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Bilateral vocal fold paralysis in infants: tracheostomy or not?

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Abstract
Tracheostomy has, in the past, been performed in the majority of children under one year with bilateral vocal fold paralysis. We present our experience of 11 cases over a ten-year period during which tracheostomy was avoided whenever possible. Ten cases were managed conservatively but in the youngest a tracheostomy was required. Full bilateral vocal fold mobility developed in all cases at a mean age of 11.5 months (range 5–26 months). Our experience suggests that the airway can commonly be managed expectantly without a tracheostomy.

Key words: Vocal cord paralysis; Infants; Tracheostomy

Introduction

Bilateral vocal fold paralysis in infants is an uncommon but potentially life-threatening condition (Holinger *et al.*, 1976). Previously the majority of cases have been managed with a tracheostomy (Cohen *et al.*, 1982; Gentile *et al.*, 1986; Tucker, 1986; Swift and Rogers, 1987; Narcy *et al.*, 1990). It has, however, been our policy to avoid tracheostomy where possible, and we present our experience of 11 cases over a ten-year period.

Materials and methods

In the decade June 1982–June 1992, from a population catchment area of 750 000, 109 children under the age of one year required direct laryngoscopy because of stridor.

The direct laryngoscopy was performed under a light general anaesthetic with the patient breathing spontaneously during the examination. The speculum of the laryngoscope was placed in the vallecula to prevent splinting and vocal fold mobility was assessed by two or more surgeons. Bronchoscopy was performed in all patients to evaluate the tracheobronchial tree and cricoarytenoid joint mobility confirmed by direct palpation with a rigid metal suction tube.

Eleven patients (10 males : 1 female) had bilateral vocal fold paralysis and were subsequently examined at two-monthly intervals wherever possible. They form the basis of this report (Table I).

Results

Stridor was the presenting symptom in all 11 cases and five also had feeding difficulties. Mean age at diagnosis was 3.6 months (range 2 days–9 months).

An associated aetiology was identified in six cases. Two pregnancies were complicated by maternal medical problems: diabetes in one and epilepsy requiring anticonvulsant medication in the other. In the latter the neonate experienced convulsions for the first two weeks of life, and was born with the multiple congenital anomalies of micrognathia, bilateral clindactyly and deep eye folds. One child was born prematurely at 35 weeks and another sustained birth trauma resulting in a brain stem haemorrhage. Duane's syndrome, consisting of deficient horizontal ocular motility due to congenital absence of the abducens nerve, and developmental delay occurred in one case. The sixth case experienced viral encephalitis in the third month of life.

Ten children were managed conservatively without a tracheostomy but in the youngest a tracheostomy was performed because of its pre-operative clinical status. Full bilateral vocal fold mobility was subsequently demonstrated in all cases at a mean age of 11.5 months (range 5–26 months). Decannulation of the tracheostomy was achieved at nine months.

Discussion

The incidence of bilateral vocal fold paralysis in infants is unknown as the reports from North America (Holinger *et al.*, 1976; Cohen *et al.*, 1982; Gentile *et al.*, 1986; Tucker, 1986) and France (Narcy *et al.*, 1990) have been from tertiary referral centres with an ill-defined catchment area. Our paediatric airways practice serves a population of 750 000 and in the decade studied 11 (10 per cent) of the infants, under one year, undergoing direct laryngoscopy for stridor had bilateral vocal fold paralysis. This represents a prevalence of 0.75 cases per million per year. The condition, therefore, does not have a high prevalence but is a significant cause of stridor in children under one year.

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TABLE I

Patient no.	Sex	Presenting symptoms	Associated aetiology	Age at diagnosis	Treatment	Age at which vocal fold mobility confirmed
1	M	Stridor	P	3 months	Conservative	8 months
2	M	Stridor	BH	8 months	Conservative	14 months
3	M	Stridor feeding difficulty		4 months	Conservative	9 months
4	M	Stridor	EM–MCA	9 months	Conservative	16 months
5	M	Stridor		5 months	Conservative	7 months
6	M	Stridor feeding difficulty	DS	3 weeks	Conservative	5 months
7	M	Stridor feeding difficulty		2 days	Tracheostomy	9 months
8	M	Stridor		2 weeks	Conservative	12 months
9	M	Stridor feeding difficulty	DM	2 months	Conservative	12 months
10	F	Stridor	VE	6 months	Conservative	10 months
11	M	Stridor feeding difficulty		6 weeks	Conservative	26 months

P = premature; BH = brain stem haemorrhage; EM = epileptic mother; MCA = multiple congenital anomalies; DS = Duane's syndrome; DM = diabetic mother; VE = viral encephalitis.

The diagnosis is a subjective one and therefore, where immediately available, a second opinion is valuable. Splinting of the folds must be avoided by placement of the anterior lip of the laryngoscope in the vallecula and cricoarytenoid joint mobility is confirmed by direct palpation with a rigid metal sucker. In adults it is unrealistic to attempt to accurately designate a paralysed vocal fold as being anything other than lateral or paramedian (Willatt and Stell, 1989). In the adult the difference between these two positions is 2–3 mm and differentiating between them can be difficult. In the smaller infant larynx it is even more so. Determining the position of the fixed fold in infants is therefore prone to inaccuracy and is not performed in our practice.

The incidence of an associated aetiological factor varies from 54 per cent (Swift and Rogers, 1987) to 89 per cent (Tucker, 1986). Our series (54 per cent) is in agreement with the lower figure. The commonest associated factor is a neural tube defect. Interestingly, these two lower figures are the most recent and report, predominantly the 1980s decade during which amniocentesis and alpha-fetoprotein analysis to detect neural tube defects *in utero* were introduced. The lower incidence of associated factors in bilateral vocal fold paralysis may reflect the increased elective abortion rate in foetuses with neural tube defects.

A systematic approach is required if associated factors are to be identified. A full history must be taken with particular attention to maternal health during pregnancy, complications during labour and delivery, and neonatal health. A full paediatric examination may usefully be supplemented by a neurology opinion to identify additional subtle neurological signs. A plain chest X-ray can identify intrathoracic anomalies and a barium swallow may detect pharyngo-oesophageal dysfunction, gastro-oesophageal reflux and congenital vascular disorders. Computed tomography and magnetic resonance imaging, particularly of the posterior fossa, can diagnose intracranial disorders. Central nervous system disease, especially Arnold Chiari malformation with meningomyelocoele, birth trauma and surgery are the most commonly reported aetiologies.

Management of bilateral vocal fold paralysis in infants can be considered under two headings: (i) the airway and (ii) the paralysed folds themselves.

The condition is potentially life-threatening and early intervention to protect the airway has previously been practised. There is, therefore, a high incidence of tracheostomy varying from 54 per cent (Swift and Rogers, 1987) to 100 per cent (Tucker, 1986). Paediatric tracheostomy itself, however, is not without problems and has twice the morbidity and mortality rate of adult tracheostomy (McRae *et al.*, 1984). Complication rates of 20–49 per cent and mortality rates of 2–8.5 per cent have been reported (Tepas *et al.*, 1981; Wetmore *et al.*, 1982; Gilmore and Mickelson, 1986; Line *et al.*, 1986; Kenna *et al.*, 1987). We, therefore, have adopted a conservative policy and it has been possible to successfully manage 90 per cent of cases without a tracheostomy.

Narcy *et al.* (1990) advocate the use of the lateralization procedures of artytenoidectomy (Woodman, 1946) or arytenoidopexy (King, 1939) to improve the airway in refractory cases, and report a success rate of 62 per cent. However, they did not wait longer than nine months for spontaneous recovery before intervening surgically, and, as our series indicates, recovery can occur two years after diagnosis and a further case has been reported as apparently recovering after nine years (Emery and Fearon, 1984). Furthermore, lateralization is an irreversible procedure and any improvement in airway is at the expense of the voice. Lateralization has not achieved widespread acceptance.

Authors agree that vocal fold mobility spontaneously recovers in a significant number of cases but the percentage is unclear due to short observation periods and large numbers lost to follow-up. In our series all subjects were examined at regular intervals until vocal fold mobility could be endoscopically demonstrated. No cases were lost to follow-up and spontaneous recovery eventually occurred in all although requiring up to two years to do so.

Tucker (1986), using the posterior cricoarytenoid muscle re-innervation technique he pioneered (Tucker, 1976), intervenes early and reports a success rate of 50 per cent. This does not compare favourably with spontaneously recovery.

Conclusions

Bilateral vocal fold paralysis in children under one year does not have a high prevalence but is a significant cause of stridor in this group. The development of full bilateral mobility can take two years to develop but in the meantime the airway can commonly be managed expectantly without a tracheostomy. Occasionally, in the neonate the pre-operative clinical status requires a tracheostomy.

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