A CASE REPORT ON A 4-MONTH OLD FEMALE WITH SUBGLLOTTIC HEMANGIOMA TREATED WITH PROPRANOLOL

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Introduction
• The subglottis is the narrowest portion of the pediatric airway; airway hemangioma in this area induces the most respiratory embarrassment.1
• Propranolol is a non-selective beta-blocker that was serendipitously discovered in 2008 to treat proliferating hemangiomas.2
• This report aims to describe the first reported case of infantile airway hemangioma successfully treated with propranolol in the Philippines.

Case History
• A 4-month old female presented with stridor at three weeks of age; worsened with feeding and not relieved by prone position.
• Initial flexible laryngoscopy showed erythema and swelling over the right subglottic area, fully mobile vocal cords, no masses.
• She was initially treated as a case of Laryngomalacia, given eight doses of intravenous corticosteroids and was maintained on continuous positive airway pressure.
• Persistence of difficulty of breathing resulted to eventual tracheostomy at 2 months of age.

Diagnostics
• Both suspension laryngoscopy and computed tomography scan were done at 4 months of age.
• A Grade 3 stenosis (99% obstruction) from level of true vocal cords down to 1.5 cm inferiorly was identified.

Figure 1. Left: Direct laryngoscopy revealing smooth, reddish, soft, compressible submucosal bulge on the right subglottic area almost completely obstructing the airway. Right: A CT scan revealed a soft tissue density at the Right glottic and subglottic area. The right vocal cord was displaced medially and there was no clearly discernable mass.

Therapeutics
• The patient was treated with propranolol with an initial dose of 0.15mg/kg (0.6mg/kg) twice daily, eventually increased to 0.4mg/kg (1.7mg/kg) twice daily.
• The preparation used was a 4.28/ml oral solution.

Results
• After five months of treatment, multiple episodes of loud cry were noted by the parents.
• Repeat laryngoscopy revealed a subglottic narrowing of < 50% (Cotton-Myer Gr. I), with passage of 3.2 mm flexible scope beyond the subglottis.
• At 11 months of age, the patient was successfully decannulated.

Discussion
• Maintaining airway patency, promoting lesion regression, avoiding tracheostomy and minimizing therapeutic side effects until the lesions enter the natural phase of involution are the treatment goals for patients with infantile hemangioma.
• Treatment options for subglottic infantile hemangioma are steroids, laser ablation, surgery, tracheostomy, and more recently, propranolol.
• A randomized controlled trial by Kim et al. (2017) involving 34 patients from a single institution has shown that therapeutic effects of propranolol were not inferior to that of steroid and that there were no significant difference in terms of safety outcomes between the two treatment options.3
• A dose of 2 mg/kg/day is the most commonly reported dose in the literature. The drug is usually started at a dose of 1 mg/kg/d to 2 mg/kg/d divided 2 to 3 times a day and then increased to 2 mg/kg/d to 3 mg/kg/d over several days to a week.4,6
• Its advantages over other modalities include being non-invasive, of rapid onset, avoids tracheostomy, prolonged steroid therapy, manipulation of subglottic tissues or prolonged periods of intubation.

Conclusion
• Airway hemangioma remains a challenging disease entity in the field of Otorhinolaryngology.
• Propranolol is now considered a safe and effective alternative treatment and may prove to be the best medical treatment for subglottic hemangioma in the future.

Figure 2. The patient with tracheostomy tube at 2 months of age.

Figure 3. The patient at present (4 years old).

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References