Conservative Management of a Trans-Spatial Infantile Subglottic Hemangioma

Abstract

- Objectives: The purpose of this presentation is to discuss the role of propranolol in treatment of a severe infantile hemangioma (IH) affecting the airway.
- Study Design: Case report and literature review.
- Methods: This is a case report describing a five-week-old patient with a subglottic hemangioma presenting with stridor and respiratory distress.
- Results: The diagnosis was made based on history, exam, direct laryngoscopy and bronchoscopy (DLB), and imaging studies. Patient was intubated and medical management began after imaging and biopsy was performed. The patient responded well to standard therapy with propranolol and is now asymptomatic.
- Conclusions: Subglottic hemangiomas are a uncommon cause of respiratory distress in patients, but may still be managed conservatively with propranolol even with extension into multiple deep neck space compartments.

Introduction

- Infantile hemangiomas (IH) are the most common benign tumor of childhood and most common tumor in the head and neck.
- While the prevalence of IHs may be as high as 10%, airway hemangiomas are significantly less common.
- Subglottic hemangiomas are typically asymptomatic at birth, but present in the first two months of age with biphasic stridor, respiratory distress, and feeding difficulties.
- Patients may even present with a history of "recurrent croup" during the neonatal period.
- Previously proposed treatment options of IH include beta blockers, systemic and intra-lesional steroids, laser, tracheotomy, or open surgery depending on the extent of the tumor.
- Over the past decade, management of airway hemangiomas has transitioned from primarily surgical to medical.
- Beta blockers including propranolol, timolol, and nadolol have all proven efficacious for rapid treatment of IH.
- While the mechanism of action of beta blockers is not fully understood, studies have proposed propranolol may work by promoting vasoconstriction, inhibition of vasculogenesis and angiogenesis (via inhibition of VEGF production), and inactivation of the renin-angiotensin system.1
- Side effects of medications are rare, but include hypotension, bronchospasm and hypoglycemia.2
- Some patients may require adjunct treatment with intralesional steroid injection or CO2 laser treatment after treatment with beta blockers.3
- Subglottic hemangiomas extending to the mediastinum may recur even after multiple laser treatments, presenting as a concerning problem for patients with more extensive disease.4

Case Report

- Five week old (ex 34 week gestation age) girl presented to the emergency department with progressively worsening stridor.
- Patient had no history of intubation or NICU stay.

Figure 1. Direct Laryngoscopy Pre-Treatment. Direct Laryngoscopy view of subglottic hemangioma (a) Inspiration (b) Exhalation

Figure 2. CT Images. CT results of hemangioma extending from skull base to mediastinum

Figure 3. MRI Images. MRI results of hemangioma extending from skull base to mediastinum

Results

- The diagnosis was made based on history, exam, DLB, and imaging studies.
- DLB: 90% subglottic stenosis (Figure 3)
- Prompting intubation for airway management
- CT Neck: large vascular mass extending from skull base to superior mediastinum; concern for kaposiform hemangioendothelioma (Figure 3)
- MRI neck: vascular mass extending suprahyoid neck to mediastinum, right deviation of trachea; no intralesional flow voids (Figure 3)
- Concern for extent of tumor with possible need for intervention prompted transfer to tertiary care pediatric hospital.
- Patient underwent ultrasound guided biopsy consistent with IH and started on propranolol.
- Patient responded well and was extubated in OR 72 hours after treatment initiation (Figure 4).
- The patient is now asymptomatic.

Figure 4. Direct Laryngoscopy Post-Treatment. Direct Laryngoscopy view of hemangioma after 3 days of propranolol (a) Glottis (b) Subglottis

Discussion

- Otolaryngologists must have a high index of suspicion for airway hemangioma when evaluating a newborn patient with stridor and history of progressively worsening stridor.
- Subglottic hemangioma should be suspected when patients manifest cutaneous hemangiomas in the "beard" distribution.
- Subglottic hemangiomas have an increased risk of airway compromise and death if left untreated.
- Literature evaluating treatment of airway hemangiomas across multiple hospitals has demonstrated similar efficacy in treatment of airway hemangiomas compared to surgical intervention, but with reduced risk of complications.3
- Irrespective of the size of airway hemangiomas, propranolol is considered first line treatment.3

Conclusions

- This case demonstrates that a large subglottic hemangioma extending into deep neck spaces and mediastinum may still be managed successfully with propranolol without the need for surgical airway management.

References


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