Speech and language development in children with acquired subglottic stenosis

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Communication Development

Most children with acquired subglottic stenosis require a tracheostomy. This paper describes the communication skills of children with tracheostomies before and after decannulation.

There are also a small number of children with acquired subglottic stenosis who do not require a tracheostomy. They are rarely referred for speech therapy. Quiney *et al.* (1986) in a retrospective review described this group as all having normal speech development. It should be noted, however, that the lack of comment of a speech delay in the medical notes is inconclusive. Unless there is an ongoing study with a speech therapist routinely assessing the speech and language development of this group, one cannot conclude categorically that this group is always free of problems in their speech development.

A review of the studies of communication skills in children with tracheostomy shows that this population has a wide range of different pathologies and abilities. The majority of the studies have been case studies in which the deficits and therapeutic implications in voice and language acquisition have been recognised. There have been no studies specifically on the communication skills of children with acquired stenosis. The dearth of studies can be explained in part by the fact that this group has only become a clinical problem in the last two decades, as more advanced techniques have enabled younger pre-term neonates to survive.

Only three studies (Simon and Handler, 1981; Ross, 1982; Kaslon and Stein, 1985) have attempted to describe the effects of tracheostomy on language acquisition.

The work of Simon and Handler has probably been the most comprehensive and is confirmed by our clinical observations. They studied 23 children, all of whom had had a period of aphonia, but of a wide range of duration, 4 months to 4 years, and with diverse aetiological factors requiring the tracheostomy. Some of the children had prolonged hospitalization in association with ventilation.

They found that children decannulated in the prelinguistic stage (that is, the period before the normal onset of spoken language) revealed speech and language skills commensurate with intellectual function. They did observe however some problems in later voice quality and breath support. This finding suggests that children can omit audible pre-speech practice for language development, and then move into expressive speech and language, all other factors being equal. This has also been found by Lenneberg (1962) and Hellyer and Farmer (1982).

Children decannulated in the linguistic period, however, show language delay and a delay in the acquisition of a mature sound system known as the phonological system. Those cannulated for more lengthy periods demonstrated more severe problems in these areas.

Children with acquired subglottic stenosis frequently require a tracheostomy that is long-term. Gould and Graham (1985) comment that the condition of acquired subglottic stenosis can cause greater problems in clinical management than congenital subglottic stenosis or laryngeal web. These lengthy cannulations have important implications for communication development, since usually cannulation does extend into the linguistic period.

The potential communication problems that this group may present can be summarised as follows:

1. Aphonia

In the first year of life these infants are often aphonic: the infant has no means of making audible sound, for example, crying, and vocal play, such as babbling, that are important precursors to the development of imitation and to subsequent language development. There is a disruption in the important turn taking games between mother and baby. A mother may find it difficult to pitch her language stimulation at the right level as she is receiving aberrant feedback from the child. The effect of this aphonia if it extends over the age of 12 to 15 months is to inhibit the development of oral language and speech.

2. Language

There is the possibility of a severe delay in the development of expressive language, for example, vocabulary, content, grammar, and its use; Kaslon and Stein (1985) studied the acquisition of voice, speech and language in ten tracheostomized children and documented not only a delay in expressive language but

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https://doi.org/10.1017/S0022215100600464 Published online by Cambridge University Press

found that to a lesser degree, receptive language was also affected.

3. Phonology

There is the possibility of a delay in the development of phonology, or the sound system required for speech.

4. Voice in the linguistic period

Voice may be completely absent or when present be of variable quality and efficiency, and may not be adequate for oral communication.

Associated problems

These children are also at considerable risk of:

1. Behaviour problems, either associated with a lag in neurodevelopmental maturity, as a consequence of impaired communication, or related to the disruption in the normal parent child relationship related to the child's nursing needs. In addition Rutter (cited by Clark and Clark, 1976) has shown that the repeated admissions to hospital that occur in this population are significantly related to behaviour problems. Freeland *et al.* (1974) in describing long-term tracheostomized children found that there was a higher than normal incidence of behavioural problems and a greater risk of social isolation.

2. Risk of the child becoming passive in his communication, and not being pushed to his potential.

3. Risk of the child relying on idiosyncratic gesture, so that he is understood only by the close family.

Variables that affect language development

The risk to the development of communication in these children cannot be solely attributed to the tracheostomy, as speech and language development is contingent upon many other factors. Some of the most pertinent factors are listed here:

1. Prematurity

The prevalence of delayed speech and language in the absence of mental retardation or severe hearing loss appears to be high in very low birth weight (VLBW) infants. Incidence figures range from 15-35 per cent of VLBW infants with delayed speech and language development at 2 years of age, compared with a prevalence of 2-3% in normal healthy full-term infants at 3 years of age (Fitzhardinge and Ramsay, 1973). Cupples (1985) said that the population of premature infants is a new group for whom prevention of speech and language disorders is a high priority. Ellison *et al.* (1983) found that a history of intubation, or respiratory distress syndrome were two of the variables placing the infants at greater risk of neurological deficit. The general finding of increased neurodevelopmental problems in this high risk group can extend into intellectual and school performance. It is important to appreciate that these children may present with subtle deficits in subtests, for example, arithmetic or abstract reasoning tasks, and yet fall within the average on a test of overall intellectual functioning.

2. Hearing

It is known that 5–14 per cent of the premature population will have sensorineural deafness. Premature infants are also at risk from secretory otitis media.

3. Environmental attitudes

There are many social factors that affect language development and these have been described elsewhere (Rutter and Martin, 1972); for example, social class and number of siblings. Long-term hospitalization should also be mentioned. Parental attitudes and coping strategies to the tracheostomy can affect the mother-child interaction and language stimulation.

Therefore in trying to examine the effects of tracheostomy on language development it is important to account for the other disabling factors. Very often a multifactorial aetiology accounts for a communication handicap at any one time, and these factors may change over time.

The literature is unanimous that early speech therapy intervention is effective in the treatment of children considered at risk for speech and language delay, such as hearing loss, cerebral palsy and cleft lip and palate. Children with acquired subglottic stenosis should be included in this group (Harlor, 1983).

Kaslon *et al.* (1978) showed that tracheostomized children were rarely referred for speech therapy. Handler *et al.* (1983) advised referral of any child identified as requiring a long-term tracheostomy. It is appropriate to refer shortly after diagnosis and tracheostomy, so that the family then know that there is a specialist involved who is overseeing the development of the child's communication skills.

The work of the speech therapist covers the following:

1. Preventative

Parents are counselled on the development of prelanguage skills, and how to optimize language stimulation. This involves encouraging the child's liberal use of gesture and pointing, imitation skills and oral motor movements, and if voice becomes possible, how to reinforce its use.

2. Therapeutic

The speech therapist may either work directly with the child or in an advisory capacity with the care giver. Programmes are devised according to the child's needs (Sell and Phippen, 1984). For example, the aphonic child is introduced to a formal sign system such as Makaton, so that communication skills are maximized. When there is voice, therapy may be required to establish oral language particularly when there has been a period of aphonia. Children may require ongoing help with the development of language and phonology sometimes until they are five or six years. Work on child's breathing and voice production may be indicated at any stage. Occasionally an electrolarynx is advised.

3. Diagnostic

The speech therapist through her assessment indentifies the significant aetiological factors in a communication disorder. The role of the tracheostomy in the communication problem is identified. it may not necessarily be the key factor.

4. Liaison

Liaison with the otolaryngologist is important so that the therapy programme is planned and modified as the child's medical condition changes. In particular, she will be anxious to explore possible tube modifications to allow phonation. The hospital based speech therapist then liaises with the local speech therapist.

Voice quality following rib graft reconstruction procedures

In these children it was observed that frequently the child was successfully decannulated but left with a permanent voice disorder. Indeed the review by Quiney *et al.* (1986) descibes how combined anterior and posterior rib grafts may succeed in widening the subglottic lumen, but possibly at the expense of voice quality. The arytenoids are moved apart by the rib graft that is inserted between the edges of the posteriorly split cricoid cartilage and as a result adduction of the vocal cords is impeded.

A pilot study was set up earlier this year with the following aims:

1. Systematically to measure the voice quality of children who had undergone rib graft laryngotracheoplasty and had later been successfully decannulated.

2. To assess anatomical and functional aspects of the vocal folds using fibreoptics and stroboscopy.

The population studied consisted of 16 patients, 9 boys and 7 girls. Their age range was 2.5–20 years with a median age of 5.5 years. All the patients had subglottic stenosis—three were congential in nature, and 13 were acquired. Three patients had also had tracheooesophageal fistula. Due to the nature of the unit the sample probably represented the more severe end of the spectrum of the disease. All the patients had been decannulated at least 4 months. Each child was matched for age and sex with a control.

The procedure consisted of:

a. Audio and video recording of a standard speech sample.
b. Electrolaryngographic recording.
c. Fibreoptic examination.
d. Parent and child questionnaires.

The speech therapist assessed the voice quality using a subjective perceptual framework based on the GRBAS scheme (Hirano, 1981) and the objective information of the electrolaryngographic recording. A Voiscope was used incorporating a laryngograph which is linked up to the BBC microcomputer (MacCurtain and Fourcin, 1982). This displays soft tissue contact variation at vocal fold level, either of the true of false vocal folds. It provides information from a frequency analysis, the regularity/irregularity of the vocal fold movement, and visual feedback during therapy.

A. On the basis of the perceptual and objective assessments of the speech therapists the voice was classified as:

Acceptable	31%
Moderately abnormal	38%
Severely abnormal	31%

therefore 69 per cent were classified as having a disordered voice.

The perceptual assessment showed the following:

1. Volume was reduced throughout the sample, even in the "acceptable voice" group. This suggests that both air pressure and the onset of vocal fold closure were inefficient.

2. Pitch was low. The voice was perceived as "heavy" and "thick" with a reduction in intelligibility.

3. Quality was rough, with elements of breathiness.

B. The objective assessment from the laryngographic information quantified the perceptual findings. The characteristic trends of the frequency analysis were:-

Mode

The favoured mode was found to be abnormally low—within 100-130 Hz range, whereas the normal child can be predicted to have a pitch range of 300-350 Hz.

Range

The range of frequencies used corresponds to the perception of intonation. The range of frequencies was judged inadequate in 31 per cent of the patients.

Distribution

The voice was dominated by low frequencies.

Vocal fold control

70 per cent had difficulty vibrating consecutively at the same frequency (whereas normal speakers vibrate twice or thrice at the same frequency) throughout the utterance. This random speed of vibration means that the voice does not sound clear. There was a scatter of incongruous frequencies outside the band needed for effective voice.

Vocal fold action

1. 70 per cent have random control of the vocal folds.
2. There is reduced soft tissue contact throughout the sample. This gives inefficient excitation of the airstream.
3. 31 per cent lacked sharp onset of closure. (Sharp onset of closure is required for "clear" voice).

C. Fibreoptic findings (see Kearns *et al.*, this supplement.) Of particular interest is the number of fibreoptic examinations that were possible—over half of the sample tolerated this procedure and as young as 4

https://doi.org/10.1017/S0022215100600464 Published online by Cambridge University Press

years. The smaller number of child questionnaires completed was related to age.

4. Parent and child questionnaires.

The parents' questionnaire was designed to detect parental degree of satisfaction with the residual voice quality and decannulation. When the child was old enough, his opinion was also sought. The results of the parent's questionnaire are as follows:-

1. 93 per cent classified speech as different from that of other children.
2. 50 per cent are happy with voice.
3. 46 per cent classified voice as a problem.
4. 66 per cent optimistic that voice would not cause problems for the future. 20 per cent felt it would cause minor problems only.
5. 80 per cent would proceed with surgery again despite the resultant voice.
6. 20 per cent would prefer not to have had surgery, i.e. remain cannulated in the hope of having a strong voice since there would be no rib graft intervention.

Seven children completed the questionnaire. Only one admitted that he had a small problem with his speech, although 4 would like to have an improved voice if possible. All the children preferred to have a weaker voice and no tracheostomy.

Observations

1. The experience of this study revealed that visual feedback is effective. We found potential for improvement in those children with aperiodic vibration.

2. That stroboscopy was not suitable for this study. The highly abnormal vibrations of the vocal fold area could not be displayed adequately, because of technical reasons. In some patients it was difficult to identify the sound source because of the masking effect of the arytenoids (see Kearns *et al.*, this supplement).

3. That this type of study is best conducted where one of the clinicians is closely acquainted with the family. It is essential that the characteristic voice of the child is recorded.

Conclusions

Sixty-nine per cent of these children have a permanent voice disorder following these rib graft procedures.

Children with acquired subglottic stenosis are at considerable risk of having communication and behaviour problems.

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