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Management of tracheobronchial obstruction in infants using metallic stents: Long-term outcome

Ling LEUNG[1] **MBBS(HK), MRCS**

Patrick Ho Yu CHUNG[1] **FRCSEd(Paed), FHKAM(Surg)**

Kenneth Kak Yuen WONG[1*] **PhD, FRCSEd(Paed), FHKAM(Surg)**

Paul Kwong Hang TAM[1] **ChM, FRCS, FRCPCH**

[1] **Division of Paediatric Surgery, Department of Surgery, The University of Hong Kong, Queen Mary Hospital, Hong Kong**

***Corresponding author:** **Dr. Kenneth Kak Yuen WONG**

Division of Paediatric Surgery

Department of Surgery

The University of Hong Kong

Queen Mary Hospital

Pokfulam Road

Hong Kong

Tel: (852) 2855 4850

Fax: (852) 2817 3155

Email: kkywong@hku.hk

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Management of tracheobronchial obstruction in infants using metallic stents: Long-term outcome

Abstract

Introduction:

Tracheobronchial obstruction, although uncommon in the paediatric age group, remains a challenging problem. We review the long term outcome of endoscopic metallic stenting in infants with tracheobronchial obstruction.

Materials and methods:

Medical records of all paediatric surgical patients who underwent tracheobronchial metallic stenting in our centre were reviewed retrospectively from 1996 to 2014. Patients' demographic data, including etiology, associated anomalies and nature of obstruction were reviewed. Outcome measures include complications such as re-stenosis, granulation tissue, stent migration, fractured stent, maximal tracheal diameter achieved, weaning of ventilator and growth at interval follow-up.

Results:

Twelve balloon-expandable metallic stents were placed in the trachea (n=10) and/or bronchi (n=2) of 5 patients with a median age of 13 months (range 5 - 30 months). Etiology of the airway obstruction included congenital tracheal stenosis (n=4), giant cervical and superior mediastinal lymphatic malformation with tracheobronchomalacia (n=1). Seven complications were reported (3 patients developed granulation tissue, 2 patients had re-stenosis, 1 stent migrated, 1 stent fractured). All patients survived and were in good condition with a median follow-up of 16 years (range 11 – 18 years). Three

patients weaned off ventilator and oxygen.

Conclusions:

Endoscopic stenting with metallic stent has satisfactory long term outcome in treating infants with tracheobronchial obstruction.

Keywords:

tracheal stenosis; airway obstruction; metallic stent; self-expandable stent; tracheobronchomalacia

Article

Introduction

Airway obstruction in paediatric patients remains a challenging problem among clinicians. Obstruction can be due to various extraluminal, intramural and intraluminal diseases. Congenital tracheomalacia and stenosis are the most common causes. A number of surgical treatment options have been advocated. Tracheoplasty can be done either by tracheal resection and anastomosis or by repair using costal cartilage or pericardial patch. [1,2] However, these operations may be difficult for small children with respiratory failure and have a high risk of operative mortality. Aortopexy has been shown to be effective in cases of short-segment tracheomalacia. [3-5] Endoscopic stenting has been shown to reduce the need for high risk surgical procedure and prolonged ventilator dependence in children with diffuse tracheomalacia. In difficult population, endoscopic stenting has been recommended as an alternative therapeutic option. [6-17] Currently, there is only scanty information regarding the long term outcome of this procedure. The

purpose of this study is to review our experience in treating congenital tracheobronchial obstruction with endoscopic stent placement.

Materials and methods

Medical records of all paediatric surgical patients who underwent tracheobronchial stenting in our centre were reviewed retrospectively from 1996 to 2014. Patients' demographic data, including etiology, associated anomalies and nature of obstruction were reviewed. Outcome measures include complications such as re-stenosis, granulation tissue, stent migration, fractured stent, maximal tracheal diameter achieved, weaning of ventilator and growth at interval follow-up.

Endoscopic stenting was offered to patients who had significant recurrent stenosis following repeated dilatations. As the number of patients with this condition was very small, our ENT colleagues did not have the expertise in performing tracheoplasties. As a result, we were only able to offer endoscopic stenting to patients. In addition it had the advantage of being minimally invasive. Selection of the stent type depended on the location of obstruction, the patient's age and body size as well as the availability of the stent at the time of procedure. Prior to stent placement, the type, location, severity, and length of the obstruction were assessed by bronchoscopy and computer tomography. In all of our patients, metallic balloon-expandable Palmaz stents (Johnson & Johnson Interventional Systems, Warren, NJ) were placed using a Storz rigid paediatric bronchoscope (Karl Storz, Germany) under general anaesthesia. The site for stenting was determined by simultaneous use of endoscopy and fluoroscopy. The stent, together with a angioplasty balloon catheter, were passed through the bronchoscope. Under fluoroscopic

control, the balloon was inflated by normal saline and the position of expanded stent was confirmed. (**Figures 1&2).** The tracheal mucosa was visualized by rigid bronchoscopy. The balloon was deflated and the catheter was removed.

Results

Five patients presented with respiratory distress in the neonatal or early infantile period. Patient demographics, the anatomy of tracheobronchial obstruction, number and location of the stents and complications were summarized in Table 1. Etiology of the airway obstruction included congenital tracheal stenosis (n=4), giant cervical and superior mediastinal lymphatic malformation with tracheobronchomalacia (n=1). One patient (patient 4) had concomitant multiple congenital anomalies including cerebral atrophy and severe hydrocephalus. The diameter of the obstructed segments ranged from 1 mm to 2 mm and the length from 14mm to the entire trachea.

The median age at first stenting was 13 months (range 5 - 30 months). Four patients had undergone rigid bronchoscopies and balloon dilatations to expand the airway before stent placements, ranging from 7 to 19 sessions. One patient (patient 5) with extensive tracheobronchial compression due to giant cervical and superior mediastinal lymphatic malformation was initially managed by open excision of the lesion, which subsequently recurred. A total of twelve Palmaz stents were placed in the trachea (n=10) and/or bronchi (n=2) in 5 patients. Four patients required placements of more than one stent, these were placed during repeated bronchoscopic procedures. All stents, except one fractured stent in patient 5, remained in place. There was no peri-operative mortality.

Long term outcome

A summary of long term outcome was presented in table 2. All patients needed regular bronchoscopic assessments and balloon dilatations to maintain luminal patency. A median of 25 bronchoscopies (range 21 – 55) and 14 balloon dilatations (range 9 – 40) had been performed. Gradual increase in tracheal diameter was noted. The median maximal tracheal diameter achieved was 13 mm (range 8 – 17 mm).

All patients survived and were in good condition with a median follow-up of 16 years (range 11 – 18 years) from their initial presentation. Three patients weaned off ventilator and oxygen. Two patients require home oxygen support in the form of positive airway pressure. All except one patient with profound mental retardation had satisfactory exercise tolerance for normal daily activity. Three patients had normal body weight and height, whereas two patients remained below the 3rd percentile.

Patient 1

No complications occurred throughout the eighteen years post-stenting. He remained ventilator free and enjoyed normal exercise capacity and growth.

Patient 2

In patient 2, two stents were inserted to distal trachea. Stent migration into the left lower lobe bronchus was noted six years post-stenting, which was managed conservatively. She was ventilator free and had an exercise tolerance of three flights of stairs.

Patient 3

In patient 3, re-stenosis proximal to the stent occurred 2 months after placement of stent and intensive care was required. It was managed by bronchoscopic dilatations and dexamethasone injection. On subsequent bronchoscopic reassessments, airway patency was maintained except granulation tissue was seen requiring removal by endoscopic cauterization twice. He had mild mental retardation, otherwise he enjoyed normal exercise capacity.

Patient 4

He had multiple congenital anomalies with profound mental retardation, hydrocephalus and obstructive sleep apnea. He developed a total of 12 episodes of pneumonia after bronchoscopies. This could result from his co-existing gastroesophageal reflux disease and he eventually underwent laparoscopic fundoplication and gastrostomy. Reassessment bronchoscopy at 4 months post-stenting showed granulation tissue at sub-glottic area and was removed. He was wheelchair bound and required nocturnal continuous positive airway pressure ventilation at home.

Patient 5

In patient 5, Palmaz stent was inserted at one and a half year of age. Three months later, a second stent was inserted in view of a malacic segment of the trachea above the first stent. However, it was complicated by stent fracture and was managed by balloon dilatation and adrenaline injection. Reassessment bronchoscopy showed obstructing granulation tissue over both bronchi requiring bilateral bronchial stents insertion. During her 35^{th} bronchoscopy, granulation tissue was found to cause a ball-valve effect, and there were

migration and fracture of the tracheal stent. Granulation tissue and the stent fragment were removed by suction and forceps until patency of the tracheal lumen was confirmed. Despite her difficult clinical condition and stormy hospitalizations, she had normal growth and intelligence. She required bilevel positive airway pressure ventilation at home, nonetheless, her normal daily activity was not affected.

Discussion

The first report regarding the use of tracheobronchial stents in children dated back to the late 1980s. [6,7,18] Endoscopic stents had been used in various clinical conditions including airway malacia or stenoses, either due to external compression or structurally abnormal airway walls. Its use in post-operative stenosis after lung transplantation had also been reported. [13] Such procedures were usually performed in combination with surgery for the treatment of severe bronchomalacia or for the prevention of post-tracheoplasty re-stenosis.

In 1995, Zinman [19] showed that tracheal stenting improved ventilatory mechanics in infants with tracheobronchomalacia. The use of vascular mesh metal prosthesis (Palmaz) in 16 children was first introduced by Filler et al. [9] The stents were reported to be well tolerated for up to 6 years.

Different classes of stents have been developed for airway stenosis. They can be broadly classified into plastic and metallic. For plastic stents, standard silicone stent is easily removable while Dumon stent is flexible. [20] Silicone stent has major limitations of being easily collapsible, prone to migration and interruption of mucociliary clearance. Its use in infants is not well tolerated because of frequent obstruction by secretions. The use

of Polyflex stent, a self expanding silicone device, had been reported. However, migration and mucus impaction occurred in all 12 patients with stenoses. [21]

Concerning metallic stents, the main division is balloon-expandable and self expanding types. These were originally intended for angiographic applications in adults. Palmaz stent is made of stainless steel and is balloon expandable. The risks of obstruction by mucus and migration were less than that of silicone stents. Palmaz stents had been shown in an experimental trial to provoke an inflammatory reaction and epithelialization. [22] When compared to self expanding stents, Palmaz stent had the advantage of expansion under direct observation. Balloon dilatation was performed first to a satisfactory size, followed by insertion of the Palmaz stent to maintain luminal patency. A major disadvantage of Palmaz stent was difficult and risky adjustment or removal.

Wallstent, on the other hand, is made of thin wire which allows flexibility. As a result, it is most frequently seen in relief of vascular compression by self-expanding properties. Balloon expanding is contraindicating as it may lead to vascular erosion. [11,21,23] However, self expanding stents might subject the tracheal wall to expanding forces, leading to significant airway damage.

Nitinol stent composed of a nickel-titanium alloy with ‘‘shape memory effect’’ had been studied in a few case series. In the recent series by Siegel et al, [24] six out of seven patients underwent stenting as a salvage procedure following open attempts at airway reconstruction. Four patients remained decannulated with their stent in place. Complications included stent migration (23%), re-stenosis (29%), edema (29%), and granulation (57%). The authors concluded nitinol stents were reserved only as a salvage procedure in severely complicated airways.

Recently, new stents that are more biocompatible were being investigated. [25,26] Biodegradable materials had been used experimentally for stenting of tracheobronchial stenosis since 1998. Polydioxanone is a biodegradable polymer that exhibits some shape memory and dissolves by 15 weeks. In a study by Vondrys et al, eleven stenting procedures were performed in 4 patients. Three patients needed repeated stenting after stent absorption. One patient died after withdrawal of care, the 3 survivors were in good ventilatory condition. [27] There was also a study on the use of Rapamycin-coated stents to prevent granulation formation [28]. More clinical application of these new stents is needed to justify the use.

Complications had been reported after Palmaz stents were inserted. Filler et al [9] reported granulation tissue in six out of seven children stented for malacia. Two patients required repeated endoscopic excisions and dilatations, two patients underwent placement of additional stents. The granulation tissue was non-obstructing in two patients and they were managed conservatively. Furman et al [11] reported major complication of epithelialization arising from the use of metallic stents, which complicates stent removal. Other complications including complete erosion of the tracheobronchial wall have been reported [29,30]. Antón-Pacheco et al [31] reported three out of twelve patients with tracheal Palmaz stents showed prominent granulation tissue with clinical significance. In addition, stent migration and stent fracture are not uncommon [12].

Although our series was among the few that had zero mortality, stent related mortality was reported to be 12.9% by Nicolai [13]. Santoro et al [8] reported two out of three neonates who underwent Palmaz stenting for tracheobronchomalacia died of sepsis after the procedure. Recurrent granulation tissue was potentially fatal, Maeda et al [14]

reported 5 infants with tracheal stenosis, one died from recurrent granulation tissue obstructing the trachea and intractable pneumonia after 9 months of palliation. Geller et al [16] reported 3 mortalities from tracheal haemorrhage and 1 from pulmonary complications among 9 patients who underwent Palmaz stent insertion for severe tracheomalacia. In that particular series, seven out of nine patients had co-existing cardiac disease.

Experience in using silicone stenting had been reported by various centers over the world. However, the long-term effect of balloon-expandable metallic stents in children is unknown.

Throughout the past ten years, we have applied the technique on five patients whom we thought will benefit from stenting. In general, the indication for considering long term stenting in this series is those who have significant recurrent stenosis following repeated dilatations. Other indications include extrinsic compression not correctable by surgery or focal malacia. Compared to other studies we had an acceptable complication rate. [10-12,32] The most common complication was granulation tissue formation, which occurred in three of our patients. Patient 5, the girl with a giant cervical and superior mediastinal lymphatic malformation with tracheobronchomalacia, was particularly challenging to manage. Having a total of five stents placed to maintain airway patency, she eventually developed granulation, stent migration and fracture, necessitating multiple bronchoscopic removals of granulation tissue and stent fragments. The reason why some patients remained granulation free with normal epithelialization while others developed recurrent granulation formation was yet to be determined. In patient 5, although the stent was fractured and migrated, removal would be extremely difficult due to granulation

formation. It had been reported by an experimental study by Fraga et al [22] that attempts to remove stent could be fatal, the stent was found to be incorporated into the fibrotic tracheal wall and could not be removed even after death. Okuyama et al [33] had reported one successful removal of stent requiring cardiopulmonary bypass and reconstruction using slide tracheoplasty. They concluded stent removal by rigid bronchoscope should be regarded as a dangerous and possibly a fatal procedure.

Despite the relatively low complication rate, our patients required a median of 25 bronchoscopies post stenting under general anesthesia. With concerns about the effects of general anaesthesia on the developing brain, we believe stenting should be reserved for complex situations.

Our study was one of the few published series with no reported stent related mortalities. [7,13,15] In view of high risk nature of this procedure, all our bronchoscopies were performed by surgeons, paediatric anaesthetists, operating theatre nurses and radiographers that had previous experiences. Paediatric intensivists were consulted for provision of post-operative support. We believed a multidisciplinary team approach was the cornerstone of our satisfactory outcome.

Our study also had the longest follow duration (18 years) published to date. All children survived and their normal activities were not affected, except one patient who had profound mental retardation and was wheelchair bound.

Given the rarity of this condition, our experience in this procedure is still limited. However, the fact that all patients still survive with reasonably good quality of life has given us much encouragement. Complications can be managed accordingly. The patients only need to undergo regular bronchoscopic assessments with occasional dilatations. We

believe, endoscopic stenting has proved itself being a treatment option to manage tracheobronchial obstruction, a potentially fatal condition in infants, with satisfactory long term outcome.

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Conflict of Interest

The authors declare that they have no conflict of interest.

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Figure legends

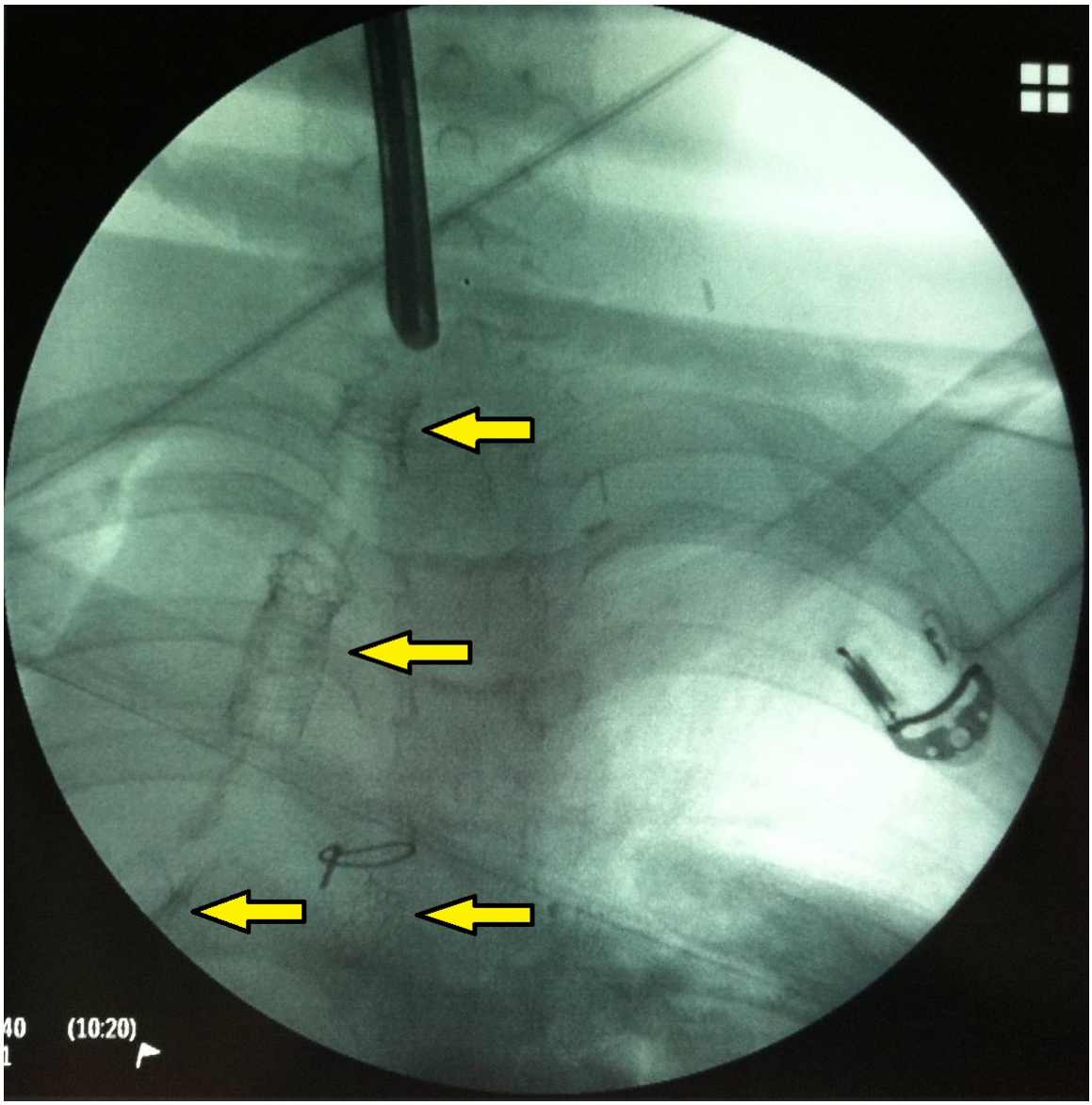

Fig. 1 - Fluoroscopy film showing multiple stents placed in trachea and both bronchi, pointed by arrows.

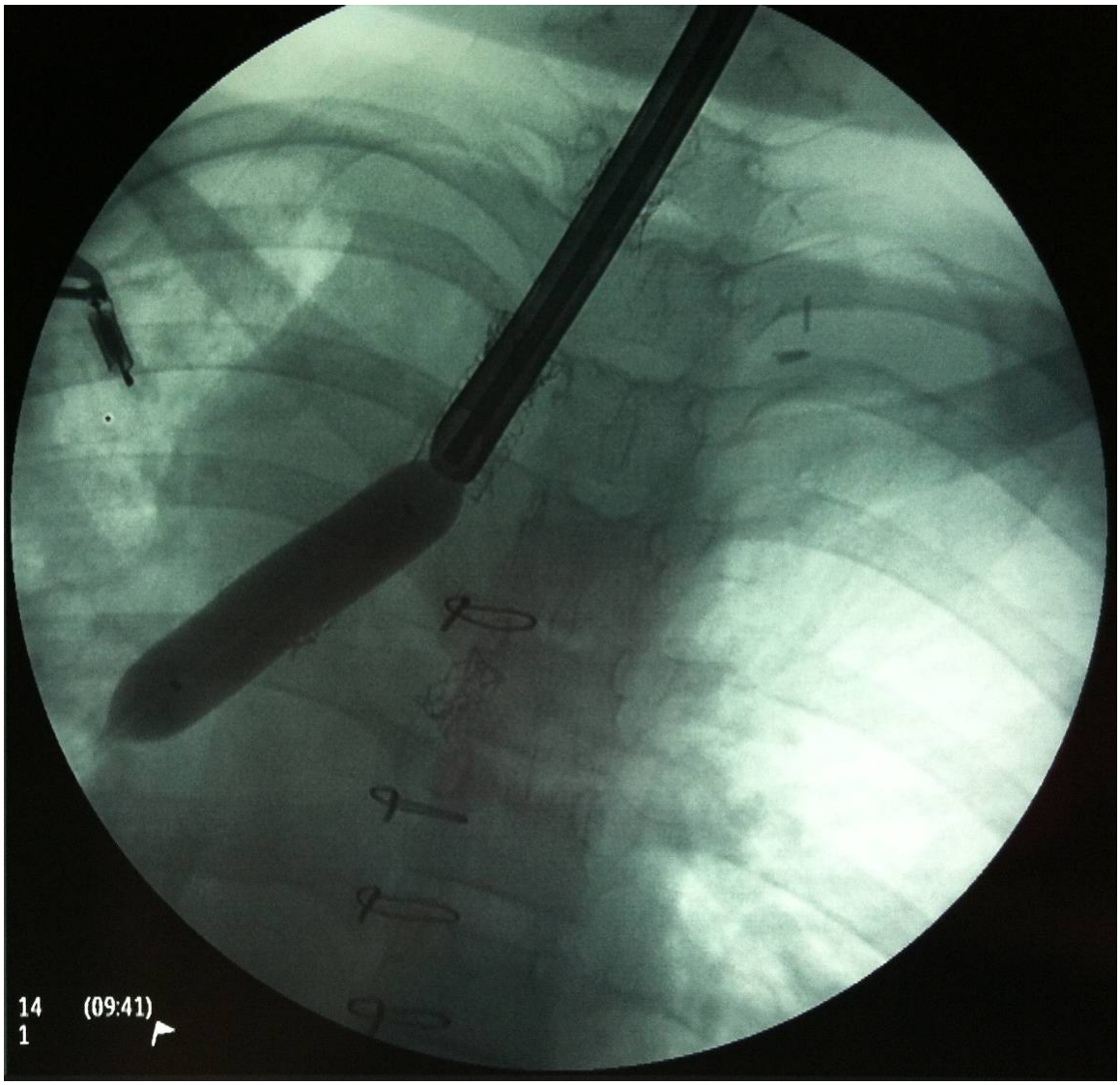

Fig. 2 - Fluoroscopy film taken during a balloon dilatation session

Table 1. Patient demographics, number and location of stent(s) and complications

Patient	Present Age (years)	Age at first stenting (months)	Primary disease	Anatomy of the stenotic/malacic segment	Number and location of stent(s)	Complications
1	18	5	Congenital tracheal stenosis	Entire lower half of trachea extending down to carina	1 in distal trachea	Nil
2	16	14	Congenital tracheal stenosis	Right bronchus was branching directly from the trachea. Stenotic trachea below that branching down to the carina and both mainstem bronchi	2 in distal trachea	Re-stenosis
3	17	13	Congenital tracheal stenosis	14mm segment of tracheal stenosis 20mm above the carina with complete cartilage rings	2 in distal trachea	Re-stenosis Granulation tissue
4	11	11	Congenital tracheal stenosis	20mm segment of tracheal stenosis 20mm above the carina with complete cartilage rings	2 in distal trachea	Granulation tissue
5	14	30	Giant cervical and superior mediastinal lymphatic malformation Tracheobronchomalacia	Tracheobronchomalacia due to compression from lymphatic malformation	1 in distal trachea 2 in proximal trachea 1 in right bronchus 1 in left bronchus	Granulation tissue Stent migration Stent fracture

Table 2. Long term outcome

Patient number	Total number of bronchoscopies	Number of subsequent dilatations	Maximal tracheal diameter achieved (mm)	Oxygen requirement	Exercise tolerance	Growth	Follow up duration (years)
1	21	9	14	Nil	Normal	Normal	18
2	29	16	8	Nil	3 flights of stairs	Below 3rd percentile	16
3	21	14	13	Nil	Normal	Below 3rd percentile	17
4	25	11	12	Nocturnal continuous positive airway pressure ventilation	Wheelchair bound due to profound mental retardation	10th percentile	11
5	55	40	17	Home bilevel positive airway pressure ventilation	Normal	Normal	14