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Laryngeal stenosis and laryngomalacia treated with a silicone stent in a prematurely delivered child: a case report

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Abstract

Background. Preterm birth is one of the most significant risk factors for infants' laryngeal stenosis (LS), which can occur as laryngomalacia (LM). LS can be defined as a partial or circumferential narrowing of the endolaryngeal airway and may be congenital or acquired. This condition cause various symptoms such as stridor, dyspnea and respiratory distress, therefore early intubation is required. Most of the patients with LS or LM outgrow their disease, but the rest of them need surgical help.

Objectives. The aim of this article is to present a successful surgical treatment for preterm infant with LS.

Methods*:* case presentation and an analysis of literature. Research of articles in "PubMed", "Google Scholar" databases with keywords used as follows: "Laryngeal stenosis", "Laryngomalacia", "Pediatric stent", "Premature delivery", "Endolaryngeal microsurgery".

Case presentation. 3-year-old male presented the hospital with diagnosed LM and for laryngeal stent insertion surgery. The patient was born at 26 gestational weeks and was treated for sepsis during the neonatal period, was intubated at birth because of severe respiratory distress, had 6 endolaryngeal microsurgeries (EM) and received Kenalog injections 5 times. Laryngeal stent was inserted during the last EM procedure. Patient undergone general anesthesia, the American Society of Anesthesiologists classification (ASA) was evaluated as III. The operation was successful without any complications. After three months the stent was removed and the patient's difficulty of breathing regressed.

Conclusion. In respiratory distress caused by LM and LS an insertion of a stent can be as equally or more effective than laryngoplasty. Regression of the respiratory distress symptoms after removal of the stent is considered a successful outcome.

Keywords: laryngeal stenosis, laryngomalacia, pediatric stent, premature delivery, endolaryngeal microsurgery.

Abbreviations

LS: Laryngeal stenosis

LM: Laryngomalacia

EM: Endolaryngeal microsurgery

ASA: American Society of Anesthesiologists classification

ENT: Ear, nose and throat

MAC: Minimal alveolar concentration

LTP: Laryngotracheoplasty

Background

Preterm infants often face a variety of organ developmental failures such as respiratory abnormalities. One of the most frequent conditions is laryngomalacia. Respiratory issues caused by laryngeal abnormalities in neonates may be life-threatening, therefore we should found the best treatment to avoid complications [1]. Bronchoscopy is the best method to diagnose respiratory abnormalities including LM and LS [2].

Treatment of airway stenosis in pediatric patients is an indication for stent insertion surgery [3]. Today there are two types of stents: metallic stent and silicon stent. It has been proved that treatment of airway stenosis with silicon stent implantation is as effective as the metallic stent implantation [2]. The Montgomery Laryngeal Stent is a molded silicone prosthesis designed to conform to the normal endolaryngeal surface. It is used for the prevention and treatment of LS when the glottic stenosis involves any of glottic area singularly or in combination. This article reviews several treatment methods of airway stenosis encountered during the neonatal period and solutions in a later age.

Case report

We present a case of a 3-year-old male, who was born at 26 gestational weeks and was treated for sepsis during the neonatal period. The patient was quickly intubated at birth because of severe respiratory distress, although the mother antenatally received dexamethasone injections for fetal lung maturity. At two months of age a bronchoscopy was performed, because of persisting dyspnea. During the procedure LM and LS were diagnosed and the patient was referred to an ear, nose and throat (ENT) surgeon for tracheostomy. A tracheostomy was performed, no complications of anesthesia or surgery were noted. The patient had no complaints after general anesthesia or operation and was discharged from the hospital one day after the procedure. At 1 year and 2 months of age he again was admitted to ENT department, because of persisting dyspnea. The anesthesiologist evaluated the patient condition as II ASA. The child then underwent general anesthesia and was mechanically ventilated through tracheostomy tube. EM was performed to remove the granulation tissue from the larynx and a biopsy was taken of the granulation tissue from the larynx to specify the changes. Also, the patient received his first Triamcinolone 40ml (Kenalog) injection. The biopsy came back as chronic inflammation with signs of fibrosis. After surgery and Kenalog injection the patient for a short time was having no complaints of shortness of breath. However, three months later dyspnea reoccurred and the boy was again admitted to ENT department. The patient again underwent general anesthesia (ASA class was evaluated as III), EM and Kenalog injection and after one day at the hospital was discharged with no complaints. But persistent dyspnea still was present after some time and after that multiple EM procedures (6 in total) were done at: 1 year and 11 months, 2 years, 2 years and 1 month and 2 years and 3 months of age. Multiple Kenalog injections (5 in total) were also given at: 1 year and 11 months, 2 years, 2 years and 1 month. All times the patient underwent general anesthesia and was mechanically ventilated through tracheostomy tube without any complications. However, because of difficulty of breathing and persistent dyspnea it was decided that the patient would undergo one more operation and a laryngeal stent would be inserted. At the age of three a silicone Montgomery laryngeal stent was placed during EM procedure. The patient underwent general anesthesia (ASA class was evaluated as III), induction reached with Sevoflurane (Sevoflurane minimal alveolar concentration (MAC) was kept at 1.0), 25μg of Fentanyl and 3mg of Mivacurium. The operation was completed successfully and no complications were observed. Post operation analgesia was given in form of Ketoprofen 30mg and Acetaminophen 300mg intravenously. The patient was discharged after one day without any dyspnea or difficulty of breathing. After three months he was admitted to ENT department for removal of the stent. During that time there were no complaints of dyspnea or difficulty of

breathing noted. The stent was removed under general anesthesia without any complications. In three-month follow-up the patient's difficulty of breathing has regressed, no complications of stent extraction were observed.

Discussion

LM is caused by floppiness of the laryngeal tissues above the vocal cords (the supraglottic larynx). In children born with LM the aryepiglottic folds are shorter than normal so that it resembles the Greek letter omega (Ω). Studies have identified three types of LM and each of them have special surgical treatment [1,4]. During inspiration the tissues above the vocal cords fall in towards the airway and cause partial obstruction. This creates stridor for newborns and infants, which occurs in 45–75% of cases [4]. In most cases, children with LM outgrow their airway problems. However, part of them encounter some difficulties, for example feeding problems, regurgitation, coughing/choking, failure to thrive, and significant respiratory distress [4].

LM can be suspected or presumptively diagnosed by anamnesis and physical exam. It is necessary to know a full birth history, information about any surgical procedures or intubations the patient has underwent [5]. Diagnosis must be confirmed with laryngoscopy while the child is conscious. The procedure is preformed using a flexible nasopharyngoscope [1].

Most cases can be treated with conservative methods. If no severe condition is present and the infant has only mild or moderate stridor without feeding problems, after the diagnosis of LM patient must be monitored for proper development and growth. However, 10 to 20 % of cases with LM are faced with severe symptoms that result in poor feeding or poor weight gain, pauses in the breathing (apnea) or cyanosis. These patients often require surgical treatment [5]. Surgery for this condition is called supraglottoplasty and it can be performed using several techniques, counting the use of a laser, cold steel, laryngeal microdebrider or coblator [6]. In our case during the bronchoscopy both LM and LS were diagnosed and the patient was directed for tracheostomy because of his life-threatening condition.

LS is the second most common cause of respiratory distress in newborns. It is also the leading laryngotracheal anomaly that requires tracheostomy in patients younger than 1 year. Classification of LS can be provided as congenital or acquired and these can further be characterized by clinical, anatomical and histopathological features [7]. Clinically LS depends on the grade of the obstruction of airway [8]. The symptoms may present from mild stridor to respiratory failure: dyspnea, hyperventilation, retractions of suprasternal and subcostal muscles, apnea, severe hypoxia, cyanosis. The most crucial risk factor for LS is preterm delivery [7]. Bronchoscopy is the preferred procedure to diagnose LS or any congenital airway malformation [2]. In severe cases of LS tracheostomy is the first-choice method of symptom relief, but in some rare cases a stent can be used as a temporary solution of symptom control for children who have a high risk of intraoperative fatality and have contraindications for surgery and general anesthesia [8-13]. Children who were diagnosed with LS, whether they had to undergo tracheostomy procedure or not, should receive systemic steroid injection during surgery [8]. In our case the patient was born prematurely at 26 gestational weeks and was quickly intubated. At two months of age LM with LS was diagnosed using bronchoscopy and the patient underwent tracheostomy operation. Kenalog injections were used alongside EM procedures. The stent was inserted after failure to control the symptoms with EM and Kenalog injections.

Airway stents have first been described in the year 1965 by W.W. Montgomery, where he presented his signature T-Tube silicone stent [14]. In the years to follow J.F. Dumon introduced his own dedicated comprised of molded silicone tracheobronchial stent [15]. There were different variations of silicone and metallic based stents used throughout the years and in the late 1990s self-expandable metallic stents were presented [16]. Although and ideal airway stent is still not made, there are quite a few diverse choices available today, but each of them need to be carefully and individually selected. A few indications for airway stenting can be found in the literature: palliative care for patients with advanced cancer; benign airway stenosis relief; fistula of the respiratory tract therapy; post lung-transplant anastomosis treatment and treatment of airway stenosis in pediatric patients [16, 17].

Laryngeal and tracheal stents in pediatric patients are most commonly used as postoperative tools. Their main cause is to aid in the healing of surgically reconstructive tissue or added grafts in the trachea and larynx, prevent aspiration and keep granulation tissue formation as low as possible [9, 18]. Furthermore, stents should not cause any difficulties while being

examined or removed and their care should not interfere with tracheotomy tube daily cleaning routine [18]. Stents as the stand-alone treatment of congenital malformations of the respiratory tract are very rarely used. Most of them are used after a successful laryngotracheoplasty (LTP) [9, 10, 19]. Kumar et al. published a study of 39 pediatric patients with laryngotracheal stenosis, who underwent airway stenting with Montgomery T-tube. Out of those patients 32 were successfully decannulated and after 6-month follow-up are doing well, 3 patients are still on stenting and 3 patients died to comorbid diseases unrelated to stenting [10]. In the case described above a Montgomery laryngeal stent was used molded to easily adapt to endolaryngeal mucosa and consisting of radio-opaque silicone. The stent is made atraumatic and flexible as possible in order to minimize damage to soft tissue.

Stent insertion can be associated with short and long-term complications [19, 13]. There are no clear guidelines for stent management after its placement, thus every case should be handled individually. The most commonly reported complications include granulation tissue formation and trachea obstruction, stent migration, difficulty of removing the stent and once in a while erosion of the respiratory tract [13, 16, 19, 20]. A study of 100 pediatric patients, who were diagnosed with serious airway obstruction received stent insertion, found that silicone-based stents were more likely to migrate than metallic stents (39.2% vs 4.1%) and are more liable to granulation tissue formation (11.6% vs 0.8%). Furthermore, after stent insertion 80 patients reported to have clinical improvement, for 17 mechanical ventilation was no longer needed and 3 had no significant clinical improvement [21]. There is also a reported case of tracheal stent erosion, which formed an arterio-tracheal fistula in an 18 months old female that ended fatally [20]. A few case reports suggest *Pseudomonas aeruginosa* and *Staphylococcus aureus* colonization were found on the stent after a successful LTP. The authors recommended antibiotic prophylaxis that cover these two microorganisms, their choice was oxacillin and ciprofloxacin. In both cases the stent was removed without any adverse effects [19, 22]. Other complications of stent placement include the following: stent fracture, airway rupture and halitosis; all of which are rarely reported, especially in pediatric patients [17]. Good patient management and follow up are needed in order to evaluate stents clinical effect. Recommendations in the literature state that the stent should be in place for at least 2 – 3 months and it should eliminate dyspnea or need for mechanical ventilation. Furthermore, fiber bronchoscopy or computer tomography scan should be used to confirm the airway is not obstructed and the stent should be easily removed by using flexible bronchoscopy [16]. In our reported case there were no complication reported of stent insertion and removal. The stent was in place for 3 months and has completely disposed of any difficulty of breathing or dyspnea. Granulation tissue formation was a problem before stent placement, hence the multiple EM.

Conclusion

In conclusion, LM and LS can both be fatally associated conditions that can cause severe respiratory distress in a prematurely born child. Although the primary treatment is LTP, but as an alternative a stent placement may have outcomes that are as equally or even more effective. Potential risk factors that need to be taken into account are: granulation tissue formation, obstruction of the trachea, migration of the stent and erosion. In addition, the patient must be closely monitored during follow-up examination in order to early detect adverse events. A stent should not be kept in place longer than 3 months. Regression of the respiratory distress symptoms after removal of the stent is considered a successful outcome.

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