



Management of infantile subglottic hemangioma with T-tube placement and propranolol

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TO THE EDITOR,

Subglottic hemangioma (SGH) is a rare benign vascular tumor that accounts for 1.5% of all congenital laryngeal lesions. Usually, the onset of respiratory symptoms occurs soon after birth, during the proliferative phase of the disease.⁽¹⁾ Since most lesions undergo regression during early childhood,⁽²⁾ treatment focuses on the prevention of long-term complications.

SGH can lead to airway obstruction when left untreated, with mortality rates approaching 50%.⁽³⁾ In this scenario, tracheostomy is considered a life-saving procedure, although it may cause life-threatening complications, particularly related to the risk of airway obstruction and decannulation during postoperative care. Because of the scarcity of reports, there are no treatment guidelines proposed for SGH. An alternative approach by means of silicone T-tube placement has been used in order to achieve and maintain airway patency. The following case reports describe the management and postoperative outcome in three infants with SGH. Propranolol has become an accepted first-line treatment for SGH, with its benefits outweighing its risks.⁽⁴⁾

A 7-month-old male infant with recurrent respiratory infection and stridor for five months had a history of intubation and tracheostomy at three months during an episode of airway obstruction at another hospital. He presented with inspiratory stridor, and the remaining physical examination was uneventful. A contrast-enhanced neck and chest computed tomography (CT) showed a faint lateral enhancement at the vocal fold (VF) level, suggestive of SGH. A suspension laryngoscopy under general anesthesia confirmed a left-sided laryngotracheal vascular tumor causing obstruction of the larynx and subglottis. Endoscopic findings were consistent with hemangioma. While still under anesthesia, an 8 mm silicone T-tube was placed supraglottically through the tracheostomy. The patient had mild transient dysphagia with aspiration, requiring nasogastric tube feeding, with complete resolution within five days postoperatively, enabling him to resume oral intake. Propranolol (1 mg/kg/day, *per os*) was initiated and, within 3 postoperative weeks, the daily dose was increased to 3 mg/kg/day, with good tolerance. The patient was discharged from the hospital one week after the procedure with no symptoms or signs of aspiration. Currently, he remains on propranolol with a favorable outcome, and treatment is scheduled for two years.

A 6-month-old female infant was referred to our institution with recurrent respiratory infections and stridor since birth. She developed respiratory failure twice, requiring intubation. CT showed a faint enhancement suggestive of airway obstruction due to SGH (Figure 1B). A suspension laryngoscopy was performed, revealing an SGH right below the VF, causing obstruction of the larynx and subglottis (Figure 1A). In the postoperative course, the patient presented with sustained dyspnea and stridor, which ultimately evolved to respiratory failure and intubation, followed by a tracheostomy after 10 days. One year later, the tracheostomy cannula was withdrawn, and an 8 mm silicone T-tube was placed with its upper end in the supraglottis (Figure 1C), without signs of aspiration. Propranolol (1 mg/kg/day) was then started and increased to 3 mg/kg/day within 14 days and continued for two years. Sixteen months later, the T-tube was removed, and the patient underwent closure of a persistent tracheostomy orifice when she was nine years old.

A 6-month-old female infant was admitted to the emergency department with respiratory failure. She was born at 34 weeks and evolved with gastroesophageal reflux and recurrent respiratory infections. Aside from stridor, physical examination was unremarkable. Neck and chest CT showed a heterogeneous and vascularized mass in the larynx. Rigid bronchoscopy revealed a pulsatile mass causing airway obstruction, suggestive of SGH on the left. Treatment was initiated with propranolol (1 mg/kg/day) and increased in a stepwise progression to 3 mg/kg/day, which was maintained for two years. A rigid bronchoscopy and CT scan at 1 year showed a regression of the SGH. The patient remains on outpatient follow-up without recurrence.

SGH is often diagnosed during the first months of life. The three cases described presented with acute respiratory failure, diagnosed as SGH, and had favorable outcomes. Cases 1 and 2 required tracheostomy and were successfully treated thereon with a T-tube and propranolol, whereas the third was successfully treated with propranolol alone. The patients remained on propranolol for 2 years, monitored by a multidisciplinary team, including a pediatric cardiologist and a thoracic surgeon. During the regular follow-up visits, blood glucose levels, vital signs, and cardiac function were monitored, and the patients had no recurrence.

SGHs are usually found right below the VF, are often unilateral, and may involve both sides. Such tumors

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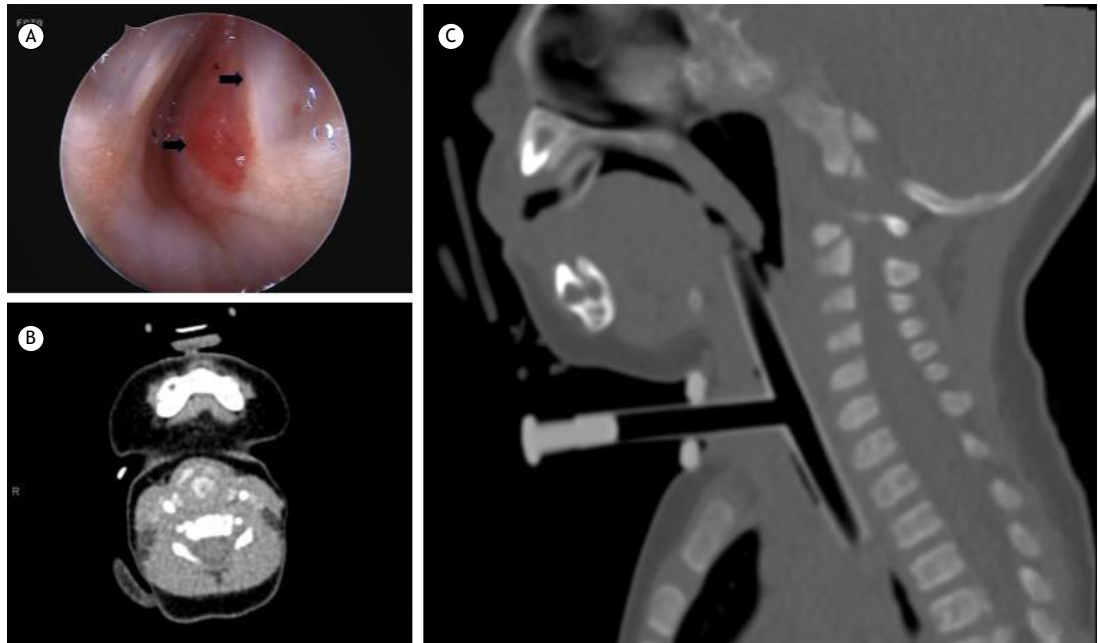


Figure 1. A. Laryngoscopic image showing right subglottic hemangioma (arrows), with airway obstruction. A (arrow): Right vocal cord; B (arrow): Subglottic hemangioma obstructing the upper airway. B. Contrast-enhanced axial CT scan showing obstructive subglottic hemangioma with characteristic intense immediate enhancement after IV contrast injection. C. Sagittal CT scan showing silicone T-tube placement, with the upper end sitting above the vocal folds.

present a surface color varying from pink to blue, are easily compressible, and project into the airway, factors that are necessary for their diagnosis.⁽⁵⁾ Cases 1 and 2 progressed to airway obstruction, and tracheostomy was performed before endoscopic evaluation, as recommended.⁽⁶⁾ However, a mortality rate of 40% to 60% has been reported due to tracheostomy complications, particularly obstruction or accidental decannulation.⁽⁶⁾ The rationale for managing these patients is by stenting with a silicone T-tube and oral propranolol administration.⁽⁷⁾ The vertical branch of the T-tube supports the tracheal lumen, while its horizontal branch anchors the tube to the tracheostomy stoma, preventing T-tube displacement.⁽⁸⁾ The main purpose of both the tracheostomy and the T-tube is to maintain airway patency while awaiting the resolution of the SGH.

In 2008, the treatment of SGH received a new medical approach with the use of propranolol,⁽⁴⁾ reducing

the number of tracheostomies performed in these patients in 2012 compared to 2003.⁽⁹⁾ An initial dose of 1 mg/kg/day orally is recommended and, if well tolerated, can be increased to 2 or 3 mg/kg/day. It is unknown how long children with SGH should remain on propranolol, but studies suggest its maintenance until the proliferative phase is completed.⁽¹⁰⁾ During treatment with propranolol, insofar, we have not yet observed either cardiac arrhythmias or other side effects of the drug.

It can be concluded that obstructive SGH in children can be managed safely and effectively by long-term oral propranolol and airway stenting by means of a silicone T-tube when a tracheostomy is required.

AUTHOR CONTRIBUTIONS

All authors participated in the drafting and revision of the manuscript, as well as in the approval of the final version.

REFERENCES

- Holinger PH, Brown WT. Congenital webs, cysts, laryngoceles and other anomalies of the larynx. *Ann Otol Rhinol Laryngol* 1967; 76:744–52. <https://doi.org/10.1177/000348946707600402>.
- Drolet BA, Esterly NB, Frieden IJ. Hemangiomas in children. *N Engl J Med* 1999; 341:173–181. <https://doi.org/10.1056/NEJM199907153410307>.
- Perkins JA, Duke W, Chen E, Manning S. Emerging concepts in airway infantile hemangioma assessment and management. *Otolaryngol Head Neck Surg* 2009; 141:207–212. <https://doi.org/10.1016/j.otohns.2009.04.013>.
- Denoyelle F, Leblouanger N, Enjolras O, Harris R, Roger G, Garabedian EN. Role of propranolol in the therapeutic strategy of infantile laryngotracheal hemangioma. *Int J Pediatric Otorhinolaryngol* 2009; 73:1168–72. <https://doi.org/10.1016/j.ijporl.2009.04.025>.
- Ferguson CF, Flake CG. Subglottic hemangioma as a cause of respiratory obstruction in infants. *Ann Otol Rhinol Laryngol* 1961; 70:1095–112. <https://doi.org/10.1177/000348946107000414>.
- Rahbar R, Nicollas R, Roger G, Triglia J, Garabedian E, McGill TJ, et al. The biology and management of subglottic hemangioma: past, present, future. *Laryngoscope* 2004; 114:1880–1891. <https://doi.org/10.1097/01.mlg.0000147915.58862.27>.
- Tamagno M, Bibas BJ, Minamoto H, Alfinito FS, Terra RM, Jatene

- FB. Subglottic and mediastinal hemangioma in a child: treatment with propranolol. *J Bras Pneumol.* 2011; 37(3):416-8. <https://doi.org/10.1590/s1806-37132011000300021>.
8. Prasanna Kumar S, Ravikumar A, Senthil K, Somu L, Nazrin MI. Role of Montgomery T-tube stent for laryngotracheal stenosis. *Auris Nasus Larynx* 2014; 41:195-200. <https://doi.org/10.1016/j.anl.2013.10.008>.
 9. Espahbodi M, Yan K, Chun RH, McCormick ME. Management trends of infantile hemangioma: A national perspective. *Int J Pediatr Otorhinolaryngol.* 2018; 104:84–87. <https://doi.org/10.1016/j.ijporl.2017.10.044>.
 10. Maturo S, Hartnick C. Initial experience using propranolol as the sole treatment for infantile airway hemangiomas. *Int J Pediatr Otorhinolaryngol.* 2010; 74(3):323-5. <https://doi.org/10.1016/j.ijporl.2009.12.008>.