The expandable metal stent for tracheal obstruction

A E Boothroyd, R Edwards, A J Petros, R Franks

Abstract

A 10 month old boy with stridor persisting from birth was found to have tracheal narrowing secondary to myofibromatosis of the tracheal wall. An expandable metal stent was positioned across the tracheal stenosis with immediate clinical improvement. There were no complications relating to stent insertion. The child remained clinically asymptomatic and repeat bronchoscopy at nine months' follow up showed that the stent had become completely endothelialised with no evidence of granuloma formation.

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Keywords: trachea, stent, myofibromatosis.

The use of endoscopically inserted expandable metal stents for tracheobronchial strictures has been reported in adults with good relief of clinical symptoms and few complications. The adult cases described have been mainly for patients with malignant disease but there are also reports of successful stenting of benign lesions such as post-transplantation anastomotic strictures and fibroinflammatory stenosis. There are very few published reports of endoscopic placement of expandable stents in the malacic segment of infants and neonates.[1] However, with the developments in the management of the neonatal airway more infants with airway collapse are surviving.[2] This report describes an infant with isolated myofibromatosis of the distal trachea which was successfully treated with a Gianturco stent.

Case report

A 10 month old infant presented with inspiratory stridor since birth. He had been thoroughly investigated for failure to thrive at the referring hospital with no conclusion. During feeding and sleeping, peripheral saturations fell to 88%. Rigid bronchoscopy revealed a severe anteroposterior narrowing of the trachea from about 3 to 1 cm above the carina. Echocardiography and computed tomography failed to reveal a cause for the tracheal stenosis. Magnetic resonance demonstrated increased signal within a thickened area. The anteroposterior diameter of the trachea at this level was 1–2 mm (fig 1). No anomalous vessels were seen. The mass was noted to be contiguous with the ascending aorta and transverse arch. Bronchoscopic biopsy was therefore considered unsafe.

Via a median sternotomy the upper trachea appeared normal. The lower third was thickened and biopsy specimens were taken from the thickened tracheal wall. Histology revealed dense hyalinised fibrocollagenous tissue indicative of myofibromatosis. Resection of the involved tracheal segment was considered but not felt to be a practical option. A modified Gianturco-Rosch Z-stent (Wm Cook Ltd), was introduced bronchoscopically, and under fluoroscopic control deployed across the stenosis. The unconstrained stent diameter was 14 mm. There was an immediate improvement in ventilatory function with a fall in the arterial carbon dioxide tension from 9·33 to 5·33 kPa, the patient was extubated without difficulty and his stridor resolved. Frontal and lateral chest radiography (figs 2A and 2B) showed the stent well expanded and to lie within the distal trachea across the stenosis. At nine months of follow up the child's respiratory status remains stable and significant improvement in weight and developmental assessment has occurred. A repeat bronchoscopy was performed and showed that the stent had become completely endothelialised with no evidence of granuloma formation.

Discussion

Infantile fibromatosis is an unusual condition generally presenting in the newborn period. Approximately half of the affected infants have solitary lesions and the remainder have more widespread visceral involvement.[3] However, unless the nodules obstruct bronchi or bowel or involve a vital organ such as the heart they do not pose a threat to life.

Although spontaneous regression of lesions is normal, the clinical features of extreme tracheal narrowing in this patient required intervention. Resection of the long involved segment was considered inappropriate. It was felt that in such

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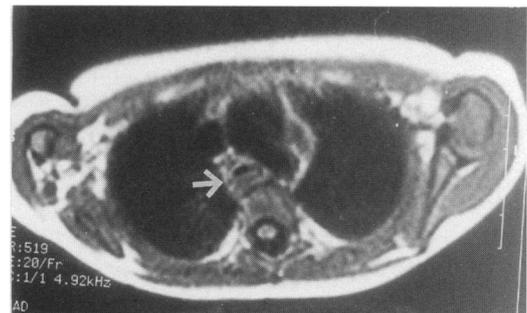R Edwards

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Figure 1   T1 weighted magnetic resonance image showing thickening of the distal tracheal wall (arrow). The area of abnormal signal is contiguous with the ascending aorta and transverse arch.
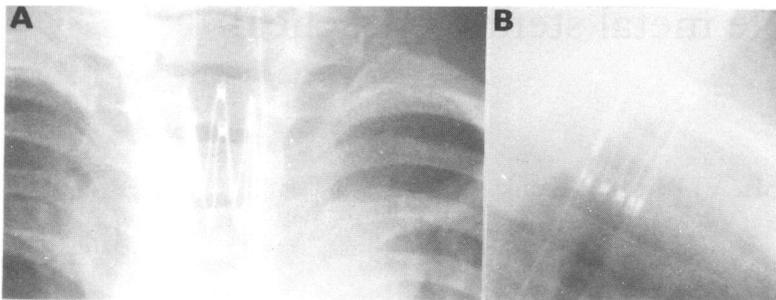
Figure 2 (A) Frontal and (B) lateral chest radiographs demonstrating the stent expanded within the tracheal lumen.

a small trachea any further intervention would be very difficult and lead to problems later. Tracheal stenting achieved good symptomatic relief and does not preclude further dilatation or surgical resection if required.

Our difficulty in imaging the lesion was not unique as both computed tomography and angiography have limitations in delineating the extent of disease.[4] Magnetic resonance is invaluable in identifying fibromatosis but signal intensities may vary within a focal lesion, or between disease sites. The appearances on magnetic resonance reflect the composition and cellularity of the lesions with the low signal areas representing hypocellularity and dense collagen deposition.[5]

The first clinical use of expandable metal stents in tracheobronchial stenosis due to malignant disease was reported by Wallace *et al* in 1986,[6] and since then, they have also been used in the treatment of a variety of benign conditions including postoperative strictures, tracheomalacia, relapsing polychondritis, and fibroinflammatory strictures.[7–9] A self expanding polymorphic woven stent used in the second of two case reports in neonates[1] was successful for six months. A 'zigzag' expandable stent constructed specifically for the first neonate also proved successful in maintaining patency, though the neonate died from other causes. Mair *et al* clearly identify the problem of progressive tracheobronchomalacia and its life threatening nature. They tentatively suggest that expandable stents may reduce the need for other high risk surgical procedures.[9]

Animal studies have shown that the stent struts are partially covered by mucosal proliferation 4–6 weeks after placement. Microscopically this consists of pseudostratified, ciliated columnar epithelium and stratified squamous cells. Although epithelial hyperplasia does occur, this does not result in significant luminal stenosis.[10] However, in the presence of active inflammation, delayed stent insertion has been advised, as a granulomatous

proliferation has necessitated stent removal.[11] One of the two neonates described by Mair *et al* died.[1] Emergency bedside bronchoscopy confirmed a patent stent and no airway collapse or obstruction. Postmortem histological examination revealed a foreign body inflammatory response and microscopic examination revealed sloughing of adjacent respiratory cilia and squamous metaplasia.[1] These authors have subsequently indicated that significant granulation formation after stent insertion may necessitate subsequent surgery and led them to explore other options including homograft tracheal transplantation. Others also report that granulomata formation and migration can cause long term problems. The potential difficulties of a child outgrowing a stent also have to be addressed in the future (Dr Ethan Phelan, Deputy Director, Department of Radiology, Royal Children's Hospital, Melbourne, personal communication to RE).

The combination of these histological features plus the low surface area of the stent suggests that mucociliary clearance is unlikely to be impaired, thus reducing the risk of stent blockage. For the same reason stent removal is probably impractical after 4–6 weeks, and in this case a larger stent diameter was chosen to accommodate growth. There is no specific paediatric tracheobronchial stent currently available and further improvements in stent design are likely to be necessary.

Tracheobronchial stenting provides another therapeutic option in a difficult clinical situation but requires careful consideration as the long term effects are not known.

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