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Insertion of two tracheobronchial stents in an infant with congenital tracheobronchial stenosis —A case report—

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Abstract

Airway management during anesthesia is mandatory but it is very difficult in certain diseases, especially when a tracheoplasty and bronchoplasty are involved. Tracheal stenosis is caused by congenital anomalies, trauma, physical compression, infection, intrabronchial granuloma, tumor, etc. Among these, congenital tracheobronchial stenosis is neither common nor easy to manage for the corrective surgery. This 11 month old boy was born with a congenital complete circular tracheal cartilage and pulmonary arterial sling, which caused tracheal stenosis. We present our anesthetic experience of successful management using two tracheobronchial stents in this infant, who received tracheobronchoplasty (3 times) under cardiopulmonary bypass, bronchoscopic examination and bougienation (7 times), and had a single tracheal stent inserted on one occasion, together with a review of the literature.

Keywords: Congenital tracheobronchial stenosis; Two tracheobronchial stents

Introduction

The occurrence of congenital stenosis of the trachea has been documented in the literature for almost a century. Despite the rarity of this malformation, certain types of the anomalies are amenable to complete correction, which assumes even greater significance when it is realized that this lesion is almost uniformly lethal if untreated.[1] Tracheal reconstructive surgery is widely accepted for adults and older children. However, the fear of postoperative stricture formation and impaired tracheal growth may have hindered the adoption of this procedure in infants and small children, and successful reconstructive surgery for obstructive lesions of the intrathoracic trachea in infants and small children has been recently reported in the literature.[2]

In addition, a small number of anesthetic reports have described the use of two tracheobronchial stents. The purpose of this report is to present an anesthetic management option for the insertion of two tracheobronchial stents in an infant with congenital tracheobronchial stenosis.

Case Report

A male infant was born on March 30th, 1999, and was hospitalized within one week due to respiratory difficulty. He had not any other symptoms except intercostal retractions after discharge. On July 31st, 1999, he was referred to the ENT department for an airway examination by the ophthalmologist who had attended for an operation for congenital blepharoptosis. The computed tomogram revealed a right tracheal bronchus, stricture in distal bronchus, pulmonary artery sling in carina, bilateral superior venae cavae, and cystic lesions in midesophagus. Whilst waiting for the elective surgery, he was admitted to pediatric intensive care unit (PICU) via emergency room due to the development of sudden severe respiratory difficulty on November 12th. An emergency operation was performed due to progressive respiratory difficulty ($PaCO_2$: 109 mmHg) on the same day. During anesthesia induction, an endotracheal tube (internal diameter 3.5 mm) could not be advanced into the subglottis. Intraoperative

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bronchoscopy revealed the complete circular tracheal cartilages. Left pulmonary artery reimplantation and tracheoplasty with pericardial patch just above the carina were performed under the cardiopulmonary bypass (CPB). Postoperative $PaCO_2$ was 40-148 mmHg and weaning trials from ventilatory support failed twice.

On December 30th, tracheoplasty was undertaken again at the same site with cartilages excised from the right tracheal bronchus under CPB. Postoperatively the hypercarpnia did not improve ($PaCO_2$: 40-136 mmHg) and he suffered from a large amount of sputum and hypoxemia. Ventilator weaning trials failed twice.

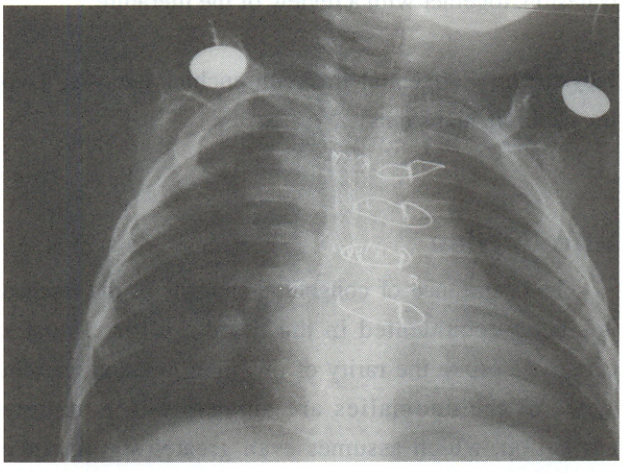
On January 11th, 2000, tracheoplasty was undertaken with pericardial flap under CPB because of the inflammation and necrosis in implanted cartilages found by bronchoscopic examination. No symptom improvements were observed after surgery.

On February 1st, 2000, a bronchoscopic examination revealed that granulation tissue growth had caused constriction proximal to the carina. We tried to widen the trachea and bronchus using bougienation method with Endobronchial Tube Replace Obturator (MettRo-19.0- 8.0: 10 Fr, 80 cm, Cook Critical Care, USA) four times. On February 21st, 2000, bronchoscopic examination revealed tracheal narrowing at the carina level and granulation tissue growth at both main bronchus inlets. We tried bougienation with 11 Fr and 12 Fr but no improvement was obtained. We realized that its cause was respiratory difficulty due to expiratory obstruction and it could not be treated with pericardial flap only.

On February 24th, 2000, straight single intratracheal stent (Niti-S, internal diameter 8 mm, length 4 cm, Taeung, Korea) was inserted into trachea. This stent was expandable, web-shaped, and made with silicon membrane and wire mesh. After insertion, symptoms improved temporarily, but the symptoms of obstruction persisted ($PaCO_2$: 48-65.9 mmHg). On February 25th, 2000, bronchoscopic examination revealed that anterior-posterior diameter had reduced at the left main bronchus due to granulation tissue growth. Bougienation was tried on further two occasions, again without any improvement.

On February 29th, 2000, two stents, one for each main bronchus from trachea, were inserted. Anesthetic induction was undertaken with thiopental 75 mg, vecuronium 1 mg (body weight 6 kg) and maintained with 100% O_2 and isoflurane. Initially the previous inserted stent was removed via bronchoscope, through which a guide wire was inserted to the right main bronchus with fluoroscopic guidance, and then bronchoscope was removed. The bronchoscope was re-inserted to the left main bronchus. As the pateint was being ventilated through the bronchoscope inserted in left main bronchus, a single web-shaped stent (Niti-S, nitinol, internal diameter 5 mm, length 4 cm, Taeung, Korea) was inserted into the right main bronchus along the guide wire. This guide wire then removed, and another guide wire re-inserted into the left main bronchus through bronchoscope, and a same size left stent inserted alomg the guide wire. The lung was intermittently ventilated with pulse oximetry monitoring during the procedure. Subsequently, the fluoroscope revealed that the stents were in good position and the trachea diameter had enlarged (Fig. 1.). The patient was transported to PICU with artificial ventilation.

Fig. 1 This chest X-ray shows two stents positioned from the lower trachea to the bilateral main bronchus (Niti-S, nitinol, ID-5 mm, length-4 cm, Taewoong, Korea) at the postoperative 1 day.



The endotracheal tube was extubated after confirming no dyspnea on postoperative day one ($PaCO_2$: 48-65.9 mmHg). On March 11th, 2000, the patient was transferred to ward after 10 days of PICU care. A CT scan revealed that the stents were in a good position, but they seemed to be relatively large compared with the

patient trachea and bronchi. Considering the possibility of constriction of trachea and bronchus, we decided to replace the stents with smaller versions and then later confirm the nature of the reaction to both stents.

On March 15th, 2000, the original stent was replaced with Niti-S stent (internal diameter 4 mm, length 3 cm). Intraoperative bronchoscopic examination revealed that there were no specific findings in the original site. No problem occurred during postoperative PICU care, and the endotracheal tube was removed on the same night (pulsed oxymeter 100%, respiratory rate 34/min).

Thereafter, the patient was in ward under observation with frequent bronchoscopy and exchange of stents at any time upon the development of sign or symptoms of airway obstruction.

Discussion

Extensive congenital tracheal stenosis continues to be a challenging problem. Tracheal reconstruction using a pericardial or cartilaginous patch graft is one of treatment methods but requires long-term intubation for stenting, and postoperative care is complicated by the formation of extensive granulation tissue over the composite graft.[3] Other therapeutic methods include tracheobronchial stenting,[4] tracheal resection and pulmonary artery reimplantation,[5] expandable metallic airway stents,[6] balloon catheter dilatation,[7] and the Montgomery T-tube method.[8]

The respiratory care during tracheal manipulation and stent change is also challenging to the anesthesiologists. Several methods are used, including using small endotracheal tube for bronchial intubation,[9] jet ventilation,[10] and using CPB.[11] We used intermittent ventilation via bronchoscopy with pulse oxymetry monitoring without any problems.

The insertion of airway stents is extremely useful for treating neonates and infants with localized tracheomalacia and bronchomalacia, because the stenting procedure is sufficiently simple and has minimal morbidity. Stenting should be considered even in children with incurable congenital heart disease and severe pulmonary symptoms.[4]

Filler et al[4] reported upon 5 year experience of stent insertion into infants and children with a variety of major airway obstructions. In their study, 30 balloon expandable stents were inserted into the trachea (n = 18) and bronchi (n = 12) of 16 infants, aged 1 week to 26 months (median, 9 months), with three types of serious airway obstruction. Three types were the tracheal or bronchomalacia, the stricture at the site of surgical repair of stenosis, and the airway compression due to enlarged pulmonary arteries associated with severe congenital heart disease or mediastinal lymphangioma. The problems reported in their study were obstructive granulations, migration of stent, and full-thickness bronchial erosion found in autopsy. They also reported that a child referred in the literature for tracheal stent removal after laser resection of granulations died at attempted removal because the stent was intimately adherent to the tracheal wall because of inflammatory reaction. The airway was completely occluded during the manipulation of the stent.[4]

Therefore, it is essential to examine the patient status thoroughly beforehand and to discuss the anesthetic plan with surgeon prior to operation because lethal airway obstruction might occur during stent exchange or removal. In particular, high frequency positive pressure ventilation and extracorporeal circulation should be prepared for airway obstruction before stent exchange.

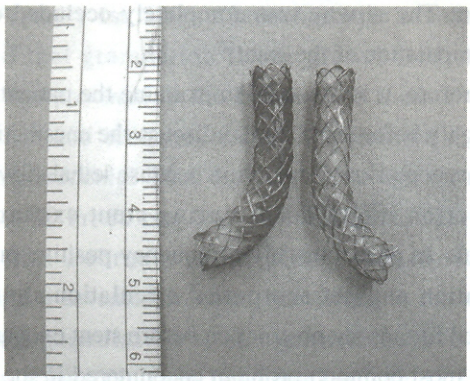
The most common problem encountered in the use of stents in the airway is the development of obstructing granulation tissue over the stent. Short of stent removal, granulations can be treated by scraping and suctioning through a bronchoscope. Nebulized salbutamol and steroids can also give some temporary relief.[4] Stent migration may occur especially in case that the external diameter of stent is smaller than the trachea or bronchus.

In our case, the major problem of the tracheobronchial stent was also the obstruction with granulation tissue and secretion. The paitent had intermittent nasotracheal suctioning but the airway obstrucion could be treated by scraping and suctioning through a bronchoscope only. Granulation tissue seemed to grow over the stent in about 3 weeks, but no specific difficulty was experienced in exchanging stents. Although the possibility of two stents to be interlocked always existed, we thought that the reason why interlocking did not occur was using the web-shaped stents made with silicon

membrane and wire mesh. We did not experience the intimately adhesion to the tracheal wall because the obstructive symptoms might develop earlier than severe inflammatory reaction. We also thought that the reason why the stents did not migrate was enough diameter of the stents. The figure 2 shows the exchanged stents with intrastent obstruction due to granulation tissue growth.

Because congenital trachea or bronchus obstruction is very rare, few reports describe the anesthetic management of the insertion of two stents into both bronchi. We report upon our anesthetic experience of the insertion of two stents and successful airway maintenance and subsequent periodic exchange.

Fig. 2 One of two removed intratracheobronchial stents (Niti-S, nitinol, ID-5 mm, length-4 cm, Taewoong, Korea) is obstructed by granulomatous plug.



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