned for associated posterior subglottic stenosis. All patients had been previously treated with systemic corticosteroids (SCs).

Five patients had undergone previous carbon dioxide laser treatment, and 1 patient had previously been treated with intubation and intralesional steroid injection. The length of follow-up ranged from 6 to 78 months. A granuloma developed at the excision site in 3 patients, 2 of whom had successful endoscopic removal. The third patient was briefly unavailable for follow-up and returned with an anterior web at the site of the granuloma. Another patient, who had a laryngofissure, developed an anterior glottic web. Both webs were endoscopically treated. One of these patients remains minimally symptomatic with a residual web. One patient required postoperative steroid therapy for supraglottic and residual glottic disease.

## COMMENT

The management of subglottic hemangiomas has been an area of controversy for several years. The disease is rare, with 695 cases having been reported in the English-language literature between 1913 and 2002. Subglottic hemangiomas vary enormously in size, position, growth, and behavior, making the collection of prospective data in matched patients very difficult. There are a number of retrospective studies detailing experience with different treatment modalities.

## SC THERAPY

In general, children with subglottic hemangiomas can be classified into 3 groups: (1) those who respond well to a

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Abbreviations: ETT, duration of endotracheal intubation; NA, not applicable.

*This patient underwent decannulation of the preexisting tracheotomy 6 weeks after surgery.
short course of SCs and have no further problems, (2) those who partially respond to SC therapy and require prolonged therapy or further treatment measures, and (3) those who do not respond to SC therapy. This variable response is illustrated well in a series reported by Narcy et al., in which 26 infants were treated with SCs alone. Seven patients responded to a short course (mean, 22 days) of SCs and required no further treatment. Fourteen patients had recurrent symptoms when the initial dosage of SC therapy was decreased; their symptoms were controlled with prolonged SC therapy for up to 12 months (average, 4.5 months). Sixteen patients were resistant to SC therapy and were intubated for 9 to 12 days. After intubation, 9 patients were asymptomatic and 1 patient was reintubated. Five patients went on to further therapy. In contrast, in a review of the various treatment modalities by Bitar et al., 102 patients were treated with SC therapy, with limited success. Twenty-five patients (mean age, 9 months) responded to SC therapy. Adverse effects, including Cushing syndrome, hypertension, and growth retardation, were seen in 13% of cases. Rahbar et al. found similar results in their retrospective review.

In most institutions, SC therapy is the first line of treatment, because approximately 25% of patients will respond to it and require no further treatment. The highest success rates are seen in cases involving small lesions, and the response is seen within a few weeks. Prolonged SC therapy increases the risk of adverse effects in a significant number of patients, and other treatment modalities should be considered.

CARBON DIOXIDE LASER TREATMENT

Treatment of subglottic hemangiomas with the carbon dioxide laser is a well recognized and successful option. Serial laser treatment is often required. Even in experienced hands, the reported subglottic stenosis rate varies from 6% to 25%. A review of the recently published series found that a mean of 2 procedures and a range of 1 to 5 procedures were required. Because subglottic stenosis as a complication of laser treatment of subglottic hemangiomas is a significant problem, conservative use of the laser is advised. A recent review of the literature showed an overall success rate of 89% (81 patients). However, a retrospective study of 36 patients from a single institution found higher failure rates in circumferential and bilateral disease, suggesting that success rates are best in low-grade stenosis, with failure rates rising in Cotton-Myer stenosis grade 3 and 4 lesions.

LOCAL STEROID INJECTION AND INTUBATION

Treatment with local steroid injection and intubation has been reported to be successful, although multiple injections are generally required (range, 1-12 injections) along with prolonged periods of intubation. Hoeve et al. reported an average intubation of 37 days in 14 successful cases (out of a total of 18 cases) and an average intubation of 54 days in the failed cases. One patient was intubated for 129 days. The prolonged length of intubation and long stay in the intensive care unit may be the reasons that this technique has not gained widespread popularity.

TRACHEOSTOMY ALONE

The use of tracheostomy alone for the treatment of subglottic hemangioma has been advocated. The advantage of this approach is the avoidance of subglottic or laryngeal manipulation, minimizing the chance of the complications that are seen with open and endoscopic resections. Tracheostomy, however, has its own complications, including accidental decannulation, obstruction, stomal granulation, tracheocutaneous fistula, infection, and mortality rates of approximately 1%. A wide range of figures have been reported for tracheostomy mortality, depending on the indications for surgery and the level of tracheostomy care available. Also, the presence of a tracheostomy carries a social stigma, and the effect of caring for a tracheostomy-dependent child is difficult to evaluate.

OTHER MODALITIES

Success has been reported for multiple different treatments including interferon alfa-2a, which has been limited by adverse effects, most significantly spastic diplegia. Bleomycin injections have also been advocated.

OPEN RESECTION AND LARYNGOTRACHEAL RECONSTRUCTION

There are several small series that have reported the results of open resection. Rahbar et al. published a multi-institutional series involving 25 patients (mean age, 4 months) with a mean airway narrowing of 82%. Previous procedures or corticosteroid therapy had failed in 21 patients. One patient required tracheostomy placement for a failed resection, and 3 patients needed further endoscopic treatment for granulation. The mean age of the 22 patients in the present single-institution series was 19.1 months. All patients had undergone previous SC therapy or surgical interventions. Seven patients had received preoperative tracheostomy tube placement. Twenty-one procedures were performed in a single stage. Two patients had postoperative granulation tissue formation and underwent successful endoscopic removal. One patient developed an anterior web, which was successfully divided endoscopically. One patient developed granulation tissue and an anterior web, which was partially treated and remains mildly symptomatic. Overall, combined with the cases reported by Rahbar et al., 45 (96%) of 47 patients were successfully treated with open resection.

Multiple successful treatments have been advocated for subglottic hemangiomas. The wide variety of treatment options shows that there are a number of ways to reduce the size of a space-occupying lesion in the airway. To our knowledge, there are no prospective structured trials comparing different treatment modalities. In view of the rarity of the condition and the wide range of size and behavior of the lesions, evidence from large prospective studies is unlikely.
Treatment is aimed at relieving the airway obstruction as rapidly as possible with minimum adverse effects and inconvenience for the patient and family. Our institutional policy is to treat all patients initially with a short course of corticosteroids. A significant number of patients will respond completely and require no further treatment. Partial responders or nonresponders are considered for carbon dioxide laser treatment. To avoid subglottic stenosis, we believe that the use of the laser should be limited to smaller noncircumferential lesions. Larger lesions or persistent lesions are treated by open excision to avoid a tracheostomy using the techniques that have been developed for single-stage laryngotracheal reconstruction, and cartilage grafts are inserted to expand the cricoid ring to prevent subglottic stenosis when judged necessary. This highly effective, definitive treatment approach can avoid years of tracheostomy dependence.

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Correspondence: Shyan Vijayasekaran, FRACS, Department of Pediatric Otolaryngology, Cincinnati Children’s Hospital Medical Center, 3333 Burnet Ave, Cincinnati, OH 45229-3039 (shyan.vijayasekaran@cchmc.org).

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REFERENCES