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Laryngeal pathology at school age following very preterm birth

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ABSTRACT

Introduction: Intubation injury resulting in laryngeal pathology is recognised as a possible complication of preterm birth, yet few published studies have examined such pathology and its relation to voice outcomes. This study reports on the results of prospective laryngeal function examinations of a cohort of very preterm children, all of whom presented with significant dysphonia at school age.
Materials and methods: The laryngeal pathology of 20 very preterm children, born between 23 and 29 weeks gestation, was examined under halogen and stroboscopic conditions. Laryngeal structure and function were assessed using a rigid laryngoscope or a flexible nasendoscope. The approach was selected based on the age and/or likely compliance of the child.
Results: Nineteen children were found to have structural laryngeal pathology. Fourteen children presented with a chink to the posterior glottis and all demonstrated at least a mild degree of supraglottic hyperfunction. Other common findings were arytenoid prolapse and vocal fold immobility. More isolated findings included posterior scar band, vocal fold atrophy, arytenoid oedema and growth on the vocal folds. One child who presented with structural laryngeal pathology was never intubated.
Discussion: Supraglottic hyperfunction was common to all participants, regardless of the nature and extent of underlying structural laryngeal pathology. Posterior glottic chink was the most common pattern of incomplete vocal fold closure. These data support the hypothesis that very preterm children adopt supraglottic tightening to compensate for underlying laryngeal pathology. The mechanism underlying laryngeal damage in the child who was not intubated is unclear.
Conclusions: Voice quality of very preterm children is affected by both laryngeal structure and function. A trial of behavioural voice treatment is recommended to evaluate any therapeutic response in this population.

1. Introduction

Dysphonia is defined as disruption in voice quality, and can affect individuals across the lifespan [1]. Dysphonia may arise from organic causes, or maladaptive use of the vocal mechanism, or both. Individuals with organic dysphonia may adopt maladaptive compensatory voicing strategies and while maladaptive use of the voice can result in structural changes to the larynx, both of which result in further impairments to voice quality [2,3].

Advances in imaging technology have led to increased and more accurate diagnosis of laryngeal pathology [4]. Laryngeal damage has been identified as a potential complication of endotracheal intubation following preterm birth [5]. Potential causes of structural injury include: initial placement of the endotracheal tube, movement of the tube in situ, infection and tissue growth around the tube [6]. Serious, long-term laryngeal complications of prolonged endotracheal intubation in infancy include: subglottic stenosis, acquired subglottic cysts, posterior glottic furrow, vocal fold scarring, cricoarytenoid joint fixation and traumatic vocal fold avulsion [7–12]. Moderate to severe airway abnormalities post-extubation were identified in up to 23.7% of preterm infants, and following intubation for as little as 24 h [4,12].

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Dysphonia has been reported in preterm children, and associated with frequency of intubation, female gender, birth weight, gestational age and emergency intubation [13–15]. Dysphonia in adulthood, associated with left vocal cord paralysis following surgical ligation of patent ductus arteriosus in extreme prematurity, has also been documented [13]. Children with laryngeal injury following neonatal intubation may adopt compensatory supraglottic hyperfunction, which is known to cause further laryngeal damage beyond the initial injury, with associated disturbances in voice quality [9,11]. However, the majority of studies of laryngeal pathology pertain to infants with few reports of voice quality. This is problematic for several reasons. Long-term voice outcomes cannot be extrapolated or predicted from infant cries due to physiological differences in infant larynges [16]. Fibrosis and stenosis can develop up to twelve months post-extubation and thus may not be reflected in voice quality in infancy [7]. Finally, lack of standardised reports of voice quality may result in underestimation of the true morbidity of dysphonia in this population. Whilst the correlation between laryngeal pathology and functional voice outcomes is imperfect, an understanding of the nature and extent of laryngeal pathology in preterm children will elucidate the mechanisms underlying disordered voice production in this population.

1.1. Aims and hypotheses of this study

It is hypothesised that very preterm children who undergo endotracheal intubation in the neonatal period are at high risk of long-term laryngeal injury, and that such injury will be associated with dysphonia severity. Further, it is hypothesised that very preterm children with laryngeal injury are more likely to adapt maladaptive strategies to initiate and sustain phonation, which will manifest as supraglottic hyperfunction resulting in a strained vocal quality.

The aim of the study was to determine the nature and extent of laryngeal pathology in this cohort. Prospective examination of the laryngeal structure and function of a cohort of very preterm children at school age with moderate to severe dysphonia was conducted.

2. Materials and methods

The study design and methodology has been described elsewhere and is unchanged [17]. A brief summary of participant characteristics and assessment methodologies is set out below.

2.1. Participants

Two hundred and fifty children underwent clinical voice assessment to determine the incidence, presentation and severity of dysphonia in very preterm children across two studies. Participants were recruited from a total of 1851 NICU discharges born at $\leq$32 weeks gestation and were aged between 4 years and 11 months and 15 years and 10 months at the time of initial assessment. Each study was approved by the Princess Margaret Hospital Human Research Ethics Committee. The pilot study investigated voice quality in children born at $<$25 weeks gestation. Due to the small number of children, all NICU discharges were invited to participate. The second study investigated voice outcomes in children born at $\geq$32 weeks gestation, who were stratified according to gestational age and number of intubations recorded on their NICU discharge summary. After exclusion of children with known disabilities likely to preclude successful assessment and those residing $>$200 km from the study centre, participants were randomly selected.

Across the two incidence studies, ninety four participants presented with dysphonia that was greater than mild in severity. Three were lost to contact between the study phases; 35 declined further assessment as their parents were not concerned about their child's voice quality. The families of 25 children refused nasendoscopic evaluation. The families of 31 children consented and 20 children have undergone the procedure, with an additional 2 refusals at the time of procedure and one unsuccessful attempt due to the small stature of the child.

2.2. Clinical assessments

Each participant underwent a clinical voice assessment by a speech pathologist with post-graduate experience in the assessment and treatment of paediatric voice disorders. The assessment consisted of a perceptual evaluation with the consensus auditory perceptual evaluation of voice (CAPE-V) [18], acoustic analysis of the voice signal with the acoustic voice quality index (AVQI) [19] and a caregiver-proxy quality of life report, the paediatric voice handicap index [20]. The CAPE-V is rated by a trained listener, on a visual analogue scale, where 0 represents normal voice and 90 represents severe dysphonia. A severity score of greater than 0 is considered dysphonic, with increasing score reflecting increased severity of disturbance to the voice. The AVQI is a new, objective assessment measure and is presently used for discriminating normal from dysphonic voices in children. The threshold for pathology of the AVQI in paediatric voice is 3.46, with a higher score representing greater disruption in the voice signal [21]. On the pVHI, children with normal voices score $\leq$2.

2.3. Laryngeal assessments

Participants were aged between 6 years and 6 months and 17 years and one month at the time of laryngeal examination. Oral (rigid) approach or nasal (flexible) approach was selected by the administering otorhinolaryngologist based on the age and likely compliance of the participant, in consultation with the family where appropriate. Co-phenylcaine was administered via the nares bilaterally prior to the introduction of the scope into the respiratory tract. Task instructions were administered by a speech pathologist with post-graduate experience in the assessment and treatment of paediatric voice disorders. Speech targets were elicited to facilitate completion of the Stroboscopy Evaluation Rating Form and included sustained phonation of a close-front vowel at comfortable pitch and loudness, at maximum loudness, at lowest and highest pitch, ascending and descending pitch glides, rote speech and inhalation phonation [22].

All participation was carried out under informed caregiver consent and child assent where appropriate. This research was conducted in accordance with the Code of Ethics of the World Medical Association (Declaration of Helsinki).

3. Results

Participant medical, demographic and voice characteristics are described in Table 1. Thirteen participants were female. Whilst participation in this study was voluntary, the larger number of female participants reflects the higher incidence of dysphonia found in females in this cohort [14]. Variables included were those with a demonstrated link to dysphonia following preterm birth. One participant underwent PDA ligation reflecting the rare occurrence of this procedure in very preterm infants in this region. All participants experienced enteral feeding via nasogastric tube. A comparison of characteristics of participating and non-participating children is presented in Table 2.