Propranolol Treatment for an Isolated, Obstructing Subglottic Hemangioma

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ABSTRACT

Objective: To present a patient with an isolated subglottic hemangioma who responded successfully to propranolol therapy

Methods: (1) Retrospective review of patient records and endoscopic documentation; (2) Review of the medical literature pertinent to the use of propranolol in subglottic and airway hemangiomas.

Results: A 6-month-old, previously full-term male with no history of prior intubations was admitted for evaluation of respiratory distress, biphasic stridor and recurrent ‘croup’. Microlaryngoscopy and bronchoscopy revealed a subglottic hemangioma obstructing 90% of the subglottic airway. Propranolol and prednisone therapy was immediately initiated following cardiology evaluation. The patient’s stridor and respiratory distress rapidly resolved, and a repeat airway evaluation 3 days following the start of propranolol showed 80% airway obstruction. Four days later, the hemangioma had contracted further to 60% obstruction. The patient remained asymptomatic and the subglottic hemangioma slowly decreased in size over the ensuing 4 months after the patient was weaned from prednisone. No additional therapy was necessary for management of this patient’s subglottic hemangioma.

Conclusion: Propranolol should be considered for first-line therapy in the treatment of isolated subglottic hemangiomas. Although most patients with subglottic hemangiomas demonstrate a positive response to propranolol, the rate and degree of response can be unpredictable, with some patients still requiring more traditional therapies or surgical intervention.

INTRODUCTION

Hemangiomas are the most common benign tumor of infancy. The natural course of infantile hemangiomas has been well documented, with most becoming apparent within the first two months of life and entering a proliferative phase with variable growth. Hemangiomas located within the subglottis can cause progressive and potentially life-threatening airway obstruction1. Subglottic hemangiomas have been managed with a variety of treatment modalities, including systemic and intralesional steroids, laser therapy, systemic chemotherapy, open resection, and tracheostomy2,3.

Propranolol, a non-selective beta blocker, was introduced as a novel modality for the treatment of cutaneous hemangiomas in 20083, and is now being readily used as an alternative, non-surgical treatment for airway hemangiomas. Although most authors report positive results, it remains apparent that all patients with airway hemangiomas do not respond in a predictable fashion.

CASE PRESENTATION

A 6-month-old, previously full-term male with no history of prior intubations was admitted for respiratory distress and biphasic stridor. Prior to otolaryngological evaluation, he had multiple admissions and emergency room visits for recurrent ‘croup’. The patient was taken to the operating room where microlaryngoscopy and bronchoscopy revealed a subglottic hemangioma obstructing 90% of the subglottic airway (Figure 1).

Propranolol and prednisone therapy was immediately initiated following cardiology evaluation. A baseline electrocardiogram was obtained, and propranolol was slowly titrated up to 2 mg/kg/day divided into 3 doses. The patient’s stridor and respiratory distress rapidly resolved. A repeat airway evaluation 3 days following the start of propranolol therapy showed a slight decrease in the size of the subglottic hemangioma (Figure 2). Four days later, the hemangioma had contracted further to 60% obstruction (Figure 3). The only side effect experienced by the patient was transient diarrhea.

The patient remained asymptomatic and the subglottic hemangioma slowly decreased in size over the ensuing 4 months (Figure 4). No additional therapy was necessary for management of this patient’s subglottic hemangioma. His care was transferred to a pediatric otolaryngologist in another state after his family moved for social reasons. At his last follow-up visit, he was asymptomatic. He is currently 1.5 years old and remains on propranolol therapy.

DISCUSSION

The utility of propranolol in the treatment of hemangiomas was serendipitously discovered when cutaneous hemangiomas dramatically improved after propranolol treatment was started in patients with concurrent cardiac disease4. The application of this therapy to the management of subglottic and airway hemangiomas has changed the treatment paradigm. A thorough review of the literature reveals multiple case series with successful results using propranolol as an adjuvant treatment to other medications or surgery1,2,4. In addition, there have been case reports demonstrating positive outcomes using propranolol as the sole treatment for airway hemangiomas5. The mechanism of action of propranolol is unclear, but theories include vasoconstriction, inhibition of angiogenesis, and induction of cellular apoptosis1,5,6.

A standardized universal protocol has not been established, but most authors recommend obtaining a baseline electrocardiogram and initial hospitalization2. An initial dose of 0.5-1 mg/kg/day is started, increasing to 2-3 mg/kg/day divided into 2-4 doses as tolerated1,4,6. Therapy is continued through the proliferative phase or until no further improvement occurs, then gradually tapered. If rebound growth occurs, propranolol therapy can be restarted. Serious side effects are rare and include bradycardia, hypoglycemia, and hypotension1,5. Further studies are needed to determine dosing strategies and long-term effectiveness of propranolol for the treatment of hemangiomas. Responses to propranolol can vary so close observation and repeat endoscopies may be necessary. Some subglottic hemangiomas do not respond and may still require more conventional therapies.

CONCLUSIONS

Propranolol should be considered for first-line therapy in the treatment of isolated subglottic hemangiomas. Close monitoring of these patients is imperative because the rate and degree of response can be unpredictable. Although most patients with subglottic hemangiomas demonstrate a positive response to propranolol, some will still require more traditional therapies or surgical intervention.

REFERENCES