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Case Report

Interesting case of subglottic hemangioma presented with stridor

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ABSTRACT

Subglottic hemangioma is a very rare infantile form of hemangioma as compared to cutaneous hemangioma but if left undiagnosed or untreated can be life-threatening. Here we report one case of preterm who presented with sudden onset of inspiratory stridor and respiratory distress in OPD in a previously normal child. The patient was initially misdiagnosed as croup which did not improve after nebulized adrenaline and steroids later on direct laryngobrochoscopy showed subglottic stenosis. A CT scan was done later which revealed subglottic hemangioma which showed complete remission in symptoms after starting oral propranolol. A careful approach towards diagnosing and managing the sudden onset of stridor with probable structural cause is necessary for a favorable outcome.

Keywords: Subglottic hemangioma, Stridor, Subglottic stenosis, Propranolol

INTRODUCTION

Infantile hemangiomas occur in 4% to 5% of the pediatric patients.[1] A subglottic hemangioma is a rare form of hemangioma that affects only 1.5% of congenital abnormalities of the airway and can cause severe obstruction of the airway.[2] It has female predominance and they usually present with some form of respiratory distress more commonly biphasic stridor before 6 months of age.[3]

It is divided into three phases (proliferative, involuting, and involuted).[4] It is usually associated with the presence of a cutaneous hemangioma, but a localized subglottic hemangioma can occur without the presence of a cutaneous hemangioma.[5]

The diagnosis is usually made clinically along with direct evidence from laryngobronchoscopy.[3] Sometimes, the hemangioma appearance on direct laryngobronchoscopy is atypical or can be missed so radiological diagnosis should be used. Computed tomography (CT) scans provide better visualization of the lesion and also the degree of luminal narrowing.[6]

Most patients with subglottic hemangiomas have a good prognosis when diagnosed and treated early and a high mortality rate if diagnosis and treatment are delayed.[7]

CASE REPORT

A one and half month-old patient was brought in OPD with complain of difficulty in breathing and stridor. On admission, the patient was febrile (99.9°F) and had tachypnea (respiratory rate: 68/minute), tachycardia (heart rate: 198/min), and SpO_2: 90% on room air. On examination, inspiratory stridor was heard with moderate chest retraction. A clinical diagnosis of acute severe croup was made, and the patient was managed with heated humidified nasal oxygen, and nebulization with adrenaline and budecortisone. A single dose of dexamethasone (0.6 mg/kg) was administered but there was no significant improvement. Routine investigations (hemograms, C-reactive protein, and blood culture) were within a normal range including chest X-ray.

In view of no improvement in her stridor and retraction, upper airway anomalies were suspected, for which a larnygobronchoscopy was done which revealed Subglottic stenosis type 1. To confirm the diagnosis, a

Contrast-enhanced computed tomography (CT) scan was also performed which revealed avidly enhancing multi-lobulated submucosal soft tissue density lesion involving subglottic trachea causing localized marked luminal narrowing as described suggestive of subglottic hemagioma.

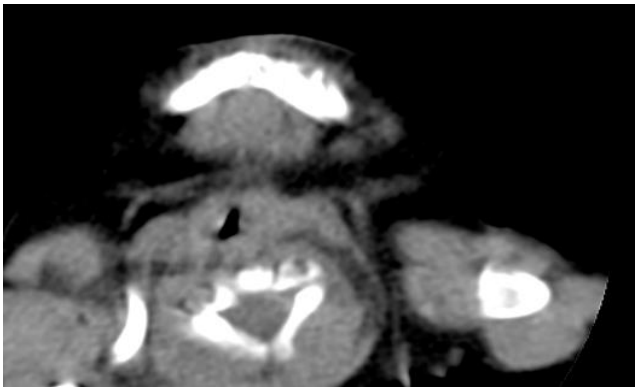

Figure 1: Pre-treatment CECT image showing hemangioma.

Oral propranolol was started at an initial dose of 1 mg/kg/day, which was gradually increased to 2 mg/kg/day in three divided dosages. The patient was closely monitored for HR, BP, and RBS. Clinically patient got better after 2 days of propranolol. Subsequently, the patient was discharged and was called for follow-up weekly for 1 month and then every month. CT scan was repeated after $3^{1/2}$ months which showed a residual very small component of previously noted hemangioma. The patient had normal weight gain along with no recurrence of symptoms and continued on oral propranolol.

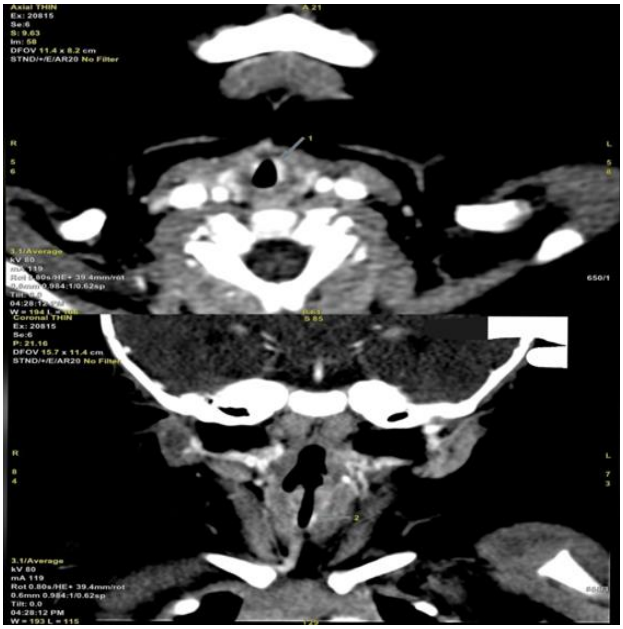

Figure 2: Post-treatment CECT showing tiny hemangioma.

DISCUSSION

A subglottic hemangioma is a common benign vascular growth of infancy that can cause severe obstruction of the airway and its incidence is 4% to 5% of the pediatric population.[1]

A Subglottic hemangioma has a well-defined pattern with a proliferative phase beginning in the first 3 months of life, and then an involution phase at around 1 year of age. Most infants get symptomatic during a proliferative phase in the form of stridor which may progress to respiratory distress. A subglottic hemangioma is often mistaken for a more common condition such as croup.[8]

A subglottic hemangioma mostly occurs in isolation but the presence of cutaneous hemangiomas, especially in the "beard" distribution in an infant along with respiratory distress, may invoke a clinical suspicion of it.[9]

Many treatment options are studied with steroids being used most commonly throughout the decade. Due to its adverse effects and less effective outcome, now steroids are replaced by propranolol as a drug of choice for all hemangiomas including subglottic ones.[10]

A protocol for the dosing of propranolol has been well established with treatment starting with 1 mg/kg/day with a gradual increase in dosing to 2-3 mg/kg/day. A close monitoring of HR, BP, and RBS should be done. The great Ormond Street guidelines advise treatment for 12 months and then tapering over 4 weeks. These children should be monitored closely thereafter for recurrence.[11]

In our case, propranolol was effective in symptomatic relief with no undesirable side effects.

CONCLUSION

A subglottic hemangioma is rare form of hemangioma which with early diagnosis and treatment has a favorable outcome. A direct laryngobronchoscopy along with CT/MRI can aid in its diagnosis. Propranolol has the upper hand on steroids for the management of subglottic hemangioma in terms of adverse effects and better outcomes.

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