Respiratory failure due to tracheobronchomalacia

Ph Collard, L Freitag, M S Reynaert, D O Rodenstein, Ch Francis

Abstract
A case is described of tracheobronchomalacia progressing to extensive tracheomalacia, complicated by episodic choking, recurrent pulmonary infections, and irreversible hypercapnic respiratory failure. A Y-shaped tracheobronchial stent was placed endoscopically to splint the trachea open, with excellent clinical and physiological improvement. New stent designs may provide long term palliation in selected cases of diffuse tracheal collapse or stenosis, and offer an alternative to surgical repair.

(Thorax 1996;51:224-226)

Keywords: tracheobronchomalacia, tracheobronchial stent, respiratory failure.

Tracheomalacia is a condition caused by weakness of the tracheal wall due to softening of the supporting cartilages. Initially it causes dynamic collapse during forced expiration and cough, which can be prevented by continuous positive airway pressure (CPAP). In the most severe cases the trachea becomes distorted and permanently narrowed.

Case report
A 52 year old man was diagnosed as having tracheobronchomalgy in 1982. In 1983 he received prolonged assisted ventilation for acute respiratory failure from bilateral bronchopneumonia. Fibreoptic bronchoscopy showed expiratory collapse of an enlarged trachea during spontaneous breathing, but the patency was maintained with a CPAP of 10 cm H₂O. Between 1984 and 1986 he had three further episodes of acute respiratory failure necessitating mechanical ventilation. Between 1987 and 1990 the patient practised postural drainage twice daily and his condition remained stable.

In 1991 he developed difficulty in eliminating secretions despite postural drainage and he
Tracheobronchial stenting

Flow-volume loop before (hatched line) and after (solid line) tracheal stenting.

required readmission for infections and distress; a further fibreoptic bronchoscopic examination revealed the trachea to be large in the lateral axis and considerably distorted and narrowed in the anteroposterior axis, with permanent collapse of its entire intrathoracic portion. The length of the stenosis was 13 cm with little variation during forced inspiratory-expiratory manoeuvres. In May 1991 severe obstructive sleep apnoea developed (apnoea/hypopnoea index of 74) and he needed a CPAP apparatus at 9–10 cm H₂O to relieve sleep related obstruction. His daytime condition did not improve and he required courses of intravenous antibiotic treatment for pulmonary infections. Bronchial drainage was somewhat facilitated by CPAP application during physiotherapy. A trial with oral steroids reduced the sputum quantity but pulmonary function tests were not improved.

In April 1992 he deteriorated with ongoing bronchial suppuration and severe chronic hypercapnia. He was mechanically ventilated several times; nasal access bivalve positive airway pressure (BiPAP) was ineffective in improving his arterial blood gases. In July 1992 a 14 cm long Y-shaped stent was inserted into the trachea through direct laryngoscopy under general anaesthesia (Dynamic Stent, Rüscher, Kernen, Germany). The stent was made of horseshoe-shaped steel clasps incorporated in a silicone body with a flexible posterior membrane. It was put into position with forceps, which were used to grasp the bronchial limbs from inside, and a laryngoscope. The inverted Y rested on the carina, its limbs extended for 2 cm into each main bronchus, and it was firmly maintained between the anterior and posterior walls of the trachea.

Breathing and sputum clearance subsequently became easy without postural drainage. The patient is fully rehabilitated into his social and professional life, arterial blood gas tensions are normal, and the pulmonary function tests now show no obstructive component and an increased vital capacity (figure). Polysomnography has confirmed the persistence of obstructive sleep apnoea and he still requires CPAP at night. He is stable 40 months after stenting (table).

**Discussion**

In tracheobronchomalacia excessive dynamic compression of the major conducting airways and loss of laminar airflow cause increased airways resistance and effort of breathing, prolonged expiration, and distal air trapping. Due to an inefficient cough mechanism, retention of secretions with recurrent pneumonia, bronchiecstasis and lung scarring occur.

 Nasal CPAP prevents dynamic collapse in patients with tracheomalacia. Intermittent CPAP during the day also results in clinical improvement with easier sputum production, reduction of medical care, and better exercise tolerance. A pressure of 10 cm H₂O provides optimal splinting of the airways in most patients.³

The management of diffuse tracheal stenosis is a difficult problem. Complications and failure rates of surgical techniques tend to be unacceptable high. The use of endoscopically placed stents provides a convenient and much less invasive alternative. Expanding self-stents have been used in adults with tracheobronchomalacia and chronic respiratory insufficiency.⁶ However, these metal stents can cause granuloma formation, severe haemoptysis, or tracheobronchial rupture.⁵ Dumon’s dedicated tracheobronchial stents, made of moulded silicone, have been used widely for obstruction due to tumours, but are not well suited for diffuse tracheobronchomalacia.⁷

The Y stent appears to be particularly good for diffuse tracheal collapse.⁴ The method for

### Arterial blood gas tensions at noon air and results of pulmonary function tests

<table>
<thead>
<tr>
<th></th>
<th>Before stenting</th>
<th>1 month after stenting</th>
<th>1 year after stenting</th>
<th>2 years after stenting</th>
</tr>
</thead>
<tbody>
<tr>
<td>( \text{PaCO}_2 ) (kPa)</td>
<td>10.0–15.0</td>
<td>6.0</td>
<td>5.1</td>
<td>5.5</td>
</tr>
<tr>
<td>( \text{PaO}_2 ) (kPa)</td>
<td>4.0–6.7</td>
<td>9.1</td>
<td>11.3</td>
<td>9.1</td>
</tr>
<tr>
<td>PEF (l/s)</td>
<td>1.66 (20)</td>
<td>5.43 (67)</td>
<td>6.59 (81)</td>
<td>7.16 (88)</td>
</tr>
<tr>
<td>FEV₁ (l)</td>
<td>1.02 (24)</td>
<td>2.8 (66)</td>
<td>3.12 (73)</td>
<td>3.01 (71)</td>
</tr>
<tr>
<td>VC (l)</td>
<td>3.50 (50)</td>
<td>3.73 (62)</td>
<td>4.27 (70)</td>
<td>4.38 (72)</td>
</tr>
<tr>
<td>Vmax50 (l/s)</td>
<td>0.75 (18)</td>
<td>3.02 (71)</td>
<td>2.47 (58)</td>
<td>2.55 (60)</td>
</tr>
<tr>
<td>Vmax60 (l/s)</td>
<td>1.55 (20)</td>
<td>3.62 (47)</td>
<td>3.91 (53)</td>
<td>4.47 (58)</td>
</tr>
</tbody>
</table>

Values in parentheses are percentages of predicted.

\( \text{PaCO}_2, \text{PaO}_2 \) = arterial carbon dioxide and oxygen tensions; \( \text{PEF} \) = peak expiratory flow; \( \text{FEV₁} \) = forced expiratory volume in one second; \( \text{VC} \) = vital capacity; \( \text{Vmax50} \) = maximal expiratory flow at 50% of FVC; \( \text{Vmax60} \) = maximal inspiratory flow at 50% of FVC.

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Commentary: yet more to see down the bronchial tree?

Martin R Hetzel

These three case reports illustrate the range of rare conditions that may be encountered from time to time at diagnostic bronchoscopy. Many may at least superficially resemble bronchial carcinomas, but lead to surprises when full laboratory results become available from bronchoscopic biopsy samples. The case described by Collard et al illustrates the usefulness of direct inspection at bronchoscopic examination in evaluating the dynamics of the airway wall.

In the case of pulmonary actinomycosis described by Hsieh et al bronchoscopic examination gave the diagnosis by demonstrating a cavitation lesion which could be entered with the bronchoscope and provided positive microbiology samples. Interestingly, however, the authors were cautious in making a diagnosis of actinomycosis alone and were concerned at the possibility of a fungal infection – presumably because of the radiographic characteristics of the meniscus sign and the previous diagnosis of old tuberculosis.

Pulmonary actinomycosis is very rare and many cases are only diagnosed retrospectively from resected specimens (sadly the author’s personal experience!) unless stronger clues such as sinus formation onto the chest wall with characteristic sulphur granules are present. The most common misdiagnosis is of bronchial carcinoma and, while some patients may not suffer too much if a resected “cancer” turns out to be actinomycosis, it is of course a tragedy if this rare but treatable disease is completely missed. Confusion with tuberculosis is a further problem, both from the radiological appearance and because Actinomycosis israelii can stain acid fast. Moreover, some reports have described cases of colonisation of tuberculous cavities. Actinomycosis is more common in the lower lobes but upper lobe lesions occur and are then more likely to mimic tuberculosis. In the case described here one presumes that the initial diagnosis of tuberculosis was erroneous. It is interesting to note that rifampicin has been tried in the treatment of actinomycosis so, if this drug was included in the three-drug regimen used, it might have held the disease under partial control during the previous period of empirical treatment for tuberculosis.

This patient was susceptible to pulmonary actinomycosis from two risk factors – namely, dental sepsis and diabetes. Because of the anaerobic conditions required for actinomycosis to flourish, response to penicillin and other appropriate antibiotics tends to be slow. Surgery alone is unlikely to cure, although surgical drainage is usually recommended where there is abscess formation. Some cases are cured by antibiotic treatment alone. In this case the authors imply that they elected for immediate surgery because of a tentative diagnosis of fungal infection although, somewhat paradoxically, they actually had proof of actinomycosis from their bronchoscopic samples. One might speculate on the possibility of curing this patient with prolonged antibiotic therapy and postural drainage. However, because of the possibility of a chronic infection in a diabetic subject, sacrificing some viable lung at lobectomy was probably still the best management option.

As reported by Farrell et al, primary malignant melanoma of the bronchus is very rare and many previous case reports have not fully
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