The Journal of Laryngology and Otology
May 1997, Vol. **111**, pp. 478–481

Acquired subglottic cysts in the low birth weight, pre-term infant

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Abstract
Although subglottic cysts have previously been reported as a cause of airway obstruction in the neonate, they have previously been considered to be a relatively rare cause. Cystic narrowing of the subglottis has been associated with endotracheal intubation. With improving survival of pre-term infants the incidence of the condition could be expected to rise. Prior to 1996, only 58 cases had been reported in the literature. We believe that the true incidence of the condition has been considerably under-reported. Over a six-month period our unit diagnosed five cases of compressible cysts in the subglottis in low birth weight, pre-term infants. All patients underwent diagnostic microlaryngobronchoscopy and vaporization of the cysts by CO_2 laser. Three children required more than one procedure. In all cases a satisfactory airway was achieved. The pathogenesis, diagnosis and treatment of the condition is discussed.

Key words: Cyst, subglottic; Laryngeal diseases; Intubation, intratracheal; Infant, newborn

Introduction

Acquired subglottic stenosis is the most common serious long-term consequence of neonatal intubation, particularly in the low birth weight infant (Cotton and Evans, 1981; Cotton and Meyer, 1984). The pathological changes result in submucosal fibrosis forming a rigid stenosis (Hawkins, 1977, 1978). Subglottic cysts have previously been reported as a cause of airway obstruction in the neonate, but have always been considered to be a relatively rare cause (Mitchell *et al.*, 1987). Cystic narrowing of the subglottis has previously been associated with endotracheal intubation and with improving survival of pre-term infants the incidence of the condition could well be expected to rise. Following the establishment of a paediatric airway service at Guy's and St Thomas' Hospitals we have identified five cases of cystic narrowing of the subglottis within the first six months of the service.

Materials and methods

The cases presented represent children with subglottic cysts investigated and treated at Guy's and St Thomas' Hospitals. Over a six-month period from August 1995 to February 1996, 28 children underwent microlaryngobronchoscopy for upper airway compromise. Fifteen children were found to have subglottic narrowing. In 10 cases the stenosis was rigid but in five cases compressible cysts were discovered.

Patients

Case 1

A seven-month-old Afro-Caribbean male child had been born at 32 weeks gestation with a birth weight of 980 g. During the neonatal period the child had been intubated for 24 hours. At three months of age he presented with a major apnoeic episode which required ventilation for 48 hours. Stridor was noted in the initial period following extubation. He suffered two further bouts of croup with stridor. A diagnostic microlaryngobronchoscopy was performed which revealed a cystic subglottic stenosis with the cyst positioned posteriorly (Figure 1). CO_2 laser vaporization of the cyst was performed and a good airway achieved.

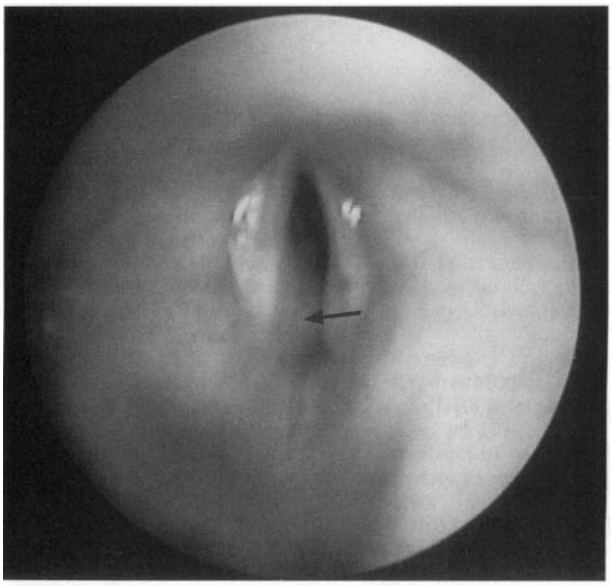

Fig. 1
Endoscopic view of the infantile larynx revealing a subglottic cyst (arrowed).

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Accepted for publication: 27 February 1997.

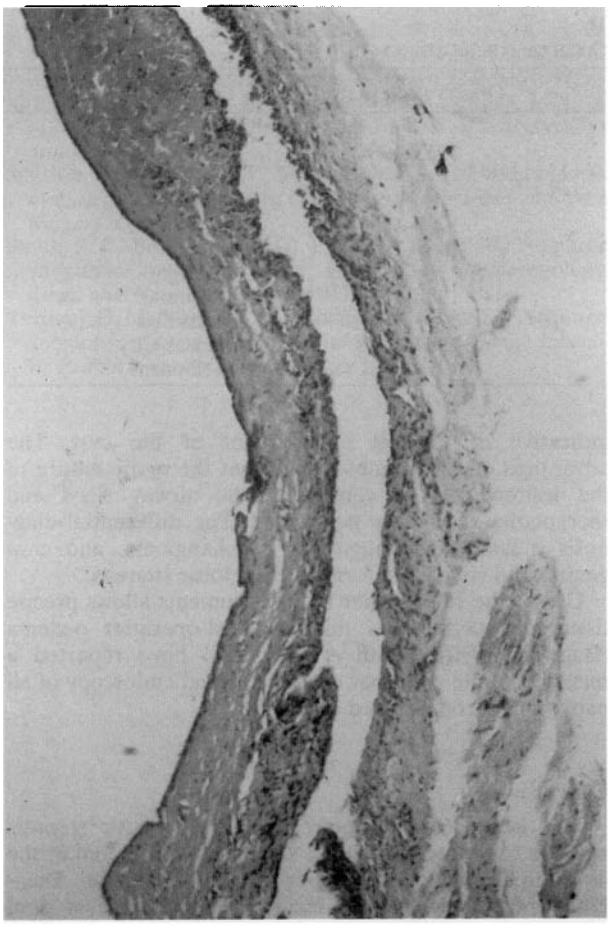

FIG. 2
Cyst wall, comprising non-ciliated cuboidal and columnar epithelium. (H & E; × 40)

Case 2

A nine-month-old Afro-Caribbean male child had been born at 27 weeks gestation with a birth weight of 1100 g. During the neonatal period the child had been intubated for 48 hours. Initial presentation followed three episodes of severe laryngotracheobronchitis with stridor. Microlaryngobroncoscopy revealed bilateral subglottic cysts. The left side was larger than the right. The cysts were vaporized by CO_2 laser. An excellent airway was achieved. Six months after the initial procedure the child had suffered one episode of croup and a repeat endoscopy revealed a recurrent cyst which was ablated.

Case 3

A 10-month-old Caucasian female child had been born at 29 weeks gestation with a birth weight of 1340 g. During the neonatal period the child had been intubated for two weeks. Initial presentation followed an episode of croup with associated respiratory distress and stridor. Microlaryngobroncoscopy revealed a subglottic cyst on the left side. The cyst was ablated using the CO_2 laser. This child has subsequently been well with no further episodes of stridor.

Case 4

An 18-month-old Caucasian male child had been born at 28 weeks gestation with a birth weight of 1285 g. During the neonatal period the child had been intubated for 24 hours. At 10 weeks he had presented with pyloric stenosis and required ventilation for two days after this procedure. Presentation to our unit followed a severe croup with stridor that was managed conservatively. Subsequent microlaryngobroncoscopy revealed bilateral subglottic cysts. Initial decompression was with a sickle knife. Follow-up endoscopy has shown recurrence of the cysts and two laser ablations have been performed.

Case 5

An 11-month-old Caucasian male child had been born at 28 weeks gestation with a birth weight of 1100 g. During the neonatal period the child had been intubated for five days but was well at discharge. He was readmitted aged four months and 11 months with respiratory distress and stridor. On his second admission an emergency tracheostomy was performed. He was then transferred to our unit for investigation. Microlaryngobroncoscopy revealed a bilobed right subglottic cyst and a smaller cyst on the left side. The cysts were vaporized with the CO_2 laser. Repeat endoscopy eight weeks later revealed a small recurrent cyst on the left side which was ablated with the laser. A check endoscopy four weeks later showed a good airway and the child was successfully decannulated. There have been no further episodes of stridor.

Histopathological analysis of a cyst wall revealed non-ciliated cuboidal and columnar epithelium (Figure 2).

In our series the age of presentation ranged from seven months to 18 months (Table I). In all our cases, a history of upper airway compromise led to a diagnostic microlaryngobronchoscopy being performed. Our practice is to re-endoscope the infant after four to six weeks to check for recurrence. In three of the five cases, residual cystic tissue required treatment. The one case that had undergone tracheostomy prior to transfer to our unit was successfully extubated after ablation of the cysts. No other cases required tracheostomy.

Discussion

Subglottic cysts were virtually unknown until recent years. The gradual increase in the number of cases is shown in Table II. Sporadic reports in the literature suggested great rarity confirmed by studies by DeSanto *et al.* (1970) who reported only one case in a review of 238

TABLE I
CLINICAL DETAILS OF CASES

Sex	Gestation	Birth weight	Intubations	Time intubated	Age at presentation
Male	27 weeks	1100 g	1	3 days	9 months
Male	32 weeks	980 g	2	3 days	7 months
Female	29 weeks	1340 g	1	14 days	10 months
Male	28 weeks	1285 g	2	3 days	18 months
Male	28 weeks	1100 g	1	5 days	11 months

Mean age at presentation	11 months
Mean gestational age at birth	28.8 weeks
Mean birth weight	1161 g

https://doi.org/10.1017/S0022215100137697 Published online by Cambridge University Press

TABLE II
OVERALL INCREASE IN NUMBER OF CASES OF SUBGLOTTIC CYSTS

Author	Publication year	No. of cases	Observation period	Location
Wigger and Tang	(1968)	1		New York
DeSanto *et al.*	(1970)	1	1945–1964	Mayo clinic
Chamberlain	(1970)	1		Tennessee
Dagan *et al.*	(1979)	1		Israel
Couriel and Phelan	(1981)	3	1979–1980	Melbourne
Mitchell *et al.*	(1987)	5	1969–1984	London
Toriumi *et al.*	(1987)	12		Chicago
Smith *et al.*	(1990)	9	1982–1988	Cincinnati
Triglia *et al.*	(1991)	1	1990	Marseilles
Downing *et al.*	(1993)	13	1988–1990	Kansas City
Smith *et al.*	(1994)	11	1979–1981	Melbourne

cases of laryngeal cysts over a 20-year period. A review of 15 years experience at Great Ormond Street from 1969–1984 yielded only five cases. Mitchell *et al.* (1987) did note that all their cases of subglottic cysts had been ventilated in the neonatal period. In recent years other authors have produced larger series and have noted the association with neonatal ventilation (Downing *et al.*, 1993; Smith *et al.*, 1994). Our experience does suggest a greater incidence of the pathology. There are a number of possible reasons for this. Firstly, the increasing survival of infants who are extremely premature may represent a true increase in the absolute numbers of cystic subglottic stenosis without an increase in the relative numbers of cases amongst pre-term infants who are ventilated. Secondly, the condition may be more widespread than previously recognized, but in its less severe form may resolve without significant compromise to the infant. Downing *et al.* (1993) examined 153 pre-term infants prospectively at discharge from a neonatal intensive care unit. Eleven cases of subglottic cysts (7.2 per cent) were noted. Only one child was completely asymptomatic. All the others exhibited stridor, a hoarse or abnormal cry or obstructive apnoea at the time of bronchoscopic examination. Downing *et al.* (1993) included two other cases that were picked up during investigation for airway compromise. In seven cases the cysts were observed without intervention and there was a gradual improvement in the airway over the next 12 months. In the other six cases: one cyst was marsupialized, there were two deaths and tracheostomy was required in three cases. Thus it may be seen that, although the condition may resolve spontaneously, there is a range of severity. The low rate of intervention in the study by Downing *et al.* (1993) is a reflection of the method of investigation. He used flexible bronchoscopy, whereas our practice is to perform a microlaryngobronchoscopy using rigid instruments which allows treatment of the cysts at the time of diagnosis.

Although cysts may be congenital in origin, all our cases had undergone endotracheal intubation in the early neonatal period. Joshi *et al.* (1972) examined 172 infants dying of respiratory distress syndrome. They found that 63 per cent of the larynges had mucosal or submucosal necrosis. Hawkins (1978) found that intubation for six days or more increased the incidence of mucosal damage to 93 per cent in the subglottic region. The subglottic mucosa has abundant mucous glands and most authors favour a hypothesis of mucocoele formation following trauma to the subglottic mucosa. Erosion, scarring and inflammation results in sealing of the ducts of submucosal glands which results in cyst formation (Wigger and Tang, 1968; Downing *et al.*, 1993; Smith *et al.*, 1994).

Infants with subglottic cysts may present with biphasic stridor and upper airway obstruction. A chronic or episodic presentation may also occur which may be indicative of gradual enlargement of the cyst. The advantage of rigid endoscopy is that the cystic nature of the lesions can be confirmed, the airway sized and therapeutic endoscopy performed. The differential diagnosis includes haemangioma, lymphangioma, and congenital and soft tissue forms of subglottic stenosis.

Use of the laser rather than instruments allows precise tissue destruction and minimal post-operative oedema (Bagwell, 1990). Smith *et al.* (1994) have reported a recurrence rate of 10 per cent and repeat endoscopy of all patients is recommended.

Conclusion

The incidence of acquired cystic subglottic stenosis appears to be increasing and should be considered in the differential diagnosis of upper airway obstruction. Diagnosis and management is facilitated by the use of rigid endoscopes and tracheostomy may be avoided in all cases where subglottic cysts are the only pathology.

Acknowlegements

The authors wish to thank the staff of the paediatric intensive care unit at Guy's Hospital for their expertise and support in the management of all our cases.

References

Bagwell, C. E. (1990) CO_2 laser excision of pediatric airway lesions. *Journal of Pediatric Surgery* **25:** 1152–1156.
Chamberlain, D. (1970) Congenital subglottic cysts of the larynx. *Laryngoscope* **80:** 254–259.
Cotton, R. T., Evans, J. N. G. (1981) Laryngotracheal reconstruction in children: five year follow-up. *Annals of Otology, Rhinology and Laryngology* **90:** 516.
Cotton, R. T., Meyer, C. M. III (1984) Contemporary surgical management of laryngeal stenosis in children. *American Journal of Otolaryngology* **5:** 360–368.
Couriel, J. M., Phelan, P. D. (1981) Subglottic cysts: a complication of neonatal endotracheal intubation? *Paediatrics* **68:** 103–105.
Dagan, R., Leiberman, A., Strauss, R., Bar-Ziv, J., Hirsch, M. (1979) Subglottic mucocele in an infant. *Pediatric Radiology* **8:** 119–121.
DeSanto, L. W., Devine, K. D., Weiland, L. H. (1970) Cysts of the larynx: Classification. *Laryngoscope* **80:** 145–176.
Downing, G. J., Hayen, L. K., Kilbride, H. W. (1993) Acquired subglottic cysts in the low birth weight infant – Characteristics, treatment and outcome. *American Journal of Diseases of Children* **147:** 971–974.
Hawkins, D. B. (1977) Glottic and subglottic stenosis from endotracheal intubation. *Laryngoscope* **87:** 339.
Hawkins, D. B. (1978) Hyaline membrane disease of the neonate: prolonged intubation in the management; effects on the larynx. *Laryngoscope* **88:** 201–224.

Joshi, V. V., Mandavia, S., Stern, L., Wiglesworth, F. W. (1972) Acute lesions induced by endotracheal intubation. *American Journal of Diseases of Children* **124:** 646–649.
Mitchell, D. M., Irwin, B. C., Bailey, C. M., Evans, J. N. G. (1987) Cysts of the infant larynx. *Journal of Laryngology and Otology* **101:** 833–837.
Smith, J. D., Cotton, R. T., Myer, C. M. III. (1990) Subglottic cysts in premature infants. *Otolaryngology – Head and Neck Surgery* **116:** 479–482.
Smith, S. P., Berkowitz, R. G., Phelan, P. D. (1994) Acquired subglottic cysts in infancy. *Archives of Otolaryngology, Head and Neck Surgery* **120:** 921–924.
Toriumi, D. M., Miller, D. R., Holinger, L. D. (1987) Acquired subglottic cysts in premature infants. *International Journal of Pediatric Otorhinolaryngology* **14:** 151–160.
Triglia, J. M., Portaspana, T., Cannoni, M., Petch, A. (1991) Subglottic cyst in a newborn. *Journal of Laryngology and Otology* **105:** 222–223.
Wigger, H. J., Tang, P. (1968) Fatal laryngeal obstruction by iatrogenic subglottic cyst. *Journal of Pediatrics* **72:** 815–820.

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https://doi.org/10.1017/S0022215100137697 Published online by Cambridge University Press