Long-term management and outcomes of tracheobronchial stent by flexible bronchoscopy in infants <5 kg: A 13-year single-center experience

Kao-Hsian Hsieh, Ya-Ling Chou, Wen-Jue Soong, Yu-Sheng Lee, Pei-Chen Tsao

Department of Pediatrics, Tri-Service General Hospital, National Defense Medical Center, Taipei, Taiwan, ROC; Department of Pediatrics, Taipei Veterans General Hospital, Taipei, Taiwan, ROC; Institute of Emergency and Critical Care Medicine, School of Medicine, National Yang-Ming University, Taipei, Taiwan, ROC; Department of Pediatrics, National Yang-Ming University, School of Medicine, Taipei, Taiwan, ROC; Children’s Hospital, China Medical University, Taichung, Taiwan, ROC

1. INTRODUCTION

Tracheobronchial (TB) narrowing in infants, due to either congenital or acquired aetiologies, can compromise breathing and result in significant and even life-threatening respiratory distress. Traditionally, in patients who fail medical management, surgical interventions such as tracheostomy, tracheoplasty, bronchoplasty, thoracotomy and extracorporeal life support may be needed. These alternatives are all quite invasive, expensive and associated with further adverse effects. TB stent placement can rapidly provide durable and stable intraluminal support to maintain sufficient patency and improve quality of life. Since 1990, metallic stent placement, which has become an option in children, provides several advantages in select patients as a less-invasive management approach. Balloon-expandable metallic stents (BEMSs) have the unique benefit of further expansion for still-growing TB lumens in paediatric patients. Although therapeutic flexible bronchoscopy (TFB) with different instruments has been reported for decades, information on their application in BEMS placement and associated management of small infants remains scarce. Most studies on paediatric stent placement to date have involved rigid endoscopy (RE), endotracheal tube (ET) and/or mechanical ventilator support while the patients required transport to the operating room with complicated life-supporting equipment. Furthermore, these studies enrolled small number of cases and provided insufficient details on metallic stent placement, maintenance management, retrieval and long-term outcomes.

Abstract

Background: Tracheobronchial (TB) lumen narrowing may require prolonged positive-pressure ventilation, endotracheal tube intubation or even surgical interventions. Therapeutic flexible bronchoscopy (TFB) of balloon-expandable metallic stent (BEMS) placement and subsequent forced, laser and balloon dilatation management might be less invasive and helpful. This study aimed to analyse the placement, follow-up management with TFB and long-term outcomes in small infants with BEMS.

Methods: This retrospective study reviewed the medical records and associated TFB videos of infants with a maximum body weight (BW) of 5.0 kg who had TB BEMS placement from January 2005 to December 2017 at our institution. All TFB procedures were supported with a novel noninvasive ventilation, nasopharyngeal oxygen with intermittent nose closure and abdominal compression.

Results: Forty-one BEMSs were placed in 24 infants. The mean BW and mean age were 4.0 ± 0.7 kg and 4.9 ± 2.4 months, respectively. There were 20, 8 and 13 stents located in trachea, carina and main-stem bronchi, respectively. Seven infants with 13 stents died without obvious stent-related mortality. Seven stents in five infants were successfully retrieved by rigid endoscopy (RE). At placement, the diameters of 28 tracheal and 21 bronchial stents were 7.5 ± 1.1 (4-10) and 5.4 ± 0.9 (4-8) mm, respectively. These implanted BEMSs could be gradually and significantly (p < 0.01) expanded. At the end of the follow-up period, all the remaining 21 stents in 12 infants were functional. The diameters of the 14 remaining tracheal and 13 remaining bronchial stents were 9.6 ± 2.0 (8-14) and 7.2 ± 1.4 (4-10) mm, respectively.

Conclusion: BEMSs are practical and effective in selected small infants with benign TB narrowing and can be safely implanted and managed with TFB, and finally retrieved by RE.

Keywords: Balloon dilatation; Balloon-expandable metallic stent; Flexible bronchoscopy; Nasopharyngeal oxygen with intermittent nose closure and abdominal compression.
At our institution, a referral centre for complicated and difficult paediatric airway diseases, many children were referred from both local and overseas tertiary centres for failed weaning following prolonged ET or positive-pressure ventilation (PPV) support. The standard management at our institution includes TFB with a noninvasive ventilation (NIV) technique, which we define as nasopharyngeal oxygen with intermittent nose closure and abdominal compression. This technique provides ventilation without using ventilation bags, masks, artificial airways or ventilators. Since 1997, we have gradually developed and employed this TFB-NIV for many airway procedures including TB BEMS implantation and subsequent management. This technique for BEMS placement and long-term outcomes in paediatric patients were reported recently. However, its implementation and outcomes in small infants have not been elucidated.

The purpose of this study was to report our experience using TFB-NIV in the management of TB BEMS including placement, maintenance, long-term outcomes and retrieval in small infants.

2. METHODS

This retrospective study included infants who fulfilled the following criteria: (1) a maximum body weight (BW) of 5.0 kg at the time of BEMS placement, (2) placement of BEMS between January 2005 and December 2017 and (3) available medical records and associated BEMS videos for review.

The following data were collected from the review of medical charts and associated videos: case number, age, sex, BW at the time of placement, symptoms, indication, major underlying aetiology, the stent diameter at the placement and the final balloon dilatation plasty (BDP), surveillance management, retrieval, complications, cause of mortality, the last FB before the end of study and long-term outcomes of these implanted stents and patients. The study was approved by the institutional review board of Taipei Veterans General Hospital (TVGH IRB No.: 2018-04-008AC).

All BEMS-associated procedures were performed in the endoscopy room of the paediatric intensive care unit with all instruments necessary for monitoring and resuscitation. All infants were in supine position, with the bronchoscopist standing by the infant’s head. Procedural sedation was titrated with intravenous midazolam (0.3-0.5 mg/kg), ketamine (1.0-2.0 mg/kg) and atropine sulfate 0.01 to 0.02 mg/kg (maximum 0.4 mg). Vital signs including heart rate, respiration and oxygen saturation by pulse oximetry (SpO2) were monitored continuously, and blood pressure was recorded intermittently using noninvasive methods from the initiation of the procedure until 4 hours after the procedure. Following the conclusion of the procedure, the infants were admitted to the paediatric intensive care unit for continuous care.

In the current cohorts, an FB with a short working length (25-36 cm) and an outer diameter of 3.2 to 5.0 mm (Olympus; HYF-V, ENF-VQ, ENF-V2 and ENF-VT2), with or without an inner channel, was introduced via the nostril of the infant. A brief summary of the NIV technique, reported previously, is as follows. A basic continuous, heated and humidified pure oxygen flow (1.0 L/kg/min, maximum 5.0 L/min) was supplied via a nasopharyngeal catheter to fill the upper airway cavity. An optional manoeuvre of assisted inspiration and expiration was performed as follows: the infant’s mouth was firmly closed with scopist’s index finger of right hand. Inspiration was assisted by nose closure (with thumb and mid-finger) accompanied with application of pressure to the cricoid. Expiration was assisted by the release of nose and cricoid manoeuvre with simultaneous abdominal compression. This assisted ventilation was optionally performed at a rate of 10 to 20 cycles per minute based on the patient’s monitoring. The scopist executed both the FB and nose closure (release) manoeuvre, whereas an assistant delivered the abdominal compression (release). No artificial ventilation bags, masks, airway tubes or mechanical ventilators were used during the entire FB procedure.

Indications for stent placement were severe TB malacia or narrowing with frequent life-threatening episodes and prolonged ET or PPV support, which could not be weaned. Stent placement was the last resort before more invasive surgical approaches. After stent implantation, subsequent TFB interventions were individually scheduled in periods of 2 to 6 months to inspect and manage associated complications. These TFB interventions included forceps debridement and laser ablation to remove granulation, BDP to debulk granuloma as well as to expand the stent to fit the growing TB lumen. All of these interventions were aimed to maintain the lumen and the patency of these implanted stents. During surveillance of management, the stent lumens could be gradually expanded and measured simultaneously using BDP. For each implanted stent, the final diameter was the balloon diameter used for the latest BDP. Surveillance periods varied based on the stent outcomes and started from the time of stent placement until 6 months after stent retrieval, death or the end of the study period.

Data were presented as means ± SD, medians with ranges and/or binomial percentages, as appropriate. Stent diameters were compared using paired samples t test, and a p value <0.05 was considered to indicate statistical significance.

3. RESULTS

The clinical characteristics of the study cohort are summarised in Table 1. There were 24 consecutive infants with 41 implanted stents during the 13-year period of this retrospective study. In all cases (100%), BEMS was successfully placed in the assigned location. At the time of stent placement, the median BW was 4.0 kg (range, 2.3-5.0 kg), and the median age was 4.8 months (range, 0.3-10.2 months).

The most common underlying aetiology of TB narrowing requiring stent placement was cardiovascular disease in 11 (45.8%) infants, followed by iatrogenic malacia in 6 (25.0%) infants with prolonged use of ET and PPV, tracheoesophageal fistula in 4 (16.7%) infants with severe tracheomalacia and idiopathic aetiology in 3 (12.5%) infants. After the restoration of the TB lumen with BEMS, dramatic respiratory improvement was achieved immediately with rapid weaning from respiratory support in all infants (100%). None of the infants underwent further surgical intervention for TB lumen problems.

The locations of the implanted stents are shown in the Fig. 1. Lower trachea was the leading location in 18 stents (43.9%), followed by the left main bronchus in 10 stents (24.4%). Eight carinal stents (19.5%) were placed using only one long-length...
(30-40 mm) stent, which was placed distally from one main bronchus, passed through the carina and ended in the lower trachea. The carinal stents were utilised to correct malacia in the pericarinal region. Their lower tracheal and bronchial portions were expanded separately by BDP using different-sized balloons to fit their luminal dimensions.

The outcomes of the infants and the stents are summarised in Table 2. At the end of the follow-up period, seven infants (29.2%) with 13 stents (31.7%) were dead due to the following causes: cardiopulmonary failure in four infants with seven stents, negative attitude of the parents in two infants with cerebral palsy with four stents and pulmonary bleeding in one infant with two stents. There was no apparent stent-related mortality. Seven stents (17.1%) in five infants (20.8%) were retrieved as they were no longer needed in the implanted lumens which had been judged well developed. Twenty-one stents (51.2%) remained in 12 infants (50%) at the end of the follow-up.

The mean surveillance period for all implanted stents was 39.9 ± 36.9 (range, 27.6, 1.4-144.8) months. The mean surveillance periods were 11.7, 52.9 and 52.9 months for the infants who expired, for those whose stents were removed and for those who retained the stents until the end of the study, respectively.

The main stent-associated problems included the following: obstructive granulation formation that caused lumen narrowing, stent distortion and/or loosening from the mucosal wall. All these could be successfully managed with the technique of TFB-NIV. All stent-associated infections or pneumonia could also be controlled appropriately.

At the time of placement, the diameters of 28 tracheal stents (including the tracheal portions of the eight carinal stents) and 21 bronchial stents (including the bronchial portions of the eight carinal stents) were 7.5 ± 1.1 (4-10) and 5.4 ± 0.9 (4-8) mm, respectively. The diameters of the remaining 14 tracheal (7 carinal) and 13 bronchial (7 carinal) stents were 9.6 ± 2.0 (8-14) and 7.2 ± 1.4 (4-10) mm, respectively, suggesting that the implanted BEMSs could be gradually and significantly expanded (p < 0.01).

### 4. DISCUSSION

To the best of our knowledge, this study with the largest cohort of small infants is the first to investigate management and associated long-term outcomes of BEMS implantation using the novel TFB-NIV technique. Several studies that reported TB BEMS in children either included small number of cases or in older age-groups. Their difficulties may bound by small-calibre airways, inappropriate respiratory support and susceptibility to hypoxia during the applications of traditional modalities including RE, ET intubation or while in transportation. Procedural sedation, intra-TB instrumentation and manipulation may further deteriorate the already present cardiopulmonary compromise. However, as previous reports in using the TFB-NIV technique, these BEMS-associated procedures can be managed in a safe, simple and effective manner.

All of the infants, included in the current study, had depended on ET or PPV before the stent placement. This NIV, nasopharyngeal oxygen with intermittent nose closure and abdominal compression, using pure oxygen flow and PPV was able to replace the original respiratory support without the constraints of a face mask or other invasive artificial airways such as laryngeal mask airway, ET and ventilator. Nasopharyngeal oxygen insufflation is simple and has already been widely used in clinical practise, with the advantages of easy accessibility, improved comfort, less oxygen flow and less invasiveness. Nose closure and abdominal compression can assist PPV and expiration. Additionally, abdominal compression can also contribute

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**Table 2**

Outcomes of tracheobronchial stents* in infants with a maximum body weight of 5.0 kg

<table>
<thead>
<tr>
<th>Clinical data</th>
<th>Number, %</th>
<th>Mean ± SD or median, range</th>
</tr>
</thead>
<tbody>
<tr>
<td>Stents</td>
<td>41 (100)</td>
<td></td>
</tr>
<tr>
<td>Infants</td>
<td>24 (100)</td>
<td></td>
</tr>
<tr>
<td>Outcomes, infant/stent</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Expired</td>
<td>7 (29.2)/13 (31.7)</td>
<td></td>
</tr>
<tr>
<td>Removed</td>
<td>5 (20.8)/7 (17.1)</td>
<td></td>
</tr>
<tr>
<td>Remain</td>
<td>12 (50.0)/21 (51.2)</td>
<td></td>
</tr>
<tr>
<td>Stent diameter, mm</td>
<td></td>
<td></td>
</tr>
<tr>
<td>At placement</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Tracheal</td>
<td>28</td>
<td>7.5 ± 1.1 (4-10)</td>
</tr>
<tr>
<td>Bronchial</td>
<td>21</td>
<td>5.4 ± 0.9 (4-8)</td>
</tr>
<tr>
<td>Remained</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Tracheal</td>
<td>14</td>
<td>9.6 ± 2.0 (8-14)*</td>
</tr>
<tr>
<td>Bronchial</td>
<td>13</td>
<td>7.2 ± 1.4 (4-10)</td>
</tr>
<tr>
<td>Stent surveillance period, mo</td>
<td></td>
<td></td>
</tr>
<tr>
<td>All</td>
<td>41</td>
<td>39.9 ± 36.9</td>
</tr>
<tr>
<td>Expired</td>
<td>13</td>
<td>27.6, 1.4-144.8</td>
</tr>
<tr>
<td>Removed</td>
<td>7</td>
<td>11.7 ± 11.4</td>
</tr>
<tr>
<td>Remaining</td>
<td>21</td>
<td>52.9 ± 36.0</td>
</tr>
<tr>
<td></td>
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<td>46.7, 6.6-144.8</td>
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*Balloon-expandable metallic stent, *p* < 0.01.

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to circulation, which was proven to be effective in cardiopulmonary support and resuscitation in both animals and adult humans. This NIV technique has been already demonstrated to be a safe, simple and effective respiratory support and rescue approach during various paediatric FB procedures. More advancement, during the TFB-NIV, the bronchoscopists may also have option to control the pressure intensity of the PPV and to dynamically adjusting to facilitate diagnosis as well as treatment.

After BEMS placement, regular medical management such as daily saline and steroids nebulisation with antigastroesophageal reflux regimens are routinely recommended. Scheduled once every 2 to 6 months, clinical and FB assessment of the approachable airway was performed with immediate and appropriate TFB, if indicated. The TFB-NIV technique used for the surveillance and management of BEMS-associated problems such as granulation formation, repair, expansion and retrieval were enough before they worsened significantly.

The mesh structure of the BEMS, compared with other silicon or membrane stents, aids in retaining the mucociliary function and allows airflow if the stent is placed over the branches. BDP indeed has served several functions in the current study. First, BDP was used to enlarge the stent openings over the bronchial orifices appropriately. Second, the luminal diameter of the BEMS could be further expanded to match the growing airways. Third, distorted stent structures could be repaired or remodelled without replacing with a new stent. Fourth, the stent-associated granulomas could be compressed or debulked simultaneously. No significant luminal obstruction was experienced in the carina or the regions of bifurcation after BDP in the current study. Finally, BDP could also be used to destroy an implanted stent to become a foreign body before its retrieval.

The indications for stent retrieval included nonfunction, destruction beyond repair, associated serious complications, stent placement of more than 2 years and apparently visible stent portion floating over mucosa by FB. In the current study cohort, seven stents, including five tracheal, one carinal and one bronchial stent, were successfully retrieved using RE with forceps. Before retrieval, the preparatory work included the insertion of a guidewire in the gap between the floated stent and the mucosa, followed by balloon inflation (BDP) to destroy and separate the stent from the mucosa for the subsequent retrieval as fragments or whole stent. The residual portions of stent were left on site after the appropriate repair with BDP.

The current study has several limitations. First, this was a retrospective study at a single institution and there was no control group for comparison because the TFB-NIV method employed in conjunction with the BEMS has been performed effectively at our centre for near 20 years. Nonetheless, future, larger cohorts and multicentre studies are necessary for further elucidation of this methodology.

In conclusion, BEMS can be an effective and conclusive therapeutic alternative to more invasive surgical interventions for TB malacia and narrowing. TFB-NIV is a safe, convenient and effective technique for BEMS placement and subsequent management in small infants. This approach also allows for successful retrieval of the implanted BEMS by RE once indicated.

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REFERENCES