Current management and outcome of tracheobronchial malacia and stenosis presenting to the paediatric intensive care unit

Abstract

Objective: To identify factors associated with mortality and prolonged ventilatory requirements in patients admitted to our paediatric intensive care unit (PICU) with tracheobronchial malacia and stenosis diagnosed by dynamic contrast bronchograms.

Design: Retrospective review.

Setting: Tertiary paediatric intensive care unit.

Patients: Forty-eight cases admitted to our PICU over a 5-year period in whom a diagnosis of tracheobronchial malacia or stenosis was made by dynamic contrast bronchography (1994–1999).

Interventions: Conservative management, tracheostomy and long-term ventilation, surgical correction, internal or external airway stenting.

Measurements and results: Recording of clinical details, length of invasive ventilation and appearance at contrast bronchography. Five groups of patients were defined: isolated primary airway pathology (n = 7), ex-premature infants (n = 11), vascular rings (n = 9), complex cardiac and/or syndromic pathology (n = 17) and tracheoesophageal fistulae (n = 4). The overall mortality was 29%. Median length of invasive ventilation in survivors was 38 days and in patients who died 45. Mortality was highest in the patients with complex cardiac and/or syndromic pathology (p = 0.039 Cox regression analysis) but was not related to any other factor. Patients with stenosis required a significantly longer period of ventilatory support (median length of ventilation 59 days) than patients with malacia (39 days).

Conclusions: Length of ventilation and bronchographic diagnosis did not predict survival. The only factor found to contribute significantly to mortality was the presence of complex cardiac and/or syndromic pathology. However, patients with stenosis required longer ventilatory support than patients with malacia.

Key words Tracheal stenosis · Bronchomalacia · Tracheomalacia · Tracheobronchomalacia · Bronchogram · Ventilation · Mortality
Introduction

Tracheobronchomalacia is a condition of dynamic airway collapse during expiration. In tracheal stenosis, fixed airway narrowing causes airway obstruction in inspiration and in expiration. These conditions, first described in 1952 [1], may be primary, when cartilaginous rings are congenitally malformed, or secondary, due to degeneration of previously normal cartilage. In neonatal chronic lung disease, development of the tracheobronchomalacia tree may be abnormal and the cartilage may be weak [2]. Extrinsic airway compression by a vascular ring or by enlarged great vessels in the presence of a large shunt can result in permanent airway narrowing [3]. Tracheo-oesophageal fistulae are often associated with malacia of a portion of the trachea at the point of insertion of the fistula. Airway abnormalities may develop in recurrent aspiration syndromes and in acquired cartilaginous disorders such as relapsing polychondritis. The severity of disease depends on the length, location and degree of narrowing in the affected airway segments.

Although primary isolated tracheobronchomalacia rarely requires ventilatory support and is often self-limiting, infants with tracheobronchomalacia or stenosis with abnormal bronchograms who require ventilatory support have been reported to have a high morbidity and mortality, often related to airway obstruction and respiratory failure [4]. This has not been our experience. We therefore reviewed children admitted to our intensive care unit with tracheobronchomalacia and stenosis, diagnosed at contrast bronchography, to determine which factors affected outcome.

Materials and methods

Following local ethics committee approval, the notes of 48 patients, who had been admitted to our intensive care unit between 1994–1999 and in whom a diagnosis of tracheobronchomalacia or stenosis had been made at contrast bronchography, were reviewed. Information collected included underlying diagnosis, length of invasive ventilation around the time of diagnosis, surgical interventions, mortality, time between diagnosis and the present day for survivors and time between diagnosis and death for those who died. Underlying diagnostic categories were defined as isolated primary airway pathology or airway pathology associated with neonatal chronic lung disease, complex cardiac and/or syndromic pathology, vascular rings or tracheo-oesophageal fistulae.

Radiographic data were also reviewed. Bronchography was performed as previously described [5]. The patients were lightly sedated, intubated and were breathing spontaneously. The endotracheal tube was maintained in a high position with the tip in the subglottic region. Opacity was obtained by bolus injections of 0.5–2 ml of contrast medium (Omnipaque, Nycomed, Birmingham, UK), injected into the airway via the endotracheal tube through a 3.7 Fr angiography catheter with a single end-hole. Contrast dispersal was obtained by hand ventilation. Up to five affected sites were identified (trachea, left main bronchus, right main bronchus, bronchus intermedius and peripheral bronchi). A pressure monitor was connected to the airway circuit via a Y connector and when collapse was found positive end-expiratory pressure (PEEP) was applied and opening pressures noted. Airways were defined as stenosed when the narrowing was fixed and unaffected by PEEP up to 25 cmH₂O and malacic when the narrowing was dynamic and improved with positive airway pressure. Infants with tracheal stenosis with distal malacia were classified as tracheal stenosis for the purposes of this analysis since the numbers of patients with both forms of pathology was small (n = 10). Our bronchograms are not stored as cine-films or videos but as static images, so no attempt was made at retrospective blinded severity scoring as this was felt to be unlikely to yield any useful information.

Data were analysed using SPSS 8.0 for Windows (SPSS, UK). Analysis of survival and length of invasive ventilation was performed using Cox regression analysis. The Kruskal-Wallis test was used to determine whether differences in the length of follow-up in survivors were significant.

Results

Of the 48 patients, 19 were female. The median age at diagnosis was 137 days (interquartile range 62–213 days). The number of patients in each group was as follows: isolated primary airway pathology (7), ex-premature infants (11), vascular rings (9), complex cardiac and/or syndromic pathology (17) and tracheo-oesophageal fistulae (4). The overall mortality was 14/48 (29 %). Median length of follow-up in survivors was 439 days and was not significantly different between any of the groups (p = 0.635, Kruskal-Wallis test). Outcome data is presented related to diagnosis and to operative procedure (Tables 1 and 2).

Isolated primary airway pathology

Seven patients with primary airway pathology were identified. The median age at diagnosis was 66 days. One term infant was referred for surgical management of congenital subglottic stenosis, tracheobronchomalacia being diagnosed when weaning difficulties were encountered following surgery. Invasive ventilation was defined in this group as the time from first intubation for airway obstruction to death or weaning to either extubation or continuous positive airway pressure (CPAP) or biphasic positive airway pressure (BiPAP) via a tracheostomy. The median length of invasive ventilation of survivors was 60 days. Five patients had isolated tracheobronchomalacia and two had tracheal stenosis with airway malacia distal to the stenotic segment. Two patients with mild tracheobronchomalacia were extubated, one after a cricoid split procedure for congenital subglottic stenosis, the other requiring no intervention. The latter patient was excluded from the statistical analysis as he had severe combined immune deficiency and died from graft versus host disease following bone
marrow transplantation. One patient with severe focal tracheomalacia had a resection and end-to-end anastomosis and was then extubated to room air. The remaining four patients had a variety of surgical procedures. Three of these four patients survived but all continue to require CPAP or BiPAP via a tracheostomy, either at home or in hospital. One patient with multiple areas of malacia affecting all major bronchi died post-operatively due to airway obstruction.

Neonatal chronic lung disease

Eleven ex-premature infants (median gestation 28.5 weeks) with bronchopulmonary dysplasia and one term baby with chronic lung disease due to meconium aspiration were identified. The median age at diagnosis was 199 days. All had been referred for investigation of their continued ventilatory requirements in the absence of severe parenchymal lung disease and all had tracheobronchomalacia. Two had been referred for surgical management of their subglottic stenoses, tracheobronchomalacia being diagnosed when weaning difficulties were encountered following cricoid split procedures. Length of invasive ventilation was defined in this group as the time from first failed extubation (following the period of ventilatory support for neonatal lung disease) to death, weaning to extubation, or CPAP or BiPAP via a tracheostomy. The median length of invasive ventilation in survivors was 23 days.

Table 1  Outcome by primary surgical procedure

| Procedure                              | No. | Diagnosis            | Further procedures                  | Invasive ventilation (median, days, survivors only) | Dead | Alive | Still ventilated |
|----------------------------------------|-----|----------------------|-------------------------------------|----------------------------------------------------|------|-------|-----------------|
| Aortoectomy                            | 2   | Tracheomalacia       | None                                | 38                                                 | 0    | 2     | 0               |
| Division of vascular ring              | 1   | Tracheal stenosis    | None                                | 14                                                 | 0    | 1     | 0               |
| Slide tracheoplasty                    | 4   | Tracheal stenosis    | Tracheostomy (4), Palmez stent (1)   | 61                                                 | 1    | 3     | 1               |
| Patch tracheoplasty                    | 3   | Tracheal stenosis    | Hgl stent and tracheostomy (1), Palmez stent (2) | 104                                               | 1    | 2     | 0               |
| Resection and end-to-end anastomosis   | 2   | Severe focal tracheomalacia | None (1), Palmez stent (1) | 17                                                 | 0    | 2     | 0               |
| Hgl stent                              | 3   | Focal tracheomalacia (2), tracheal stenosis (1) | Tracheostomy (1), Palmez stent (1) | 12                                                 | 2    | 1     | 1               |
| Tracheostomy and long-term ventilation | 19  | Tracheo bronchomalacia | None (18), Palmez stent (1) | 42                                                 | 8    | 11    | 7               |
| No surgical intervention              | 13  | Tracheo bronchomalacia | None                             | 29                                                 | 2    | 11    | 0               |
| Died on table during surgery           | 1   | Tracheal stenosis    | None                                | 34                                                 | 1    | 0     | 0               |

Two infants were extubated to room air without any intervention. The remaining nine patients all required tracheostomy for long-term ventilation with either CPAP or BiPAP. One patient died of viral encephalitis and was excluded from the statistical analysis. One patient, who had four major bronchi affected by disease, died following an episode of airway obstruction. Of the seven survivors, three have been weaned from ventilatory support and two have been decannulated. All survivors have been discharged from hospital.

Vascular rings

Nine patients with vascular rings were identified, two with a double aortic arch and seven with pulmonary artery slings. One patient had isolated tracheobronchomalacia, two had isolated tracheal stenosis and six had tracheal stenosis with distal bronchomalacia. All of these patients presented in infancy with airway obstruction, with a median age at diagnosis of 115 days. Length of invasive ventilation was defined in this group as the time from first intubation for airway obstruction to death, weaning to extubation or CPAP or BiPAP via a tracheostomy. The median length of invasive ventilation of survivors was 48 days.

All patients required division of vascular rings. Additional primary procedures included slide tracheoplasty (3) and pericardial patch tracheoplasty (2), aortoectomy (1) and placement of a Hgl external stent (1). One pa-
### Table 2 Outcome by diagnosis

| Diagnosis                  | Airway pathology | No. | Initial procedure                                                                 | Invasive ventilation (median, days, survivors only) | Dead | Alive | Still ventilated |
|---------------------------|------------------|-----|----------------------------------------------------------------------------------|----------------------------------------------------|------|-------|-----------------|
| Primary airway            | Tracheobroncho-  | 5   | Tracheostomy (2), no intervention (2), resection and end-to-end anastomosis (1)  | 50                                                 | 1    | 4     | 1               |
|                           | malacia          |     |                                                                                  |                                                    |      |       |                 |
|                           | Tracheal stenosis | 2   | Slide tracheoplasty (1), pericardial patch tracheoplasty (with Hagl stent) (1)    | 96                                                 | 1    | 1     | 1               |
| Neonatal chronic lung     | Tracheobroncho-  | 11  | Tracheostomy (9), no intervention (2)                                            | 23                                                 | 2    | 9     | 4               |
| disease                   | malacia          |     |                                                                                  |                                                    |      |       |                 |
|                           | Tracheal stenosis | 0   |                                                                                  |                                                    |      |       |                 |
| Vascular rings            | Tracheobroncho-  | 1   | Division of ring                                                                  | 14                                                 | 0    | 1     | 0               |
|                           | malacia          |     |                                                                                  |                                                    |      |       |                 |
|                           | Tracheal stenosis | 8   | Aortoexcision (1), pericardial patch tracheoplasty (2), slide tracheoplasty (3)  | 55                                                 | 2    | 6     | 1               |
| Complex cardiac or        | Tracheobroncho-  | 17  | Tracheostomy (8), Hagl stent (2), resection, end-to-end anastomosis (1) and no intervention (6) | 44                                                 | 8    | 9     | 1               |
| syndromic                 | malacia          |     |                                                                                  |                                                    |      |       |                 |
|                           | Tracheal stenosis | 0   |                                                                                  |                                                    |      |       |                 |
| Tracheo-oesophageal fistul| Tracheobroncho-  | 4   | No intervention (3), aortoexcision (1)                                           | 34                                                 | 0    | 4     | 0               |
|                           | malacia          |     |                                                                                  |                                                    |      |       |                 |
|                           | Tracheal stenosis | 0   |                                                                                  |                                                    |      |       |                 |

A patient died of a pulmonary hypertensive crisis during surgery. Three were extubated shortly after surgery. The two patients who had pericardial patch tracheoplasties were extubated and discharged from hospital after placement of Palmaz stents. The three patients who had slide tracheoplasties all required tracheostomy for long-term ventilation. One of these died from airway obstruction post-operatively and two were discharged home on chronic ventilation but are now off ventilatory support.

**Complex cardiac and/or syndromic pathology**

Seventeen infants with complex pathology, 15 cardiac (7 of whom had multiple anomalies) and 2 syndromic, were identified. These infants had a range of abnormalities and most had been admitted to PICU following cardiac surgical procedures. All had tracheobronchomalacia. Tracheobronchomalacia had been suspected in most of these infants when post-operative weaning from ventilation was difficult and in some infants with shunts was thought to be due to enlarged great vessels compressing the airway. The median age at diagnosis was 76 days. Length of invasive ventilation was defined as the time from first intubation for airway obstruction or cardiac surgery to death or weaning to either extubation or CPAP or BiPAP via a tracheostomy. The median length of ventilation in survivors was 44 days. Five infants, who are still alive, were extubated and required no intervention for airway management. Of the remaining 12, airway included tracheostomy for long-term ventilation (8), Hagl external stent (2), resection and end-to-end anastomosis (1) and no intervention (1). Five patients who had tracheostomies died, two of viral pneumonia, one of hypertrophic obstructive cardiomyopathy and two of airway obstruction. Both patients with Hagl stents died, one of acute respiratory distress syndrome (ARDS) following cardiac surgery and the other, who had multiple anomalies, of progressive respiratory failure when intensive care was withdrawn. One other patient with multiple anomalies died of progressive respiratory failure when...
intensive care was withdrawn. The patient who had a resection and end-to-end anastomosis had further problems with airway obstruction, was extubated following insertion of a Palmaz stent and is now at home. Of the three survivors with tracheostomies, one has been decannulated and two remain on CPAP but are at home.

Tracheo-oesophageal fistula

Four patients with tracheo-oesophageal fistulae were identified. All had tracheomalacia, median age at diagnosis being 128 days. Length of invasive ventilation in this group was defined as the interval from first intubation for airway obstruction to weaning to extubation. The median length of ventilation was 34 days. Two patients were extubated without any airway intervention, one was extubated following repair of his tracheo-oesophageal fistula and one required aortoectomy before extubation was possible. All patients survived and none required long-term ventilation. A large number of patients with tracheo-oesophageal fistulae who have been treated with aortoectomy in our institution have not been included in this analysis as their airway pathology did not require bronchography for diagnosis.

Table 3 Causes of death (CPAP continuous positive airway pressure, ARDS acute respiratory distress syndrome, RSV respiratory syncytial virus)

| Sex | Diagnostic category | Airway pathology | Interventions | Cause of death |
|-----|---------------------|------------------|---------------|----------------|
| M   | Neonatal           | Tracheobronchomalacia | Tracheostomy, CPAP | Airway obstruction |
| M   | Neonatal           | Tracheobronchomalacia | Tracheostomy, CPAP | Viral encephalitis |
| M   | Vascular ring      | Tracheal stenosis  | Died on table during surgery | Airway obstruction |
| F   | Vascular ring      | Tracheal stenosis  | Division of ring, slide tracheoplasty, tracheostomy CPAP | Airway obstruction |
| F   | Primary airway     | Tracheobronchomalacia | Extubated to room air | Graft versus host disease following bone marrow transplantation |
| F   | Primary airway     | Tracheal stenosis  | External stent, redo stent, tracheostomy CPAP | Airway obstruction |
| F   | Complex cardiac    | Tracheobronchomalacia | Hagle stent, Palmaz stent | Airway obstruction, limitation of care due to other lesions |
| M   | Complex cardiac    | Tracheobronchomalacia | Hagle stent | ARDS post cardiopulmonary bypass |
| M   | Complex cardiac    | Tracheobronchomalacia | No intervention | Airway obstruction, limitation of care due to other lesions |
| M   | Complex cardiac    | Tracheobronchomalacia | Tracheostomy CPAP | RSV pneumonia |
| F   | Complex cardiac    | Tracheobronchomalacia | Tracheostomy CPAP | Hypertrophic obstructive cardiomyopathy |
| F   | Complex cardiac    | Tracheobronchomalacia | Tracheostomy CPAP | Influenza A pneumonia |
| F   | Complex cardiac    | Tracheobronchomalacia | Tracheostomy CPAP | Airway obstruction |
| M   | Complex cardiac    | Tracheobronchomalacia | Tracheostomy CPAP | Airway obstruction |

Statistical analysis of factors associated with death

Fourteen (29%) infants died (Table 3). Twelve died of cardiorespiratory causes, seven of airway obstruction, one during tracheal surgery, two of viral pneumonia, one of ARDS following cardiopulmonary bypass and one of hypertrophic obstructive cardiomyopathy (HOCM). Two infants died of non-cardiorespiratory causes, one after bone marrow transplantation and one of viral encephalitis, and were excluded from the statistical analysis. Cox regression analysis was used to identify factors associated with death in the remaining 46 patients. Patients were defined by diagnostic category (cardiac and/or syndromic and others), length of invasive ventilation, bronchographic diagnosis (i.e. tracheobronchomalacia or tracheal stenosis) and number of major airways affected. Patients with complex cardiac and/or syndromic pathology were at significantly higher risk of death than those in any other group (p = 0.039) (Fig. 1) but other factors were not found to be significant.
Statistical analysis of factors associated with prolonged invasive ventilation

Cox regression analysis was used to identify factors associated with prolonged invasive ventilation. Four patients who died during the initial period of invasive ventilation were excluded from this analysis as length of ventilation was limited by death in these infants. Patients were defined by diagnostic category, number of major airways affected and bronchographic diagnosis (stenosis or malacia). Length of invasive ventilation was not significantly influenced by the number of major airways affected or by the diagnostic category. However, patients with tracheal stenosis were likely to require longer invasive ventilation (median 59 days) than patients with tracheobronchial malacia (median 39 days) and this difference was statistically significant ($p = 0.037$) (Fig. 2).

Discussion

Mild tracheobronchial malacia or stenosis classically presents in early childhood with wheeze, cough, stridor and recurrent lower respiratory tract infections. It usually resolves spontaneously as cartilaginous development of the airway occurs [6, 7]. More severe lesions present in early infancy with respiratory failure, episodes of profound cyanosis, dying spells, reflex apnoea and inability to wean from mechanical ventilation following preterm delivery or surgical procedures and often present to intensive care. Although mortality and morbidity in the group requiring intensive care are widely assumed to be poor, there has only been one large series published to date. In that series, from Melbourne, 62 patients with tracheobronchomalacia or stenosis who presented between 1986 and 1995 were reviewed and those ventilated for longer than 3 weeks with abnormal bronchograms had a mortality of 100% [4]. However, since then there have been many advances in the management of the child with critical airway lesions, including the use of external [8] and internal [9, 10] airway stents and advances in tracheal surgery [11, 12, 13]. Our experience over the last 5 years suggests

![Fig. 1 Survival curves of different diagnostic groups](image1)

![Fig. 2 Length of invasive ventilation by bronchographic diagnosis (survivors only)](image2)
that the majority of infants with these lesions can now survive, even following prolonged ventilation.

Diagnosis

The most important aspect of diagnosis is a high index of suspicion in the groups of infants outlined above. The association of airway abnormalities with tracheoesophageal fistulae and vascular rings is well recognised. However, although there have been reports of tracheobronchial malacia and stenosis occurring in association with both neonatal chronic lung disease [2] and congenital cardiac disease (due to either enlarged great vessels or conduits) [14, 15, 16], the associations are not widely recognised and it is likely that many patients with clinically important airway pathology remain undiagnosed.

A diagnosis of tracheobronchomalacia requires assessment of the trachea and bronchi throughout the respiratory cycle with demonstration of dynamic airway collapse in expiration. Contrast bronchography is currently the investigation of choice for ventilated infants in our institution [5]. It enables assessment of the trachea and the more distal bronchi, unlike bronchoscopy, and since it is performed in real time with the infant spontaneously breathing, the airways are assessed throughout the respiratory cycle. It is simple to perform in infants who are already intubated and is also useful in diagnosing fixed airway stenosis. Methods have been suggested for quantitative severity scoring of bronchographic data [17] but, in our experience, clinical assessment of the child is still paramount in determining which infants should be referred for long-term ventilation and/or surgical management. Bronchoscopy is still necessary to identify complete tracheal rings and examination of the airway under direct vision by the surgeon at operation has been more useful than imaging in deciding which surgical approach is most suitable for several patients.

Distal malacic lesions often present after proximal lesions have been treated. This is particularly the case in infants with neonatal chronic lung disease who may be referred for treatment of subglottic stenosis and in infants with vascular rings following surgery. In these infants, areas of distal malacia should, therefore, be searched for prior to any intervention in order to guide management.

Management

Infants requiring intensive care may need a variety of management approaches. Treatment should be individualised. Infants with very mild disease may require no intervention. Moderate airway malacia often requires tracheostomy and long-term home ventilation [18], which improves pulmonary mechanics and prevents dynamic collapse [19, 20] until the airway grows sufficiently to allow ventilatory support to be withdrawn. However, some infants with severe localised proximal disease benefit from surgical management. Our current strategy is patch tracheoplasty and repair of associated lesions in long segment tracheal stenosis and resection with end-to-end repair in short segment tracheal stenosis. Slide tracheoplasties are now rarely performed. Patients developing restenosis or malacia are treated with internal wire mesh Palmaz stents or external Gore-Tex Hagl stents and, if necessary, tracheostomy and long-term ventilation. Mild tracheomalacia, particularly if associated with a tracheo-oesophageal fistula, is treated with aortopecty [21]. Severe short segment tracheomalacia is treated with resection and end-to-end anastomosis and long segment severe tracheomalacia is treated with external Gore-Tex Hagl stents. Patients developing restenosis or malacia are treated with balloon dilatation, internal wire mesh Palmaz stents or tracheostomy and long-term ventilation. The management of these infants is evolving and the use of stents rather than surgery as a primary intervention in malacic airways is likely to increase if their design improves.

Outcome

In our patient population, survival was significantly affected by one factor among those studied – the existence of associated complex cardiac and/or syndromic pathology. Although most patients died of respiratory causes (Table 3), included amongst the respiratory deaths in the group with complex cardiac and/or syndromic pathology are two deaths following withdrawal or limitation of care and three due to parenchymal lung disease rather than airway obstruction. Clearly the existence of significant non-airway pathology in these patients may have contributed to their “respiratory” deaths. Length of invasive ventilation and bronchographic diagnosis had no impact on outcome. Our data are at odds with the series from Melbourne in which there was a much higher mortality. There are two possible reasons for this. Firstly, our patients might be significantly different from those reported in the previous study. Although there was a similar increased male to female ratio in both our series (29/48 patients male) and in the Melbourne series (38/62 patients male), more patients in the Melbourne study had congenital cardiac disease (41/62 patients) compared to our study (17/48 patients). Secondly, the patients reported from Melbourne are from an earlier epoch and thus may not have benefited from recent advances in the management of critical airway obstruction.

Length of invasive ventilation in survivors was significantly longer if tracheal stenosis was present. This re-
reflects the tendency for these patients to be managed with tracheal surgery, the need for prolonged ventilation following operation and the tendency for restenosis requiring further interventions. Length of ventilation was not affected by number of affected major bronchi or by underlying diagnosis.

We recognise the need to study respiratory and neurodevelopmental outcome and quality of life in our long-term survivors and their families. This will be the subject of future research.

In conclusion, it is possible for infants with tracheobronchial malacia or stenosis to survive episodes of prolonged ventilation in the presence of major bronchographic abnormalities. Mortality is more dependent on pathology outside the airway than it was in the past. The improved outcome in recent years probably reflects advances in intensive care and surgical management, including new surgical procedures and the use of internal and external airway stents. The problems of infants with critical airway lesions are often extremely complex and are best managed by a multidisciplinary team.

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