

Abstract

Objectives:

When assessing paediatric dysphonia there are different approaches that can be taken in gathering a subjective view of the impact voice difficulties have on a child. Most valid questionnaires require parent proxy reporting while it has become increasingly important to gather the views of children themselves. This study reports a pilot study of an adaptation to the Pediatric Quality of Life Questionnaire (PVRQoL).

Methods:

24 parent and child dyads were recruited from a tertiary paediatric voice clinic. Children were aged between 3;08 and 15;03. Parents completed the existing PVRQoL questionnaire while their children were given a child adapted version. Follow up completion of the child questionnaire was conducted after a two week period.

Results:

There was a good correlation between the two time periods when children completed the adapted PVRQoL and also between parent and child responses. Of particular interest however was the different ratings on individual items by parents and their children with parents tending to overestimate the extent to which their children may be emotionally affected by their voice disorder.

Conclusions:

This study shows that children have much to tell about their own voice related quality of life so our conclusion is that they should be asked. The PVRQoL when adapted for use with children offers an additional insight that can be gathered in a relatively short timeframe and be considered with other assessments of vocal function.

Background

In the UK, children are normally referred to hospital ear, nose and throat (ENT) clinics by a general practitioner (GP) seeking assessment. Assessment of patients with voice disorders takes place in the voice clinic. From here, patients may undergo surgical intervention and/or speech and language therapy (SLT). SLT often takes place in the community setting. Comprehensive evaluation of voice in children can, and should, include evaluation of vocal function and the subjective impact of voice difficulties^{1 2}. A number of valid and reliable tools exist for the subjective impact of voice in adults including for example, the VOS³, VHI⁴ and VRQoL⁵ that have been further developed for the paediatric voice patient using parental-proxy report forms (the PVOS⁶, PVHI⁷ and PVRQoL⁸). Table 1 below outlines the main characteristics of these adult self-assessment questionnaires along with their parental-proxy counterparts for use in the paediatric population.

<INSERT TABLE 1 HERE>

Further detail about how the role of self-assessment in the evaluation of paediatric voice disorders has been published elsewhere by the authors². The main concerns raised in that review were that parent proxy rating scales for voice disorder in children are insufficient without associated self-assessment by the children themselves. This notion is well supported in the literature^{9 10} and has led to the recent development of a 31 item Paediatric Voice Symptom Questionnaire (PSVQ) exploring a range of voice related symptoms suitable for children aged 6 years and older and their parents¹¹. This questionnaire was designed and tested on 333 children and their parents in a test-retest design. Participants included those presenting with voice problems, and a control population and the findings contribute to the notion that parent-proxy and child self-assessment of voice impact should be continued in the paediatric voice clinic.

Parent and Child Responses to the Paediatric Voice Related Quality of Life Questionnaire

In a voice assessment clinic there is the need for a tool that is quick to administer given the multi-faceted approach to assessment. In our paediatric voice clinic, a typical consultation incorporates case history taking and direct laryngoscopic observation (where the child is amenable to this). Additional voice function analysis using perceptual and acoustic parameters is also considered depending on time. To add to this a lengthy questionnaire proves to be troublesome for parent and child alike. Thus there is the need for a valid and reliable short tool for subjective impact evaluation that can be asked of both children and their parents.

As outlined above, the VRQoL and PVRQoL are both 10 item questionnaires that provide a quick evaluation of voice related quality of life. The other suitable questionnaires appropriate for the paediatric voice clinic are either designed for a specific population (e.g. PVOS is designed to evaluate outcomes associated with otolaryngological surgery) or contain more items (e.g. PVHI has 23 and PVQS has 31) thus requiring more time to administer. This would suggest that there is scope for exploring the value of the PVRQoL in relation to both parent-proxy and child self-administration.

In the PVRQoL there are two domain scores (physical functioning and social-emotional) and a total score, with a higher value indicating higher voice related quality of life. There are six items relating to physical function domain (PF) and four items to the social-emotional domain (SE). There is well documented evidence surrounding the development and validation of the PVRQoL Error! Bookmark not defined. in addition to the sensitivity and specificity to voice related impact from parental proxy reports¹². It is unknown to what extent children report similar or different concerns when they answer the PVRQoL questions through self-assessment.

The aim of this study was to develop and validate the PVRQoL for use by children with voice disorder and their parents. The following specific research questions were defined:

Parent and Child Responses to the Paediatric Voice Related Quality of Life Questionnaire

1. Is there a correlation between the three domain scores (PF, SE and Total) derived from an adapted child version of the PVRQoL in two time conditions?
2. Is there a correlation between parent and child domain scores (PF, SE and Total) on the PVRQoL?
3. To what extent is their correspondence on parent and child PVRQoL individual item responses?

Method

Developing the Questionnaire: A series of adaptations were made to the format of the existing questions in the PVRQoL to make it presentable to children. Vocabulary simplifications replacing the terms “anxiety” and “depression” with “worried” and “sad” respectively were made following pilot evaluation with two children aged 10 and 6 years following discussion with the first author about what each question meant. In these discussions, the two children suggested simpler words that would give a similar interpretation. The numbered likert rating scale which represents the extent to which a particular question response is “not a problem” to “problem is as bad as it can be” was replaced with a series of faces with “not a problem” indicated by an extremely happy face to “problem is as bad as it can be” indicated by a sad face with tears, with a range of pictorial graded differences in between.

Permissions: Ethical approval was granted by the West of Scotland Research Ethics Service and management approval for the study granted by the health board which hosts a dedicated monthly paediatric voice clinic service.

Participants: All patients over the age of 3 attending the paediatric voice clinic during a 6 month period were invited to participate in the study. During this period, 24 parent and child dyads were recruited. All children were subsequently diagnosed with dysphonia (18 had vocal fold nodules, 3

Parent and Child Responses to the Paediatric Voice Related Quality of Life Questionnaire

had muscle tension dysphonia, 2 had vocal fold cysts and 1 had previously had laryngotracheal reconstruction surgery). There were 15 boys aged 3;08 – 12;03 and 9 girls aged 6;09 – 15;03.

Procedures: Each dyad completed the parent and child versions of the questionnaires independently of each other at the time of their routing clinic appointment. Children were supported when completing the questionnaire by the first author (referred to as 'time 1'). Support given to the children involved reading aloud each question to the child and asking them to either circle or colour in the corresponding face that best represented their response. A further 19 children completed the child questionnaire during a follow up phone call that was arranged to take place two weeks after the initial clinic appointment ('time 2'). Doing this by phone reduced additional travel time associated with the repeat questionnaire so as to avoid unnecessary additional burden on participating families. Parents were not required to repeat the questionnaire at 'time 2'.

All follow up phone calls were pre arranged and conducted by the first author. Each child had a blank copy of the questionnaire at home to assist in the phone call consultation. Following a short general conversation, the child was asked if he or she recalled completing the questionnaire at the clinic and if they had the new one at home in front of them. Each question was read aloud by the first author verbatim along with a description of each of the smiling faces. Each child was asked to say which of the smiling faces they felt answered the question for them on that day.

Statistical analysis: The numerical domain scores were calculated for each questionnaire.

Correlation between the domain scores for the two time periods was made using MedCalc (Version 13.0.2). Intraclass correlation coefficient (ICC) was calculated for the domain scores in the time 1 and time 2 child questionnaires. Concordance correlation coefficient (CCC) was calculated to compare the domain scores in the parent and child (time 1) questionnaires. Further analysis using

weighted Kappa was undertaken to compare the categorical rating for each of the ten items in the parent/child comparison.

Results

Descriptive information regarding the scores yielded from the preliminary analysis is shown in table 2 below. Although a small sample size, the dataset was split into age bandings in order to compare scores by age group using ANOVA. No significant difference was observed in any of the domain scores. Further, comparison of the domain scores by gender also yielded no significant difference. There was no association of domain scores by age or gender.

<INSERT TABLE 2 HERE>

Graphs illustrating the three domain scores from the time 1 vs time 2 and the parent vs child comparisons are shown in figures 1 and 2 below. ICC for the child time 1 and time 2 domain scores ratings were: SE $r = 0.85$, PF $r = 0.77$, and Total $r = 0.87$. CCC for the parent and child domain scores were: PF $r = 0.67$, SE $r = 0.72$ and Total $r = 0.76$.

<INSERT FIGURE 1 HERE>

<INSERT FIGURE 2 HERE>

Individual item analysis using the weighted Kappa calculation was completed on each dyad's ratings, the results of which are shown in table 3. While overall parent/child agreement was found in the totalled domain scores, the extent to which there was agreement on the individual items was more varied, and this was more so in all of the PF items and in one of the SE items. Close inspection of the individual responses to each of the items revealed that parents tended to rate voice impact lower

than their child except on two questions: “...sometimes not know what will come out when begins speaking” and “..sometimes gets depressed [sad] (because of voice)” where the child rating was lower than their parent’s corresponding rating.

<INSERT TABLE 3 HERE>

Discussion

This pilot study has shown that there is a good correlation over time when children complete the adapted PVRQoL. Of particular interest is the difference in individual item ratings between parents and children. The parents and their children in our study had different interpretations on aspects of physical functioning impact associated with voice difficulties and differences in understanding and/or opinion about emotions relating to mood. Parents had a tendency to overestimate the extent to which their children may be emotionally affected by their voice disorder. It is however noted that there is no way of evaluating if the vocabulary simplifications may have impacted on these findings. That there was poorer agreement in the ratings for the PF domain items than the SE domain items conflicts with that in the general health related QoL literature, where there tends to be a greater agreement in aspects of physical function over social-emotional function¹³. It might be surmised that questionnaires like the VRQoL are so specialised in relation to the impact of voice difficulties that it can be difficult to correspond its PF items with general physical functioning items in other health related questionnaires. Exploratory analysis of the small corpus of data did not reveal any significant difference in scores in relation to age or gender, and this may be limited as a result of sample size.

It is currently recognised in the health-related QoL literature, that low parent-child agreement reduces as children mature¹⁴. Notwithstanding this, it is possible that self-assessment of severity may be impacted by the child’s own perception of their vocal function - it is feasible that younger

children will have a less sophisticated self- perception of their own voice quality than older children. Further investigation of this is warranted so that clinicians can have a greater understanding of what matters to children at different ages and provide treatment that is tailored according to their different social and educational experiences at the time of intervention.

This was a small scale study and is worthy of extension and replication to further enhance its findings. Specifically it would be worthy to evaluate whether or not completion of this type of self- assessment by children and their parents pre and post treatment can contribute to intervention outcome measures. During the design phase of the study several means of gathering the time 1 and time 2 data were explored. In planning this study, the authors were cognisant that children who were attending the clinic, some with extensive travel, should not be asked to attend unnecessarily for the purposes only of developing a clinical tool. While it would have been more robust to collect time 1 and time 2 questionnaires both face to face, over two clear time periods before the initial voice evaluation appointment, it was not possible to do this within the constraints of current patient care delivery, and ethical considerations relating to this were paramount.

Conclusion

This study shows that children have much to tell about their own voice related quality of life so our conclusion is that they should be asked. The PVRQoL when adapted for use with children offers an additional insight that can be gathered in a relatively short timeframe and be considered with other assessments of vocal function. The PVRQoL may have value as an outcome indicator and our recommendation is that further investigation be undertaken to explore its sensitivity and specificity in relation to therapy outcome measures.

Acknowledgements

The authors are grateful for the willing participation of all of the children and their parents for in this pilot study.

References

- ¹ Cohen W, Wynne DM, Kubba H, & McCartney E. Development of a minimum protocol for assessment in the pediatric voice clinic: Part 1: Evaluating vocal function. *Logopedics Phoniatrics Vocology*. 2012;37(1): 33-38.
- ² Cohen W, Wardrop A, Wynne DM, Kubba H. & McCartney, E. Development of a minimum protocol for assessment in the pediatric voice clinic: Part 2: subjective measurement of symptoms of voice disorder. *Logopedics Phoniatrics Vocology*. 2012;37(1):39-44.
- ³ Gliklich RE, Glovsky RM, Montgomery WW. Validation of a Voice Outcome Survey for unilateral vocal fold paralysis. *Arch Otolaryngol Head Neck Surg*. 1999;120:153-8.
- ⁴ Jacobson B, Johnson A, and Grywalsky C. The Voice Handicap Index (VHI): development and validation. *Am J Speech Lang Path*. 1997;6:66-70.
- ⁵ Hogikyan ND and Sethuraman G. Validation of an instrument to measure voice-related quality of life (V-RQOL) *Journal of Voice*. 1998;13:557-569
- ⁶ Hartnick CJ. Validation of a pediatric voice quality of life instrument: the pediatric voice outcome survey. *Arch Otolaryngol Head Neck Surg*. 2002;128:919-922.
- ⁷ Zur KB, Cotton S, Kelchner L, Baker S, Weinrich B, Lee L. Pediatric Voice Handicap Index (pVHI): a new tool for evaluating paediatric dysphonia. *Int J Pediatr Otorhinolaryngol*. 2007;71:77-82.
- ⁸ Bosely ME, Cunningham MJ, Volk MS, Hartnick CJ. Validation of the Pediatric Voice-Related Quality-of-Life Survey. *Arch Otolaryngol Head Neck Surg*. 2006;132:717-720.
- ⁹ Connor, N.P., Cohen, S.B., Theis, S.M., Thibeault, S.L., Heatley, D.G. and Bless, D.M. Attitudes of children with dysphonia. *Journal of Voice*. 2006;22:197-209.
- ¹⁰ Verduyckt, I., Remacle, M., Jamart, J., Benderitter, C. and Morsomme, D. Voice related complaints in the pediatric population *Journal of Voice*. 2011;25(3):373-380.
- ¹¹ Verduyckt, I., Morsomme, D. and Remacle, M. Validation and standardization of the Pediatric Voice Symptom Questionnaire for dysphonic children and their parents. *Journal of Voice*. 2011;25(3):373-380.
- ¹² Merati AL, Keppel K, Braun NM, Blumin JH, Kerschner JE. Pediatric voice-related quality of life: findings in healthy children and in common laryngeal disorders. *Ann Otolaryngol Rhinol Laryngol*. 2008;117:25 9-62.
- ¹³ Eiser C and Morse R Can parents rate their child's health-related quality of life? Results of a systematic review. *Quality of Life Research*. 2001;10:347-357.
- ¹⁴ Ramil, L, Rodríguez López, A., López-Aguilà, S. and Alonso J. Parent-child agreement on health-related quality of life (HRQOL): a longitudinal study. *Health and Quality of Life Outcomes*. 2013;11:101.

Parent and Child Responses to the Paediatric Voice Related Quality of Life Questionnaire

	Title	Citation	Brief description
Adult	Voice Symptom Scale (VOS)	Gliklich RE, Glovsky RM, Montgomery WW. Validation of a Voice Outcome Survey for unilateral vocal cord paralysis. <i>Arch Otolaryngol Head Neck Surg.</i> 1999;120(2):153-158.	Validated in relation to outcome measures associated with unilateral vocal cord paralysis. 5 questions, 5 point rating scale Validated on 56 patients with UVCP against generic medical outcomes rating scale and evaluation of vocal function.
	Voice Handicap Index (VHI)	Jacobson BH, Johnson A, Grywalski C, Silbergleit A, Jacobson G, Benninger MS, Newman CW. The Voice Handicap Index (VHI) development and validation. <i>Am J Speech Lang Pathol.</i> 1997;6(3):66-70.	Designed to evaluate the psychosocial impact of voice disorder across three domains – functional, physical and emotional. 30 questions, 5 point rating scale. Validated on 63 voice patients in test-retest conditions. Also available as short form in the VHI 10 (Rosen CA, Lee AS, Osborne J, Zullo T, Murry T. Development and validation of the Voice Handicap Index-10. <i>The Laryngoscope</i> 2004;114(9):1549-1556.)
	Voice Related Quality of Life (VRQoL)	Hogikyan ND, Sethuraman G. Validation of an instrument to measure Voice-Related Quality of Life (V-RQOL). <i>J of Voice.</i> 1999;13(4):557-569	Designed to measure voice related quality of life in two domains – social emotional and physical functioning. 10 questions, 5 point rating scale. Validated on 109 voice patients and 22 controls.
Parent-proxy	Pediatric Voice Symptom Scale (VOS)	Hartnick CJ. Validation of a pediatric voice quality of life instrument: the pediatric voice outcome survey. <i>Arch Otolaryngol Head Neck Surg.</i> 2002;128:919-922.	Validated in relation to surgical outcomes associated with pediatric otolaryngological problems Uses same 5 questions and 5 point rating scale from VOS, adapted for parent proxy reporting. Validated on 108 parents of children who had received surgical intervention associated with otolaryngological problems.
	Pediatric Voice Handicap Index (VHI)	Zur KB, Cotton S, <u>Kelchner L</u> , <u>Baker S</u> , <u>Weinrich B</u> , <u>Lee L</u> . Pediatric Voice Handicap Index (pVHI): a new tool for evaluating paediatric dysphonia. <i>Int J Pediatr Otorhinolaryngol.</i> 2007;71:77-82.	Adaptation of the VHI for parental proxy reporting. 23 questions , 5 point rating scale Validated on a group of parents of children with otolaryngological problems and a control group.
	Pediatric Voice Related Quality of Life (PVRQoL)	Bosely ME, Cunningham MJ, Volk MS, Hartnick CJ. Validation of the Pediatric Voice-Related Quality-of-Life Survey. <i>Arch Otolaryngol Head Neck Surg.</i> 2006;132:717-720	Validated in relation to a range of otolaryngological disorders including voice problems Uses same 10 item questionnaire and 5 point rating scale as VRQoL with wording adapted for parent proxy reporting. Validated with 120 parents of children with otolaryngological problems.

Table 1 Overview of the most commonly used adult self-assessment questionnaires in the voice clinic and their parent-proxy counterparts

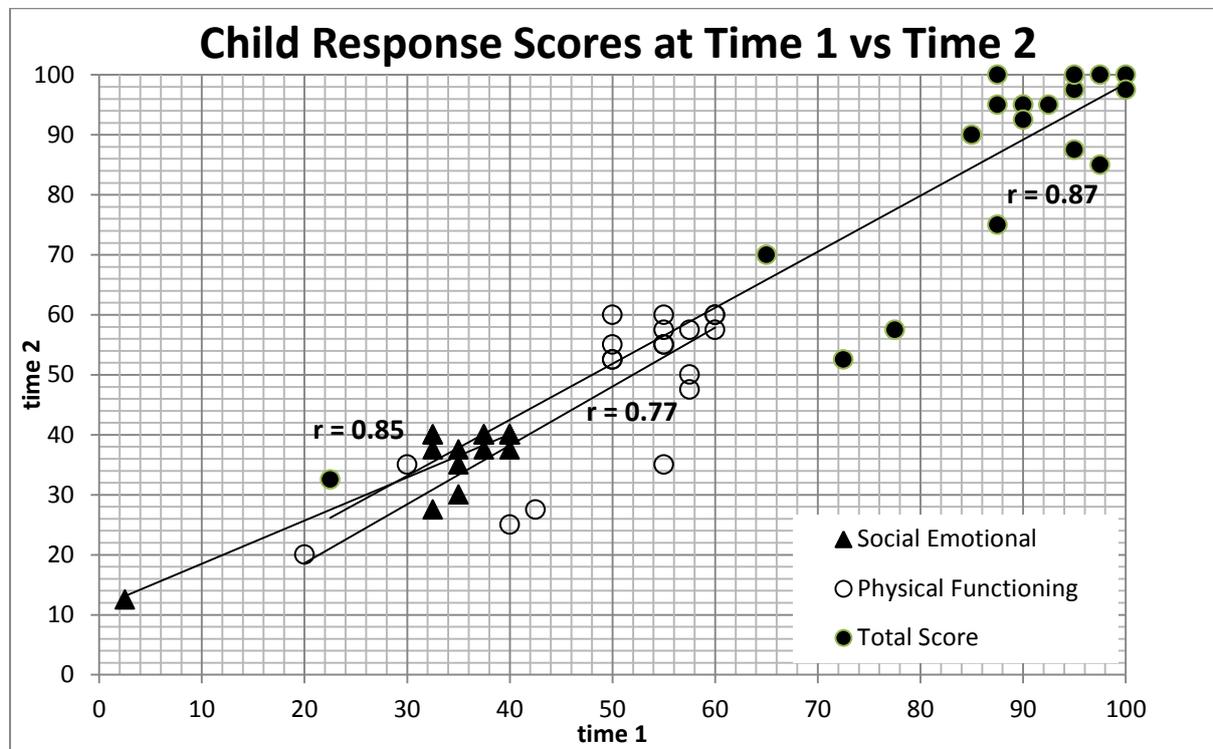


Figure 1 Scatterplot showing scores on the three domains derived from the responses from the children at time 1 and time 2

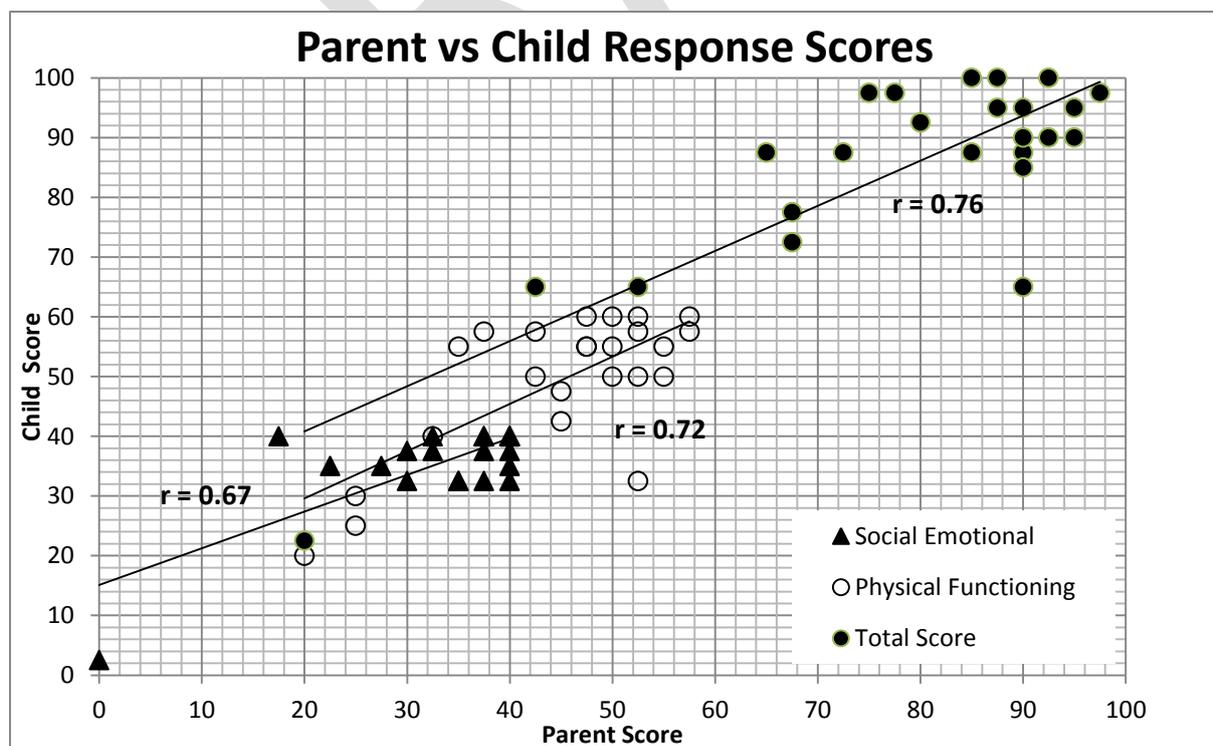


Figure 2 Scatterplot showing scores on the three domains derived from the parent and child responses

Domain	Value	Girls Age range: 6;09 - 15;03			Boys Age range: 3;08 - 12;03		
		Parent n = 9	Child Time1 n = 9	Child Time2 n = 7	Parent n = 15	Child Time1 n = 15	Child Time2 n = 12
Social Emotional	\bar{x}	29.44	33.89	34.29	36.33	37.00	37.92
	sd	13.85	12.06	10.28	3.99	3.30	3.67
	range	0 – 40	2.5 – 40	12.5 – 40	27.5 – 40	32.5 – 40	27.5 – 40
Physical Functioning	\bar{x}	44.72	45.28	48.21	45.00	51.67	48.75
	sd	13.89	14.00	16.94	8.61	9.76	11.21
	range	20 – 57.5	20 – 60	20 – 60	25 – 52.5	32.5 – 60	25 – 60
Total Score	\bar{x}	74.17	79.17	82.50	81.33	88.67	86.67
	sd	27.04	23.39	26.69	11.76	12.06	14.28
	range	20 – 95	22.5 – 97.5	32.5 - 100	52.5 – 92.5	65– 100	52.5 – 100

Table 2. Mean, sd and range of domain scores from the parent and child time 1 and child time 2 data by gender

Question [wording in square brackets indicates the vocabulary change for the child version of the questionnaire]	Domain category	Parent / child comparison n = 24
<i>...sometimes not know what will come out when begins speaking</i>	PF	0.209
<i>...trouble speaking loudly or being heard in noisy situations</i>	PF	0.320
<i>...runs out of air and needs to take frequent breaths when talking</i>	PF	0.333
<i>..trouble using the telephone or speaking with friends in person</i>	PF	0.353
<i>..has to repeat....to be understood</i>	PF	0.365
<i>..trouble doing schoolwork (because of voice)</i>	PF	0.377
<i>..sometimes anxious [worried] or frustrated (because of voice)</i>	SE	0.172
<i>...avoids going out socially (because of voice)</i>	SE	0.455
<i>..sometimes gets depressed [sad] (because of voice)</i>	SE	0.541

Parent and Child Responses to the Paediatric Voice Related Quality of Life Questionnaire

<i>...become less outgoing (because of voice)</i>	SE	0.574
---	----	-------

Table 3. Weighted Kappa analysis of the parent and child ratings for each PVRQoL item

DRAFT