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Long-term quality of life in children after open airway surgery for laryngotracheal stenosis

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ABSTRACT

Objectives: The purpose of this study is to evaluate the long-term health related quality of life (HRQoL) in a cohort of children surgically treated for laryngotracheal stenosis (LTS).
Study design: Prospective cohort study.
Methods: Parents of children between 4 and 18 years at follow-up completed the Child Health Questionnaire Parent Form (CHQ-PF50). Children between 11 and 18 years at follow-up completed the Child Health Questionnaire Child Form (CHQ-CF87). Biographical and pre-operative data were extracted from the hospital records. Post-operative measurements consisted of the Bruce treadmill test and pulmonary function testing (PFT).
Results: Fifty-four parents completed the CHQ-PF50; twenty-one children completed the CHQ-CF87. The CHQ-PF50 was significantly worse than the norm population on the subscales physical functioning, role functioning: emotional/behavior, general health perceptions, family activities, parental impact: emotional, and time. CHQ-CF87 was significantly worse than the norm population on physical functioning and better on mental health. After multivariate analysis, presence of co-morbidities and glottic stenosis are the most important pre-operative factors for worse scores on general health. As post-operative measurements, the Bruce treadmill test and peak expiratory flow (PEF) correlate well with HRQoL physical subscales.
Conclusions: At long-term follow-up after treatment for LTS, deficits in HRQoL may still exist. Presence of co-morbidities and glottic stenosis are important negative factors for long-term HRQoL. The Bruce treadmill test and peak expiratory flow on pulmonary function testing correlate well with physical subscales on HRQoL. A long-term multidisciplinary follow-up with assessment of HRQoL is advised in patients treated for LTS.
Level of evidence: 2B, individual prospective cohort study.

1. Introduction

Acquired laryngotracheal stenosis (LTS) is a life-threatening complication of prolonged intubation for which a tracheostomy is often necessary. Children with laryngotracheal stenosis often have co-morbidities; congenital syndromes, bronchopulmonary dysplasia and tracheomalacia are common [1]. Open airway surgery in the form of laryngotracheal reconstruction (LTR) or cricotracheal resection (CTR) has proven its value in the treatment of LTS over the past decades. A number of studies have been published on different treatment modalities, treatment algorithms and surgical outcome [1–5]. Surgical outcome usually focuses on decannulation: the number of patients who have been successfully relieved of their tracheal cannula after surgery. In a recent study, we were the first to report on long-term functional outcome in which we objectively assess the patency of the reconstructed airway [6]. We found that glottic involvement of the stenosis and the presence of co-morbidities gave less favorable long-term functional outcome.

To our knowledge, there are no reports on health-related quality of life (HRQoL) in children treated for LTS. This is remarkable, since generic and disease specific quality of life data

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are in abundance in parents and children with other diseases with large impact and long-term deficits [7–10]. Also, the impact on everyday life of caregivers with children with a tracheostomy has been reported in various previous studies, but long-term HRQoL after successful decannulation has never been reported [11,12].

Health-related quality of life measurements give a comprehensive assessment of the patient's health status or health-related quality of life on physical, psychological and social domains and further completes clinical assessment of a patient's well-being. Generic measurements assess the patients HRQoL in a non-disease specific way and are applicable in a wide variety of population samples [13].

The aim of this study is to describe long-term HRQoL in a cohort of children successfully treated for LTS and identify additional predictors for HRQoL.

2. Material and methods

This study was part of a large follow-up study of which the methods have been previously described [6]. All children who underwent open airway surgery (LTR or CTR) in the time period of September 1994 until September 2009 for acquired, post-intubation LTS in the Erasmus Medical Center, Sophia Children's Hospital, Rotterdam were approached for this study. Only children who were without a tracheostomy and above the age of four at time of the study were included. The ethics committee approved the study (MEC-2010-298) and informed consent was obtained from all subjects.

2.1. Predictor variables: functional health status

2.1.1. Biographical characteristics and pre-operative characteristics

Biographical data comprised of age at time of follow-up and gender. Medical records were checked for age at time of surgery in years, presence of pre-operative tracheostomy, presence of co-morbidity, Cotton-Meyer grade of stenosis, glottic involvement of the stenosis and presence of congenital syndromes.

2.1.2. Medical status at follow-up

2.1.2.1. Spirometry. During the follow-up visit children performed pulmonary function testing (PFT), which included forced expiratory and inspiratory flow volume loops. The methods for these measurements are extensively described in our previous publication [6]. Results are given in continuous standard deviations or as percentages in the case of FiV1/VCmax.

2.1.2.2. Bruce treadmill test. A motor-driven treadmill test was performed according to the Bruce protocol to voluntary exhaustion while heart rate and O_2 saturation were measured in children who were able to comply with the protocol [14]. The methods for this test are extensively described in our previous publication [6]. Results are given in standard deviation (SD) scores. These SD scores are distributed as continuous data.

2.2. Outcome measure: health-related quality of life

HRQoL in children was assessed with the Child Health Questionnaire (CHQ). The Child Health Questionnaire Parent Form (CHQ-PF50, age 4–18 years) was filled out by parents about their child, and Child Health Questionnaire Child Form (CHQ-CF87, age 11–18 years) was filled out by children about themselves.

The Child Health Questionnaire Parent Form (CHQ-PF50) is a generic quality of life instrument measuring subjective health status on both physical and psychosocial domains [13]. Parents were asked to complete 50 items regarding physical and psychosocial concepts, which can be divided into 11 multi-items scales and 2 single-items questions (Family Cohesion and Change in health) (Table 2). Subscale scores range from 0 to 100, where higher scores indicate a better HRQoL.

Raat et al. described satisfactory psychometric properties (subscale Cronbach's α ranged from .39 to .96; mean .72) for the CHQ-PF50. For the CHQ-PF50 normative data were derived from a representative sample of 353 Dutch schoolchildren [13].

The Child Health Questionnaire Child Form (CHQ-CF87) is a questionnaire with 87 items regarding physical and psychosocial concepts. The CHQ-CF87 consists of 10 multi-items scales and 2 single-items questions (Family Cohesion and Change in health) (Table 2). The CHQ-PF50 and the CHQ-CF87 have comparable scales with the exception that the CHQ-PF50 contains the subscales Parental Impact: Emotional and Parental Impact: Time of the CHQ-PF50. The CHQ-PF50 subscale Role Functioning: Emotional/Behavior is a combination of the CHQ-CF87 Role Functioning: Emotional and Role Functioning: Behavior. The CHQ-CF87 has satisfactory psychometric properties (subscale Cronbach's α ranged from .56 to .90; mean .80). The CHQ-CF87 normative data contained a representative sample of 444 Dutch schoolchildren [15].

2.3. Statistical analyses

Children with complete data for medical history, present medical status, and parent-reported HRQoL were included in the statistical analyses. Comparisons between complete cases ($n = 65$) and non-complete cases ($n = 11$) for age at time of follow up and age at surgery were done using Mann–Whitney U tests. Pearson's χ^2-tests were used to test differences in distributions of gender, presence of pre-operative tracheostomy, presence of co-morbidities or congenital syndrome, Cotton-Myer grade of stenosis and glottic involvement of the stenosis.

Due to small sample sizes, comparisons between CHQ-PF50 and CHQ-CF87 were done using non-parametric Wilcoxon signed-rank tests. Comparisons between CHQ-PF50 and CHQ-CF87 means (SDs) and normative data were done with Students' t tests and Cohen's d were calculated.

A two-stage strategy was followed to test the predictive power of functional health status on each subscale of parent-reported HRQoL in multiple linear regression analyses. In phase 1, each pre-operative and post-operative functional health variable was associated with each CHQ-PF50 subscale (univariate analyses). When the association was significant ($p < .05$), the variables were forced simultaneously into a cluster analysis (i.e. pre-operative cluster and post-operative cluster). Functional health variables that were not significant ($p < .05$) in the final model were removed (backward elimination procedure), then the total explained variance (R^2) was calculated. Regression analysis was not performed on the CHQ-CF87 results due to small numbers.

To check multicollinearity, the variance inflation factor (VIF) was calculated. For each model, the average of the VIFs of the entered functional health variables was around 1, which is expedient. Continuous functional health variables (x-axis) were regressed on CHQ-PF50 scales (y-axis) in scatter plots to check the linearity assumption. The scatter plots presented no other than linear relationships for continuous variables. Statistics were conducted using SPSS version 21.0.

3. Results

3.1. Baseline characteristics and pre-operative characteristics

Between September 1994 and September 2009 a total of 80 children were treated with open airway surgery (LTR or CTR) for