Wolters Kluwer Health | Lippincott Williams & Wilkins

The Laryngoscope Founded in 1896 Official Journal of the Triological Society

 Volume 109(8), August 1999, pp 1281-1286

Surgical Removal of Subglottic Hemangiomas in Children

[Independent Papers]

Van Den Abbeele, Thierry MD, PhD; Triglia, Jean-Michel MD; Lescanne, Emmanuel MD; Roger, Gilles MD; Nicollas, Richard MD; Ployet, Marie-Josèphe MD; Garabédian, Eréa-Noël MD; Narcy, Philippe MD

From the Departments of Pediatric Otorhinolaryngology (T.V.D.A, P.N.), Robert Debré Hospital, Paris, Pediatric Otorhinolaryngology (J.-M.T., R.N.), La Timone Hospital, Marseille, Pediatric Otorhinolaryngology (E.L., M.-J.P), Clocheville Hospital, Tours, and Pediatric Otorhinolaryngology (G.R., E.-N.G), Armand-Trousseau Hospital, Paris, France.

Editor's Note: This Manuscript was accepted for publication March 22, 1999.

Send Correspondence to Thierry Van Den Abbeele, Service d'ORL Pédiatrique, Hôpital Robert Debré, 48 Boulevard Sérurier, 75019 Paris, France. E-mail: thierry.van-den-abbeele@rdb.ap-hop-paris.fr

Abstract

Objective: To examine the indications and the results of surgical excision of severe subglottic hemangiomas.

Design: Retrospective study and case series.

Setting: Four academic tertiary care centers of pediatric otolaryngology.

Patients: Twenty children were included from 1991 to 1997. All presented with severe subglottic hemangiomas resistant to classical treatments such as corticosteroids and/or CO_2 laser.

Intervention: Six children were operated on using laryngotracheal reconstruction and prolonged stenting by a reinforced Silastic roll and 14 children were operated on by a single-stage laryngotracheoplasty and post-operative intubation in a pediatric intensive care unit.

Results: All patients were successfully decannulated or extubated and have been free from recurrent hemangiomas.

Conclusion: The surgery of severe subglottic hemangiomas is a reliable technique in selected patients and should be considered in corticoresistant or corticodependent, circular, or bilateral hemangiomas.

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INTRODUCTION

Subglottic hemangiomas are rare tumors that account for fewer than 5% of all congenital laryngeal anomalies.[1] They usually develop within the first 6 months of life and are revealed by symptoms of abnormal breathing. These tumors typically develop in the three classical phases of cutaneous hemangiomas, with an initial period of rapid growth during the first few months of life, stabilization for several months, and a third phase of slow regression over months to years. Recent developments in the area of pediatric laryngology have greatly improved the diagnosis of these tumors, but the therapeutic strategy remains controversial because although the natural history of these tumors results in complete regression, severe forms are sometimes life-threatening. The ideal treatment should preserve the normal anatomy of the larynx and have a high success rate and few complications. Most

of the authors have used high doses of corticosteroids, but these are not always effective and their prolonged use can retard growth and cause hypertension or a Cushingoid appearance.[2,3] Tracheostomy alone is considered to be the standard method, but complications, including death, are not unheard of.[4] CO_2 laser surgery has recently been described as a method of choice,[5] but cases of subsequent subglottic stenosis have also been reported.[6] Radiation therapy, cryotherapy, and electrocautery [7] have been proposed (for a review see the report by Davidoff and Filston [8]). Successful results with interferon have been reported in selected cases. The intralesion injection of corticosteroids followed by endotracheal intubation for 3 to 4 weeks has been advocated recently, but in this study, some children required more than 10 injections and/or were intubated more than 3 months.[9,10] Surgical excision is not a new method, since it was first described in 1949 by Sharp et al.[11] for one case of tracheal hemangioma and in 1974 by Evans and Todd [12] to treat three cases successfully by his original technique of "castellated" laryngoplasty, but tracheostomy and stenting were necessary. The recent improvements in the assessment of laryngotracheal stenosis and especially single-stage laryngotracheoplasty (SS-LTR) also indicate that surgical treatment may be suitable for subglottic hemangiomas. A few papers have reported some cases of children successfully treated by SS-LTR since 1989.[13-17] The present study describes the successful surgical treatment of 20 recent cases of severe hemangiomas in four departments of pediatric otolaryngology in France and illustrates the evolution of the surgical techniques over the past few years.

MATERIALS AND METHODS

A total of 20 patients underwent surgery for subglottic hemangioma at the Robert Debré Hospital in Paris (n=7), the Timone Hospital in Marseille (n=8), the Trousseau Hospital in Paris (n=3), and the Clocheville Hospital in Tours (n=2) between 1991 and 1997. The records of these patients were reviewed to determine the morphological appearance of the hemangiomas, the sex, age at initial presentation, age at the time of surgical management of the patients, and any previous treatment.

The surgical techniques used to remove the hemangiomas in the four centers were similar. Briefly, the larynx was opened by a vertical incision through the cricoid cartilage that was extended downward to the first tracheal ring and upward to the thyroid cartilage. A sterile endotracheal tube was placed in the tracheal lumen via the lower part of the laryngeal opening, or via a separate lower incision through the third and fourth tracheal rings. The hemangioma was then dissected out using an operative microscope and middle ear instruments; particular care was taken to find a plane between the hemangioma to preserve the mucosa. There was no significant bleeding during removal of the tumor. However small arterial pedicles were found passing through the cartilage in some cases, and these were removed using microbipolar cautery. The first few cases had previously been tracheotomized for several months, and were operated on to remove the tracheostomy. Two of these children had an associated severe subglottic stenosis. The surgical technique used was classical laryngotracheal reconstruction with an anterior cartilage graft and postoperative stenting with reinforced Silastic for a few weeks. This technique has gradually evolved to single-stage procedures since 1992 to avoid a tracheostomy. The histograms in Figure 1 show the changes in surgery from 1991 to 1997.

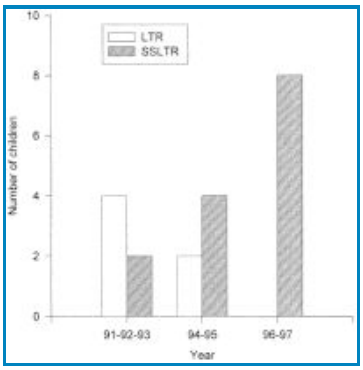

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Fig. 1. Histograms illustrating the evolution of the surgical procedures from 1991 to 1997.

RESULTS

Patients' Clinical Features

The classic predominance of female patients with subglottic hemangiomas was particularly marked, the sex ratio being 5:1. The mean age at surgery was 10 months, with extremes of 2.5 to 40 months. Figure 2 shows the age of the children with a peak between 6 and 18 months. Figure 3 summarizes the topography of the subglottic hemangiomas. Most of them were bilateral

(40%) or circumferential (30%) (Fig. 4). More than half were associated with extralaryngeal hemangiomas. The narrowing of the subglottic area was often difficult to quantify according to the classification of Cotton,[6] but exceeded 80% in two thirds of the cases. One case was a complete grade 4 after CO_2 laser therapy, and another case was a grade 3 probably due to tracheostomy. The mobility of the larynx was normal in all cases, except for one child who had a postoperative unilateral paralysis after the surgical treatment of a giant cervical hemangioma that required ligaturing of the external carotid artery.

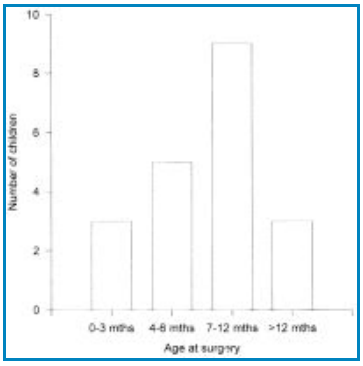

Fig. 2. Distribution of the children by age.

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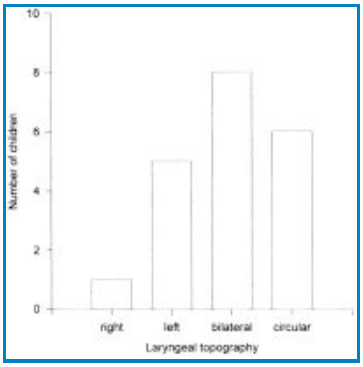

Fig. 3. Topographic localization of the hemangiomas.

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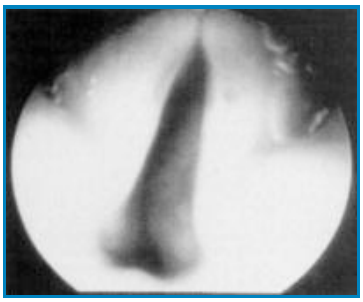

Fig. 4. Endoscopic view of a bilateral subglottic hemangioma before surgery.

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Table I summarizes the previous treatment undergone by these children. All were given corticosteroids as initial therapy.

Corticotherapy was for longer than 1 month in 70% of cases and longer than 3 months in 30% of cases. Growth retardation (70%) and Cushingoid appearance (45%) were the most frequent side effects. Severe hypertension was reported in only 25% of the children. The other treatments were CO_2 laser (5 cases), intubation for 7 to 10 days (7 cases), and tracheostomy (5 cases). We separated the analysis of the anatomical and functional results in two categories according to the surgical technique used.

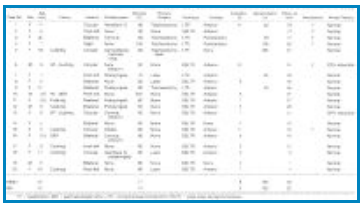

TABLE I. Clinical Characteristics of Patients.

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Laryngotracheoplasty With Tracheostomy and Silastic Calibration

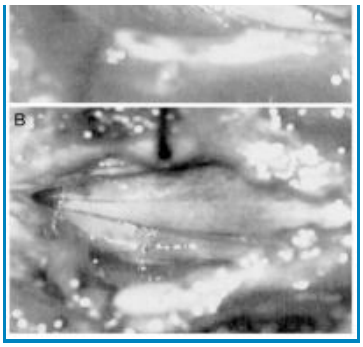
The six children treated by this technique included five who had an existing tracheostomy and were operated on at 19 ± 13 months to remove the tracheostomy. A cricoid enlargement was necessary in the five children by the means of an anterior cartilage graft and two of them by an additional posterior graft. The last case did not respond to sequences of CO_2 laser treatment and required a tracheostomy. Surgical excision was performed at the same time, and this child did not require a cartilage graft. The Silastic stent was removed after 36 ± 10 days in five cases and all were successfully decannulated 74 ± 25 days after surgery. The last child (case 5, see Table I) had suffered from right laryngeal palsy, underwent 6 months of stenting with a Montgomery silicone T-tube and five sequences of CO_2 laser treatment before the surgical excision, and was decannulated 15 months after surgery. This child had a tracheal extension of the hemangioma involving four tracheal rings and three sequences of CO_2 laser were necessary to treat the tracheal part of the tumor after subglottic excision.

These six children have now been followed-up for a mean period of 60 months. No recurrence has occurred. The mean time of steroid therapy before surgery was 85 days, and 48 days of progressive reduction for endocrinological purposes after surgery. Postoperative complications were limited to one case of left laryngeal palsy and three cases of subglottic granulations requiring a CO_2 laser sequence in two of them. The subglottic airways of all the children are now normal.

Single-Stage Laryngotracheoplasty

Fourteen children were treated using this new technique (Fig. 5). Three of them had been previously treated by CO_2 laser and six others intubated for 8 to 15 days with a recurrence or without significant improvement. The children were operated on at a mean age of 7 ± 3 months. An anterior cartilage graft was used in 12 of 14 cases. The two other cases did not require a cartilage graft, because the laryngeal lumen was large enough after removal of the angioma. All children were successfully extubated after 7.5 ± 3 days. Five children were intubated for only 4 to 5 days, illustrating the trend to reduce the duration of intubation. The mean follow-up is 27 months (6 mo-6 y). The mean time on corticosteroids before surgery was 84 days, and 20 days after surgery. No severe postoperative complications were found. Three cases showed subglottic granulations, but CO_2 laser treatment was required in one case. The three children that presented postoperative inflammatory granulations were intubated longer than 10 days and were not given gastroesophageal reflux therapy. The subglottic area was normal in 12 of 14 children and there was no more than 20% to 30% narrowing of the subglottis in the two remaining children. Two children had a severe recurrence of cervical or facial hemangiomas several months after surgery but without any significant reduction of the airway patency, indicating the complete removal of the "tumor" during surgery (Fig. 6).

Fig. 5. Perioperative view of the subglottic hemangioma. **A.** Before dissection of the hemangioma. This is a macrolens photograph showing bilateral hemangiomas that were excised and there remains a strip of normal posterior tracheal wall. Note the extension from the subglottic area to the two first tracheal cartilage rings. **B.** After dissection. The mucosa has been replaced over the



cricoid cartilage

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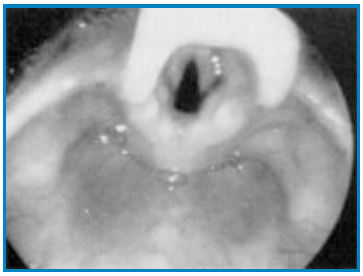

Fig. 6. Endoscopic postoperative view of the subglottic area.

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DISCUSSION

This study describes a large number of patients with subglottic hemangioma that were treated surgically. It confirms the good results obtained by others. The success of this approach is owing to the recent improvements in the surgical techniques used to manage subglottic stenosis in children. SS-LTR was not a routine technique at the beginning of our study, and the severity of the cases with presence of a tracheostomy persuaded us to perform classical LTR with Silastic stenting. SS-LTR has since become the technique of choice for treating subglottic stenosis. This remarkable development has many beneficial consequences for the surgical treatment of subglottic hemangiomas. First, tracheostomy is no longer necessary. Second, the period of postoperative stenting can be reduced from 2 months to 1 week and perhaps less. Third, there is less risk of postoperative complications with SS-LTR. Granulations were the most frequent and occurred in 50% of the LTRs, but they occur in only 21% of the SS-LTRs. The mean stenting time seems a crucial factor of subglottic granulations, because the mean intubation time was significantly longer in cases of granulations (12.3 ± 2.3 d, n=3) than in the other children (6.3 ± 1.5 d, n=11, $P < .0001$). These results indicate the benefit of reducing the postoperative intubation time, and thereby minimizing the risk of spontaneous extubation. A recent study reported SS-LTR in subglottic stenoses with immediate extubation after surgery.[18] Our patients all spent their postoperative periods in pediatric intensive care units, and the children were mildly sedated for 2 to 3 days, with no pharmacological paralysis. Some authors reported recently two cases of surgical excision of subglottic hemangiomas without interposition of an anterior cartilage graft. Only three of our children were operated on without any cartilage graft. The outcomes of these cases were good, but this sample is too small to evaluate the risk of subglottic stenosis with or without graft. The use of cartilage in cases of major injury to the mucosa during the surgery or a previous laser vaporization probably prevents the occurrence of a subglottic stenosis. We found that a previous laser therapy did not lead to postoperative complications.

Our mean follow-up is now longer than 3 years and there has been no laryngeal recurrence of the hemangiomas. Two children developed new cervical hemangiomas, but the subglottic area was not involved, suggesting that the surgical removal was complete. Prolonged corticoid treatment was stopped in all cases and the mean postoperative period on corticosteroids was less than 1 month. Thus our experience demonstrates that open laryngeal surgery is a reliable method for managing severe subglottic hemangiomas. Although classical LTR with tracheotomy and prolonged stenting could raise some criticisms in a disease that regressed spontaneously, the new technique of SS-LTR with short-term intubation (5 days and perhaps less) has many advantages and does not require tracheostomy. This technique provides a rapid (less than 1 week) and definitive solution to the

challenging problem of severe subglottic hemangiomas. Most of our cases were bilateral or circular hemangiomas. However, six cases of our series were unilateral hemangiomas and required a surgical removal. These cases had no response to laser surgery and/or had a large surface of implantation and a high risk of subglottic stenosis after laser surgery. This approach should be considered for all cases that require a tracheostomy, such as bilateral or circular subglottic hemangiomas, corticodependence, corticoresistance or side effects of corticosteroids, when laser or other treatments have failed.[17] We believe that the surgical excision of subglottic hemangiomas will continue to benefit from the future refinements of subglottic stenosis surgery.

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