

Niel Khangel S. Reyes, MD

Department of Otorhinolaryngology
Head and Neck Surgery
Armed Forces of the Philippines Medical Center

Hypopharyngeal, Supraglottic and Subglottic Stenosis after 1-Week Intubation

Laryngeal stenosis is a partial or complete narrowing of the endolarynx and has many etiologies. Common causes of laryngeal stenosis are iatrogenic (prolonged intubation, laryngeal surgery), external neck trauma, congenital, burns, ingestions, infection, and inflammation (gastroesophageal reflux or Wegener's). Laryngeal stenosis secondary to trauma usually affects the posterior endolaryngeal region in adults and the subglottic region in children.¹

Patients with mild to moderate laryngeal stenosis are usually asymptomatic and if otherwise, majority of the presenting signs and symptoms are mainly related to the airway, feeding and voice resulting to marked respiratory distress, dysphagia/odynophagia and altered voice, respectively.

We present a case of hypopharyngeal, supraglottic and subglottic stenosis occurring 1 week after intubation.

CASE REPORT

A 3-year-old boy from Ormoc City was admitted in our institution for dysphagia of 2 months.

Three months prior to admission, he was treated for hypersensitivity reaction after eating shrimp and crab. The boy experienced sudden onset perioral swelling with bluish discoloration, dyspnea, severe drooling and vomiting of previously ingested food immediately after taking a vitamin supplement syrup that had been preceded by the dinner of crustaceans. He was immediately brought to a primary hospital in Ormoc City but was not relieved by nebulization and unrecalled intravenous medications.

The boy was eventually transferred to a tertiary hospital in Ormoc City. During this time, perioral swelling, dyspnea and cyanosis with associated severe drooling persisted. He was also noted to have stridor, was intubated and subsequently admitted to the intensive care unit for 7 days with an impression of severe hypersensitivity reaction secondary to crustacean ingestion. He was fed via a nasogastric tube (NGT). His condition eventually improved and the endotracheal tube was removed after 7 days. According to the relatives, there was significant increase in expectoration of saliva hours after removal of the endotracheal tube, allegedly occurring almost every minute, accompanied by drooling. There was no fever, no difficulty of breathing, no aspiration, no vomiting episodes and no easy fatigability noted at this time. However, the boy was noted to have dysphagia associated with frequent coughing and expectoration of saliva on intake of both fluids and solid food following removal of the NGT. His relatives denied any episodes of dyspnea, vomiting, cyanosis, easy fatigability or fever, and he was subsequently discharged after 1 month of confinement.

One month and three weeks prior to admission, he still presented with dysphagia, frequent spitting of saliva and now with associated wheezing and weight loss on follow-up at the hospital. He was referred to a pediatric pulmonologist in Cebu City for further evaluation and management.

One month before admission, a thickened epiglottis was seen on neck and chest CT-scan and "*acquired subglottic stenosis, post intubation*" was diagnosed, for which direct laryngoscopy was recommended. The relatives did not consent, and the boy was discharged on Betamethasone + Dexchlorpheniramine syrup for 5 days, Montelukast Na oral granules for 30 days, and Amoxicillin suspension and Salbutamol syrup for 7 days.

Nineteen days prior to admission, with persistence of the previously-mentioned symptoms, the boy was brought to our outpatient service and subsequently admitted.

Correspondence: Dr. Niel Khangel S. Reyes
Department of Otorhinolaryngology
Head and Neck Surgery
Armed Forces of the Philippines Medical Center
7th Floor Armed Forces of the Philippines Medical Center
V. Luna Avenue, Quezon City 0840
Philippines
Phone: (632) 426 2701 local 6172
Email: nielkhangel_reyes@yahoo.com
Reprints will not be available from the author.

The author declares that this represents original material, that the manuscript has been read and approved by the author, that the requirements for authorship have been met by the author, and that the author believes that the manuscript represents honest work.

Disclosures: The author signed a disclosure that there are no financial or other (including personal) relationships, intellectual passion, political or religious beliefs, and institutional affiliations that might lead to a conflict of interest.

On admission, he was ambulatory and not in cardiorespiratory distress. He still had symptoms of increased expectoration and drooling with associated dysphagia for both liquids and solid foods. Because of difficulty inserting a nasogastric tube, he had to subsist on small, frequent sips of fluids consisting mostly of milk and water. Fluid thickeners improved swallowing. Initial flexible nasopharyngolaryngoscopy revealed a thickened epiglottic area obscuring the vocal cords. A modified barium swallow and airway fluoroscopy showed aryepiglottic fold thickening and non-persistent episodes of narrowing at the supraglottic and glottic areas. A trace of nasopharyngeal regurgitation was also noted without any gross tracheal aspiration.

Flexible nasopharyngolaryngoscopy by a laryngologist revealed a normal nasopharynx, absent epiglottis, absent pyriform sinuses, stenotic hypopharynx and supraglottis, normal vocal cord structure and mobility with grade 1 subglottic stenosis. (Figures 1 A-D) Possible laser surgical release of fibrosis and removal of strictures was recommended, with close observation until then. Meanwhile, due to persistent dysphagia and significant weight loss, he underwent gastrostomy and was started on supplements for nutritional build up.

DISCUSSION

Laryngeal stenosis is a partial or complete cicatricial narrowing of the endolarynx and may be congenital or acquired.¹ Trauma is the most common cause of acquired laryngeal stenosis, both in children and in adults, and is classified as external laryngeal trauma or internal laryngeal trauma. The latter is more commonly due to iatrogenic causes, especially prolonged endotracheal intubation.² Approximately 90% of cases of acquired chronic subglottic stenosis in infants and children occur secondary to endotracheal intubation. The reported incidence of stenosis after intubation ranges from less than 1% to 8.3%.¹ In our case, the patient initially presented with persistent dysphagia with associated excessive drooling and increased expectoration after intubation for 1 week.

A study by Gallo *et al.* of 70 patients with laryngeal stenosis revealed that the causes of stenosis may be numerous (including intubation, autoimmune disease, iatrogenic) and multiple areas of the airway can be involved.³ The reported incidence of tracheal stenosis following laryngotracheal intubation ranges from 6% to 21%.³ They further explained that erosion and mucosal necrosis occur within hours of

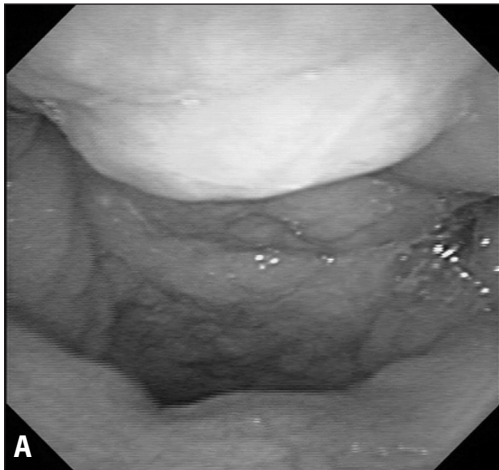


Figure 1A. Normal nasopharynx

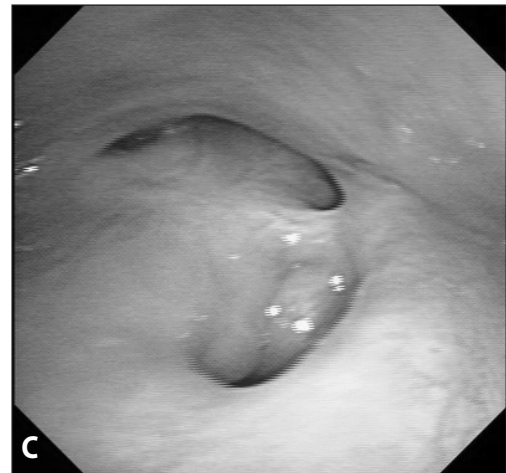


Figure 1C. Absent pyriform sinuses; stenotic hypopharynx and supraglottis

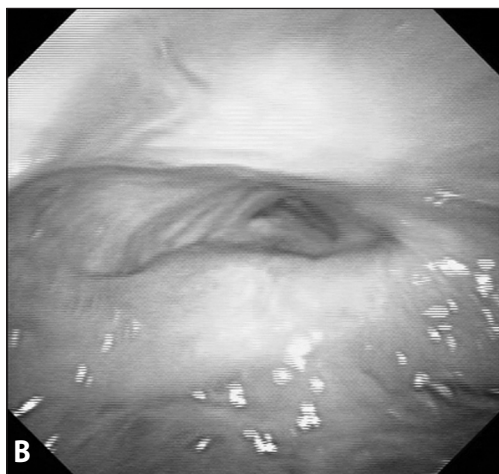
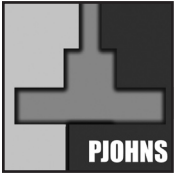


Figure 1B. Absent epiglottis



Figure 1D. Normal vocal fold structure and mobility with Grade 1 subglottic stenosis



endotracheal intubation and full thickness injury exposes cartilage with development of perichondritis if the tube is not withdrawn within a week. Re-epithelialization of the edges of the ulceration follows and healing is completed within 4 weeks. The previous site of the ulceration is usually marked with fibrosis and metaplastic squamous epithelium. The risk of tissue damage and development of laryngotracheal stenosis increases depending on the severity of the ulceration and if the healing process is delayed by secondary infection.³ Duration of intubation and size of the endotracheal tube are the most important factors in the development of laryngeal stenosis, but no definite safe time limit for endotracheal intubation has been established. Severe injury has been reported after 17 hours of intubation in adults and 1 week after intubation in neonates. The area most commonly injured in children is the subglottic region.¹

Initial evaluation when suspecting laryngeal stenosis includes radiographic evaluation to aid in assessing the degree and length of stenosis. Computed tomography scanning (CT) and magnetic resonance imaging (MRI) scanning are not standard techniques to assess the laryngotracheal airway but may be used as an adjunctive diagnostic technique to help determine the length of the stenosis or concurrent vascular compression.⁴ Initial Computed Tomography (CT) scans prior to admission revealed only thickening of the epiglottis with narrowing of the subglottic region. The extent of involvement was not determined until flexible nasopharyngolaryngoscopy revealed absence of the epiglottis and pyriform sinuses, hypopharyngeal and supraglottic stenosis and grade 1 subglottic stenosis with normal vocal cord structure and mobility. Flexible and rigid endoscopy should be a part of assessment as they allow direct inspection of the dynamic laryngeal and hypopharyngeal airway.⁴ As illustrated in this case, fluoroscopy is also helpful in studying tracheal dynamics.¹

The basic techniques for management of laryngeal stenosis include endoscopic and external methods. Wiatrak suggested that conservative management or a “wait-and-see” approach may be considered.⁴ The management of laryngeal stenosis in infants and young children should be conservative, since in the majority of cases, the stenosis will improve with laryngeal growth.⁵ While such management may be considered in our case, it is rarely successful for acquired laryngeal stenosis. Endoscopic methods including balloon dilatation and laser assisted excision are options, although the former is only beneficial in cases of early, soft stenosis, before mature, firm stenosis has developed.⁴

Open surgical methods include expansion and resection surgery. Open surgical procedures should only be recommended when it has been established by careful endoscopic assessment that the laryngeal lumen has not increased in size.⁵ According to Gallo *et al.*, tracheal resection and anastomosis is considered the treatment of choice for tracheal stenosis.³ However, this approach may not be applicable when the glottis and/or the subglottis are also involved. Moreover, it may not be feasible due to the extent of the stenosis, underlying disease and general health of the patient.³ While grade 1 stenosis may be managed by open surgery, it may also be amenable to endoscopic techniques.

The challenge for this case includes correction of the laryngeal area, ensuring stability of airway and improving general status and health with the least invasive management possible. The use of stents offers another management option for laryngeal stenosis. Alshammari and Monnier used laryngotracheal stents on 65 patients during open surgery and endoscopy to keep the airway expanded after surgical reconstruction or trauma. However, they also reiterate that stents should be avoided unless absolutely necessary since there are potential risks for mucosal injuries, ulcerations, granulation tissue formation and subsequent restenosis.⁶ According to Zanetta *et al.*, there is no ideal stent for the treatment of subglottic stenosis in children and that it can act as a foreign body in the reconstructed airway causing difficulties for feeding and in voice production.⁷

Our proposed method for addressing the laryngeal stenosis is to attempt laser excision to correct the affected areas and hopefully improve the feeding status while ensuring stability of the airway. At present, nutritional build up is the initial target in preparation for the contemplated procedure.

Laryngeal stenosis in the pediatric population is one of the most controversial topics in pediatric otolaryngology. There are various techniques available for management of laryngeal stenosis. Therapeutic procedures range from repeated dilatation, prolonged laryngeal stenting with or without the use of steroids, the use of carbon dioxide laser to create an airway with or without tracheostomy (through a laryngeal mask airway), to early tracheostomy and open surgery.^{5,8} However, feasibility of the technique, invasiveness, as well as possible outcome are some of the problems a physician may encounter. We should always consider individualizing our management according to pathologic findings, patient's age, degree and consistency of stenosis and importantly, the general condition of the patient. Echoing Evans, one could at least give the parents of pediatric patients a reasonably accurate prognosis, and the hope that their child can be restored to normality.⁵

REFERENCES

1. Zalzal GH, Cotton RT. Glottic and subglottic stenosis. In: Flint PW, Haughey BH, Lund VJ, Niparko JK, Richardson MA, Robbins KT *et al.*, editors. Cummings Otolaryngology Head and Neck Surgery. 5th edition. Philadelphia: Mosby; 2010. p. 2912-2924.
2. Khalid AN, Goldenberg D. Surgical management of upper airway stenosis. In: Flint PW, Haughey BH, Lund VJ, Niparko JK, Richardson MA, Robbins KT *et al.*, editors. Cummings Otolaryngology Head and Neck Surgery. 5th edition. Philadelphia: Mosby; 2010. p. 943-952.
3. Gallo A, Pagliuca G, Greco A, Martellucci S, Mascelli A, Fusconi M, De Vincentiis M. Laryngotracheal stenosis treated with multiple surgeries: experience, results and prognostic factors in 70 patients. *Acta Otorhinolaryngol Ital.* 2012 Jun; 32(3): 182-188.
4. Wiatrak BJ. Glottic and Subglottic Stenosis in Children. In: Ossoff RH, Shapshay SM, Woodson GE, Nettekville J, editors. The Larynx. Philadelphia: Lippincott Williams and Wilkins; 2003. p. 451-469.
5. Evans JNG. Stenosis of the Larynx. In: Kerr AG, Groves J, Evans JNG, editors. Scott-Brown's Otolaryngology Volume 6. 5th edition. London: Butterworth; 1987. p. 34.
6. Alshammari J, Monnier P. Airway stenting with the LT-Mold for severe glotto-subglottic stenosis or intractable aspiration: experience in 65 cases. *Eur Arch Otorhinolaryngol.* 2012 Dec; 269(12): 2531-2538.
7. Zanetta A, Cuestas G, Rodriguez H, Tiscornia C. A Novel laryngeal stent in the treatment of subglottic stenosis in children. *Acta Otorrinolaringol Esp.* 2014; 65(2): 120-122.
8. Vorasubin N, Vira D, Jamal N, Chhetri DK. Airway management and endoscopic treatment of subglottic and tracheal stenosis: The laryngeal mask airway technique. *Ann Otol Rhinol Laryngol.* 2014 April; 123(4): 293-298.