Phoniatrics

Preliminary considerations on the application of the Voice Handicap Index to paediatric dysphonia

Considerazioni preliminari sull'applicazione del Voice Handicap Index alla disfonia pediatrica

A. SCHINDLER[1], P. CAPACCIO[2], P. MARUZZI[1], D. GINOCCHIO[1], A. BOTTERO[1], F. OTTAVIANI[1]
[1] Department of Clinical Sciences L. Sacco, University of Milan, Italy
[2] Department of Otorhinolaryngology and Ophthalmology, University of Milan, Italy

SUMMARY

Dysphonia is a common paediatric condition. Adult voices are usually evaluated using a set of minimal basic measurements including: endoscopic examination, aerodynamics, perception, acoustics, and self-assessment by the patient. The Voice Handicap Index is the most widely used self-assessment tool, but its use in the paediatric setting has never been reported. Aim of this study was to report Voice Handicap Index ratings in a group of dysphonic children, multi-modally assessed before and after voice therapy. The study involved 28 children (16 female, 12 male, mean age 10.9 years (range 6-12)) presenting chronic hoarseness due to vocal fold nodules (18 cases), unilateral localised oedema (6 cases) or recurrent laryngeal paralysis (4 cases). All received voice therapy for 5-6 months, and underwent voice assessments based on video-endoscopy ratings (size of nodule/oedema or glottic closure in the case of recurrent laryngeal paralysis), maximum phonation time, GIRBAS scale, spectrograms and a perturbation analysis. All patients also completed the Voice Handicap Index. Aerodynamic, acoustic, perceptual and self-assessment data, before and after voice therapy, were compared using Wilcoxon's test and Student's *t* test. Correlations between the Voice Handicap Index domains were measured by means of Pearson's correlation coefficient. Post-treatment measurements showed that the nodules/oedema had decreased in size in 18 children following therapy, and two subjects with recurrent laryngeal paralysis showed improved glottic closure. Mean maximum phonation time increased slightly, but the difference was not significant. There was a general reduction in perceptual severity, but this was only significant for parameters G, B and A. Spectrographic analysis showed no significant improvement and, although the mean perturbation analysis values improved, only the difference in jitter values was significant ($p = 0.016$). Voice Handicap Index was applicable in all cases, and showed a clear and significant improvement ($p = 0.0006$). The correlations between the three Voice Handicap Index factors were close; no correlation was found between the functional domain and the physical and emotional domains. The Voice Handicap Index is a useful tool in children with dysphonia, but an adapted version validated for paediatric patients is essential.

KEY WORDS: Dysphonia • Children • Diagnosis • Self-assessment

RIASSUNTO

La disfonia in età pediatrica è una condizione frequente. La valutazione della voce negli adulti è solitamente ottenuta attraverso un approccio multimodale; un insieme di misure minime di base per la valutazione della voce comprende l'esame endoscopico, la valutazione percettiva, acustica, aerodinamica e un'autovalutazione da parte del paziente. Il Voice Handicap Index (VHI) è lo strumento di autovalutazione più utilizzato al mondo, ma la sua applicazione in età pediatrica non è mai stata descritta. Scopo dello studio è riportare i punteggi del VHI in un gruppo di bambini disfonici valutati con un protocollo multidimensionale prima e dopo la terapia logopedica. 28 soggetti con disfonia cronica (16 femmine, 12 maschi) con un'età media di 10,9 anni (range: 6-12 anni) sono stati inclusi nello studio; 18 avevano noduli vocali, 6 un edema localizzato unilaterale e 4 una paralisi laringea. Tutti i bambini sono stati sottoposti a trattamento logopedico per un periodo compreso fra i 5 ed i 6 mesi. È stata eseguita una valutazione videoendoscopica delle dimensioni dei noduli/edema e della chiusura glottica nel caso di paralisi laringea. È stato misurato il massimo tempo di fonazione (MPT). Sono stati registrati gli spettrogrammi ed è stata eseguita un'analisi di perturbazione del segnale. Le voci sono state valutate percettivamente attraverso la scala GIRBAS. Tutti i soggetti hanno compilato il VHI. Le valutazioni aerodinamica, acustica, percettiva e autovalutativa prima e dopo il trattamento logopedico sono state confrontate attraverso i test di Wilcoxon e lo Student's t-test. Le correlazioni fra i domini del VHI sono state ottenute attraverso il test di Pearson. I noduli e l'edema sono diminuiti di dimensioni in 18 bambini dopo la terapia, ed è stata riscontrata una migliore chiusura glottica in due soggetti con paralisi laringea. Il MPT medio è aumentato leggermente ma la differenza non è risultata statisticamente significativa. Non è stato osservato alcun miglioramento significativo all'analisi spettrografica; per quanto concerne l'analisi di perturbazione del segnale i valori medi sono migliorati, ma solo la differenza nel valore di Jitter è risultata statisticamente significativa (p = 0.016). Nella valutazione percettiva è stata riscontrata una riduzione generale della gravità, ma la differenza si è rivelata statisticamente significativa solo per i parametri G, B e A. Il VHI è stato applicabile in tutti i casi. I valori del VHI hanno mostrato un evidente e significativo miglioramento (p = 0,0006). Le correlazioni fra i tre fattori del VHI sono state nel complesso elevate, essendo le uniche eccezioni rappresentate *dalla correlazione fra i domini funzionale da un lato e fisico ed emotivo dall'altro. Il VHI è apparso come uno strumento utile nei bambini con disfonia; un adattamento e una validazione per l'età pediatrica sono tuttavia indispensabili.*

PAROLE CHIAVE: ***Disfonia • Bambini • Diagnosi • Autovalutazione***

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Introduction

Paediatric dysphonia is not uncommon, and approximately 6-9% of all children have voice problems [1]. The most frequent causes are vocal fold nodules, which have been found in 38-78% of children evaluated for chronic hoarseness [2], but other vocal fold lesions, such as localised oedema and irregularity at the junction of the anterior and middle third of the vocal fold, have recently been found in 13.3% of the 617 children examined [3]. Common adult laryngeal diseases, such as recurrent laryngeal paralysis, or vocal fold polyps or cysts, are only seldom found in children.

Adult voices are usually assessed multi-modally. One evaluation, recently proposed, is based on the five aspects of perception, endoscopic examination, acoustics, aerodynamics, and a subjective self-assessment [4], a set of measurements that is not yet commonly used for children. Although the development of flexible fibre-optic endoscopes has made paediatric laryngeal examinations an everyday practice also in the doctor's surgery [5], and it has been shown that computer-assisted voice analyses correlate well with perceptual evaluations, thus extending their use in the management of paediatric dysphonia [6 7], subjective ratings have only recently been introduced in paediatrics.

Self-assessments, such as the Voice Handicap Index (VHI) [8], the Voice Outcome Survey [9], the Voice-Related Quality of Life [10] and the Outcome Scale [11] are currently used throughout the world to assess dysphonia as an outcome measure [12 13], and the VHI has not only been translated into various languages and used in many countries, but has also been applied to various patient groups [14-21]. It consists of 30 questions, divided into functional, emotional and physical domains to which patients are asked to respond using a five-point scale ranging from zero (never) to 4 (always).

To the best of our knowledge, there are no published reports concerning the paediatric application of the VHI. The aim of this study was to record VHI ratings in a group of children with different laryngeal disorders assessed multimodally before and after voice therapy.

Material and methods

Patients

This retrospective study of vocal function involved 28 children with chronic hoarseness: 16 females and 12 males with a mean age of 10.9 years (range 6-12). At the time of the first assessment, a diagnosis of bilateral vocal nodules was confirmed in 18 patients (mean age 11.6 years, range 10-12), recurrent laryngeal paralysis in 4 (mean age 8.8 years, range 6-11), and unilateral localised oedema in 6 (mean age 10 years, range 7-12). The study was carried out in accordance with the Declaration of Helsinki.

Voice therapy

Voice therapy was provided by an experienced speech and language therapist in the form of 20-30 minute sessions held once a week over a period of 5-6 months, and involved the cooperation of the patient's family, educators, peers and friends where appropriate and possible. The therapeutic protocol consisted of 5 behaviour based approaches: vocal hygiene (which was aimed at making the children aware of vocal abuse situations and behaviours, and encouraging their avoidance, with their caregivers being given additional information about the importance of hydration); direct facilitation based on reducing loudness, yawn-sigh, humming, resonant voice, confidential voice, and hard glottal attack reduction or increase (depending on the child and the voice disorder), with the caregivers taking part in vocal games designed to reinforce the practised technique at home; respiration and relaxation exercises (which were used sparingly because children often consider them boring and this reduces their compliance); and, finally, a carry-over approach that involved attempts to transfer the newly learned vocal behaviours to everyday situations [22-24].

Physical examination and maximum phonation time

At the beginning and end of voice therapy, each subject underwent videolaryngoscopy (in all cases without anaesthesia) using a flexible Storz FNL-10RP2 fiberscope (Storz Endoskop Productions GmbH, Tuttlingen, Germany) or an Atmos 4450.47 70° rigid telescope (ATMOS Medizin Technik GmbH & Co KG, Leuzkirch, Germany). The subjects with nodules or localised oedema were assessed on the basis of the size of the nodule/oedema (larger/smaller/no difference), whereas those with laryngeal paralysis were assessed on the basis of glottic closure (better/worse/no difference).

Maximum phonation time (MPT) was determined by measuring three productions of an /a/ sustained for as long as possible on the basis of the oscillogram signal, the longest of which was used for further processing. The voice signal was recorded and directly stored using the Computerized Speech Lab programme (version 5.05) with a 4400 external module (CSL, Kay Elemetrics Corp., NJ, USA).

Perceptual and acoustic voice analyses, and VHI

The GIRBAS scale was used for the perceptual voice analysis, with an experienced phoniatrician and speech therapist scoring each patient for conversational speech and sustained vowels.

The Computerized Speech Lab programme (version 5.05) and Multi Dimensional Voice Programme (MDVP) (version 1.34) with a 4400 external module (CSL, Kay Elemetrics Corp., NJ, USA) were used to make the objective voice evaluations. All the voices were recorded using a microphone, positioned approximately 15 cm from the mouth, slightly below the chin in order to reduce airflow effects. Spectrograms of the sustained vowels /a/ and /i/

were recorded at FFT-1024 points ranging from 0 to 8 kHz, with a sampling frequency of 20,000 Hz. The degree of acoustic perturbation in the voice was assessed in the spectrograms using Yanagihara's classification of hoarseness [25]. On the basis of the spectrographic results, each patient's voice was classified as type 1, 2 or 3 according to Titze's recommendations [26]. Only the patients with a type 1 voice underwent perturbation analysis, for which a sustained /a/ was used with a sampling frequency of 50,000 Hz, and jitter (Jitt %), shimmer (Shim %), the noise-to-harmonic ratio (NHR), and average fundamental frequency (Fo) were calculated.
Finally, each subject completed the VHI, although the form used had not been adapted for paediatric use.

Statistical analysis
Results are given as arithmetic mean values ± standard deviation. The MPT, perturbation analysis and VHI data before and after voice therapy were compared using Student's paired t-test, and the perceptual and spectrographic data were compared using Wilcoxon's signed-rank test. A p value of < 0.05 was considered statistically significant. The correlations between the VHI emotional, physical and functional factors were measured by means of Pearson's product-moment correlation, with the VHI scores before and after voice therapy being considered together. The data were statistically analysed using the SPSS 11.5 for Windows package (SPSS Science, Chicago, IL, USA).

Results

Physical examination and maximum phonation time
After therapy, the nodules/oedema had decreased in size in 18 children, and remained unchanged in the remaining 6; better glottic closure was observed in 2 of the 4 subjects with recurrent laryngeal paralysis, with no difference being detected in the others. The mean MPT was 8.7 ± 3.1 sec before voice therapy, and 9.9 ± 2.9 sec after; the difference was not significant (p = 0.25).

Perceptual ratings, acoustic voice analysis and VHI scores
The GIRBAS scale results before and after voice therapy are shown in Table I; therapy led to a reduction in the severity of all of parameters, but this was significant only in the case of G, B and A. The mean pre-therapy spectrographic score using Yanagihara's classification was 2.6: ten children were rated as type 4, two as type 3, ten as type 2, and six as type 1. After voice therapy, the mean value decreased to 2.2 (not significant: p = 0.56), and the individual ratings were two children with type 4, ten with type 3, ten with type 2, and eight with type 1.

Table I. GIRBAS scale data: mean, standard deviation and p values before and after voice therapy.

	Before voice therapy	After voice therapy	p value
Grade	2.2 ± 0.5	1.2 ± 0.6	0.020
Instability	0.6 ± 0.5	0.5 ± 0.7	0.054
Roughness	0.7 ± 0.8	0.5 ± 0.7	0.052
Breathiness	2.0 ± 0.7	1.1 ± 0.8	0.018
Asthenicity	1.1 ± 0.8	0.5 ± 0.6	0.048
Strained	0.4 ± 0.6	0.4 ± 0.6	0.65

Table II. Perturbation analysis data: mean, standard deviation and p values before and after voice therapy (only voices with a clear harmonic structure upon sound spectrography underwent perturbation analysis).

	Before voice therapy	After voice therapy	p value
Average fundamental frequency	234.8 ± 27	226.8 ± 38	0.41
Jitter %	2.3 ± 1.4	1.1 ± 0.5	0.016
Shimmer %	7.4 ± 2.9	5.7 ± 1.9	0.12
Noise to harmonic ratio	0.15 ± 0.05	0.12 ± 0.01	0.06

Table III. VHI data: mean, standard deviation and p values before and after voice therapy.

	Before voice therapy	After voice therapy	p value
VHI total	40.8 ± 15.0	26.9 ± 10.5	0.0006
VHI physical	21.2 ± 6.7	15.7 ± 6.5	0.0026
VHI functional	10.2 ± 3.8	6.7 ± 3.8	0.011
VHI emotional	8.8 ± 6.1	4.7 ± 2.4	0.010

Table IV. Correlations (*r* values) between the three VHI factors.

	VHI total	VHI physical	VHI functional	VHI emotional
VHI total	–	0.770 (p = 0.0001)**	0.741 (p = 0.0001)**	0.855 (p = 0.0001)**
VHI physical	0.770 (p = 0.0001)**	–	0.197 (p = 0.315)	0.770 (p = 0.001)**
VHI functional	0.741 (p = 0.0001)**	0.197 (p = 0.315)	–	0.386 (p = 0.042)*
VHI emotional	0.855 (p = 0.0001)**	0.770 (p = 0.001)**	0.386 (p = 0.042)*	–

* p < 0.05; ** p < 0.01

None of the subjects showed a Titze type 3 or type 2 signal, and, therefore, all the patients' voices were analysed using the MDVP programme. The results of the perturbation analysis showed that although the mean Fo, Jitt %, Shim % and NHR values all improved after voice therapy, only the difference in Jitt % was significant ($p = 0.016$) (Table II).

VHI values, before and after voice therapy, are shown in Table III. At both assessments, the scores of the physical subscale were higher than those of the emotional and functional subscales, but there was a clear and statistically significant improvement in the total values and each of the physical, functional and emotional subscales.

The correlations between the three VHI factors (Table IV) reveal that the overall correlation values were high, ranging from $r = 0.741$ to $r = 0.855$; low correlation scores were found between the functional domain and both the emotional ($r = 0.197$) and physical ($r = 0.386$) domains.

Discussion

We assessed the changes in voice quality, after voice therapy, in a group of 28 children with dysphonia by means of endoscopic, aerodynamic, perceptual, acoustic and self-assessment ratings. Endoscopic, perceptual, acoustic and aerodynamic ratings have previously been used by various authors, but this is the first report concerning the use of the VHI as a paediatric self-assessment tool.

A number of instruments have been used to measure the status of adult patients with voice disorders [8-11] but, to the best of our knowledge, the only paediatric voice quality-of-life instrument available is the Paediatric Voice Outcome Survey (PVOS) [27], which consists of 5 questions and relies on the parents as the source of information. The PVOS has been validated in 108 children and normative data have been obtained from 385 parents, and, therefore, it is a valid and reliable means of measuring voice-related quality of life in children [27, 28]; however, the fact that it has only five questions means that it cannot address the physical, functional and emotional aspects of voice disorders in any depth.

Although children's health-related quality of life can be assessed by means of a caregiver proxy, there are a number of published general health status measures based on the self-reporting of children aged more than five years [29]. In our study, the VHI was completed by 18 children themselves; the other children had the questions simply read or explained to them by their parents or the speech therapist. We preferred to record the children's view for a number of reasons. First of all, voice quality-of-life brings into play many factors that include not only social attitudes, environmental barriers, community supports, and cultural and ethnic backgrounds, but also a subject's psychosocial traits, education, age and sex [30], and, therefore, parents' reports do not necessarily reflect their children's real opinions. Secondly, one of the main goals of voice therapy is to make patients aware of their vocal behaviour in order to reduce "abusive" behaviours such as screaming or loud speaking [23], and answering the VHI helps to draw attention to such behaviours. Thirdly, motivation is an extremely important part of voice therapy, and answering voice quality-of-life questions helps patients to become aware of their everyday difficulties, thus increasing their motivation to improve the quality of their voice. Finally, it has been found that personality structure is very relevant in children with chronic hoarseness [31], and the future, widespread use of a self-assessment instrument may provide further insights into some of the causes of voice abuse or misuse in children.

The VHI could be administered to all of the children in the study, but some of the statements (such as, "my voice problem causes me to lose income" or "I am less outgoing because of my voice problem") seemed to be inappropriate for a young age. Furthermore, some of the terms used – such as "handicapped" or "personal and social life" – were not always easily understood and had to be explained by the therapist.

Speech therapy is the therapeutic cornerstone in paediatric voice disorders, although surgery has proved to be highly effective in the immediate/short-term treatment of vocal fold nodules [32 33]. We found that endoscopic, perceptual and aerodynamic data indicated better vocal quality after voice therapy, but the difference was statistically significant only in the case of some parameters; whereas the VHI scores clearly showed an improvement in health. There has been some controversy in the literature regarding the benefits of voice therapy in the treatment of paediatric dysphonia [34], and the future use of an adapted and validated paediatric VHI may help clarify the role of behavioural therapy in children with voice disorders. Furthermore, as pointed out by various authors, voice laboratory measurements such as acoustic, aerodynamic and perceptual ratings, offer certain insights into the severity of voice impairments in comparison with an expected normal voice, but fail to indicate why patients with similar voices experience different levels of handicap [35]. By considering this issue, the VHI represents a significant new development in the field of voice dysfunction, and paediatric vocology can draw some advantage from its use in children with voice disorders.

The correlations between the different domains of the VHI were generally close, with the exception of the functional domain in which the correlations with the physical and emotional domains were poor. Adults also show close correlations between the VHI domains, but with no exceptions [36]. The poor correlations in the VHI functional domain, in children may reflect their difficulty in recognising the functional correlates of their voices, although it is also possibly due to difficulties associated with the terminology used.

In conclusion, the VHI seems to be a useful tool in children with dysphonia aged 6-12 years, although an adapted and validated paediatric version is needed. The findings emerging from this preliminary study indicate that an adaptation and validation procedure would be worthwhile.

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Address for correspondence: Dr. A. Schindler, Dipartimento di Scienze Cliniche L. Sacco, Università di Milano, via G.B. Grassi 74, 20157 Milano, Italy. Fax +39 02 39043207. E-mail: antonio.schindler@tiscali.it