

ABSTRACT

Subglottic hemangioma is a rare congenital anomaly of the airway that is potentially life threatening with a high mortality rate. Without a high index of suspicion, the diagnosis can easily be missed. Herein, we present a case of a 2-month-old girl who presented as recurrent croup, who was diagnosed with a subglottic hemangioma on direct laryngoscopy and bronchoscopy. Oral propranolol was initiated, with resolution of stridor within 24 hours of diagnosis.

INTRODUCTION

- Infantile hemangiomas are one of the most common benign tumors of infancy and affect 4-10% of infants [1,2]
- May be seen on both the skin and in the airway
- Subglottic hemangiomas are rare, cited as 1.5% of congenital anomalies
- May cause acute airway obstruction, with mortality rates approaching 50% if left untreated [3,4]
- May be difficult to diagnose as they are not easily appreciated on awake bedside laryngoscopy or suspected in absence of a cutaneous facial hemangioma, leading to a delay in diagnosis [5,6]
- Diagnosed by direct laryngoscopy and bronchoscopy; imaging (i.e. CT or MRI) may help lead to diagnosis [7]
- Treated with multiple treatment modalities: open tracheotomy, open resection, laser ablation, systemic and intralesional steroids, and most recently, systemic propranolol

CASE PRESENTATION

- 2-month-old girl with intermittent stridor and URI starting at 2 weeks of life
- Healthy, born full-term, no history of intubations or cyanotic episodes
- Worse with feeding, + adequate weight gain
- Transiently improved with racemic epinephrine and Decadron during multiple prior hospitalizations for treatment of presumed laryngotracheobronchitis
- Negative lateral neck XR, echocardiogram, Barium esophogram
- PE: afebrile, intermittent tachycardia, no desaturations, + inspiratory stridor and barking cough
- Bedside NPL exam with possible subglottic thickening, otherwise WNL with no evidence of laryngomalacia
- CBC WNL, viral panel negative
- Started on empiric Decadron and racemic epinephrine
- Persistent symptoms and no evidence of acute infection → OR for direct laryngoscopy and bronchoscopy (DLB)
- OR (day #4 of admission): DLB, right posterior partially obstructive subglottic hemangioma visualized (Figure 1); patient extubated and transferred to ICU



Figure 1. Right posterior subglottic hemangioma.

- Hematology consulted and EKG obtained to initiate propranolol hours after surgery
- Patient started on 1 mg/kg/day of oral propranolol (given TID) and titrated to 2 mg/kg/day on POD#1
- Patient with resolution of stridor within 12-24 hours of treatment with propranolol

DISCUSSION

- Natural course of infantile/subglottic hemangiomas: proliferate in first 1-3 months of life where patient becomes symptomatic with spontaneous involution by 12 months [8,9]
- Literature review/meta-analysis: 49% had other hemangiomas at time of diagnosis (i.e. beard distribution, PHACES syndrome) [10]
- Must have high index of suspicion in absence of cutaneous hemangioma; if fails medical therapy/does not improve as expected in 7-10 days → low threshold for DLB [6]
- Efficacy of propranolol has revolutionized treatment, but no standardized dosing or treatment duration guidelines exist
 - Clinical response noted almost immediately
 - Starting dose in literature varies, most sources titrate to 1-3 mg/kg/day with TID dosing regimens
 - Suggested to treat at least until 12 months of age, though age and duration varies in the literature [10]
 - Concomitant treatment with steroids shown to increase risk of treatment failure [10]
- In our experience, administering *albuterol* to patients with subglottic hemangioma causes resumption/worsening of symptoms, and we recommend it is listed as an Allergy in patients with this diagnosis

CONCLUSIONS

- Subglottic hemangiomas are a rare and potentially life threatening disease entity
- Represent a common presentation of an uncommon problem, leading to a delay in diagnosis and treatment
- Must have high index of suspicion and low threshold for direct laryngoscopy and bronchoscopy if patient fails to significantly improve with medical management
- Oral propranolol safe and effective treatment with quick resolution of symptoms
- Consider listing *albuterol* as Allergy in patients with subglottic hemangioma

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