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Subglottic haemangioma treated with propranolol – a case report

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Abstract:

Introduction. Haemangioma is the most common tumour in infancy and can occur in about 10% of the population. They are categorized into two types: “infantile” and or rare “congenital”. In some uncommon cases, the haemangioma can appear in the airway, particularly in the subglottic area, often resulting in symptoms of stridor and respiratory distress.

Clinical case. A 5-month-old girl was admitted under Ear, Nose & Throat (ENT) and paediatric intensive care unit (PICU) teams with pre-existing worsening biphasic stridor. She had previously been admitted to a different hospital for inspiratory stridor, as it was suspected she had laryngomalacia with superimposed viral infections. She had a microlaryngoscopy and bronchoscopy (MLB), which revealed a bulging compressible subglottic mass on the right and left sides, consistent with a haemangioma. Following the procedure, a 1 mg/kg/day propranolol treatment was initiated to reduce the size of the haemangioma. The does was gradually increased to 2 mg/kg/day. Six days later, she had a repeated MLB. The subglottis was reassessed, and there was a significant improvement in the size of the haemangioma - the size was reduced by approximately 80%. She was later discharged with a close monitoring for propranolol’s side effects.

Conclusion. Subglottic haemangioma can be successfully treated with 2-3 mg/kg/day of propranolol. The patient must be monitored for propranolol side effects. More research is needed to fully understand the interaction of systemic propranolol and subglottic haemangioma treatment.

Keywords. Subglottic haemangioma, stridor, propranolol.

Introduction

Haemangioma is the most common tumour in infancy and can occur in about 10% of the population. They are categorized into two types: “infantile” and rare “congenital” (1). It can arise in any part of the body in the first few weeks of life. They have a characteristic of progressive growth with spontaneous involution, which is usually completed at the age of 7-10 years (2,3). In some uncommon cases, the haemangioma can appear in the airway, particularly in the subglottic area, often resulting in symptoms of stridor and respiratory distress (4).

Clinically, determining whether a haemangioma is the cause of stridor is difficult due to more common differential diagnoses such as croup or laryngomalacia. An awake fibronasendoscopy can visualize the supraglottis and glottis areas, ruling out vocal cord abnormalities or laryngomalacia. For a definite diagnosis of subglottic haemangioma, microlaryngoscopy and bronchoscopy under general anaesthesia are required to visualize the subglottis region. (5).

The previous treatment options for subglottic haemangiomas were surgical excision, laser or conservative therapies such as corticosteroids (6). It is known that propranolol now is a more effective treatment option which was first described by Léauté-Labrèze et al in 2008 (7). Their study showed a significant reduction in the size of haemangiomas within 24 hours of initiating therapy. The mechanism by which propranolol reduces the size of haemangiomas is unknown, but theories include vasoconstriction, growth factor regulation, and apoptosis (8).

Case report

A 5-month-old girl was admitted under ENT and PICU teams with pre-existing worsening biphasic stridor. She had previously been admitted to a different hospital for inspiratory stridor, as it was suspected she had laryngomalacia with superimposed viral infections. She was referred for ENT to rule out laryngomalacia, however, her ongoing stridor became worse which was difficult to control with dexamethasone and adrenaline nebulizers. She was urgently transferred to our hospital as an airway emergency for urgent ENT review.

The child is premature, born on the 27th week. She has capillary haemangiomas on her back, neck, and one arm. She had no previous history of intubation. She is otherwise fit and well.

On admission, the patient had a respiratory rate of 40-50 breaths per minute and a heart rate of 180-195 beats per minute. She also had tracheal tug and costal recession. Her saturations were 99% on 10L of optiflow. She was also covid positive. ENT decided to perform an urgent microlaryngoscopy with a bronchoscopy due to respiratory distress

She had a microlaryngoscopy followed by a bronchoscopy, which revealed a bulging compressible subglottic mass on the right and left sides, consistent with a haemangioma. Other abnormalities in the supraglottis, glottis, trachea, and bronchus were not discovered. She has also been intubated to maintain the patent airway.

Following the procedure, a 1mg/kg/day propranolol treatment was initiated to reduce the size of the haemangioma. The dose was gradually increased to 2mg/kg/day. She also had a craniofacial magnetic resonance imaging of her head and neck to see if there were other haemangiomas, which turned out to be normal.

She had repeated MLB done 6 days later. Subglottis was reassessed which had a significant improvement in the size of haemangioma - the size was reduced by approximately 80 percent. She was clinically improving, therefore both PICU and ENT teams decided to extubate the patient two days later.

Six days later, she had a repeated MLB. The subglottis was reassessed, and there was a significant improvement in the size of the haemangioma - the size was reduced by approximately 80%. Because she was clinically improving, the PICU and ENT teams agreed to extubate the patient two days later.

The patient was successfully extubated. The patient's vital signs were stable after extubation. Her respiratory rate remained between 30 and 36 breaths per minute, while her heart rate ranged between 103 and 116 beats per minute. After dexamethasone weaning, her breathing was normal on air, with saturations ranging from 96 to 100 percent.

She was sent home two weeks later with regular follow-up appointments with the ENT and paediatric teams because she was still on propranolol to control the size of the haemangioma. After one month of using propranolol, it was discovered that she had no side effects and her repeated MLB was normal with minimal appearance with subglottic haemangioma.

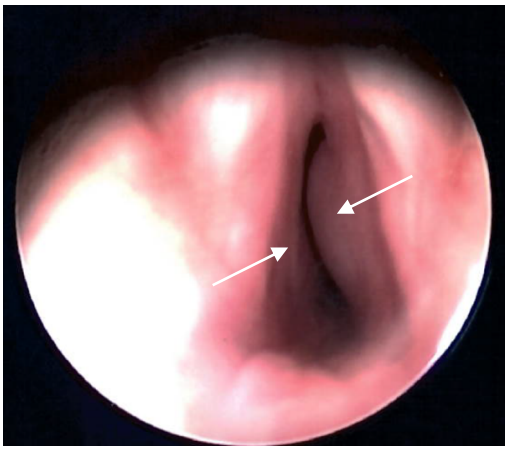

Figure 1. Bulging compressive masses on the right and left with haemangioma appearance

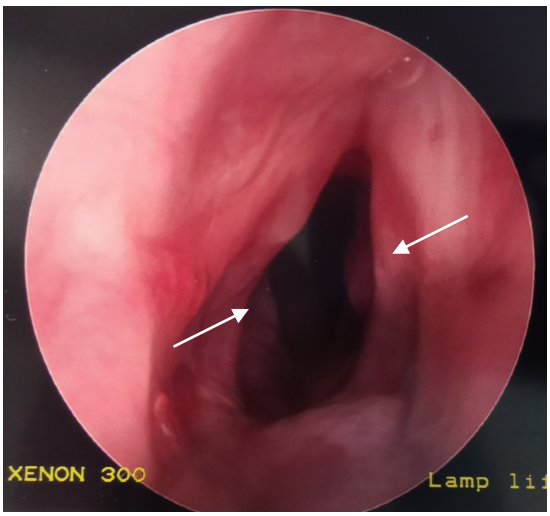

Figure 2. Repeated MLB shows significant improvement in size of haemangioma.

Discussion

Since the initial report by Léauté-Labrèze et al (7) describes propranolol use for treating haemangiomas, there are only a few case reports describing the efficiency and potential side effects of using this medication.

The first case report addressing the efficiency of propranolol in treating subglottic haemangioma was described by Denoyelle et al in 2009 (9). Their study showed that subglottic haemangioma can be successfully treated with 2-3 mg/kg/day of propranolol. Our patient was initially treated with 1mg/kg/day with a good response. The dose was gradually increased to 2mg/kg/day. More research is needed to fully understand the interaction of systemic propranolol and subglottic haemangioma treatment (5).

Before starting treatment with propranolol, patients should be screened for possible risks. The physician should examine cardiovascular and pulmonary systems and include determinations of pulse and blood pressure. Propranolol should not be given to patients with a history of cardiogenic shock, sinus bradycardia, hypotension, second- or third-degree heart block, heart failure, bronchial asthma, and hypersensitivity propranolol. Routine electrocardiogram screening is also recommended before the initiation of propranolol.

Ji et al (11) and Léaute-Labrèze et al (12) did systematic reviews to evaluate the intolerable side effects of propranolol treatment. They found that the most common side effect was severe sleep disturbances. The other side effects were: severe agitation, respiratory disorders, and symptomatic hypoglycaemia, cold peripheries. The most serious side effects such as atrioventricular block, bradycardia, hypotension, bronchospasm, bronchial hyperactivity, and hypoglycaemia were managed by reducing the dose/discontinuing propranolol.

In addition to the safety and efficacy of propranolol therapy in haemangiomas, a higher response rate can be obtained when treatment is initiated early and with a prolonged course of therapy. Propranolol therapy should be continued until there is no more response for two consecutive months, regardless of the age at which therapy was initiated or the duration of treatment (13).

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