Tracheobronchomalacia: follow-up study of 100 children treated with airway stenting

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Abstract

OBJECTIVE: We described a cohort of 100 children with a wide variety of airway obstruction who underwent stent positioning in the last 7 years. The study examined the outcomes of this treatment in the largest series of paediatric patients reported in the literature with special concern over safety and clinical effectiveness.

METHODS: We performed a retrospective analysis of 100 consecutive paediatric patients who underwent stent insertions between January 2005 and May 2012. Statistical analysis was performed and exact likelihood was used.

RESULTS: A total of 235 stents were placed for severe airway obstruction. One hundred and twelve silicone stents (cylinder, hourglass or Y-shaped), 120 metallic stents (covered Nitinol stents, expandable coronary and vascular stents) and 3 biodegradable polydioxanone (PDS) stents were used. Eighty patients presented clinical improvement after stent insertion, 17 were weaned off mechanical ventilation and 3 showed no significant clinical improvement [95% confidence interval (CI) 0.1–8.0%]. Complications were different according to stent type. In our cohort, no fatal stent-related complications have been observed. At follow-up (median 41.4 months, range 1.1–145.4) complete resolution was registered for 60 patients (66%; 95% CI 55–76%), 17 are still under treatment, 9 were lost to follow-up, 8 underwent surgery and 6 died of causes not stent related.

CONCLUSION: Airway stenting represents a conservative treatment before more invasive surgical procedures and can be very effective when performed in selected children and in specialized centres by physicians experienced in rigid and flexible bronchoscopy.

Keywords: Airway stent • Tracheobronchomalacia • Tracheobronchostenosis • Silicone stent • Metallic stent • Self-expandable stent • Cohort study

INTRODUCTION

Tracheobronchomalacia is a rare condition in children but associated with significant morbidity and mortality [1]. Children have a small airways size, and therefore, a significant and persistent decrease in diameter related to wall abnormalities, extrinsic compression or intraluminal causes might lead to fatal consequences [2]. Patients with severe airway obstructions require corrective treatments to guarantee a sufficient airstream. Several surgical and non-surgical treatments are available. An attractive non-surgical option is endoscopic airway stenting. The role of airway stenting is well established in adults, whereas the paediatric experience is still limited to small series and associated with high morbidity and mortality [3–6].

In the present paper, we report the follow-up of the cohort of patients treated in the period 2005–2012 at the Respiratory Endoscopy Unit of ‘A. Meyer’ Paediatric Hospital (Florence, Italy) for endoscopic stent placement with special concern over safety and clinical effectiveness.

MATERIALS AND METHODS

The study enrolled all 100 patients who had an airway stent placement from January 2005 to May 2012 at the Respiratory Endoscopy Unit of ‘A. Meyer’ Paediatric Hospital (Florence, Italy). Once ethics approval was obtained, patients’ clinical records were retrospectively reviewed to collect age, sex, associated diseases, type of airway obstruction, outcomes after stent placement and endoscopic follow-up. Stents were evaluated in terms of type, brand, placement technique, complications and permanence time in the airway. Primary diagnostic tool was bronchoscopy. Chest X-rays, magnetic resonance imaging or computed tomography (CT) were performed on an individual basis.
Technical success was defined as successful placement in the appropriate site in one bronchoscopic session. Clinical success was considered in terms of clinical improvement when the child was weaned off mechanical ventilation or there was a dramatic improvement in respiratory symptoms or a reduction of respiratory infection rate in the subsequent period after the procedure. The follow-up included a first bronchoscopic examination 3 weeks after stent placement to verify that the device was in the proper position and then each month for the first 3 months. For silicone and covered Nitinol stents, further bronchoscopies were performed every 2, 4 or 6 months depending on the tendency to develop granulation tissue. For stainless steel metallic stents, bronchoscopic follow-up continued every 4–6 months up to 1 year, at prefixed times at 1, 3 and 5 or 7 years of age to calibrate stent diameter according to airway growing and then until the final calibration. Polydioxanone (PDS) stents were endoscopically monitored every month until the complete reabsorption. Extra bronchoscopies were performed on an individual basis. The Endoscopic Unit provided 24/24 emergency bronchoscopies in case of complications. Complete resolution was considered when the airway lumen maintained >70% of its diameter after silicone stent removal or when stainless steel metallic stent did not need further calibrations and it was completely epithelialized and free from granulation tissue. Silicon stents were removed when no longer needed or when major complications occurred without any evident benefit of the device. Patients were considered lost to follow-up if they missed clinic appointment and were not contactable by telephone for at least 12 months after the last examination.

Indications for stent placement

Indications for stenting were malacia and stenosis of trachea and bronchi. Malacia, primary or secondary to oesophageal atresia, tracheoesophageal fistula (TOF), protracted endotracheal intubation, tracheostomy, external compression due to cardiovascular abnormalities or skeletal disorders, was diagnosed by airway endoscopic inspection under general anaesthesia without positive end expiratory pressure and treated when airway lumen was reduced more than 70%. When lumen reduction was within 50 and 70%, stent placement was indicated if failure to wean off ventilation, apnoea episodes, frequent respiratory infections (>3 pneumonia/year), severe respiratory distress or failure to thrive occurred.

Airway were classified as stenosed when the narrowing was fixed and unaffected by positive airway pressure. Congenital tracheal stenosis showed complete tracheal cartilage rings and congenital bronchial stenosis was associated with dysplastic cartilage. Acquired tracheobronchial stenoses were due to reconstructive tracheobronchial surgery, inflammatory process (i.e. endotracheal tube irritation and inhalational injury) or lung transplant anastomosis. Stenting was executed in symptomatic patients with moderate or severe fixed stenosis according to the classification reported by Burden et al. [7].

In case of acquired stenosis, laser incision and tracheobronchial balloon dilatation were the first line treatments in order to widen the airway, diode laser (Dornier MedTech) was used to make four radial cuts at the cardinal points of the stenotic circumference and if necessary, a balloon dilatation was performed with a dilating catheter passed into the narrowed segment of the tracheobronchial tree. Repeated balloon dilatations were usually required to achieve a sufficiently wide airway lumen. Stenting was used in case of relapsing stenosis needing a measure to better stabilize the airway. Before stent placement, a dilatation was often performed: only in case of severe tracheal relapsing stenosis treated with hourglass-shaped stents, the stenotic segment was not dilated and the narrowed part of the device was placed in the stenotic segment.

We did not perform stenting before surgical correction in children with vascular compression or with complete tracheal rings.

Stent types

Tracheobronchial stents used in our patients can be classified into four groups: soft silicone stents, metallic balloon expandable stents, self-expandable covered stents and polydioxanone (PDS) stents. Among silicone stents, Poliflex (Rusch) and Dumon (Novatech) were used; Dumon stents were cylindrical, hourglass- or Y-shaped. Stent diameter varied from 6 to 14 mm and length from 20 to 40 mm. Metallic stents are made of a stainless steel mesh. We used vascular stents (Jostent, Abbott; Palmaz, Johnson&Johnson) with an expansion range within 4 and 12 mm and a length of 12 or 17 mm in children principal bronchi and coronary stents (Multilink, Abbott) with a diameter of 3–4 mm and a length of 8 mm in preterm main bronchi or in lobar bronchi of the older child. Self-expandable polyester covered Nitinol stents (Silmet), a special biocompatible and resistant material that changes shape as a response to an applied stress, were used with a diameter ranging from 6 to 12 mm and a length from 20 to 40 mm. PDS stents are made of polydioxanone, a biodegradable polymer belonging to the polyester family. Its monofilament exhibits some shape memory and tends to coil.

Stent type was selected according to endoscopic indication. Silicone or covered Nitinol stents, both easily removable, were preferred in case of tracheomalacia or tracheostenosis and stainless steel metallic stents in case of bronchomalacia or bronchostenosis. Different stent types were inserted if both tracheal and bronchial disease coexisted.

Technical considerations

Before stent placement, informed consent was obtained from child parents outlining all potential risks (including airway perforation, infection, dislocation, obstruction, respiratory distress and death). Once the area requiring stent insertion was visualized by flexible bronchoscopy (Karl Storz, Germany; Ø 2.5/3.7/5.2 mm), we assessed the distance of the stenosis/malacia from the dental arches and the length of the affected segment by rigid bronchoscope. In case of stenosis, stent external diameter corresponded to that of the largest bronchoscopy tube passing through the stenosis; in case of malacia the diameter was chosen 2–4 mm larger than the measured value of the diameter of the trachea or bronchi. Stent placement was performed in the operating theatre using a rigid bronchoscope (Karl Storz, Germany; Ø 3.5/4/4.5/5.5 mm) under general anaesthesia (sevoflurane inhalation ± propofol i.v.) and radiological control.

No specific insertion device is currently available to place silicone stents in paediatric patients. Therefore, the paediatric rigid bronchoscope was needed to insert the device. The bronchoscope was introduced in the trachea under fluoroscopic control and a radiopaque marker was placed on the thorax of the child in the corresponding segment to treat. Then, the stent was lubricated, folded
along the longitudinal axis, inserted at the distal end of the rigid bronchoscope (Fig. 1) and placed within the airway at the level previously established. Since the patient could not be ventilated during device placement, prior to stent insertion the patient was ventilated with 100% oxygen for 3–4 min to guarantee a sufficient oxygenation during manoeuvre. Apnoea of ≏2 min was well tolerated. Under continuous fluoroscopic control, the rigid bronchoscope was moved forward with gentle turning movements at the level of the radiological marker. Then, a rigid optical device maintained the stent in the right position while the rigid bronchoscope was withdrawn. At the end of the procedure, a bronchoscopic examination confirmed the correct location of the device. In case of incomplete deployment, the stent was expanded by means of forceps, rigid optical instrument or angioplasty balloon. Maintenance of silicone stent included laser removal of granulation tissue (Fig. 2) and substitution with a larger size to follow airway growth.

Metallic balloon expandable stents were inserted in the airway mounted on an appropriate-size balloon catheter and self-expandable stents through their own delivery system. Vascular and coronary stents were provided wrapped around an angioplasty collapsed balloon: they were placed through the rigid bronchoscope in the airway segment and expanded under radiographic control by filling a balloon with a standard angioplasty syringe equipped with a pressure gauge. The selected balloon diameter corresponded with the desired diameter of the bronchus. Then, the catheter was removed by deflating the balloon and a bronchoscopic examination immediately after the placement confirmed the correct location of the device (Fig. 3). It is worth mentioning the insertion technique of coronary stents in lobar bronchi because they require a careful measurement of the distance between the affected lobar bronchus and the next lobar ramification. In some cases, because of the shortness of the narrowed lobar segment compared with the device length, the tip of the wire mesh was turned over the orifice of the bronchus by forceps.

In infant upper lobar bronchi, we often used a soft tipped guidewire (Ø 0.25 mm; St Jude Medical®, MN, USA) to reach the lesion and to provide a pathway for the stent which was moved forward over it. These devices were adapted during airway growth and over-expanded to reach a maximum diameter of 6 mm to guarantee adequate patency. Maintenance of metallic expandable stents included calibrations and dilatations both performed with balloon dilatation under fluoroscopic control using a rigid bronchoscope. We used the term ‘calibration’ to refer to dilatations required to adequate stent dimension to the progressive growth of the child’s airway, while we used balloon ‘dilatation’ to refer to the procedure performed in case of stent ovalization and loss of adherence or granulation tissue formation both to ri-expand the ovalized stent and to compress granulation tissue against the airway wall. The balloon diameter was chosen according to the size of the stent and to the desired diameter of the bronchus. The dilating catheter was passed into the stent inserted in the selected position and the balloon inflated with diluted contrast material for a maximum inflation time of 60 s. Then, the catheter was removed by deflating the balloon and a bronchoscopic examination immediately after the dilatation confirmed the correct placement of the device.

Calibrations, dilatations and laser removal of minor granulations with diode did not require hospital admission.
PDS stents were manually inserted by rigid bronchoscope. A postoperative chest X-rays film provided radiographic documentation. After stent insertion patients were admitted to the sub-intensive care unit for −48 h to undergo a course of systemic corticosteroids and to perform chest X-rays 24 h after the procedure. Inhalation therapy with nebulized adrenaline and β2-agonist were usually administered.

**Statistical analysis**

A descriptive statistical analysis was performed to summarize the variables of interest. Pearson’s $\chi^2$ test of independence and exact test was performed on tables of frequency classifying stent type and indication, complications and patient outcomes (StatXact® version 10th). Eventually, we computed a test on the equality of proportions to compare the proportion of the main complications of silicone and metallic stents. When appropriate, confidence intervals (CIs) were calculated using exact likelihood. The level of significance was set to 5% two-sided. Results for quantitative variables are expressed as mean ± standard deviation or median (range).

**RESULTS**

**Population**

The study enrolled 100 consecutive patients (63% males) who underwent airway stenting. Forty-one patients were hospitalized in the intensive care unit: 17 presented difficult weaning off mechanical ventilation after surgery for congenital heart disease (CHD) ($n = 3$), vascular ring ($n = 3$), TOF or oesophageal atresia ($n = 7$), poli-trauma ($n = 1$), congenital diaphragmatic hernia ($n = 1$) and tracheal hypoplasia ($n = 2$); 24 showed respiratory distress after extubation. The latter group included children with congenital lobar emphysema ($n = 1$), lung hypoplasia ($n = 1$), VACTERL syndrome ($n = 1$) and children in the postoperative period [CHD ($n = 2$); vascular ring ($n = 9$); laringotracheal cleft ($n = 1$); TOF or oesophageal atresia ($n = 4$) and tracheal hypoplasia ($n = 5$)]. Twenty-seven patients were transferred by the ordinary paediatric ward because of recurrent lower respiratory tract infection (LRTI) ($n = 3$), chronic cough ($n = 3$), radiological signs of chronic atelectasis or hyperinflation ($n = 10$), stridor ($n = 5$), nocturnal apnoea ($n = 1$) or persistent asthma ($n = 5$).

The remaining patients ($n = 26$) were referred to our outpatient clinic to perform a bronchoscopic assessment because of radiological signs of chronic atelectasis or hyperinflation ($n = 2$), stridor ($n = 5$), recurrent LRTI ($n = 6$), oxygen desaturation ($n = 1$) and chronic cough ($n = 12$). Furthermore, 2 patients with cystic fibrosis were referred to evaluate stenotic lesions after lung transplant at the site of bronchial anastomosis and 4 patients to consider weaning off tracheostomy.

Our population data including primary disease, endoscopic findings and outcomes are reported in Table 1.

**Stents**

Overall, we placed 235 stents (a median of two stents per patient; range 1–11). Patient and stent characteristics are summarized in Table 2. Considering stent location, 40 patients required stenting in the trachea, 51 in the bronchial tree and 9 in both sites. Bronchial stents were placed in left main stem bronchus (45%), middle lobar bronchus (19%), left upper lobe bronchus (15%), right mainstem bronchus (12%), left lower lobe bronchus (4%), intermediate bronchus (4%) and right lower lobe bronchus (1%).

Endoscopic indications and stent complications are respectively reported in Tables 3 and 4. Choice between silicone and metallic stent depended on endoscopic indication—exact Pearson $\chi^2$ (15 degrees of freedom) = 292.2; $P < 0.001$. Silicone stents were mainly placed in trachea; stainless steel metallic stents were mainly placed in bronchi. Complications were different according to stent type—exact Pearson $\chi^2$ (25) = 175; $P < 0.001$. No complications were reported in 24/112 silicone stents and in 21/120 metallic stents. Silicone stents were more prone to granulation tissue formation [major granulations 11.6 vs 0.8%; a difference of 10.8% 95% confidence interval (CI) 5–17%; $P = 0.0006$; minor granulations 27.6 vs 12.5%; a difference of 15.2% 95% CI 5–25% $P = 0.0038$] and dislocation (39.2 vs 4.1%; a difference of 35.1% 95% CI 25–45% $P < 0.0001$) compared with metallic stents. Compared with the other metallic stents, covered Nitinol stents showed more granulation tissue formation: 50 vs 8.5%, a difference of 41.5% 95% CI 15–68% $P < 0.0001$.

**Silicone management**

Each stent developing granulation tissue ($n = 13$) required a mean of 1 laser treatment (range 1–6). Of the 112 silicone stents, 44 (39%) were substituted following their dislocation: 36 were replaced with a larger one and 8 with a covered Nitinol stent to obtain greater stability. Thirteen stents (12%) were removed and substituted because of obstructing granulation tissue and 13 (12%) because of dislocation: 36 were replaced with a larger one and 8 with a PDS stent. Of the 112 silicone stents, 44 (39%) were substituted following their dislocation: 36 were replaced with a larger one and 8 with a covered Nitinol stent to obtain greater stability. Thirteen stents (12%) were removed and substituted because of obstructing granulation tissue and 13 (12%) because of dislocation: 36 were replaced with a larger one and 8 with a covered Nitinol stent to obtain greater stability. Of the 112 silicone stents, 44 (39%) were substituted following their dislocation: 36 were replaced with a larger one and 8 with a covered Nitinol stent to obtain greater stability. Thirteen stents (12%) were removed and substituted because of obstructing granulation tissue and 13 (12%) because of dislocation: 36 were replaced with a larger one and 8 with a covered Nitinol stent to obtain greater stability. Of the 112 silicone stents, 44 (39%) were substituted following their dislocation: 36 were replaced with a larger one and 8 with a covered Nitinol stent to obtain greater stability.

**Metallic stents**. Each stent developing granulation tissue ($n = 13$) required a mean of 1 laser treatment (range 1–4) laser treatment. A mean median of 3 (range 1–16) calibrations/dilatations per stent were required. Five covered Nitinol stents were removed and substituted: 4 because of dislocation and 1 because of exuberant granulation tissue formation. One not mucolized Multilink stent was removed after 22 days because of dislocation and was substituted by a Jonmed stent.

**Outcomes and follow-up**

After stent insertion (on average 2–3 days), 80 patients presented clinical improvement and 17 were weaned off mechanical ventilation. Three patients, 2 with congenital lobar emphysema and 1 with a surgically corrected vascular ring, showed no significant clinical improvement (3%; 95% CI 0.1–8.0%) (Table 1).

Outcomes and stent type are reported in Table 5. Complete resolution at follow-up was registered for 60 patients (66%; 95% CI 55–76%), 17 are still under treatment, 9 were lost to follow-up, 8 underwent surgery and 6 died.

Patients with both metallic and silicone stents showed a more severe clinical condition and needed stenting in multiple positions. The percentages of resolution were, respectively, 73% (95%
<table>
<thead>
<tr>
<th>Primary disease</th>
<th>Endoscopic findings at first assessment</th>
<th>n</th>
<th>Clinical result after stenting</th>
<th>Definitive outcomes</th>
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<tbody>
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<td>Congenital lobar emphysema</td>
<td>MLB m + LULB m</td>
<td>1</td>
<td>Clinical improvement</td>
<td>Resolution</td>
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<tr>
<td></td>
<td>LULB s</td>
<td>1</td>
<td>Clinical improvement</td>
<td>Resolution</td>
</tr>
<tr>
<td></td>
<td>LULB m</td>
<td>1</td>
<td>Clinical improvement</td>
<td>Resolution</td>
</tr>
<tr>
<td></td>
<td>LULB m</td>
<td>4</td>
<td>2 clinical improvement, 2 not improved</td>
<td>2 resolution, 2 surgery</td>
</tr>
<tr>
<td>Congenital diaphragmatic hernia</td>
<td>MLB m</td>
<td>1</td>
<td>Clinical improvement</td>
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<tr>
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<td>LMSB m</td>
<td>1</td>
<td>Clinical improvement</td>
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<tr>
<td>Prematurity, BPD</td>
<td>LULB m</td>
<td>2</td>
<td>2 clinical improvement</td>
<td>1 resolution, 1 lost to follow-up</td>
</tr>
<tr>
<td></td>
<td>LMSB m</td>
<td>1</td>
<td>Clinical improvement</td>
<td>Dead</td>
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<tr>
<td></td>
<td>RMSB s</td>
<td>1</td>
<td>Clinical improvement</td>
<td>Resolution</td>
</tr>
<tr>
<td>Collagenopaty</td>
<td>TM + LMSB m + RMSB m</td>
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<td>Clinical improvement</td>
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<td>Associated with identified syndromes</td>
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<td>2</td>
<td>2 clinical improvement</td>
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</tr>
<tr>
<td></td>
<td>LULB m</td>
<td>1</td>
<td>Clinical improvement</td>
<td>Resolution</td>
</tr>
<tr>
<td></td>
<td>MLB s</td>
<td>3</td>
<td>3 clinical improvement</td>
<td>3 resolution</td>
</tr>
<tr>
<td></td>
<td>LMSB m</td>
<td>1</td>
<td>Clinical improvement</td>
<td>Resolution</td>
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<tr>
<td></td>
<td>LLLB m + LMSB m + LB m</td>
<td>1</td>
<td>Clinical improvement</td>
<td>Under treatment</td>
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<tr>
<td></td>
<td>LMSB m</td>
<td>2</td>
<td>2 clinical improvement</td>
<td>2 resolution</td>
</tr>
<tr>
<td></td>
<td>RMSB s</td>
<td>1</td>
<td>Clinical improvement</td>
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</tr>
<tr>
<td>Vascular ring</td>
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<td>13</td>
<td>3 weaned off, 9 clinical improvement, 1 not improved</td>
<td>9 Resolution, 1 under treatment, 1 surgery, 1 lost to follow-up, 1 dead</td>
</tr>
<tr>
<td></td>
<td>RMB m + MLB m</td>
<td>1</td>
<td>Clinical improvement</td>
<td>Under treatment</td>
</tr>
<tr>
<td></td>
<td>LMSB m</td>
<td>8</td>
<td>8 clinical improvement</td>
<td>5 resolution, 1 lost to follow-up, 2 dead</td>
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<tr>
<td></td>
<td>RMSB m</td>
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<td>7 resolution, 4 surgery, 2 lost to follow-up</td>
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<td></td>
<td>TM + LMSB s</td>
<td>1</td>
<td>Clinical improvement</td>
<td>Surgery</td>
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<tr>
<td></td>
<td>TM + LMSB s + RMSB s</td>
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<td>Weaned off</td>
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<tr>
<td></td>
<td>TM + MLB m</td>
<td>1</td>
<td>Clinical improvement</td>
<td>Under treatment</td>
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<td></td>
<td>TM + MLB s</td>
<td>1</td>
<td>Weaned off</td>
<td>Under treatment</td>
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<tr>
<td></td>
<td>TM + LMSB m</td>
<td>1</td>
<td>Clinical improvement</td>
<td>Under treatment</td>
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<td></td>
<td>TS</td>
<td>1</td>
<td>Clinical improvement</td>
<td>Resolution</td>
</tr>
<tr>
<td>CHD</td>
<td>LMSB m</td>
<td>8</td>
<td>3 weaned off, 5 clinical improvement</td>
<td>5 resolution, 1 under treatment, 1 lost to follow-up, 1 dead</td>
</tr>
<tr>
<td></td>
<td>RMB m + LMB m</td>
<td>1</td>
<td>Clinical improvement</td>
<td>1 dead</td>
</tr>
<tr>
<td>Tracheal hypoplasia (corrected by tracheoplasty)</td>
<td>TM</td>
<td>4</td>
<td>4 clinical improvement</td>
<td>3 resolution, 1 under treatment</td>
</tr>
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<td></td>
<td>TS</td>
<td>3</td>
<td>2 weaned off, 1 clinical improvement</td>
<td>2 resolution, 1 under treatment</td>
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<tr>
<td>Tracheal traumatic rupture</td>
<td>TS</td>
<td>1</td>
<td>Weaned off</td>
<td>Resolution</td>
</tr>
<tr>
<td>Cystic fibrosis + lung transplant</td>
<td>MLB m + RLLB m</td>
<td>1</td>
<td>Clinical improvement</td>
<td>Resolution</td>
</tr>
<tr>
<td></td>
<td>LLLB s + MLB s + LULB m</td>
<td>1</td>
<td>Clinical improvement</td>
<td>Under treatment</td>
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<td>Tracheal angiomatosis</td>
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<td>Tracheostomy (endoscopic examination for decannulation)</td>
<td>IB s</td>
<td>1</td>
<td>Clinical improvement</td>
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<td></td>
<td>TS + RMSB s</td>
<td>1</td>
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<td></td>
<td>TM</td>
<td>1</td>
<td>Clinical improvement</td>
<td>Lost to the follow-up</td>
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<tr>
<td></td>
<td>TM + LMSB m + MLB m</td>
<td>1</td>
<td>Clinical improvement</td>
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<tr>
<td>Oesophageal duplication cyst</td>
<td>TM</td>
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<td>Bronchocele</td>
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<td>TM</td>
<td>1</td>
<td>Clinical improvement</td>
<td>Resolution</td>
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CI 60–86%) for silicone; 77% (95% CI 62–92%) for metallic and 15% (95% CI 4–35%) for silicone and metallic.

Technical success was achieved at first attempt in all but 3 patients in whom a metallic balloon expandable stent was placed in the erroneous position requiring removal and substitution during the same bronchoscopic session. The procedure was well tolerated in all patients.

Surgery

Conservative treatment with stenting was not effective in 8 cases who eventually required surgical intervention: 5 of the 8 patients underwent aortotruncopexy for persistent residual tracheomalacia after TOF corrective surgery, 2 of the 8 needed lobectomy of the left upper lobe for congenital lobar emphysema and 1 of the 8 a second aortotruncopexy for relapsing right bronchomalacia.

Biodegradable polidioxanone stents

Three PDS stents were implanted in 3 patients.

Patient 1. A 2-year old girl with post-haemorrhagic hydrocephalus presenting with tracheostomy and ventilator dependency showed laryngotracheal stenosis and multiple bronchial obstructions. She underwent laryngotracheoplasty, stent positioning in left main stem bronchus and intermediate bronchus with successful decannulation. After 1 month she developed severe respiratory distress due to postoperative tracheal stenosis; a silicone stent then substituted by a covered Nitinol stent was inserted in the trachea but both were removed because of major tissue granulation formation. In an attempt of reducing the granulation tissue formation rate, we placed a PDS stent (8 × 20 mm) in the trachea, but its radial force was not enough to guarantee the patency of the lumen and a tracheostomy was eventually required.

Patient 2. A 6-month old boy with CHD showed difficulty weaning off invasive ventilation in the postoperative period due to complete left lung atelectasis related to left main stem bronchus malacia. He was treated with a PDS stent (6 × 15 mm) and after 48 h was weaned off mechanical ventilation. After 4 months, the stent was completely reabsorbed but for the persistent severe left bronchomalacia shown at the endoscopic evaluation, a metallic stent was eventually inserted.

Patient 3. An ex-preterm baby with Feingold syndrome (oesophageal atresia/TOF), cervicostomy and tracheostomy presented with stenosis of the larynx, of the distal segment of the trachea and of left and right main stem bronchi related to multiple granulations. Granulation tissue in the right main stem bronchus was removed by laser ablation, the left main stem bronchus stenosis was treated with a vascular stent and the tracheal stenosis with a silicone stent. The latter was removed after 2 months because dislocated and substituted by a covered Nitinol stent removed after 3 months because of recurrent obstructing granulation tissue. A PDS stent (10 × 22 mm) was inserted in the stenotic tracheal segment but then removed after 10 days because it was not effective. The stenosis was eventually treated with a Y-shaped silicone stent that is still in place.
DISCUSSION

We described a cohort of 100 children with a wide variety of airway obstructions who underwent stent positioning in the period 2005–2012. Technical success was achieved in 100% of patients and clinical success in 66% (95% CI 55–76%). None of the deaths recorded in our population was related to stent insertion or its permanence in the airway. In particular, the patient who died because of unsuccessful intubation had the stent removed months before he underwent laryngotrachealplasty. The death...
occurred in another hospital and no information was available about the complications encountered by the colleagues at the time of intubation.

Airway stenting in paediatric population is a relatively recent technique and since 1980 a small number of cases have been published [8-13].

Compared with the largest paediatric series reported in the literature [6, 13], our series includes a greater number of patients and shows a higher percentage of success (66% 95% CI 55–76% vs 53% 95% CI 35–71%; and 52% 95% CI 34–69%). A recent review [14] reported several publications focused on the severe and even lethal stent-related complications in children with a derived mortality of 13%. These data may reflect that in most paediatric series stenting was performed as a last resort in children not clinically stable and who were too sick for other interventions [1, 4, 6, 10, 15, 16]. Our positive results may have been related to our cautious policy about where, when and how to perform a stenting. Our centre performs more than 1200 paediatric bronchoscopies/year, both diagnostic and interventional, and we consider stent placement as conservative treatment only when other medical treatments have failed (including non-invasive ventilation and respiratory physiotherapy) and before considering more invasive surgical options such as tracheostomy or tracheoplasty.

Clinical indication was carefully evaluated before stent insertion. In case of airway obstruction due to vascular compression, stenting was not performed before surgical correction because the high pressure on the mucosa may result in ischaemia and subsequent severe airway damage as reported in some cases [1, 3, 17]. We suggest stent placement only in case of remaining malacia/stenosis with failure to wean off mechanical ventilation or persistent respiratory distress after the surgical correction. We treated 24 patients with this indication and in 23 of 24 we noticed a dramatic improvement in respiratory condition as soon as the stent was inserted without any of the ensuing problems reported in the literature [3]. One of 24 patients required a second surgery after tracheal stent removal because of a persistent vascular compression on the distal trachea and on the right main stem bronchus; after aortopexy a further stenting was not needed. As for vascular compression, we did not place stents if trachea displayed complete tracheal rings because of the risk of tracheal erosion or penetration into the great vessels especially into the innominate artery. Maeda et al. [18] described their experience in 5 patients treated with tracheal stents after intraluminal balloon dilatation of complete tracheal rings with moderate results but the possible long-term risks of this procedure are not predictable. Conversely, very gratifying results with stenting have been obtained in infants with airway obstruction developed after reparative surgery for structural tracheal stenosis [19]. In our series, 7 patients underwent tracheal stenting for residual airway obstruction after tracheoplasty for tracheal hypoplasia with a significant improvement in their clinical condition and all are alive and well without respiratory symptoms. In 5 of 7 patients stents were removed during follow-up and all patients are asymptomatic from a respiratory standpoint.

The use of tracheal stents is reported in case of residual malacia with persistent respiratory symptoms or difficult weaning off ventilation after surgical correction of oesophageal atresia with congenital TOF [20]. We treated 19 patients with this condition and only 4 eventually required another surgical intervention. Three cases underwent aortotruncopexy because of the variation of anatomical relationship between airway and vascular structures probably due to oesophageal dilatation after surgical correction. In 1 patient stenting was not effective because he was found to have a relapsing TOF.

Conservative treatment of children with congenital lobar emphysema is an attractive option [21]. In our cohort, stenting was effective in 5 of 7 patients with congenital lobar emphysema and left upper lobar bronchus obstruction; 1 patient presented multiple bronchial impairment. The good result obtained in 5 patients was attributed to the fact that stenting was performed before irreversible damage to the lung (septal alveolar disruption and alveolar bleeding) occurred. In the 2 unsuccessful cases, stenting did not prevent the surgical approach. However, a word of caution is necessary because longer follow-up for patients conservatively treated for congenital lobar emphysema is required. Surgery cannot yet be excluded in the future of these patients.

Stent type must be carefully selected considering location, indication and severity of the obstruction, since each stent has different properties and advantages. The mesh of metallic stents helps to preserve mucociliary clearance and if placed over a bronchial orifice allows the airstream through its wires. Hence, in case of bronchial obstruction, we used stainless steel metallic stents. In 6 patients 2 covered Nitinol stents and 9 Dumon stents were inserted in a bronchial site because the proper size of the stainless steel metallic stent was not available at the time of the procedure.
and stents were supposed to stay in place for less than one month. In case of severe malacia of distal trachea involving carina, the Y-shaped silicon stents must be measured very carefully. In 3 patients in whom this type of stent was inserted, we personally modified the bronchial segments to avoid the obstruction of the bronchial orifices. None of these 3 patients experienced atelectasis or problems related to poor ventilation of the bronchial segment due to the stent position. In our population, only silicon or covered Nitinol stents were placed in the trachea, since they are both removable and they are not re-epithelialized. Hence, metallic uncovered expandable stents, due to their lack of elastic re-expansion, can be broken or deformed with coughing so that they should not be used in central airways [11]. In our experience, the only stainless steel metallic stent placed in trachea broke 12 years after the insertion and required the resection of the tracheal segment with subsequent tracheoplasty (unpublished data).

Considering complications, the literature reports that granulation tissue formation is more common in metallic stents than in silicone stents. In our series, the latter were more prone to develop granulation tissue. Noteworthy, Nitinol and silicone stents had a similar granulation tissue formation rate. This finding is in contrast with previous studies and leads to some considerations.

In some series where metallic stents were placed both in the trachea and bronchi, granulations were found to be very common [1, 22]. According to Pacheco et al. [12], bronchial metallic stents are probably better tolerated than those placed in trachea likely due to the fact that trachea is subject to greater forces as during coughing. In animals studies with metallic stents airway inflammation and granulations occurred more frequently in those animals in which the stents had been over-expanded [23]. Considering these findings, our lower granulation rate (minor granulation 13%, 95% CI 8–21%) compared with the rest of the literature [4, 7, 9] may be due to our policy of inserting metallic expandable stents only in bronchi and of limiting the overexpansion to the stents placed only in the lobar ones. Nevertheless, we did not find any significant correlation between coronary stents overexpansion and excessive granulation formation.

Considering silicone stents, our study included a greater number of this type of stents compared with the largest series described in the literature [13] in which the authors reported a significant granulation tissue formation necessitating bronchoscopic intervention. Fayon et al. [11] reported the use of 26 silicone stents in 14 children: granulomatous reaction was seen in 5 of 26 stents and was attributed to abnormal prosthetic mobility.

Reviewing the few data on silicone stents present in the literature and considering our large series, we hypothesize that the higher formation of granulations encountered can be due to the main properties of the silicone stent:

(i) greater mobility compared with metallic stents;
(ii) interruption of mucociliary clearance, mucous plugging and consequent bacterial growth;
(iii) prior condition of tracheal inflammation before stents insertion (either for chronic inflammation or for previous surgical or endoscopical treatments on the tracheal mucosa).

In our series, device dislocation occurred in 39% (95% CI 30–48%) of silicone stents. Only when the external diameter was larger than the airway lumen, enough pressure was exerted towards the wall to keep the silicone tube in place. When the stenosis was severe, we preferred hourglass-shaped stents and we noticed that if the narrowed part was placed in the stenotic segment, the hourglass-shaped stent was very stable and efficacious. Furthermore, silicon stents interrupt mucociliary clearance and the mucosa covered by the device becomes prone to mucus accumulation and secondary infections. Nevertheless, in our population no severe complications with luminal obstruction have been observed thanks to the close monitoring of patients, especially of patients with Y-shaped stents. In 8 patients suffering from severe and relapsing tracheal stenosis, the Dumon stent, excessively prone to dislocation, was substituted by a covered Nitinol stent. Nitinol is exceptionally bio-compatible and the special mesh structure is highly resistant to corrosion and compression minimizing the risk of trauma. Nitinol stents do not exert a significant constant pressure against the wall but if chosen larger than the actual airway size, they are less prone to dislocation [14]. For this reason, Nitinol has been proposed as the optimal material for airway stenting. In our series, we used only self-expandable covered Nitinol stents because all tracheal-bronchial lesions were benign and it would be ideal to remove the stent when no longer needed. However, the use of covered Nitinol stents in children has not been widely documented and a comparison with other experience is not suitable. In our own experience, this stent showed pronounced granulation tissue formation in 7 of 14 stents requiring removal in just 1 case. No bleeding or severe mucosa damage was described during stent removal. Our hypothesis is that even if the stent does not increase its pressure on the tracheal wall after expansion (in contrast to the self-expandable stent), when applied on the soft mucosa of paediatric airway, it results in impaired microcirculation and promotes granulations. In addition, although covered metallic stents can decrease the ingrowth of hyperplastic tissue through the wire mesh, granulation tissue may still grow over either end of the stent. An experimental study of animals comparing biocompatibility of covered and uncovered self-expandable stents in rats reported a significant high incidence of granulation tissue in the proximal and distal border both in covered and uncovered Nitinol stents (42.9 vs 40%) but mucosa inflammation was greater in covered stents [24]. Considering the few studies describing the use of covered Nitinol stents in paediatric airway obstruction, more data are needed to fully understand the real etiopathogenesis of granulations in this type of stent.

In stainless steel metallic stents ovalization was the main complication occurring in 62% (95% CI 54–71%). Nevertheless, routine ballooning of metallic stents appears dangerous and should be considered only when airway obstruction is clearly attributable to small stent size [14]. In the current series, we reported the breakage of a metallic stent placed in a bronchus of a patient affected by Jeune syndrome. Since the stent was completely re-epithelialized, the breakage was suspected because airway lumen was ovalized and stent radial force was not maintained after repeated dilatations. A thorax CT scan revealed the ovalization and the minimal longitudinal breakage of the device. No further stenting was performed considering the evolution of the chronic restrictive disease.

Although Furman et al. [9] reported a mean time of mucosalization of metallic stents of 44 days, in our population it was found to be highly variable with a range of 1.5–5.3 months. Before complete epithelialization, the stent may be removed if necessary but the procedure can be very risky and it must be performed by experienced physicians. In these exceptional cases, in order to avoid airway laceration, we clamped the device with forceps and we gently withdrew the forceps and the stent slightly folded into the working channel of a rigid bronchoscope. In our series, four stainless steel stents had to be removed: three as soon as they
were inserted because they had been placed in the wrong position and one Multilink stent after 22 days because of dislocation. In all cases, no significant bleeding was observed. Considering our experience and the serious complications reported by other authors after metallic stent removal [6], this procedure must be avoided straightforward and epithelialized stents must be considered permanent.

Some authors expressed doubts on the long life of stainless steel metallic stents because they stay in the airway for a lifetime [4]. Our protocol includes stent calibration at different ages to follow the physiological growth of the airway until the final calibration which corresponds to the largest stent diameter. Exceptional extra dilations can be executed on an individual basis considering that if the stent is over-expanded the radial force may decrease and an overlapping stent may be necessary to maintain the lumen open. In our series where the median follow-up was ~5 years, no issues related to airway growth have been noted.

Considering biodegradable stents, Vondrys et al. [25] described the first experience with PDS in children with promising results. Our very preliminary experience with the use of absorbable stents is limited to 3 patients presenting extremely severe clinical conditions. The small size and the characteristics of our case series led to unsatisfactory results. Further studies are required to establish the effectiveness of these devices in children with airway obstruction.

The present study has some limitations. It is an observational study of a single centre with no comparison with other experiences and there is no control group to compare stent intervention against other therapies. Long-term prospective multicentre studies are needed to clearly define clinical indications for airway stenting and its therapeutic value in selected patients.

**CONCLUSION**

To obtain satisfying results, stenting must always be performed in specialized centres by health care operators very experienced both in rigid and in flexible bronchoscopy. A careful consideration of the right patient is mandatory and each stent must be selected appropriately considering the expected permanence in the airway and the site where it has to be placed.

Adequate follow-up is crucial to avoid life-threatening events and to guarantee an appropriate management of the possible complication.

**Conflict of interest:** none declared.

**REFERENCES**


