The Journal of Laryngology and Otology
June 1994, Vol. **108**, pp. 474–478

Formal reintubation for incipient neonatal subglottic stenosis

JOHN M. GRAHAM, F.R.C.S.

Abstract
In a small prospective series of 10 children who presented with incipient subglottic stenosis following neonatal intubation a protocol of formal reintubation for two weeks, with sedation, enabled six of the children to avoid tracheostomy or other forms of surgery and in the remaining four it is unlikely that the trial of reintubation made the degree of laryngeal and subglottic damage worse.

Key words: Tracheostomy; Laryngostenosis; Infant, newborn

Introduction

This paper describes the result of adopting a conservative form of management for incipient subglottic stenosis in infants who have been intubated and ventilated in neonatal intensive care units. Incipient subglottic stenosis is defined as a degree of inflammatory swelling, in the larynx and subglottis, that prevents respiration without the presence of an endotracheal tube and would conventionally cause the child to have a cricoid split (Cotton and Seid, 1980; Cotton, 1985; Ochi *et al.*, 1987; Richardson and Inglis, 1991), a tracheostomy with late reconstructive surgery (Cotton, 1978; Cotton and Evans, 1981; Crysdale, 1983; Quiney *et al.*, 1986; Prescott and Vanlierde, 1989) or the more recently described immediate reconstructive surgery (Prescott, 1988; Richardson and Inglis, 1991; Stenson *et al.*, 1993).

Pathological studies (Gould and Graham, 1985; Gould and Howard, 1985; Quiney and Gould, 1985; Gould, 1988; Gould and Graham, 1989) have shown that although mucosal damage, with inflammatory changes and ulceration occurs rapidly in the first 24–48 hours after intubation, this is followed, if the tube remains undisturbed, by healing with re-epithelialization of the mucosal surfaces around the tube. Healing is often complete after four weeks, with the mucosa showing fibrosis and metaplastic squamous epithelium covering the previously ulcerated areas. Re-epithelialization even takes place directly over exposed cartilage. These pathological findings suggest that where the critical subglottic narrowing is caused by inflammatory soft tissue, and when antibiotics, steroids and other measures have not allowed extubation, then leaving an endotracheal tube undisturbed for two weeks should allow epithelialization to occur around the tube, stabilizing the soft tissue swelling and allowing extubation.

This pilot study was therefore designed to establish whether endoscopy, followed by a two-week period of undisturbed intubation to allow healing to take place, might permit successful extubation in babies who previously, in our unit, would have been managed by tracheostomy or a cricoid split (Quiney *et al.*, 1986).

Materials and methods

At University College Hospital, London (UCH) there is a regional neonatal unit containing 30 cots. Between 320 and 380 babies are admitted to the unit per year, 80–120 weighing less than 1.5 kg and 30–60 weighing less than 1.0 kg. Between 90 and 130 babies a year require intubation and ventilation: 35–60 with a birth weight of 1 kg or less. Between 1980 and 1983 the unit generated five cases of subglottic stenosis (SGS) requiring tracheostomy

TABLE I
PROTOCOL FOR FORMAL REINTUBATION

Entry	Unextubatable Other aspects stable (especially chest)
Stage 1	Microlaryngoscopy and bronchoscopy Two weeks uninterrupted nasal intubation Sedation and ventilation Nasogastric feeding and antireflux Keep chest clear
Stage 2	At two weeks: microlaryngoscopy and reintubation Extubate later in Intensive Therapy Unit Be prepared to reintubate
Stage 3a	Successful extubation
3b	Failed extubation:- If larynx no worse; and chest bad: reintubate after two weeks and try again. Tracheostomy after second failure.
3c	Larynx worse at microlaryngoscopy or baby unstable (e.g., cardiac problem): tracheostomy.

From the Royal Ear Hospital, Middlesex Outpatients Department, Cleveland Street, London W1P 5FD.
Accepted for publication: 14 January 1994.

TABLE II
NEONATAL DATA

Name	Date of birth	Weeks of gestation	Days intubated	Oral/ nasal tube	Size of tube (mm)	Birth weight (kg)	Born UCH (U) or elsewhere (E)
Failures							
TA	22.12.86	40	7	0	?	3.0	E
NF	19.04.88	27 (Twin)	120	0	?	1.7	E
SE	14.07.88	28	7	0	?	1.25	E
TC	14.10.88	30	21	0	?	1.7	E
Successes							
OF	03.07.84	26	4	0	?	0.9	E
RA	16.05.89	25	6	0	2.5	1.0	U
AA	24.01.91	40	2	0	3.5	1.2	E
NS	28.07.91	24	18	0	2.5	0.7	E
DA-S	04.06.91	40	3	0	3.5	3.5	U
RW	23.12.92	24 (Twin)	47	0	2.0	0.6	E

but since 1984 there have been no cases of SGS among babies who were born at our hospital and ventilated only in our neonatal unit.

Table I describes the protocol devised in conjunction with the neonatologists at UCH for managing babies with incipient subglottic stenosis who could not be extubated. The criteria for entry to the protocol are: (1) failure of extubation at a time when the baby's general condition, particularly the state of the lungs, would normally allow extubation; (2) that the next step in conventional management would be either tracheostomy or cricoid split or a primary reconstructive operation.

First the baby's airway is properly assessed under anaesthetic to confirm the presence of subglottic narrowing severe enough to obstruct breathing, and to exclude other causes of airway obstruction. Then the child is reintubated with a nasal endotracheal tube, of a size that fits the airway comfortably with no leak, and the child is sedated and intensively nursed for two weeks. During this time the tube must not be disturbed and movement of the tube is kept to a minimum. After two weeks there is formal extubation, with steroid cover, usually after further endoscopy under anaesthetic.

If this extubation fails then a decision is made either to perform a tracheostomy or to try another two weeks' intubation. Babies with cardiac instability may be given a tracheostomy at this point, otherwise the decision depends on the state of the chest and on whether the subglottis looks better or worse than it did two weeks earlier.

Results

Of the 10 babies who went through this protocol the trial of extubation was successful in six, was partly successful in one but failed in three. Table II shows the basic neonatal history, with gestational ages and birth weights. Only four had been intubated after birth for longer than seven days, but even brief periods of intubation have been shown histologically to cause severe damage (Gould and Howard, 1985) and most large series of laryngotracheal stenosis requiring tracheostomy include babies intubated for as little as 24 hours.

Tables III and IV show that both groups, successes and failures, include early, primary and later, secondary presentation. The numbers in this pilot study are small and no statistical analysis has been attempted. Conclusions of the study are shown in Table V.

Discussion

In the four babies in whom the method failed and tracheostomy had to be performed in (Table III) the first and third cases regained an adequate laryngotracheal lumen by spontaneous growth and had their tracheostomy closed without the need for reconstructive surgery. In the third baby (SE) it could be argued that formal reintubation had really been successful, since he was extubated, with a good airway and no stridor, for a period of two months before gastroesophageal reflux led to a succession of cya-

TABLE III
FAILURES — TRIAL OF INTUBATION: TRACHEOTOMY PERFORMED

Name	1°/2°	Wt (kg)	Corrected age	Oral/ nasal tube	Size of tube (mm)	Days intubated	Outcome	Comments
TA	2°	7.0	9/12	N	3.0	8	Tracheostomy; decannulated without reconstruction	Transglottic oedema, laryngotrachaeo-bronchitis
NF	1°	2.6	1/12	N	3.0	23	Tracheostomy; reconstruction, not yet decannulated	Twin, sub- and supraglottic oedema
SE	2°	3.6	2/52	N	2.5	15	Tracheostomy; decannulated without reconstruction	Successful extubation for 2/12: slow stridor; Severe reflux; cyanotic episodes collapsed; reintubated; tracheostomy
TC	2°	1.9	3/52	N	3.0	21	Tracheostomy; died of pneumonia	Birth asphyxia; severe cerebral damage

1° = primary presentation: never successfully extubated.
2° = secondary presentation, after extubation and discharge from hospital.

https://doi.org/10.1017/S0022215100127148 Published online by Cambridge University Press

TABLE IV
SUCCESSES — TRIAL OF INTUBATION

Name	1°/2°	Wt (kg)	Corrected age	Oral/ nasal tube	Size of tube (mm)	Days intubated	Outcome	Comments
OF	2°	6.8	7/12	N	2.5	9		No leak. Reintubated same day.
						+7	OK	Leak. No further stridor
RA	1°	1.25	−1/12	N	3.0	16		Reintubated in 2 days
						+12		Reintubated in 2 days
						+14	OK	No further stridor
AA	2°	6.0	3/12	N	3.0	15		Reintubated same day: heart slowed
						+4	OK	No more stridor (since birth)
NS	1°	0.6	−3/12	N	2.5	20	OK	
DA-S	2°	8.5	8/12	N	3.0	15	OK	
RW	2°	2.8	1/12	N	2.5	18	OK	

1°, Primary presentation.
2°, Secondary presentation.

notic episodes and tracheostomy was performed to protect his airway.

The second case (NF) does have a severe degree of laryngotracheal scarring but this is likely to have reflected a more severe degree of trauma at the time that she was referred to us, having already been intubated, with many extubations and reintubations, for 120 days. This baby's social background made it likely that management of a tracheostomy would be difficult (events have proved this assessment correct), and the period of formal reintubation was followed by an anterior and posterior cricoid split and by the use of the laser before tracheostomy was eventually performed. The fourth baby had very severe cerebral damage in addition to his subglottic stenosis and was not expected to survive. After tracheostomy he was able to be transferred to another hospital, closer to his parents' home, and at the time of his death from pneumonia had shown no improvement in his overall state.

The first and third cases (Table III) have normal phonation after decannulation, further evidence that the period of controlled reintubation did not produce significant laryngeal damage.

In the successful cases it could be asked whether, without the period of formal undisturbed intubation, these children really would have required tracheostomy or other forms of surgery. None of the published literature on SGS defines precise criteria for tracheostomy after failure of extubation in premature babies. Such babies are generally simply referred to the local ENT service, or transferred to a specialist centre, with a request for endoscopy and surgical intervention if laryngotracheal damage is confirmed. At the time of referral the second and fourth cases (Table IV) were still intubated in our neonatal intensive care unit.

TABLE V
CONCLUSIONS: 10 CHILDREN IN THE TRIAL OF FORMAL REINTUBATION

6/10	Avoided tracheostomy
2/10	Tracheostomy: decannulated later without reconstruction
1/10	120 days original intubation – rib graft at 8/12 – not decannulated at 5 years – phonates
1/10	Died of other causes

The first, third, fifth and sixth cases had originally been extubated and discharged home with mild stridor, resulting from a lesser degree of SGS. At the corrected ages of 3, 7 and 8 months respectively they developed worsening stridor after respiratory infection and had to be reintubated. The sixth case had been intubated for 47 days at the hospital where she was born, and had been extubated with mild stridor and transferred to a district general hospital near her home. The stridor had become progressively more severe, with increasing oxygen dependence and increasingly frequent episodes of more severe respiratory obstruction with hypercapnea making some form of intervention necessary.

Five of these six infants, therefore, were intubated at the time of referral to the ENT unit and in all of them endoscopy showed that the only reason for airway obstruction was swelling in the larynx and subglottis. Bearing in mind the fact that all neonatal intensive care units have enormous experience of intubating and subsequently extubating babies, it seems reasonable to accept the inevitably rather vague criterion of 'failure of extubation' as a blanket term describing the well recognized clinical situation that conventionally has led to cricoid split, tracheostomy or early reconstructive surgery.

All the babies in this study had originally been intubated with Cole pattern tubes. Although a recent study (Rivers, personal communication) showed no difference in the risk of SGS whether Cole pattern or straight-sided endotracheal tubes were used in the neonatal period, we chose to use straight-sided tubes and nasal endotracheal intubation for the period of formal reintubation in each case. The choice of the size of tube was made in consultation with the anaesthetists and the neonatologists. The tube used in each case was large enough to prevent a leak of air on positive pressure ventilation but not large enough to fit too tightly in the larynx. In most cases a leak of air around the tube appeared a day or two before the second endoscopy and extubation and was a reassuring sign.

It is important to achieve adequate sedation of the baby to prevent accidental intubation. This was achieved using various agents including chloral hydrate and trimeprazine. Ventilation was used in some cases. Two of the babies developed a mild chest infection after the first endoscopy, treated with antibiotics and physiotherapy. Clearance of secretions is also important to prevent blockage of the nasal endotracheal tube.

Subglottic or laryngotracheal stenosis (SGS) is a serious complication of neonatal intensive care. It occurs in a small proportion of infants who receive laryngeal intubation. No specific aetiological factor has been identified: gestational age, duration of intubation and birth weight, for example, all show considerable variation in the published reports of SGS (Fearon *et al.*, 1966; Abbott, 1968; Hengerer *et al.*, 1975; Holinger *et al.*, 1976; Strong and Passy, 1977; Hawkins, 1978; Cotton and Seid, 1980; Papsidero and Pashley, 1980; Crysdale, 1983; Quiney and Gould, 1985; Quiney *et al.*, 1986; Laing *et al.*, 1986; Sherman *et al.*, 1986; Albert *et al.*, 1990; Grundfast *et al.*, 1990).

Established SGS is a clear clinical and pathological entity. The airway through the larynx and subglottis is too narrow to allow respiration; this narrowing is produced by fibrotic scar tissue, often with thickening of the cricoid cartilage itself.

While a baby remains intubated the lumen of the subglottis is stented by the endotracheal tube: in failed extubation removal of this tube leads to rapid narrowing of the lumen by acute inflammatory swelling of soft tissue within the ring of the cricoid cartilage. Clinically this leads to stridor with airway obstruction: when the tube is replaced the inflammatory swelling is compressed and the lumen is held open again. Once tracheostomy has been performed, however, this swollen soft tissue organizes into mature fibrotic scar tissue. In the course of the next year or two the more fortunate infants develop a gradual spontaneous widening of the subglottic lumen and can be decannulated. In others some form of laryngotracheal reconstruction is necessary, with variable benefits to the airway and variable effects on the voice (Smith *et al.*, 1993).

The repertoire of conventional management of infants with incipient SGS has included cricoid split, tracheostomy and laryngotracheal reconstruction: immediate or late. Tracheostomy is associated with a reported risk of mortality of zero to 24 per cent (Fearon and Cotton, 1974; Holinger *et al.*, 1976; Cohen *et al.*, 1977; Pashley, 1983; Quiney *et al.*, 1986; Swift and Rogers, 1987; Campbell *et al.*, 1989; Prescott and Vanlierde, 1989), and may also deprive the developing child of phonation making it necessary to use a sign system such as Makaton. Reconstructive surgery, after spontaneous growth of the airway has failed to occur, is associated with a high incidence of abnormal phonation (Smith *et al.*, 1993).

A child with a tracheostomy is not easy to look after. Tube changes, the need for repeated suction, difficulties with feeding, lack of phonation (linked with absent cry and spoken language), repeated admissions to hospital for endoscopy and the knowledge that obstruction of the tube may lead to the death of the child, all produce high levels of anxiety and stress in the families of these children (Hazinski, 1986; Graham, 1988; Jennings, 1988; Gillinson, 1988; Campbell *et al.*, 1989). In the UK this has led to the founding of a parents' association, Aid for Children with Tracheostomies (ACT) which provides support for families in this situation; there is also a clear need for more specialist nurses, to form a link between the hospitals, the home and local medical and nursing services.

Conclusions

In a small group of 10 babies with subglottic stenosis caused by endotracheal intubation in the neonatal period, a formal trial of controlled reintubation allowed successful extubation in six cases. Of the remaining four babies who received tracheostomies, one still has her tracheostomy, one has died from causes not related to his SGS and two have been decannulated with normal phonation and without needing reconstructive surgery. This pilot study would justify a larger trial of this method on the grounds that it appears to do no harm and may allow a proportion of babies with incipient subglottic stenosis to be extubated without further surgical intervention.

Acknowledgements

I am most grateful for the help and advice of Dr Jonathan Shaw, Professor Osmund Reynolds and their colleagues in the University College Hospital Neonatal Unit, Dr Beverley Astley, Consultant Anaesthetist and Dr Steven Gould, Neonatal Pathologist, and not least to Miss Susan Shann who expertly typed the manuscript and constructed the Tables.

References

Abbott, T. R. (1968) Complications of prolonged nasotracheal intubation in children. *British Journal of Anaesthesia* **401:** 347–353.

Albert, D. M., Mills, R. P., Fysh, J., Gamsu, H., Thomas, J. N. (1990) Endoscopic examination of the neonatal larynx at extubation: a prospective study of variables associated with laryngeal damage. *International Journal of Paediatric Otorhinolaryngology* **20:** 203–212.

Campbell, J. B., Morgan, D. W., Pearman, K. (1989) Experience with the home-care of tracheotomised paediatric patients. *Archives of Otorhinolaryngology* **246:** 345–348.

Cohen, S. R., Eavy, R. D., Desmond, M. S., May, B. C. (1977) Endoscopy and tracheostomy in the neonatal pereiod: a 10-year review. *Annals of Otology, Rhinology and Laryngology* **86:** 577–583.

Cotton, R. T. (1978) Management of subglottic stenosis in infancy and childhood. *Annals of Otology, Rhinology and Laryngology* **87:** 649–657.

Cotton, R. T. (1985) Prevention and management of laryngeal stenosis in infants and children. *Journal of Paediatric Surgery* **20:** 845–851.

Cotton, R. T., Seid, A. B. (1980) Management of the extubation problem in the premature child. Anterior cricoid split as an alternative to tracheostomy. *Annals of Otology, Rhinology and Laryngology* **89:** 508–511.

Cotton, R. T., Evans, J. N. G. (1981) Laryngotracheal reconstruction in children: five-year follow-up. *Annals of Otology, Rhinology and Laryngology* **90:** 516–520.

Crysdale, W. S. (1983) Subglottic stenosis in children. A management protocol plus surgical experience in 13 cases. *International Journal of Paediatric Otorhinolaryngology* **6:** 23–36.

Fearon, B., Macdonald, R. E., Smith, C., Mitchell, D. (1966) Airway problems in children following prolonged endotracheal intubation. *Annals of Otology, Rhinology and Laryngology* **75:** 975–986.

Fearon, B., Cotton, R. T. (1974) Surgical correction of subglottic stenosis of the larynx in infants and children. Progress report. *Annals of Otology, Rhinology and Laryngology* **83:** 428–431.

Gillinson, P. (1988) Acquired subglottic stenosis in infants: a parent's view. *Journal of Laryngology and Otology* **102 (17):** 41–44.

Gould, S. J. (1988) The pathology of neonatal endotracheal intubation and its relationship to subglottic stenosis. *Journal of Laryngology and Otology* **102 (17):** 3–6.

Gould, S. J., Graham, J. M. (1985) Acquired subglottic stenosis in neonates (editorial) *Clinical Otolaryngology* **10:** 299–302.

Gould, S. J., Graham, J. M. (1989) Long-term pathological sequelae of neonatal endotracheal intubation. *Journal of Laryngology and Otology* **103:** 622–625.

Gould, S. J., Howard, S. (1985) The histopathology of the larynx in the neonate following endotracheal intubation. *Journal of Pathology* **146:** 301–311.

Graham, J. M. (1988) Acquired subglottic stenosis in infants: introduction. *Journal of Laryngology and Otology* **102 (17):** 1.
Grundfast, K. M., Camilon, F. S., Pransky, S., Barber, C. S., Fink, R. (1990) Prospective study of subglottic stenosis in intubated neonates. *Annals of Otology, Rhinology and Laryngology* **99:** 390–395.
Hawkins, D. B. (1978) Hyaline membrane disease of the neonate. Prolonged intubation in management: effects on the larynx. *Laryngoscope* **88:** 201–224.
Hazinski, M. F. (1986) Pediatric home tracheostomy care: a parent's guide. *Pediatric Nursing* **12:** 41–69.
Hengerer, A. S., Strome, M., Jaffe, B. F. (1975) Injuries to the neonatal larynx from long-term endotracheal tube intubation and suggested tube modification for prevention. *Annals of Otology, Rhinology and Laryngology* **84:** 764–770.
Holinger, P. H., Kutnik, S. L., Schild, J. A., Holinger, L. D. (1976) Subglottic stenosis in infants and children. *Annals of Otology, Rhinology and Laryngology* **85:** 591–599.
Jennings, P. (1988) Nursing and home aspects of the care of a child with a tracheostomy. *Journal of Laryngology and Otology* **102 (17):** 25–29.
Laing, I. A., Cowan, D. L., Ballentine, G. M., Hume, R. (1986) Prevention of subglottic stenosis in neonatal ventilation. *International Journal of Paediatric Otorhinolaryngology* **11:** 61–66.
Ochi, J. W., Seid, A. B., Pransky, S. M. (1987) An approach to the failed cricoid split operation. *International Journal of Paediatric Otorhinolaryngology* **14:** 229–234.
Papsidero, M. J., Pashley, N. R. T. (1980) Acquired stenosis of the upper airway in neonates, an increasing problem. *Annals of Otology, Rhinology and Laryngology* **89:** 512–514.
Pashley, N. R. T. (1983) Serial dilatation compared with elective laryngo-tracheoplasty in the treatment of acquired subglottic stenosis in children. *International Journal of Paediatric Otorhinolaryngology* **5:** 59–65.
Prescott, C. A. J. (1988) Protocol for management of the interposition cartilage graft laryngotracheoplasty. *Annals of Otology, Rhinology and Laryngology* **97:** 239–242.
Prescott, C. A. J., Vanlierde, M. J. R. R. (1989) Tracheostomy in children — the Red Cross War Memorial Children's Hospital experience 1980–1985. *International Journal of Paediatric Otorhinolaryngology* **17:** 97–107.
Quiney, R. E., Gould, S. J. (1985) Subglottic stenosis; a clinicopathological study. *Clinical Otolaryngology* **10:**315–327.
Quiney, R. E., Spencer, M. G., Bailey, C. M., Evans, J. N. G., Graham, J. M. (1986) Management of subglottic stenosis: experience from two centres. *Archives of disease in childhood* **61:** 686–690.
Richardson, M. A., Inglis, A. F. (1991) A comparison of anterior cricoid split with and without costal cartilage graft for acquired subglottic stenosis. *International Journal of Paediatric Otorhinolaryngology* **22:** 187–193.
Sherman, J. M., Lowitt, S., Stephenson, C., Ironson, G. (1986) Factors influencing acquired subglottic stenosis in infants. *Journal of Pediatrics* **109:** 322–327.
Smith, M. E., Marsh, J. H., Cotton, R. T., Myer, C. M. (1993) Voice problems after pediatric laryngotracheal reconstruction: videolaryngographic, acoustic and perceptual assessment. *International Journal of Paediatric Otorhinolaryngology* **25:** 173–181.
Stenson, K., Berkowitz, R., McDonald, T., Gruber, B. (1993) Experience with one-stage laryngeal reconstruction. *International Journal of Paediatric Otorhinolaryngology* **27:** 55–64.
Strong, R. M., Passy, V. (1977) Endotracheal intubation — complications in neonates. Archives of Otolaryngology **103:** 329–335.
Swift, A. C., Rogers, J. H. (1987) The outcome of tracheostomy in children. *Journal of Laryngology and Otology* **101:** 936–939.

Address for correspondence:
Mr. John M. Graham, F.R.C.S.,
The Royal Ear Hospital,
Middlesex Outpatients Department,
Cleveland Street,
London W1P 5FD.