

Title page

Cross-sectional follow up of voice outcomes in children who have a history of airway reconstruction surgery.

Wendy Cohen PhD BSc Hons PG Cert

University of Strathclyde, School of Psychological Sciences and Health, 40 George Street, Glasgow, G1 1QE. Tel +44 141 548 3793. Email:

wendy.cohen@strath.ac.uk

David M Wynne MB,ChB, MRCS(Glasg), FRCS(ORL-HNS), PgDip

Royal Hospital for Children, NHS Greater Glasgow and Clyde

Susan Lloyd MA Hons, MSc

University of Strathclyde

Richard B Townsley BSc MBBS DOHNS FRCS(ORL-HNS)

Crosshouse Hospital, NHS Ayrshire and Arran

Grant funding

This project was supported by generous grants from Action Medical Research, The Hugh Fraser Foundation and Jeffrey Charitable Trust.

Abstract

Objectives

This study reports vocal function in a cross-section of children with subglottic stenosis. Each child had a history of laryngotracheal reconstruction and/or cricotracheal resection surgery. Vocal function was measured using laryngoscopy, acoustic analysis, perceptual evaluation and impact of voice on quality of life.

Design

All patients aged >5 years with history of laryngotracheal reconstruction and/or cricotracheal resection surgery at the Scottish National Complex Airways service were invited to participate.

Setting

Data was gathered in the Royal Hospital for Children in Glasgow in a single out-patient appointment.

Participants

Twelve out of fifty-six former patients (aged 5 – 27) provided a voice sample and eleven consented to awake laryngoscopy. All consented for detailed evaluation of their medical records.

Main outcome measures

Acoustic analysis of fundamental frequency and pitch perturbation was conducted on sustained vowel [a]. Perceptual evaluation was conducted by four trained listeners on a series of spoken sentences. Impact on quality of life was measured using the Paediatric Voice Related Quality of Life questionnaire. Laryngeal function was descriptively evaluated.

Results

Four children had normal voice acoustically, perceptually and in relation to voice related quality of life. One of these had vocal fold nodules unrelated to surgical history. Two other children had 'near normal' vocal function, defined where most voice measurements fell within the normal range.

Conclusions

Normal or 'near normal' voice is a possible outcome for children who have had this surgery. Where there is an ongoing complex medical condition, voice outcome may be poorer.

Keywords: children, laryngotracheal reconstruction, cricotracheal resection, subglottic stenosis, voice quality

Introduction

Background

Airway narrowing from subglottic stenosis (SGS) may be congenital or acquired following intubation or neonatal laryngotracheal injury¹. Although uncommon, (incidence <0.63%²), intervention establishes an airway through bypassing the obstruction (tracheostomy) or reconstructive surgery to expand or excise stenosis³. Two open surgical techniques exist: laryngotracheal reconstruction (LTR) and partial cricotracheal resection (CTR)⁴. Primary surgical outcomes are survival or decannulation.

In adults, voice quality is reduced following LTR or CTR, particularly in women⁵. In children, voice outcome may be poor^{6,7} or good^{8,9}, with good voice related quality of life (QoL) despite persisting levels of hoarseness¹⁰. Voice quality is largely better where the resulting phonation is glottic rather than supraglottic⁷.

Voice outcome varies according to surgical procedures or how voice is evaluated⁷, with for example different tools for evaluating QoL and different techniques for evaluating voice quality. Published studies have a range of participant numbers: larger studies tend to be retrospective caseload analyses. Clinical 'good practice' proposes a range of voice parameters are evaluated in adults¹¹ and children¹².

Studies do not always encompass all parameters, with debate about the relationship between them. For example there is a weak-fair correlation between perceptual evaluation of voice quality and voice-related QoL¹³.

A challenge in reporting voice outcome in children is the comparison voice. In adults, within-subject design allows for pre and post-surgery comparison. As the age at which most children have this surgery (usually infancy) comparison is made with the

typical population, as in other aetiologies such as recurrent respiratory papillomatosis¹⁴. Few normative studies of children's voice quality exist, with only one published since 2010 providing normative data for English speaking children aged up to 18years¹⁵.

LTR and CTR procedures have been undertaken in Glasgow since the 1980s. A retrospective audit of parents' perspectives on their child's quality of life highlighted concerns about breathing, respiratory tract infections and voice quality; anxieties about independence and ability to lead a normal life¹⁶.

The aim of this study was to evaluate voice in children who were >5 years who had had LTR/ CTR.

Objectives

1. What are the laryngeal vibratory mechanisms used when producing voice observed using video nasendoscopy?
2. How do the acoustic features of voice recordings compare to a published normative acoustic dataset for an English speaking population¹⁵?
3. What is the voice quality when measured using an established perceptual descriptor (GRBAS¹⁷)?
4. What are parents' and children's perspectives on voice related QoL using established questionnaires (PVRQoL Parent¹⁸ and Child¹⁹)?

Methods

Ethical permissions

Permission for the study was granted by the National Health Service West of Scotland Research Ethics Committee and the University of Strathclyde Research

Ethics Committee. Information sheets and consent forms were specifically designed so they were appropriate for younger children using pictures to describe the study, allowing for written consent to be obtained from both the child and their parent/guardian.

Study Design

Using a cross-sectional design, participants were recruited from the Scottish National Complex Airways Management Service. All surviving children aged >5 years ($n=56$) who had undergone LTR/CTR were invited to take part through postal information leaflets.

Setting

Participants attended a single out-patient appointment at the Royal Hospital for Children in Glasgow. Each appointment incorporated laryngeal evaluation, voice recording and completion of the PVRQoL.

Participants

Fifty-six past patients who had a history of SGS and LTR were invited to participate. All had laryngofissure, with neonates tending to have rib grafts and older patients thyroid alar graft. Sixteen opted-in to the study: twelve attended, one withdrew and three failed to attend. NHS permissions were conditional on potential participants being contacted once and invited to 'opt-in' to the study.

Variables

Awake nasendoscopy was digitally recorded in eleven children using a STORZ CCD Video-Rhino-Laryngoscope model 11101VP. All clinical cases are routinely evaluated using a high definition 'chip-in-tip' nasendoscopy system, as transoral rigid stroboscopy is not as well tolerated as transnasendoscopy in our population. Audio

recordings followed a standard protocol including four attempts at the sustained vowel sound [a] for 3-5 seconds, six sentences from the CAPE-V²⁰ and a sample of spontaneous speech. Recordings were made in a sound-treated room tested for ambient noise reduction to 14dB using a Tascam DR-05 Version 2 Dictaphone Linear PCM Portable Recorder. The inbuilt stereo condenser microphone was used with a mouth-to-microphone distance of 21cm. Raw recordings were edited using Audacity 1.2 (Audacity Team, audacity.sourceforge.net) and exported as mp3 files (codec: mpga; channels: mono; sample rate: 16 kHz; bit rate: 128 kB/s.). Medical records were reviewed for medical and surgical histories.

Measurement / Statistical methods

Descriptive analysis of awake laryngoscopy followed the laryngeal function aspects of the CCHMC Endoscopy/Stroboscopy Rating Form²¹. Two otolaryngology surgeons independently rated the recorded images with follow-up discussion and consensus agreement where possible. This provided a systematic observation of phonatory function in relation to overall vibratory source, glottic closure, supraglottic activity, vocal fold and arytenoid mobility.

Acoustic analysis of the sustained vowel was evaluated using the Multi-Dimensional Voice Program Model 5105 software option for the Computerized Speech Laboratory Model 4500 (KayPENTAX). Measurement of fundamental frequency (F_0), jitter percentage (*jitt%*), shimmer percentage (*shimm%*) and noise to harmonic ratio (*NHR*) was made from the middle 3.5s of the voiced segment produced in the fourth recording of the sustained vowel. Acoustic values for each participant were compared with the age/gender matched mean and upper/lower range in the normative dataset¹⁵ (for participants >18years comparison was made with the eldest available age range).

Recordings were coded, duplicated, randomised and played to a panel of four consensus-trained listeners. Ratings were conducted using the GRBAS¹⁷ protocol rating Grade, Roughness, Breathiness, Aesthenia and Strain as 0=normal voice, 1=mild dysphonia, 2= moderate dysphonia, 3 = severe dysphonia. There was significant intra-rater reliability using Minitab's Attribute Agreement Analysis: Kendall's Coefficient of Concordance ranged intra-rater reliability between $W=0.88$ and 0.93 ($p<0.001$) and inter-rater reliability at $W=0.74$ ($p<0.001$).

Scores from the parent and child responses to the PVRQoL^{18,19} were calculated and compared to norms, where typical children score 90-100¹⁵.

Results

Participants

Table 1 summarises the medical and surgical histories, including age at time(s) of surgery and at time of participation. Medical histories were varied: six were premature, three had syndromes with medical comorbidities, seven had neonatal respiratory problems requiring ventilation and/or tracheostomy. One participant was ventilated as an adolescent, developing SGS. Participant #12 was the only child who had both CTR and LTR. The remaining eleven had LTR only. Six presented with larygologically complexity at the time of involvement in the study such as chronic cardiac conditions ($n=2$); chronic reflux condition ($n=1$); in-situ tracheostomy ($n=1$) and a surgically damaged vocal fold ($n=1$).

<insert Table 1 here>

Descriptive data

Table 2 provides descriptors of laryngeal function. Agreement occurred between both otolaryngologists in seven cases with minor differences in four cases for supraglottic activity. Two children had bilateral vocal fold nodules.

<insert Table 2 here>

Outcome data

The acoustic data is presented in table 3, showing each participant's F_0 , *jitt%*, *shimm%* and *NHR* value in comparison to age/gender matched norms.

<insert Table 3 here>

As shown in figure 1, eight participants had F_0 within the normal range (1a), seven with *jitt%* within the normal range (1b), seven with *shimm%* in the normal range (1c) and seven with *NHR* within the normal range (1d). Six (#4,6,8,9,10,11) had all acoustic measures relevant for voice quality (*jitt%*, *shimm%* and *NHR*) within the normal range, and four (#8,9,10,11) had all acoustic values within the normal range.

<insert Figure 1 here>

Main results

Table 4 summarises combined findings of the acoustic analysis, the perceptual evaluation and PVRQoL scores. The acoustic data is coded as '+' indicating 'within normal range' and '-' indicating 'out-with normal range'. The modal GRBAS rating is given along with parent and child PVRQoL values.

When all of the acoustic data lies within the normal range (#8,9,10,11) this was perceived as either 'normal' or 'mildly dysphonic' by the listeners. This is supported by at least one parent or child PVRQoL score in the normal range.

<insert Table 4 here>

Four participants presented with acoustic and perceptual data within the normal range, with minimal parental or child impact rating. The participants with vocal fold nodules tended to have acoustic and perceptual data within the normal range: one had 3/4 acoustic parameters within the normal range and perceptually identified as normal/mildly dysphonic. PVRQoL was lower, with a parent score of 40 and a child score of 62.5 showing a voice related impact on QoL. The other had normal acoustic voice parameters, a normal/mild perceptual rating of voice quality, no recognised impact in the parental PVRQoL and a mild impact in the child PVRQoL indicating minimal impact despite vocal fold nodules.

Discussion

Key results

This study gathered information about vocal function using a recommended adults¹¹ and child¹² protocol. The vibratory source in all cases was the true vocal folds. Two children had vocal fold nodules, not related to their history of LTR (each presented with risk factors²² for vocal fold nodules).

Acoustic analysis provides an objective measurement of voice quality¹¹. There are a variety of techniques for measuring the acoustic signal, though there remains debate about which is most useful²³. The aim of this study was to make a comparison with an existing published dataset¹⁵. Our findings concur with previous research that there is potential for normal voice, both acoustically and subjectively in the longer term following LTR, particularly where there is evidence of glottic phonation and relatively intact laryngeal structures⁷. Our two participants with VFN had not previously indicated concern about voice quality. Perhaps unconcern about voice quality relates to families' expectations given the LTR history.

Limitations

Though a limitation, small sample size is similar to other published prospective studies. Ethical requirements were that our participants opted to take part: it is possible that for those who did *not* opt-in the parents and/or child have no current concerns about voice.

There are other approaches to acoustic analysis (e.g. cepstral peak prominence²⁴), although no published datasets exist from a paediatric population using this technique. Alternative approaches exist for perceptual evaluation such as the CAPE-V²⁰, however GRBAS is in common clinical use in the UK. Further analysis of the data using different acoustic and perceptual analysis techniques is proposed.

Interpretation

One of the challenges in drawing conclusions about voice outcome for children who have had LTR/CTR following SGS is evaluating vocal function. Voice quality when evaluated using a multi-parametric protocol was found to be normal or 'near normal' in half of our participants. Those with ongoing complex medical conditions have compromised voice quality and voice outcome may be a lesser concern for these individuals. Where there is a good long term medical prognosis, irrespective of aetiology, this has translated to a good prognosis for voice quality in our population.

Generalisability

Evaluation of laryngeal function alone does not always identify those children for whom voice therapy is warranted, supporting the rationale for a multi-dimensional approach to voice evaluation in the paediatric population¹². Furthermore, we propose that in this clinical population, the multifactorial definition of 'good voice outcome' does not necessarily require all voice parameters rated as 'normal'. This

would corroborate findings of previous research¹³. Long term impact on voice quality arises only for those participants where there *remain* complex medical conditions. Further exploration and replication of these findings would support that assertion.

Other information

Funding

This project was supported by generous grants from Action Medical Research, The Hugh Fraser Foundation and Jeffrey Charitable Trust.

ID #	Gender	Age(s) at surgery	Medical and surgical information	Age at time of study (years)
1	F	Neonate	Born at term. 22q11DS, cardiac surgery and LTR during neonatal period. Continues to attend for cardiology.	5
2	F	12 months	Pre-term 33 weeks. Ventilated in neonatal period. SGS. LTR at 1 year. Chronic reflux treated medically	6
3	F	Neonate – 1 year	Pre-term 27 weeks, cardiac surgery, global developmental delay, SGS. Tracheostomy as a neonate, decannulated and LTR by 12 months. Continues to attend for cardiology.	6
4	F	2-3 years	Pre-term 25 weeks, SGS, neonatal cardiac surgery now discharged from cardiology. Tracheostomy followed by LTR and decannulation	12
5	F	16 years	Emergency admission and ventilation followed by LTR.	27
6	M	6 months	Pre-term 28 weeks. Ventilated during neonatal period. SGS. LTR.	6
7	M	Neonate – 2 years	Pre-term 24 weeks. SGS. Neonatal tracheostomy. LTR at age 2 with a revision within one month. Tracheostomy in situ at time of study.	6
8	M	Neonate – 2 years	Born at term. Pierre Robin sequence, cleft palate. Tracheostomy during neonatal period. LTR and decannulated at 2 years.	6
9	M	3 years	Born at term. Laryngeal cleft and tracheo-oesophageal fissure. LTR.	9
10	M	1 year	Born at term. Primary laryngeal atresia with glottic web and SGS. LTR.	11
11	M	1 year	Pre-term 32 weeks. SGS. LTR.	13
12	M	Neonate – 5 years	SGS. Tracheostomy in neonatal period. Decannulated and CTR at 2 years; residual stenosis LTR at 5 years. Atrophy of R vocal fold.	14

Table 1 – Biographical and surgical information of the study participants.

	2	3	4	5	6	7	8	9	10	11	12
Glottic Closure	Irregular	Complete	Anterior Gap	Complete	Complete	Cannot rate	Complete	Posterior Gap	Complete	Complete	Incomplete
Supraglottic Activity – Compression Pattern	Mixed	None	None	Anterior-Post	None	Lateral	Anterior-post	Mixed	Lateral	None	Lateral
			Anterior-post			Mixed			Anterior-post		Mixed
Supraglottic Activity - Degree	Moderate	None	None	Moderate	None	Severe	Moderate	Mild	Moderate	None	Moderate
			Mild								
Vertical Level Approximation	Level	Level	Level	Level	Level	Cannot rate	Level	Level	Level	Level	Off level
Vocal Fold Edge – Left	Irregular	Straight	Irregular	Straight	Lesion	Straight	Straight	Straight	Straight	Lesion	Straight
Vocal Fold Edge – Right	Irregular	Straight	Straight	Straight	Lesion	Cannot rate	Straight	Straight	Straight	Lesion	Scarring
Arytenoid Mobility – Left	Normal	Normal	Normal	Normal	Normal	Restricted	Normal	Normal	Normal	Normal	Normal
Arytenoid Mobility – Right	Normal	Normal	Normal	Normal	Normal	Restricted	Normal	Normal	Normal	Normal	Restricted
Vibration source	True vocal folds	True vocal folds	True vocal folds	True vocal folds	True vocal folds	True vocal folds	True vocal folds	True vocal folds	True vocal folds	True vocal folds	True vocal folds

Table 2 – Laryngeal function ratings following CCHMC rating form. Agreed ratings are given. Where there is any disagreement between the two otolaryngologists both ratings are provided.

Participant No.	<i>F0</i> (Hz)			<i>Jitt%</i>			<i>Shimm%</i>			<i>NHR</i>		
	Study value	Maturo et al ¹⁵ dataset		Study value	Maturo et al ¹⁵ dataset		Study value	Maturo et al ¹⁵ dataset		Study value	Maturo et al ¹⁵ dataset	
		Mean (SD)	Range		Mean (SD)	Range		Mean (SD)	Range		Mean (SD)	Range
1	248.45	263 (23.86)	220-306	5.846	1.18 (0.89)	0.43-3.59	12.24	3.11 (1.24)	1.1–5.17	0.337	0.11 (0.02)	0.07-0.14
2	208.85	277 (27.54)	229-320	7.12	1.68 (1.02)	0.31-3.33	9.69	2.69 (1.11)	1.05-4.68	0.26	0.11 (0.02)	0.07-0.13
3	260.454	277(27.53)	229-320	2.244	1.68 (1.02)	0.31-3.33	16.562	2.69 (1.11)	1.05-4.68	0.306	0.11 (0.02)	0.07-0.13
4	297.14	243 (19.7)	202-275	2.56	1.03 (0.83)	0.39-2.85	5.50	2.53 (1.32)	1.14–5.64	0.14	0.12 (0.03)	0.08-0.17
5	237.365	235 (28.67)	195-267	2.806	1.16 (0.72)	0.35-2.37	4.075	2.47 (1)	1.47–4.81	0.158	0.09 (0.03)	0.05-0.15
6	400.622	251 (33.96)	197-295	1.396	2.24 (0.73)	0.56-5.3	3.831	4.66 (2.02)	2.44-9.02	0.169	0.13 (0.04)	0.1-0.27
7	175.50	251 (33.96)	197-295	6.79	2.24 (0.73)	0.56-5.3	21.71	4.66 (2.02)	2.44-9.02	0.66	0.13 (0.04)	0.1-0.27
8	294.706	251 (33.96)	197-295	2.777	2.24 (0.73)	0.56-5.3	6.6	4.66 (2.02)	2.44-9.02	0.149	0.13 (0.04)	0.1-0.27
9	214.989	243 (21.73)	211-278	1.667	1.57 (0.86)	1.42-2.96	4.129	2.68 (0.98)	1.31-4.79	0.136	0.11 (0.01)	0.08-0.13
10	261.463	217 (22.41)	188-271	1.255	1.07 (0.49)	0.3-2.08	3.808	2.5 (0.97)	1.38-4.22	0.119	0.11 (0.01)	0.07-0.14
11	214.14	177 (51.3)	103-268	1.68	1.72 (0.97)	0.26-4.76	5.73	4.22 (3.97)	1.56-14.22	0.13	0.14 (0.05)	0.07-0.24
12	147.23	176 (33.5)	120-224	4.80	0.97 (0.63)	0.44-2.18	14.54	2.38 (0.77)	1.45-3.97	0.38	0.14 (0.02)	0.07-0.24

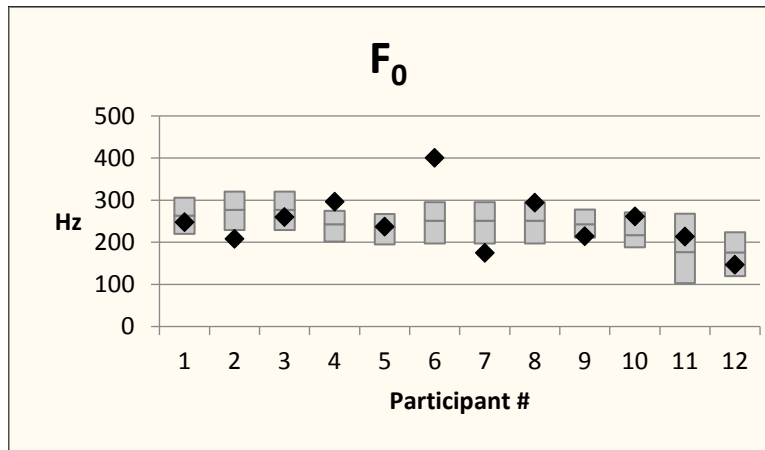
Table 3. Study participants' F0, jitt%, shimm% and NHR compared with normative dataset from Maturo et al

Gender	Participant #	Acoustic data				Modal value for each parameter of the GRBAS					PVRQOL	
		F ₀	Jitt%	Shimm%	NHR	G	R	B	A	S	Parent score	Child Score
♀	1	+	-	-	-	2	2	2	2	2	85	Not available
	2	-	-	-	-	2	1	2	2	0	47.5	92.5
	3	+	+	-	-	2	1	0	1	2	65 ¹	Not available
	4	-	+	+	+	1	0	1	1	1	72.5	90
	5	+	-	+	+	2	2	0	0	0	100	65
♂	6	-	+	+	+	1	1	0	0	1	40 ²	62.5
	7	-	-	-	-	3	3	0	0	2	75	87.5
	8	+	+	+	+	1	1	0	0	1	90	97.5
	9	+	+	+	+	1	1	0	0	0	100	100
	10	+	+	+	+	0	0	0	0	0	82.5	97.5
	11	+	+	+	+	1	0	0	1	0	100	87.5
	12	-	-	-	+	2	2	1	1	1	62.5	92.5

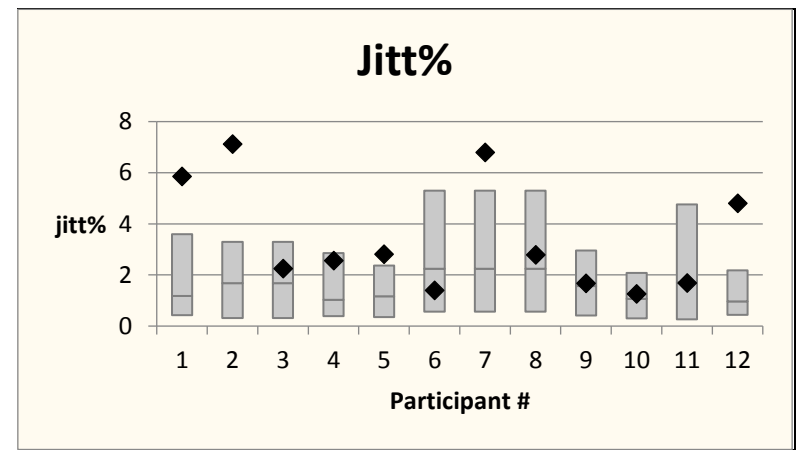
Table 4. Individual participant data presented with acoustic data (on the left), with + indicating within the normal range and – indicating not within the normal range; GRBAS modal values (centre) and PVRQOL scores (parent and child) (on the right).

¹ One question was rated as ‘not applicable’ giving missing values from the total score – had it been rated as “not a problem” the total score would be 75.

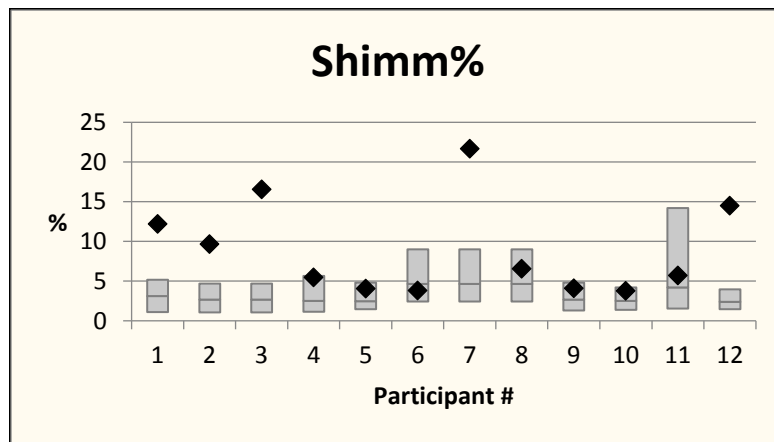
² Two questions were rated as ‘not applicable’ giving missing values from the total scores – had these been rated as “not a problem” the total score would be 60



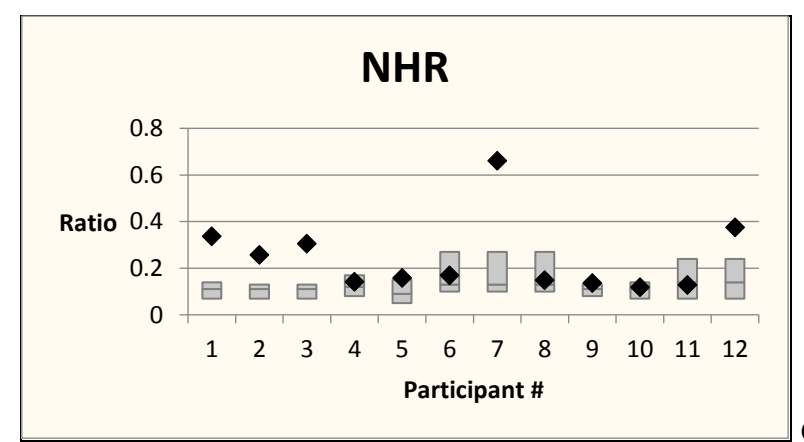
a



b



c



d

Figure 1. Charts showing each participant's acoustic value (◆) plotted against the mean, upper and lower data range from Maturo et al's normative dataset¹⁵ (the upper and lower range are shaded with the mean value denoted by a horizontal line)

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