the detection of the wall of the capsule and the normal pituitary tissue, which contributes to a successful open of the capsular wall to communicate with the subarachnoid space and protects the function of the normal pituitary. Therefore, transsphenoidal approach is a safe and effective way, but neurosurgeons should be cautious to postoperative cerebrospinal fluid leakage and intracranial infection. Autologous adipose tissue filling, artificial meningeal repairment, and large mucosal flap application are fundamental for the prevention of cerebrospinal fluid rhinorrhea.

In order to reduce CSF rhinorrhea, some scholars have tried to apply a less invasive way to treat ICAs. Oyama, K[15] performed transsphenoidal cyst cisternostomy. The bone is fenestrated from the upper third of the enlarged sellar floor to the planum sphenoidale, formingt a 1 cm long, "I" shaped opening on the dura, followed by the endoscopic exploration. Afterward, the arachnoid wall is broken up to communicate the cyst with the prechasmatic cistern, and finally the dura defect is repaired with artificial membrane. This surgical procedure has advantages of less invasive vulnus, and it seems that the CSF rhinorrhea is significantly reduced. Only 1 of the 6 patients received this treatment had a severe cerebrospinal fluid leak. In addition, since there is no direct access to the pituitary fossa, damage to normal pituitary tissue can be maximally avoided. Shim KW[16] adopted another way called transventricular endoscopic fenestration. All patients underwent the endoscopic transventricular approach with neuronavigational guidance, a burr hole was made 1 cm in front of the coronal suture and 2 to 3 cm lateral to the midline subsequently, the right lateral ventricle was tapped using a ventricular catheter inserted through the burr hole. A peel-away catheter was then inserted through the tract a small hole was made just behind the optic chiasm, closely anterior to the infundibular recess with grasping microforceps. Hence, the author could deal with the cyst in the narrow space with the help of endoscope. This surgical approach avoids the complications of the transsphenoidal approach, well-suited for cases of enormous arachnoid cysts in the sellar region extending to suprasellar without hydrocephalus. However, since the normal pituitary gland may be an obstacle for this surgery, pituitary damage arise probably.

CONCLUSION

The IACs are such rare disease in clinical practice that should be fully differentiated. The transsphenoidal approach is the main treatment, but the sellar floor reconstruction should be conducted strictly because of the high rate of CSF rhinorrhea, our experience is that the big mucosal flap repairment is probably necessary. Less invasive approach like transsphenoidal cyst cisternostomy and transventricular endoscopic fenestration are also alternative in some selected cases.

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A Life Threatening Subglottic and Mediastinal Hemangioma in an Infant

Serap Sahin Onder, MD, Ozgul Gergin, MD, and Burak Karabulut, MD

Abstract: Subglottic and mediastinal hemangioma are rare benign vascular tumors of childhood. They cause potentially life threatening condition which requires intervention. Several therapeutic options have been described in the literature with varying degrees of success and complications. We report a case of a stridulous 2-month old female infant with mediastinal and subglottic hemangioma. The child was treated with propranolol without the need for

From the Umraniye Research and Education Hospital. Department of Otolaryngology, Istanbul, Turkey.
Received November 27, 2018.
Accepted for publication December 15, 2018.
Address correspondence and reprint requests to Serap Sahin Onder, MD, Umraniye Research and Education Hospital. Department of Otolaryngology, Istanbul, Turkey; E-mail: serapsahinonder@gmail.com
The authors report no conflicts of interest.

ISSN: 1049-2275
DOI: 10.1097/SCS.0000000000005340

tracheostomy or any other surgical intervention, and with no reported side effects. Propranolol is an effective, non-invasive treatment for life threatening infantile hemangiomas compressing the airway, should be used as a firstline treatment for subglottic hemangiomas when intervention is required.

Key Words: Infant, mediastinal hemangioma, propranolol, subglottic hemangioma

Infantile hemangiomas (IHs) are cited as the most common tumor of infancy. It affects from 4% to 10% of infants.[1] Usually, they are asymptomatic at birth and appear within the first month of life, enlarge till 6 to 9 months, and involute in 3 to 7 years.[2] Obstruction of developing airway is usually present by the first and the second month of the life with stridor, respiratory distress and feeding difficulties. Subglottic and mediastinal hemangiomas are rare benign tumors that can cause life-threatening airway compromise.

Management of airway IHs can be challenging. For decades, prednisolone, at doses ranging from 2 to 5 mg/kg/day for several months, was the first line medical therapy for IHs. The other options included interferon alfa and vincristine. However, Leatute-Lebreze et al[3] first reported propranolol use for infantile capillary hemangioma in 2008. Denoyelle et al has studied about the role of propranolol for infantile laryngotracheal hemangioma.[4] Due to these case reports, propranolol has become the mainstay of the pharmacological therapy of IHs. Following days after initiation of propranolol therapy dramatic response have been reported for both cutaneous and airway IHs.[5] Propranolol use can also prevent the incidence of common adverse effects due to prolonged high-dose steroid use.

We report a case of an infant with life-threatening multiple subglottic and mediastinal IHs successfully treated with propranolol.

CLINICAL REPORT

A 2-month-old female infant was referred to our department with biphasic stridor and respiratory distress. First evaluation of her airway using rigid laryngoscopy revealed foreshortened aryepiglottic folds and right-sided subglottic multiple IHs (Fig. 1A–C). Supraglottoplasty was performed using cold steel instruments. Ultrasound of the brain and abdomen did not reveal any other lesions. Treatment with oral propranolol was started at 1 mg/kg/day; subsequently, the dose was increased progressively to 2 mg/kg/day administered in three divided doses. The patient tolerated propranolol monotherapy without adverse effects. Within 3 days her stridor resolved. However, she continued to have an intermittent biphasic stridor while crying and feeding. We have repeated the laryngoscopic examination 1 month later demonstrated %50 improvement in her proximal subglottic IHs. But there was no response in her distal IHs to propranolol treatment (Fig. 1B–D).

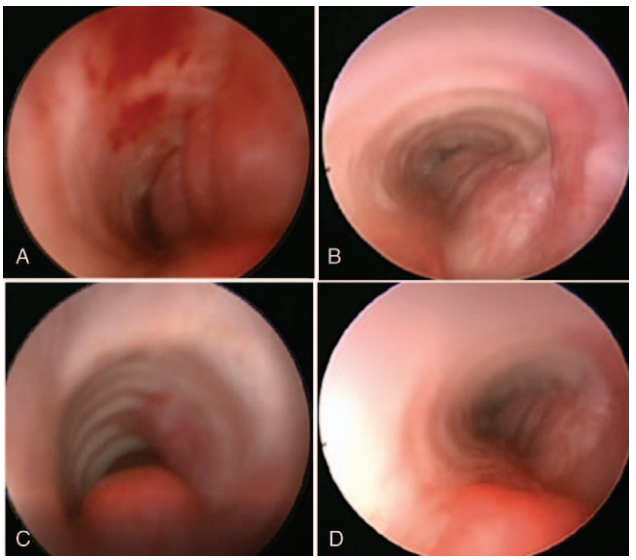

FIGURE 1. The view of distal tracheal segment hemangioma; before propranolol treatment (A), after 1 month of propranolol treatment (B), the view of posterior subglottic hemangioma; before propranolol treatment (C), after 1 month of propranolol treatment (D).

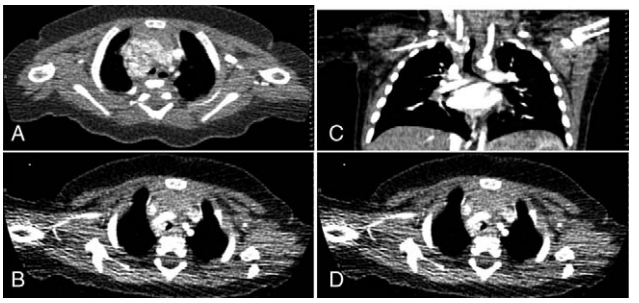

FIGURE 2. CT imaging of mediastinal hemangioma in axial views; before propranolol treatment (A), after 3 months of propranolol treatment (B). The lesion is identified by the arrow. CT imaging showing mediastinal hemangioma in coronal views; (C) before propranolol treatment, (D) after 3 months of propranolol treatment. The lesion is identified by the arrow.

Computer tomography image (CT) with contrast of the neck was obtained postoperatively, and it revealed a large mediastinal contrast enhanced mass compressing the airway confirming the diagnosis of IHs (Fig. 2A–C). The current treatment was continued as there were no feeding difficulty and cyanosis attacks. The control CT which was performed three months later after the first CT demonstrated a % 90 reduction in size of the intra-thoracic IHs (Fig. 2B–D). At 1 year follow-up, the stridor had resolved and now she has no symptom.

DISCUSSION

Subglottic and mediastinal IHs are rare entities that can be potentially life threatening because of obstruction of the airway during their proliferative period. To the best of our knowledge, only two documented cases of mediastinal and subglottic IHs had been reported in the literature until date.[6,7] Symptoms appeared the age of 2 months. Biphasic stridor, respiratory distress, cough, dysphagia, vomiting and hemoptysis are the most common symptoms.[8] Our patient had only biphasic stridor and respiratory distress and no feeding difficulty. Even though flexible fiberoptic nasolaryngeal examination performed routinely in out-patient clinical settings, these evaluations don't always shed light upon the diagnosis. Diagnostic process can be difficult especially in child with respiratory distress. Diagnosis must be confirmed by proceeding directly to laryngoscopy and bronchoscopy under general anesthesia. Biopsy is not usually necessary to establish the diagnosis. Other radiological examinations such as CT and magnetic resonance imaging (MRI) may help identify the presence of mediastinal IHs and exclude other space-occupying lesions. Investigation at this age group often requires general anesthesia or sedation. In this case, first we performed direct laryngoscopy and bronchoscopy. After the propranolol treatment for 1 month of duration the control laryngoscopic examination showed there was no response in her distal IHs. Our pediatric radiology department

offered chest and neck CT for rapid evaluation without general anesthesia. CT showed features of large intra-thoracic hemangioma compressing the distal trachea. So we concluded that, chest imaging should be kept in mind when subglottic IHs causing airway obstruction were encountered.

Various medical treatments such as systemic and intralesional corticosteroids, sclerotherapy, interferon alfa, vincristine have been proposed as primary treatment for IHs over time.[9] Steroid medications can inhibit growth of the lesion during the proliferative phase with success rates ranging from %30 to %93.[10] However, long-term management of steroids' adverse effects may be intolerable.[8] Interferon has been used with success in treating IHs but it has a significant risk for neurotoxicity. Therefore interferon should only be considered when all other traditional modalities fail.[11] Vincristine also has been reported to be effective, but there are many reports supporting this treatment modality for IHs.[12] In the cases that are not responsive to medical therapy, operative interventions may include laser ablation, tracheotomy and intralesional injection of corticosteroids. In our case; tracheotomy was not an option because of distal localization of the mediastinal lesions. Because of the huge mediastinal IHs, laser ablation was not appropriate method for this case.

Cases of mediastinal hemangiomas are extremely rare. In 2008, Tan et al reported an infant with a mediastinal hemangioma causing tracheal airway compression that was successfully treated with midline sternotomy approach.[13] Discovery of propranolol for IHs was an incidental finding in 2008. In 2008, Leaute-Labreze et al reported that propranolol which ordered for obstructive cardiomyopathy in a patient age of 1 month, has caused a decrease in size of cutaneousa nasal hemangioma of the patient.[3] Since this time the efficacy of propranolol in treating cutaneous IHs has been well established in literature. Despite the several reports about the propranolol use in airway IHs, there is no widely accepted guidelines regarding duration and optimal dosing of propranolol. Therefore, many institutions confidence in personal experience rather than scientific evidence.

Hardison et al reviewed the literature and reported a meta-analysis about the use of propranolol in the treatment of subglottic IHs. The authors suggested that treatment should be continued through at least 12 months of age and up to 18 months.[14,15] The stoppage time of treatment in our case was 12 months of age. In the same study the authors claimed that the appropriate dose of propranolol might be 3 mg/kg/day.[14] In our case we used 2 mg/kg/day propranolol and we did not encounter any side effect.

Truong et al described a case with subglottic and mediastinal IHs; multiple treatments modalities have been used such as CO_2 laser ablation, oral dexamethasone, intralesional corticosteroid injection, open submucosal resection of the subglottic hemangioma with an anterior cartilage graft reconstruction but unfortunately they reported that respiratory distress did not resolve. Treatment with oral propranolol resulted in resolution of symptoms.[6]

We first started the propranolol in a dose of 1 mg/kg/day. The patient tolerated that dose properly, therefore, we decided to proceed to the dose of 2 mg/kg/day. After supraglottoplasty inspiratory stridor decreased in 48 hours. However, expiratory stridor resisted. We suggested that this was due to resolution of supraglottic obstruction by supraglottoplasty. In control bronchoscopic evaluation at the time of first month, even propranolol has affected properly the resolution was not as much as expected due to huge mass effect of mediastinal lesion. Therefore, it took an additional month (totally 2 months after diagnosis) for regression of tracheal compressive symptoms.

Propranolol has become the standard of care in the treatment of IHs. Our case revealed that propranolol is effective as a first line treatment for life treating airway IHs If there are no response to propranolol treatment in airway IHs, chest imaging should be performed The treatment with propranolol avoids the common adverse effects of prolonged high-dose steroid use, complications of open surgery and tracheotomy. However, controlled trials and long term follow-ups are needed to determine the ideal dose and duration of the treatment.

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The Superficial Temporal Artery Island Flap: An Option for Moustache Reconstruction

Bilgen Fatma, MD, Ural Alper, MD, and Bekerecioglu Mehmet, MD

Abstract: Vascular anomalies are a group of lesions originating from blood vessels and lymphatics with varying histology and clinical