Lobular Capillary Hemangioma of the Neonatal Larynx

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Objective: To describe a previously unreported condition of the neonatal larynx.

Design: Case series of 4 neonates with an uncommon laryngeal lesion.


Patients: Four neonates in the first 10 days of life with stridor, hoarseness, and respiratory distress.

Intervention: The patients were examined using flexible fiberoptic laryngoscopy, and laryngeal lesions were identified and subsequently removed using microlaryngoscopy. Photodocumentation of the lesions was performed. Microscopic evaluation of biopsy specimens by a pathologist followed.

Main Outcome Measures: Each patient's medical record was carefully reviewed for prenatal history, birth history, neonatal history, pathologic findings, and office follow-up.

Results: All 4 neonates were deliveredatraumatically and developed symptoms of upper airway obstruction within the first few minutes to days of life. Each neonate was found to have an obstructive laryngeal lesion requiring surgical intervention. No child had other congenital abnormalities or a history of obvious laryngeal trauma. Pathologic review of each laryngeal specimen revealed inflammatory lesions with characteristic features of a lobular capillary hemangioma (or a pyogenic granuloma).

Conclusions: The diagnosis of a lobular capillary hemangioma of the larynx should be considered in the differential diagnosis of a newborn with stridor, hoarseness, or respiratory distress. The cases seem to be of congenital origin, although acquired pathogenesis cannot be ruled out. Treatment of these lesions includes microscopic surgical excision.


Neonates can have abnormalities of the larynx ranging from simple to complex. If not identified and treated appropriately and promptly, some of these abnormalities can lead to substantial morbidity that can be life altering or life threatening. Anomalies of the larynx can affect the ability to adequately respire, phonate, and protect the respiratory tract to a degree proportional to that of the abnormality. Symptoms of laryngeal obstruction include variations of hoarseness, retractions, dysphagia, aspiration, respiratory distress, a weak cry or aphonia, and stridor (depending on the location of the lesion).1-3 Most lesions of the neonatal larynx are of congenital origin, but acquired lesions can occur in neonates that may necessitate various interventions in the perinatal period. Symptoms of congenital and acquired laryngeal abnormalities can manifest immediately at birth or may be delayed.

The objective of this article is to describe 4 neonates diagnosed as having a lobular capillary hemangioma (LCH) of the larynx, also referred to as a pyogenic granuloma. Although the pathogenesis of the lesion can be congenital or acquired, pathologic review and clinical evidence lead us to believe that each case was due to a congenital cause. Lobular capillary hemangioma of the larynx is a lesion that has not previously been described in the literature, to our knowledge. This anomaly should be considered in the differential diagnosis of any neonate with characteristic symptoms of laryngeal obstruction.

REPORT OF CASES

CASE 1

The boy was born at 39 weeks weighing 4309 g from an uneventful pregnancy. He wasatraumatically suctioned at delivery because of suspected meconium aspiration but was not intubated. His Apgar scores were 9 at 1 minute and 9 at 5 minutes. Shoulder dystonia was noted. The
child seemed healthy in the newborn nursery according to hospital notes and was discharged home with his mother the following day.

Shortly after discharge, his parents noticed that, while the infant’s breathing was quiet during sleep, he had a continuous “noisy breathing” pattern while awake. In addition, the parents noticed that the noise was louder during feeding or crying and that the infant seemed to be struggling to breathe. Finally, the parents commented that his cry had been weak since birth. They were unaware of any cyanotic events. The infant was taken to his pediatrician and was referred to the pediatric otolaryngology office at 9 days of age.

On examination, the child was noted to have a weak cry, moderate inspiratory stridor, and mild substernal retractions. Flexible fiberoptic laryngoscopy revealed a large mass emanating from the right posterior larynx and obstructing 75% to 80% of the glottic inlet. The other head and neck examination findings were normal.

The next day, the child was taken to the operating room for microlaryngoscopy and bronchoscopy. A large pedunculated mass was present at the midportion of the right true vocal cord and ventricle region (Figure 1A). It was a true mass and not a cyst. The remainder of the tracheobronchial tree was normal. While the patient was under general anesthesia with spontaneous respiration, the larynx was suspended with a laryngoscope, and the mass was successfully removed using microscissors and microforceps under microscopic guidance. After excision of the lesion, a topical corticosteroid–soaked pledget was placed for 3 minutes on the raw mucosa. The specimen was sent for pathologic analysis. The child was observed overnight and was discharged home the next day breathing comfortably and quietly. The parents were given instructions to administer an antireflux medication for 3 months.

At 2-month follow-up, flexible fiberoptic laryngoscopy showed no recurrence of the mass. At 14 months...
of age, microlaryngoscopy was performed during bilateral myringotomy and tube placement to reconfirm that no recurrence was evident. At 36 months of age, microlaryngoscopy was performed during tonsillectomy and adenoidectomy, and the larynx was found to be normal.

**CASE 2**

The girl was born at 35 weeks' gestation from an uncomplicated pregnancy. Her Apgar scores were 3 at 1 minute and 8 at 5 minutes. After 10 minutes, she was thought to have poor respiratory effort, possibly due to maternal opioid dosing, as well as respiratory distress and stridor. Therefore, she was briefly and atraumatically intubated with a 3.5-mm endotracheal tube by an attending neonatologist. The neonatologist did not note any abnormalities during intubation. The neonate received naloxone hydrochloride to reverse suspected opioid toxicity, and the infant was extubated without complication 2 hours after birth. While in the neonatal intensive care unit (NICU) during the first week of life, stridor was noted with each attempt to cry; the quality of the cry was described as weak. In addition, the infant exhibited occasional retractions, desaturation episodes, and several short cyanotic spells. Because of difficulty feeding, she was placed on nasogastric tube feedings. The pediatric otolaryngology service was consulted at 1 week of age.

Physical examination revealed a healthy-appearing newborn with mild biphasic stridor. However, no retractions were present, and her oxygen saturation levels were 98% on room air. Flexible fiberoptic laryngoscopy revealed a large mass emanating from the right true vocal cord and obstructing 70% of the glottic inlet.

The following day, the infant was taken to the operating room for microlaryngoscopy and bronchoscopy. A large mass was present that emanated from the anterior portion of the right true vocal cord (Figure 1B). The ventricle was normal, and the mass was not cystic in nature. The remainder of the airway was normal. While the patient was under general anesthesia with spontaneous respiration, suspension microscopic laryngoscopy was performed. Microscissors and microforceps were used to excise the mass without difficulty, and the specimen was sent for pathologic analysis. Following surgery, the child's stridor resolved; she began to feed well and was discharged home 3 days later with antireflux medication to be administered for 3 months.

At 6 months of age, office follow-up revealed a mildly raspy quality to the infant's cry. Fiberoptic laryngoscopy showed some mucosal inflammation in the area over the anterior aspect of the true vocal cord, but no recurrence of the laryngeal mass was evident.

**CASE 3**

The girl was born at 39 weeks' gestation weighing 3910 g. Her mother had type 1 diabetes mellitus and required a cesarean delivery. Meconium-stained amniotic fluid was noted at delivery. The Apgar scores were 8 at 1 minute and 9 at 5 minutes. Mild stridor and meconium were noted at birth, and the infant was intubated with a 3.5-mm endotracheal tube by the NICU fellow (D.D.S.) for suctioning purposes. Because of persistent mild respiratory distress, the child was reintubated with the same size of tube without complication and was left intubated for 2 hours. The intubating physician (D.D.S.) noted no mention of a laryngeal mass. After extubation, continued stridor was noted, which became more pronounced during the next 3 days. A nasogastric tube was used for feeding.

In less than 1 minute, he was extubated and sent to the NICU for observation. The infant was then found to have persistent oxygen requirement and respiratory distress and was reintubated atraumatically for 8 additional hours. The intubating physician (D.D.S.) made no mention of visualizing a laryngeal mass during intubation. Following extubation, stridor, subcostal retractions, and a respiratory rate exceeding 60 breaths per minute were noted; the pediatric otolaryngology service was consulted on the first day of life.

Physical examination revealed biphasic stridor and mild substernal retractions. Flexible fiberoptic laryngoscopy showed a floppy mass in the anterior glottic inlet obstructing 50% of the airway. The other examination findings were normal.

On the second day of life, the neonate was taken to the operating room for microlaryngoscopy and bronchoscopy under general anesthesia with spontaneous respiration. A large mass was seen emanating from the anterior and midportion of the right true vocal cord and obstructing the laryngeal inlet by 50% (Figure 1C). The remainder of the airway was normal. Because the lesion was thought to be a granuloma, it was not immediately removed; instead, the child was treated for 4 days with intravenous corticosteroids and antireflux medication in the NICU. After corticosteroid treatment, the child was taken again to the operating room for microlaryngoscopy. The findings showed the same mass emanating from the right ventricle and false cord region and still obstructing 50% or more of the laryngeal inlet. The mass was excised under a microscope using microcup forceps and was sent for pathologic analysis. The child seemed healthy following surgery and was discharged home 4 days later with antireflux medication to be administered for 3 months.

At 2 months of age, office follow-up revealed no respiratory distress. The infant was feeding well, thriving, and exhibiting normal voice and cry. Flexible fiberoptic laryngoscopy showed some mild mucosal irritation but no recurrence of the mass.

**CASE 4**

The girl was born at 39 weeks' gestation weighing 4000 g to a healthy mother. Meconium-stained amniotic fluid was noted at birth, and the Apgar scores were 8 at 1 minute and 9 at 5 minutes. Because of the presence of meconium, the infant was intubated with a 4.0-mm endotracheal tube without complication in the delivery room by the NICU fellow (D.D.S.) for the purpose of suctioning.
costal retractions. Flexible fiberoptic laryngoscopy demonstrated a large mass obstructing the laryngeal inlet.

On day 4 of life, the child was brought to the operating room. While the patient was under general anesthesia and breathing spontaneously, microlaryngoscopy revealed 90% obstruction of the laryngeal airway by a mass emanating from the right false vocal cord and ventricle region (Figure 1D). The tracheobronchial tree was normal. The larynx was exposed using a Parson laryngoscope, and the mass was removed using microcup forceps and was sent for pathologic analysis. The child was left intubated and was treated with intravenous corticosteroids and antireflux medication. She returned to the operating room 2 days later for laryngoscopy to ensure that edema was not present. No substantial edema was noted, so she was extubated without complication. She had obvious improvement in her breathing and feeding and was discharged home 3 days later on a regimen of antireflux medication for 3 months. At 2-month office follow-up, flexible fiberoptic laryngoscopy showed no recurrence.

PATHOLOGIC FINDINGS FOR ALL CASES

The most obvious feature in each case was prominent vascular proliferation with a lobular growth pattern. The vascular proliferation in all cases was well circumscribed. In case 3, a fibromyxoid background was appreciated. Focally to completely ulcerated overlying squamous epithelium was present in all cases. The squamous epithelium, when present, was mildly hyperkeratotic and reactive in appearance. No notable atypia were present in any of the samples. In addition, a mild to moderate amount of neutrophilic infiltration involving the mucosa was noted. The biopsy specimen fragments from each case revealed histologic features compatible with an LCH (Figure 2).

**COMMENT**

The lesions found in the 4 cases reported herein were pathologically consistent with an LCH, also known as a pyogenic granuloma. Lobular capillary hemangioma has been proposed as a more accurate and distinguishing term than pyogenic granuloma, although they refer to the same lesion.

Physicians often use the terms pyogenic granuloma and reactive granuloma synonymously or simply use the general term of granuloma, which can be confusing; however, LCH and reactive granuloma are thought to be dis-
tinct entities. Despite their being distinct entities, LCH and reactive granuloma share several characteristics, including acute inflammation, an endothelial cell prominence, and an edematous and fibrous environment. Although the differentiation can be challenging, a reactive granuloma lacks the classic lobular growth pattern of the capillary proliferation that characterizes an LCH. In addition, LCH is typically a circumscribed lesion. A fibromyxoid background between the lobules of capillaries is also characteristic of LCH. Immunohistochemistry is not helpful in differentiating the 2 lesions.

Lobular capillary hemangioma is prevalent in children and commonly manifests as a rapidly growing, erythematous, and pedunculated or raised nodule. Studies discussing LCH have almost exclusively described lesions involving the skin and mucous membranes of the oral cavity and nasal region. In investigations of LCH in children, 76.9% of lesions were found on the head and neck, 76.7% had no history of trauma or predisposing factors, and most manifested as a single lesion. Congenital LCH in the oral mucosa has been documented previously, and the lesion is considered one of the most common pediatric benign tumors in children. Lobular capillary hemangioma has not previously been described in the larynx, to our knowledge.

Based on the lack of obvious laryngeal trauma in the cases reported herein, we suspect a congenital nature of the lesions. The first case reported in this study did not require intubation; the second, third, and fourth cases were intubated briefly (lasting minutes in 2 cases and 2 hours in the third case) andatraumatically without any noted difficulty, bleeding, or trauma (in all of the cases). In addition, the lesions were soft and movable, allowing them to be easily pushed aside as the endotracheal tube was passed. This explains the lack of reported problems by the neonatologist. The significance of the lesions’ consistently emanating from the right side of the larynx is unclear.

Laryngeal (or saccular) cysts, subglottic hemangioma, and lymphangioma extending to the larynx are lesions that can cause varying degrees of stridor, as well as acute respiratory distress. The anatomic congenital entity most similar to the laryngeal lesion presented in this report that has been well described is a congenital laryngeal cyst. Laryngeal cysts are characterized by squamous or respiratory epithelium within a fibrous stroma with endodermal and mesodermal elements. Congenital laryngeal cysts are cystic lesions, not solid, and are submucosal in location, differentiating them grossly and histologically from the lesions found in the 4 cases reported herein.

The differential diagnosis of this type of lesion would include angiofibroma, angiosarcoma, granulation tissue, hemangiopericytoma, and hemangioma. Lobular capillary hemangioma is distinguished from hemangioma clinically and histologically based on its lobular growth pattern, fibromyxoid background, overlying ulceration, and acute inflammation.

Acquired lesions involving the neonatal larynx can develop and cause similar symptoms. These most commonly are due to trauma related to intubation, nasogastric tube placement, or gastroesophageal reflux injury. Literature exists suggesting that reactive laryngeal granulation tissue is often caused by intubation, usually for a prolonged period. The incidence of postintubation laryngeal granulation tissue for all ages is 1 per 800 to 1000 intubations. Two studies describing laryngeal granulation tissue in neonates specifically posit that the findings of laryngeal granulation tissue must have been secondary to traumatic intubation, although no complications or history of trauma was noted in the reports. No mention of a possible congenital cause was made in the 2 studies.

When considering an acquired, or reactive, cause of a laryngeal lesion, several considerations need to be made. First, most intubation-induced lesions are found in adults and are rarely reported in children or neonates, especially with short-term intubations of less than 24 to 48 hours. Second, it has been surmised that the position and shape of the neonate larynx may displace mechanical traumatic forces toward the narrower subglottis and that the loose areolar tissue of the submucosa of the neonate may resist injury from intubation. Third, although mucosal granulomas can arise due to tissue overgrowth following trauma, variants often appear de novo because of special events such as hormonal changes occurring during pregnancy.

To our knowledge, no literature exists regarding a congenital or an acquired laryngeal lesion of this nature in a neonate; therefore, although a congenital argument seems logical, the cause remains unknown. None of the children had recurrence of the lesions, although we believe that the possibility exists. Our recommendation is to follow up children with a history of this lesion closely during the first few years of life and to repeat endoscopy as necessary.

CONCLUSIONS

We report 4 separate neonate cases with characteristic symptoms of a laryngeal abnormality. In each case, laryngoscopy revealed a mass that was found to be an LCH on pathologic analysis after microscopic surgical excision. On further review and follow-up, it is concluded that LCH of the larynx should be considered in the differential diagnosis of a newborn with stridor, hoarseness, or respiratory distress. Future clinical evidence is necessary to more definitively characterize the origin of these types of lesions as congenital or acquired.
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REFERENCES