

Treatment of Voice Disorders in Medically Complex Children

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ocal abuse occurring with or without inflammation of the vocal cord nodules is regarded as the primary cause for voice disorders in school-age children (Aronson, 1985), and, several different treatment programs to reduce vocal abuse are available (Blonigen, 1980; Boone, 1980; Johnson, 1985; Wilson, 1987). However, a recent investigation revealed that less familiar laryngeal pathologies are also prevalent in children (Dobres, Lee, Stemple, Kummer, & Kretschmer, 1990). In a population of 731 children studied by Dobres et al. (1990), subglottic stenosis, laryngomalacia, vocal fold paralysis, and papillomatosis constituted 53% of the laryngeal pathologies. Children with these and other organic voice disorders, frequently referred for speech pathology services, typically present with complicated medical profiles.

Clinicians dealing with such children may have limited background in laryngeal pathology and there is a paucity of literature on laryngeal pathologies in children to guide them

ABSTRACT: A recent prevalence study revealed that laryngeal pathologies, including subglottic stenosis, laryngomalacia, vocal cord paralysis, and papillomatosis, are common in children. Children with these and other organically based alterations in laryngeal structure and/or function, frequently referred to the speech-language pathologist, often present with complex medical profiles. There is, however, a paucity of information to guide the clinician in diagnosing and treating voice problems associated with such laryngeal pathologies in children. This article presents individual case studies that detail information regarding management decisions and specific treatment options that have been implemented with medically complex children with voice disorders. The case studies demonstrate that there is an opportunity to improve vocal function in medically complex children.

KEY WORDS: voice disorders, children, voice therapy, laryngeal pathologies

regarding treatment of the associated voice problems. Although intervention programs for children who are tracheotomy dependent are available (Kaslon & Stein, 1985; Simon, Fowler, & Handler, 1983; Simon & Handler, 1981; Simon & McGowan, 1989), these do not address the diagnosis and treatment of voice problems associated with laryngeal pathologies.

The purpose of this article is to outline possible treatment options for voice disorders in medically complex children. Because of the limited information available on this topic, this article includes the presentation of case studies. These studies present the profiles of two medically complex children and provide information regarding treatment for their voice problems. The case studies and ensuing discussion are viewed as the beginning point in a process whose initial goals are to (a) heighten awareness regarding the vocal problems of children that exist in conjunction with laryngeal pathologies (other than vocal nodules), and (b) define the role of the speech-language pathologist in the treatment of concurrent voice disorders.

TYPES OF VOICE DISORDERS

For purposes of this article, "voice disorders in medically complex children" refers to the problems children experience with the production of voice that are caused by anatomic and neurologic anomalies that adversely affect normal laryngeal function. The voice disorders can present as disruptions across any of the vocal parameters (i.e., pitch, loudness, and/or quality), and they range in severity from mild to severe. At the extreme end of the range, we may be confronted with a child who experiences complete loss of voice, or aphonia. The alterations in laryngeal anatomy and function may impact the patency of a child's airway and cause respiratory problems. In some cases, the respiratory problems are so severe that a tracheotomy (i.e., a surgical opening through the neck into the trachea) will be required to permit an open airway.

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Anatomic Malformations

A review of the data provided by Dobres et al. (1990) can provide a guide to the specific anatomic and neurologic pathologies that we are likely to be confronted with in children. Based on that study, the three most frequent anatomical malformations of the larynx that are diagnosed in children are: (1) subglottic stenosis, (2) laryngomalacia, and (3) papilloma.

Subglottic stenosis is a thickening of tissue in the subglottis that may be extensive enough to compromise the airway for normal respiration. It may be either congenital or acquired. The severity of this condition will vary, and surgical treatment may be necessary in more severe cases. State-of-the-art surgery for subglottic stenosis involves laryngotracheal reconstruction, which is frequently accomplished by tracheal expansion, using grafting procedures, with rib cartilage.

The incidence of voice disorders in children who have had laryngotracheal reconstructive surgery is well documented in the literature (Luft, Wetmore, Tom, Handler, & Potsic, 1989; Sell & MacCurtain, 1988; Smith & Catlin, 1991; Zalzal, Loomis, Derkay, Murray, & Thomsen, 1991). To date, however, limited objective data regarding the vocal characteristics in this population have been reported (MacArthur, Kearns, & Healy, 1992; Smith, Marsh, Cotton, & Myer, 1993; Zalzal, Loomis, & Fischer, 1993).

Laryngomalacia, which is a condition characterized by soft or abnormally flacid cartilage in the larynx, is a common congenital anomaly diagnosed in infancy. The characteristic symptom of this disorder is stridor (i.e., production of a harsh, high-pitched sound on inhalation). This condition frequently resolves without intervention by the time a child is 1 year of age (Gray, 1992; Maddern, Campbell, & Stool, 1991). Therefore, the speech-language pathologist is called on infrequently with this population of children.

Papilloma is a benign growth that may occur in the trachea or larynx and is thought to be caused by a virus. These growths tend to proliferate and can cause airway obstruction. Consequently, surgical removal of papillomas is required. Papillomas are difficult to eradicate in children and frequently require repeated surgical excisions. The presenting vocal symptom with papillomas is hoarseness (Colton & Casper, 1990).

Although the above three diagnoses are the most common non-neurological organic laryngeal disorders in children, it is worth mentioning some of the other anomalies with which we may be confronted. These include (a) cyst, (b) hemangioma, (c) laryngeal cleft, (d) web, (e) granuloma, (f) caustic ingestion, and (g) laryngeal trauma (Dobres et al., 1990).

Neurological Disorders

Vocal fold paralysis has been cited as one of the most common laryngeal anomalies in children (Holinger & Brown, 1967), and it was reported in 6.2% of the population studied by Dobres et al. (1990). Vocal cord paralysis may be congenital or it may be acquired as a result of

surgical injury, trauma, growths, tumors, vascular lesions, or inflammatory illness (Wilson, 1987). One or both of the vocal cords may be affected, and the lesion will affect either abduction (i.e., opening of the vocal folds) or adduction (i.e., vocal fold closure). When bilateral abductor paralysis causes the vocal folds to remain closed, respiratory difficulties may result and a tracheotomy may be required. Vocal fold paralysis is, however, more often unilateral (Colton & Casper, 1990). The primary presenting vocal symptoms are breathiness and hoarseness (Colton & Casper, 1990).

APPROACHING THE PROBLEM

The deviations in the laryngeal anatomy and/or function with involved children may not remain static, but may change over time as a result of continued development, the disease process, or surgical intervention. Furthermore, based on my own experience, it appears that children with organic deviations in laryngeal structure and/or function frequently exhibit conditions of vocal misuse related to increased tension of the laryngeal musculature, that is, vocal hyperfunction.

For example, increased bulk, or hypertrophy, of the false vocal cords has been diagnosed in several of the children with laryngeal pathologies whom I have seen. Thickening of the false vocal cords presumably occurs secondary to ventricular phonation (i.e., phonation occurring secondary to movement of the false vocal cords). Therefore, in these children, ventricular phonation is thought to be a form of hyperfunction.

In some instances, it appears that hyperfunctional patterns of voice production may initially have been used as a means of compensation in the absence of normal vocal fold movement, secondary to an existing laryngeal pathology. In some children, these patterns become unnecessarily habituated after the laryngeal status is improved following medical/surgical intervention. When this is the case, children may present with patterns of vocal hyperfunction that are distinct from the underlying voice disorder caused by the organic pathology. Therefore, when we work with children with anatomic and/or neurological anomalies that affect laryngeal function, we should consider the possibility that patterns of vocal hyperfunction may contribute to the existing voice disorder.

The speech-language pathologist's overall goal for treatment with the child with a voice disorder is to help him or her achieve the best voice possible to meet his or her communication needs. With any voice disorder, and especially with the medically complex child, we must consider any existing limitations occurring secondary to alterations in laryngeal structure and/or function. The knowledge that structural and/or functional limitations are probable, and the multiplicity of problems that some of our children present, may cause the speech-language pathologist to feel overwhelmed by the task at hand.

It may help if we realize that intervention for the medically complex child with a voice disorder requires a

team approach. Evaluation and management of voice disorders in these children necessitates interdisciplinary staffing and consultation between the speech-language pathologist and the otolaryngologist. The otolaryngologist will examine the child's larynx, diagnose any laryngeal problems, and prescribe treatment. The otolaryngologist's recommendations may include medical treatment, surgery, a voice evaluation by a speech-language pathologist and therapy, and/or referral to other specialists. Good communication with the otolaryngologist can aid the speechlanguage pathologist in understanding the existing laryngeal pathology and the impact of any medical/surgical intervention on the laryngeal structures and function. The speechlanguage pathologist may also consider a referral to his or her colleagues in the field who specialize in the treatment of voice disorders.

There has been a rapid expansion in the number of speech-language pathologists with a specialty in voice disorders and in voice laboratories where instrumentation is routinely employed for the assessment and treatment of voice disorders. We should not overlook the value of using these resources for consultation with children who exhibit voice disorders. And, because we are dealing with children, parents should be actively incorporated in the intervention process. Other persons who may play a role in the treatment team include the pediatrician, teacher, and school psychologist. When a child has a tracheotomy, a school or private nurse will be involved with the child and is an important member of the team.

EVALUATION

The intervention process begins with the evaluation. This is accomplished by the otolaryngologist and the speechlanguage pathologist. A laryngological examination is typically part of the otolaryngologist's medical evaluation. Laryngoscopic procedures commonly used with children are briefly discussed here. Although a review of laryngoscopic techniques is not the purpose of this article, it is important that the reader understand the differences in the information obtained with each procedure.

Indirect laryngoscopy using a laryngeal mirror sometimes provides adequate diagnostic information. However, in order to accurately diagnose laryngeal pathologies in young children, it is frequently necessary to use endoscopy (Gray, 1992). Endoscopic procedures include direct laryngoscopy, fiberoptic nasoendoscopy, and rigid oral endoscopy. Because direct laryngoscopy requires general anesthesia, structures but not function can be observed during this procedure. On the other hand, the child is awake for endoscopy done with a fiberoptic or a rigid oral scope and visualization of both laryngeal structure and function is possible. Despite the fact that information regarding laryngeal function is invaluable to the speech-language pathologist, an awake laryngological examination cannot always be conducted with medically complex children. Therefore, definitive information regarding laryngeal function may not be obtained.

The speech-language pathologist's evaluation should include the acquisition of pertinent background information and an assessment of the child's voice. Background information to be obtained includes a review of the otolaryngologist's laryngeal diagnosis, treatment plans, and any data available regarding alterations in laryngeal structure and/or function. In addition, the speech-language pathologist should consider general medical history and collect voice symptom history, voice use history, and any relevant information regarding psychosocial factors. Perceptual and instrumental assessments of voice production are routine components of the voice evaluation.

A structured evaluation protocol is a valuable aid for conducting the perceptual assessment. Colton and Casper (1990) presented a thorough history and evaluation protocol in the appendix of their text that can serve as a model for clinicians conducting a voice evaluation. Although the completion of structured tasks may not be possible with infants and toddlers, the majority of preschool and schoolage children comply with a variety of structured tasks that result in an adequate sample of vocal production. For example, tasks routinely elicited with children seen for voice evaluations at Children's Hospital in Boston include:

- · sustained production of isolated vowels,
- sustained production of /s/ and /z/,
- · counting,
- sentence imitation,
- · singing a familiar song,
- · oral reading or recitation of a nursery rhyme, and
- · a spontaneous speech sample.

With adaptations in elicitation techniques, such as clinician models, practice trials, and visual cues, assessment tasks may be completed successfully even with young children (Champley & Andrews, 1993). When possible, instrumental measures such as videostroboscopy, acoustic analysis, aerodynamic procedures, and electroglottography should be conducted as part of the voice evaluation. The information obtained from all the evaluation procedures is then used to determine whether further intervention is indicated.

The following case studies are presented as a means of illustrating the thought processes followed when voice treatment was deemed appropriate and specific intervention strategies were used in therapy with two medically complex children.

CASE STUDY 1: HR

HR, an 8-year-old female, was referred for a voice evaluation by her otolaryngologist. She presented with a diagnosis of congenital subglottic stenosis and had a tracheotomy at 3 years of age. HR had undergone multiple surgeries, including laryngotracheal reconstruction (LTR) and an LTR revision. The initial voice evaluation was conducted 11 months following LTR revision surgery, and 6 months after HR's tracheotomy was closed. At the time of the initial evaluation, HR had just completed second grade. She had no previous speech or voice therapy.

Laryngeal Examination

The anterior point of attachment for the true vocal cords (i.e., the anterior commissure) is normally a common point on the posterior surface of the thyroid lamina. Direct laryngoscopy was done with HR and, in addition to subglottic stenosis, the presence of a blunt anterior commissure and paralysis of the right vocal cord were reported. Flexible nasoendoscopy was attempted, but could not be conducted due to noncompliance.

Voice Evaluation

Perceptual assessment. Average maximum phonation time for the sustained vowel /a/, produced across three trials, was 5 seconds. HR sustained /s/ for 6.9 seconds and /z/ for 5.2 seconds, resulting in an s/z ratio of 1.3.

HR presented with extremely aberrant vocal quality. The rating scale from the Mayo Clinic Dysarthria Study (Darley, Aronson, & Brown, 1975) is a useful tool for conducting a perceptual assessment of vocal deviations and was used to rate HR's voice. Perceptually, moderate to severe deviations from normal were indicated across the parameters of pitch level, monopitch, harsh voice, audible inspiration, and overall bizarreness, with the definition for bizarreness stated as "the degree to which overall speech calls attention to itself because of it's unusual, peculiar, or bizarre characteristics" (Darley et al., 1975, p. 295). A summary perceptual description of HR's voice characterized her productions as low and rough, with little prosodic variation, and with a quality similar to esophageal speech.

Instrumental assessment. Frequency measures were obtained with the Visi-Pitch (Kay Elemetrics, Pine Brook, NJ). A speaking fundamental frequency of 142 Hz, obtained during a counting task, was significantly below the lower limits of the normal range for girls of HR's age. HR's speaking fundamental frequency, in fact, approximated the pitch of an adult male. HR demonstrated a maximum phonational range (i.e., lowest to highest note) of 78 Hz to 499 Hz. The obtained measures indicated good pitch range (i.e., greater than two octaves).

Diagnostic therapy tasks were conducted with the Visi-Pitch. A horizontal cursor was placed at various frequency levels on the Visi-Pitch and HR was asked to produce a sustained /a/ that matched the visual line. HR demonstrated the ability to match the visual line at different frequency levels. She exhibited improved measures in perturbation and consistency of voicing with increased frequency; a judgement of improvement was based on a comparison of intrasubject data for repeated production of sustained /a/. HR's best performance on this task was obtained at 266 Hz; this pitch was within normal limits for 8-year-old girls.

Considerations for Intervention

HR presented with known deviations in laryngeal structure and function, including a blunt anterior commissure and paralysis of the right true vocal cord. Although

visualization of movement of the true and/or false vocal cords was not accomplished due to HR's noncompliance with nasoendoscopy, it was questioned if she was producing voice with her false vocal cords (i.e., ventricular phonation). This was postulate as the quality of HR's voice resembled textbook descriptions of ventricular phonation (Aronson, 1985; Colton & Casper, 1990); she exhibited a very low-pitched, harsh voice with limited prosodic variation in her connected speech. Furthermore, it seemed logical that ventricular phonation may have been initiated as a compensatory behavior secondary to altered laryngeal structures and function.

At first glance, we might be tempted to dismiss any consideration for further intervention with children with known alterations in laryngeal structure and function, such as those exhibited by HR. One might question if her vocal variations were appropriate compensatory behaviors, given the presence of altered laryngeal structures and function. Her voice, while aberrant, could be deemed functional for general communication needs, with no recommendations for intervention.

However, if we review the information obtained from the evaluation, a case for further intervention can be made. Specifically, *HR* demonstrated a pitch range of greater than two octaves, as well as improved measures of pitch and consistency of voicing in response to trial therapy techniques. This information suggested a positive prognosis for achievement of more appropriate pitch relative to *HR*'s age and sex and improved vocal quality with therapy. It was recommended that diagnostic voice therapy be conducted for 2 months, with continued therapy if measurable voice improvements could be documented.

Management Program

HR was enrolled in voice therapy for 1 hour each week over a period of 10 months. The goals of treatment during therapy were to achieve higher speaking fundamental frequency, or pitch, and reduced vocal roughness. The treatment program implemented with HR was relatively traditional; training tasks began with vowel production and progressed through a hierarchy of speech tasks to spontaneous speech across a variety of situations.

Procedures employed in therapy incorporated visual feedback with the Visi-Pitch, auditory feedback through the use of a tape recorder, negative practice, and self-monitoring. HR's mother actively participated in part of each therapy session to aid in the carryover of targeted behaviors into home-practice activities. Although the visual feedback provided to HR with the Visi-Pitch was judged to be beneficial, the same basic approach could be executed in a clinic or school setting without instrumentation.

The following outline details the basic training tasks employed in HR's therapy program. Criterion established for stages one through four of the program was 80% accuracy across 10 response trials. The established criterion for connected speech was demonstration of the target behavior across 75% of the sentences/phrases produced during a 5-minute conversational sample.

· Stage 1 Target Behavior

Production of the vowel /a/ at a pitch level of 257 Hz (+/- 10 Hz) with greater than 90% consistency of voicing, as measured with the Visi-Pitch.

Training was done to the stated criterion level with the aid of a visual cue, which consisted of placing a horizontal cursor at 257 Hz on the Visi-Pitch. After this goal was achieved, the same target behavior was trained to criterion without the aid of a visual cue.

· Stage 2 Target Behavior

Clear vocal production of one syllable words initiated with a vowel or /h/ at a pitch level of 257 Hz (+/- 10 Hz), with the judgement of clear production determined by the clinician.

This training step was first conducted to criterion with the use of the horizontal cursor as a cue and then in the absence of the visual cue. HR and her mother were trained to make judgements regarding "rough" versus "clear" vocal quality that were reliable with the clinician's decision. Once HR and her mother reliably judged the quality of HR's vocal productions, regular home assignments were initiated.

Training HR and her mother to be reliable judges regarding the accuracy of HR's response behaviors was deemed important for two reasons: (1) to ensure that the presence or absence of the target behavior was accurately judged during completion of the home assignments, and (2) to shape HR's self-monitoring skills.

• Stage 3 Target Behavior

Clear vocal production of multiple syllable words and phrases initiated with a vowel or /h/ at a pitch level of 257 Hz (+/- 10 Hz), with the judgement of clear production determined by the clinician.

As was done with the previous steps, this training task was first conducted to criterion with the use of the horizontal cursor as a cue and then in the absence of the visual cue. HR and her mother were trained to judge whether HR's phrase productions were at the desirable pitch level, which was referred to as "high pitched," or if her productions were "low pitched." Once HR and her mother reliably judged whether HR's responses were high versus low pitched, home assignments incorporated their judgments regarding both pitch and quality.

Stage 4 Target Behavior

Clear vocal production of short spontaneous sentences at a pitch level of 257 Hz (+/- 10 Hz), with the judgement of clear production determined by the clinician.

Spontaneous sentences were elicited in response to a question. This training step was conducted to criterion with the computer, but without the aid of the horizontal cursor as a visual cue, and then without the computer. In the absence of the objective measure of pitch, as computed with the Visi-Pitch, perceptual judgements regarding pitch and quality were made live or following playback of an audio-taped response.

· Stage 5 Target Behavior

Clear vocal production of connected speech during oral reading and conversational speech.

Carryover of the desired vocal behaviors (i.e., high, clear voice) was targeted across a variety of activities for HR in her home and school environments. These activities included times such as recess, math class, during dinner with the family, when a friend was over, and while doing homework. Specific situations in which HR was to self-monitor her vocal use were identified during the therapy session and HR maintained charts and/or journals to indicate how she did with carryover during the designated times/activities.

Results of Therapy

Vocal improvement was documented based on instrumental measures and perceptual judgments. Pre- and post-therapy measures indicated an increase in maximum phonation time for the vowel /a/ from 5 seconds to 13 seconds. Speaking fundamental frequency increased roughly five semitones, from 142 Hz to 217 Hz. Both of these post-therapy measures were within normal limits, based on normative data reported by Wilson (1987). Additionally, HR's upper pitch range was extended from 499 Hz to 623 Hz.

Perceptual judgments regarding HR's voice were made by two experienced speech-language pathologists. Pre- and post-therapy audiotaped samples, which consisted of HR reading the passage "The trip to the zoo," were used for this purpose. Perceptually, HR's voice was judged to be improved across the dimensions previously mentioned, (i.e., pitch level, monopitch, harsh quality, and aberrancy).

CASE STUDY 2: DF

DF was referred for a voice evaluation by his otolaryngologist at 5 years of age. DF was enrolled in a special needs language-focussed preschool class at the time of his initial evaluation and he subsequently went on to a regular kindergarten class. DF presented with a spectrum of anomalies referred to as VATER association, which consists of vertebral defects, anal atresia, tracheoesophageal (T-E) fistula, and radial and renal dysplasia.

DF was born with a T-E fistula, an abornmal opening between the trachea and the esophagus, and this was repaired at 1 day of age. Bilateral abductor paralysis of the true vocal cords and tracheomalacia, which is characterized by abnormal softening of the tissues of the trachea, were documented at that time. He also presented with other anomalies that are characteristic of VATER association defects. These included aorta fixation, a left club hand, narrowing of the anus, and placement of a gastrostomy tube due to esophageal abnormalities.

When DF was almost 3 years of age, a tracheotomy was performed. At 4 years of age, he underwent a surgical procedure known as arytenoidectomy, to facilitate lateralization of the left true vocal cord. Arytenoidectomy is known to be a reliable procedure for treating bilateral abductor

vocal cord paralysis. Closure of the tracheotomy, or decannulation, is frequently accomplished following arytenoidectomy and vocal function remains satisfactory (Lim, 1985; Ossoff, Duncavage, Shapshay, Krespi, & Sisson, 1990). Reportedly, *DF* exhibited "adequate" voice with the tracheotomy prior to the arytenoidectomy; however, he was essentially aphonic following surgery.

Laryngeal Examination

Before the arytenoidectomy, fiberoptic laryngoscopy revealed that the left vocal cord was positioned just off midline with adduction and no abduction. Good adduction of the right vocal cord with moderate abduction was reported. Following the arytenoidectomy, direct laryngoscopy was completed by the referring otolaryngologist. He reported the presence of an abnormal increase in bulk of the false vocal cords (i.e., false cord hypertrophy). The otolaryngologist expressed concern that the enlarged false vocal cords compounded DF's airway difficulties and could interfere with eventual closure of his trache-otomy.

Voice Evaluation

Perceptual assessment. As a result of limited client cooperation, administration of a formal voice evaluation protocol was not completed at the time of the initial contact. Instead, the initial evaluation consisted of perceptual assessment of vocal behaviors exhibited by DF during mother-child interactions and play activities. An instrumental assessment of vocal function was conducted with DF at a later date and findings from that evaluation are detailed later in this case presentation.

At the time of the initial evaluation, DF had a tracheotomy. Although DF's otolaryngologist had prescribed use
of a one-way speaking valve (i.e., the Passy-Muir valve), DF did not use this valve. DF generally communicated in
complete sentences using well-articulated but whispered
speech. He exhibited a rapid speaking rate and frequently
spoke with his back turned toward the person with whom
he was communicating. DF intermittently exhibited
effortful phrase and sentence productions that were
characterized by weak, rough, and breathy phonation.
These symptoms were exhibited with the trach unoccluded
in conjunction with anger, singing, and attempts to use
loud voice, as well as when DF was excited. The limited
voice produced in these instances did not enhance speech
intelligibility.

Weak and breathy phonation was also perceived on a reflexive level during coughing. Occasional production of phonation on inhalation, which could be described as a squeak, was also demonstrated. Duration of inhalation phonation was short and pitch was judged to be higher than for vocalizations demonstrated on exhalation. Inhalation phonation was produced on demand, with and without the trach occluded. Attempts to elicit phonation on demand during exhalation with the trach occluded were unsuccessful.

Considerations for Intervention

DF phonated intermittently, using two different voices. One was produced on exhalation and was characterized by weak, rough, and breathy phonation. This voice was typically produced in concurrence with increased effort. Based on these findings in conjunction with the otolaryngologist's report of false cord hypertrophy, it was believed that the false vocal cords (i.e., the ventricular folds) were the source of phonation for voice produced on exhalation. Use of ventricular phonation was not desirable, even in the absence of true cord phonation, given that hypertrophy of the false vocal cords might interfere with successful closure of this child's tracheotomy.

A second voice was produced on inhalation and could be described as a squeaking sound. It was higher in pitch and of limited duration. Citations in voice textbooks indicate that voice produced on inhalation is the result of vibration of the true vocal cords (Boone & McFarlane, 1988; Colton & Casper, 1990). Based on this information, it was suspected that the true vocal cords were the sound source for vocalizations demonstrated by *DF* during inhalation. The demonstrated inhalation phonation was considered a positive prognostic indicator for stimulating true cord phonation.

It was recommended that *DF* be enrolled in voice therapy. Elimination of effortful productions that were considered hyperfunctional was identified as a primary treatment goal. Even though it was not determined if *DF* was speaking with ventricular phonation, hypertrophy of the false vocal cords was documented and it was believed that this could be caused by vocal hyperfunction. Reduction of vocal hyperfunction was believed to be an important factor for achieving resolution of the false cord hypertrophy, which was prerequisite to closure of this child's trache-otomy. Other treatment goals that were targeted were the modification of interactional patterns that were judged to impede effective communication, use of a one-way speaking valve (i.e., the Passy-Muir valve), and facilitation of true cord phonation.

Management Program

DF was enrolled in voice therapy for 1 hour each week. His mother participated in all therapy sessions and daily home assignments were completed in conjunction with therapy. The specific treatment goals and therapy strategies that were implemented are presented below.

Goal 1 Target Behavior

DF will exhibit optimal speech and interactional patterns during communication interchanges, including use of "easy speech," appropriate positioning with respect to the listener, and appropriate speaking rate.

Elimination of effortful productions that were thought to be hyperfunctional was a primary component of this treatment goal. As is the case with many children who exhibit vocal hyperfunction, *DF* intermittently demonstrated more desirable communication patterns.

In his case, desirable behaviors were characterized by non-effortful production of whispered speech. This pattern was referred to in therapy as "easy speech."

Training techniques to facilitate this target behavior consisted of the provision of positive verbal reinforcement immediately following incidental use of the targeted "easy speech." The use of verbal praise served two purposes: (1) it resulted in increased selfawareness on DF's part regarding what the desired speech pattern was, and (2) it effected increased use of the target behavior.

"Easy speech" patterns were initially shaped during play and other communication interactions with the speech-language pathologist within the therapy room. Family members were incorporated into activities in the therapy sessions and trained to systematically reinforce the target behavior. Subsequently, home assignments that targeted "easy speech" across a variety activities in the home and school environments were given. Additional modifications to enhance successful communication were addressed concurrently. These included reducing DF's speaking rate and training DF to position himself face-to-face with his listener in order to provide optimal visual cues during communication interactions. These behaviors were shaped with the training techniques described above.

Goal 2 Target Behavior

DF will wear his speech valve for 5 hours daily.

A program was implemented with the family to gradually increase use of the Passy-Muir valve on a systematic basis. DF initially wore the valve for 30 minutes each day. Increments of 30 minutes were made on a weekly basis until DF wore the valve for approximately 5 hours daily. DF was always supervised by his parents or nurse while wearing the valve. The valve was removed when DF was fed orally and for suctioning. Although use of the valve did not immediately aid in facilitating phonation, it appeared to enhance production of the voice on exhalation once this had been established through use of the procedures detailed below.

Goal 3 Target Behavior

DF will exhibit volitional control of voice production on exhalation for general communication needs.

As previously described, DF produced intermittent, phonatory squeaks during inhalation. Phonation on inhalation has been used as a therapy technique to stimulate true cord phonation with individuals with functional aphonia, ventricular phonation, and spasmodic dysphonia (Boone & McFarlane, 1988; Colton & Casper, 1990; Shulman, 1993). This technique was tried with DF and productions of isolated vowels and words on inhalation were readily facilitated. Initial attempts to get DF to match this voice on exhalation met with limited success. Training techniques for inhalation phonation as outlined by Shulman (1993) were, however, eventually executed successfully with DF. Because this is not a well known therapy

technique, I will take a few moments to explain the basic training steps followed with DF.

- 1. Production of the vowel /i/ on inhalation.
- 2. Production of /i/ followed by numbers on inhalation (i.e., /i/ 1, /i/ 2, /i/ 3, etc.).
- 3. Production of /i/ on inhalation and then immediately, on the same breath, production of a number on exhalation; matching the sound produced on inhalation.
- 4. Production of words on exhalation without strain.
- 5. Production of words, phrases, and sentences of increased length on exhalation without strain.

Results of Therapy

DF attended voice therapy 1 hour each week for a period of 4 months. The initial treatment goals for elimination of effortful productions and modification of interactional patterns were achieved. DF consistently wore his Passy-Muir speaking valve for approximately 5 hours daily. In treatment sessions, DF demonstrated voiced production of two and three syllable words on exhalation without strain at a level of 85% accuracy. Although DF's spontaneous speech remained largely aphonic, he demonstrated intermittent carryover of voiced production of short phrases on exhalation without strain. DF demonstrated very positive changes in his communication patterns in response to the intervention techniques and, even though he continued to be largely aphonic outside the therapy, persons with whom he interacted reported that his speech was easier to understand.

Voice Reevaluation

Given the complexity of this case, laryngeal function studies were pursued in order to augment perceptual information. This instrumental assessment was conducted in collaboration with Robert Hillman, PhD, director of the Voice and Speech Laboratory at the Massachusetts Eye and Ear Infirmary in Boston.

Instrumental assessment. DF remained tracheotomy dependent and used a Passy-Muir speaking valve when vocal function testing was completed. The following objective measures were obtained: (a) acoustic analysis consisting of frequency, intensity, and perturbation measures; (b) aerodynamic assessment of glottal air flow, subglottic air pressure, and glottal resistance to air flow; and (c) electroglottographic data. Attempts were made to obtain all measures without and then with the speaking valve in place. A summary of Dr. Hillman's reported findings (R. E. Hillman, personal communication, July 6, 1993) is provided below.

Acoustic data. Measurable acoustic data were not obtained without the Passy-Muir valve. With the Passy-Muir valve, acoustic signals that contained measurable segments were produced by DF during production of sustained vowels and a counting task. The obtained acoustic measures were characterized by abnormally high fundamental frequency (pitch), reduced sound pressure level (loudness), and increased levels of frequency and amplitude perturbation.

Aerodynamic data. Measures of subglottal air pressure, glottal air flow, and glottal resistance were obtained during comfortable phonation of the repeated production of /pae/. Data were collected with a pneumotachograph, which was mounted in a face mask, and a catheter placed between DF's lips. Without the Passy-Muir valve, only subglottal air pressure could be reliably measured, and this measure was two to three times greater than normal. With the speaking valve, reliable measures were obtained for subglottal air pressure, glottal air flow, and glottal resistance during comfortable phonation. Sound pressure level (i.e., loudness) and glottal air flow rate were within normal limits. Subglottal air pressure was high, as was glottal resistance.

Electroglottographic data. Electroglottography, which measures relative vocal fold contact, was obtained during phonation of /a/. Without the Passy-Muir valve, measurable electroglottographic (EGG) signals were not obtained. With the speaking valve, DF produced measurable EGG signals that had periodic segments that indicated pressed phonation.

Impressions and Recommendations

DF showed improvement with the speaking valve in place. When wearing the Passy-Muir valve, DF was able to produce consistent voicing at a laryngeal level; however, it could not be determined whether valving of the airstream was accomplished by DF's true and/or ventricular folds. Based on the obtained instrumental measures, prognosis for further development of phonatory capabilities was considered favorable. It was believed that DF would benefit from continued voice therapy with emphasis on the following:

(a) increased percentage of periodic voiced phonation during connected speech, (b) improved breath support and coordination of respiration with voicing, and (c) reduction of vocal hyperfunction.

Follow-up

A direct laryngoscopy was conducted following 4 months of therapy and revealed no indication of false cord hypertrophy, as previously documented. Decannulation was subsequently accomplished and *DF* continued in Phase II of therapy. Treatment goals were expanded to incorporate the recommendations made based on the results of the instrumental assessment.

DISCUSSION

Children with medically based voice disorders represent a heterogeneous population with regard to both the etiologies of their laryngeal pathologies and their vocal symptoms. The very diversity of this group of children preclude considering the two case studies that are presented here as representative of the population. Rather, the information offers a reference point from which we can start to discuss

the assessment and treatment of voice disorders in medically complex children.

ASSESSMENT

The Challenge of Assessing Laryngeal Function With Children

Although nasoendoscopy and rigid oral endoscopy are important tools for the visualization of laryngeal function and are used successfully with many children (Chait & Lotz, 1991), there are instances when these procedures cannot be conducted. At such times, the clinician will not have definitive information regarding laryngeal function. When this is the case, the clinician must consider all the evaluative information obtained, determine its significance, and then make his or her best judgement regarding diagnosis and implications for treatment. The case study of *HR* is used here as an example.

HR's voice was characterized by low pitch and very hoarse quality, which is typical of ventricular phonation. However, a definitive diagnosis of ventricular phonation was not possible because HR did not cooperate for nasoendoscopy. Prognosis for facilitating a more appropriate pitch, a higher pitch, was, nevertheless, judged to be positive even in the absence of this diagnostic information. This judgement was based on HR's pitch range, which was greater than two octaves, and improved measures of frequency and perturbation in response to diagnostic therapy techniques conducted as part of the assessment process.

When visualization of laryngeal function is not possible, assessments with clinical voice instrumentation are particularly valuable in determining prognostic factors and appropriate treatment goals with children. Indeed, instrumental measures of vocal function should be used whenever possible to augment assessment information, with or without successful visualization of laryngeal function. Although still relatively rare, the use of instrumentation with children, including videostroboscopy, acoustic analysis, aerodynamic procedures, and electroglottography, is documented in the literature (Glaze, 1993; Lewis, Andreassen, Leeper, Macrae, & Thomas, 1993; Stathopoulos, 1985, 1986; Zajac & Linville, 1989; Zajac, Farkas, Dindzans, & Stool, 1993).

To Treat or Not to Treat

The speech-language pathologist working with the medically complex child often faces a dilemma. Despite the fact that the child may be relying on compensatory behaviors that are actually hyperfunction, should the speech-language pathologist accept the child's voice as optimal, given the medical circumstances? Or, should the speech-language pathologist instead attempt to improve the child's voice through therapy?

Initiating treatment may seem as perilous as opening Pandora's box, but these case studies suggest that intervention is often productive. For example, easy phonation on exhalation was eventually established with *DF*—a child

who initially exhibited hyperfunction in conjunction with all phonation produced on exhalation and inhalation phonation that was described only as a "squeak." The course of treatment with this child was novel and, in fact, required the exploration and use of techniques not commonly described for children.

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TREATMENT

Considerations for Intervention

Once a decision is made to proceed and at least attempt treatment, then traditional therapeutic techniques outlined in numerous texts (Andrews, 1986; Aronson, 1985; Boone & McFarlane, 1988; Case, 1991; Colton & Casper, 1990; Stemple, 1993; Wilson, 1987) can be used as a starting point for facilitating vocal improvement. In general, therapy approaches described in these textbooks can be used effectively with children. Adaptations in the presentation of the information to the cognitive and interest level of the individual child will be necessary, but the underlying principles are the same. Use of the inhalation phonation technique is a case in point. Use of this technique has only been cited in the literature with respect to adults with voice disorders, but it was certainly advantageous with the child discussed in case study #2.

As with assessment, instrumentation can be extremely valuable in the treatment process. Visual feedback with an electroglottograph or pitch monitoring equipment, such as the Visi-Pitch, can be beneficial. These instruments may help a child to visualize deviations in his or her own vocal productions that they may not be able to perceive auditorily. The speech-language pathologist can also use these tools to provide a visual model for the child to match.

Video recordings can be used to provide visual feedback to a child who exhibits increased tension or abnormal body posture during speech. Even the use of auditory recordings can be invaluable in training self-monitoring skills. The use of instrumentation in combination with good therapy strategies allows for exciting treatment opportunities that are limited only by the creativity of the individual clinician.

Carryover is an expected outcome with the medically complicated child's voice program, as it is with any treatment program. It is, therefore, important for the speech-language pathologist to build generalization training into the management plan. This can be done through elicitation of the targeted vocal behaviors during natural communication interactions conducted across settings and with a variety of people. Collaboration with either the child's parent(s) or classroom teacher is very helpful, and perhaps necessary, for successful carryover.

CONCLUSION

The case studies presented here suggest that there is a potential for vocal improvement with medically complex children, despite alterations in laryngeal structure and/or

function. The speech-language pathologist, who plays a vital role in assessing, counseling, and treating voice disorders in medically complex children, may, however, find the presenting problems to be overwhelming. The rewards for successful management of voice disorders in the medically complex child can also be enormous when the speech-language pathologist forges ahead, obtains diagnostic data that lead to an understanding of the clinical problem, sets realistic goals, and then implements intervention strategies effectively.

REFERENCES

- Andrews, M. L. (1986). Voice therapy for children. White Plains, NY: Longman.
- Aronson, A. E. (1985). Clinical voice disorders (2nd ed.). New York: Thieme.
- **Blonigen, J. A.** (1980). Remediation of vocal hoarseness. Hingham, MA: Teaching Resources.
- Boone, D. R. (1980). The Boone voice program for children. Austin, TX: Pro-Ed.
- Boone, D. R., & McFarlane, S. C. (1988). The voice and voice therapy (4th ed.). Englewood Cliffs, NJ: Prentice Hall.
- Case, J. L. (1991). Clinical management of voice disorders (2nd ed.). Austin, TX: Pro-Ed.
- Chait, D. H., & Lotz, W. K. (1991). Successful pediatric examination using nasoendoscopy. *Laryngoscope*, 101, 1016-1018.
- Champley, E. H., & Andrews, M. L. (1993). The elicitation of vocal responses from preschool children. Language, Speech, and Hearing Services in Schools, 24, 146-150.
- Colton, R. H., & Casper, J. K. (1990). Understanding voice problems: A physiological perspective for diagnosis and treatment. Baltimore, MD: Williams & Wilkins.
- Darley, F. L., Aronson, A., & Brown, J. R. (1975). Motor speech disorders. Philadelphia, PA: W.B. Saunders.
- Dobres, R., Lee, L., Stemple, J. C., Kummer, A. W., & Kretschmer, L. W. (1990). Description of laryngeal pathologies in children evaluated by otolaryngologists. *Journal of Speech and Hearing Disorders*, 55, 526-532.
- Glaze, L. (1993). Use of patient/family education and behavior modification. In J.C. Stemple (Ed.), Voice therapy: Clinical studies (pp. 32-35). St. Louis, MO: Mosby-Year Book.
- Gray, S. (1992, July). Treatment of pediatric voice disorders.Paper presented at the University of Wisconsin Phonosurgery Symposium, Madison, WI.
- Holinger, P. H., & Brown, W. T. (1967). Congenital webs, cysts, laryngoceles and other anomalies of the larynx. Annals of Otology, Rhinology and Laryngology, 76, 744-752.
- Johnson, T. S. (1985). Vocal abuse reduction program. Boston, MA: College Hill Press.
- Kaslon, K. W., & Stein, R. E. (1985). Chronic pediatric tracheotomy: Assessment and implications for habilitation of voice, speech and language in young children. *International Journal of Pediatric Otorhinolaryngology*, 9, 165-171.
- Lewis, J. R., Andreassen, M. L., Leeper, H. A., Macrae, D. L., & Thomas, J. (1993). Vocal characteristics of children with cleft lip/palate and associated velopharyngeal incompetence. *The Journal of Otolaryngology*, 22, 113-117.

- Lim, R. Y. (1985). Laser arytenoidectomy. Archives of Otolaryngology, 111, 262-263.
- Luft, J. D., Wetmore, R. F., Tom, L. W. C., Handler, S. D., & Potsic, W. P. (1989). Laryngotracheoplasty in the management of subglottic stenosis. *International Journal of Pediatric Otorhinolaryngology*, 17, 297-303.
- MacArthur, C. J., Kearns, G. H., & Healy, G. B. (1992, April).
 The role of laryngeal structure and function in voice quality after laryngotracheal reconstruction. Paper presented at the meeting of the American Society of Pediatric Otolaryngology, Palm Desert, CA.
- Maddern, B. R., Campbell, T. F., & Stool, S. (1991). Pediatric voice disorders. Otolaryngologic Clinics of North America, 24, 1125-1140.
- Ossoff, R. H., Duncavage, J. A., Shapshay, S. M., Krespi, Y. P., & Sisson, G. A. (1990). Endoscopic laser arytenoidectomy revisited. Annals of Otology, Rhinology and Laryngology, 99, 764-771.
- Sell, D., & MacCurtain, F. (1988). Speech and language development in children with acquired subglottic stenosis. *Journal of Otology*, 17, 35-38.
- Shulman, S. (1993). Symptom modification for abductor spasmodic dysphonia. In J.C. Stemple (Ed.), Voice therapy: Clinical studies. (pp. 128-131). St. Louis, MO: Mosby-Year Book.
- Simon, B. M., Fowler, S. M., & Handler, S. D. (1983). Communication development in young children with long-term tracheostomies: Preliminary report. *International Journal of Pediatric Otolaryngology*, 6, 37-50.
- Simon, B. M., & Handler, S. D. (1981). The speech pathologist and management of children with tracheostomies. The Journal of Otolaryngology, 10, 440-448.
- Simon, B. M., & McGowan, J. S. (1989). Tracheostomy in young children: Implications for assessment and treatment of communication and feeding disorders. *Infants and Young Children*, 1, 1-9.
- Smith, M. E., Marsh, J. H., Cotton, R. T., & Myer, C. M. (1993). Voice problems after pediatric laryngotracheal recon-

- struction: Videolaryngostroboscopic, acoustic, and perceptual assessment. *International Journal of Pediatric Otorhinolaryngology*, 25, 173-181.
- Smith, R. J. H., & Catlin, F. I. (1991). Laryngotracheal stenosis: A 5-year review. Head and Neck Surgery, 13, 140-144.
- Stathopoulos, E. T. (1985). Effects of monitoring vocal intensity on oral air flow in children and adults. *Journal of Speech and Hearing Research*, 28, 589-593.
- Stathopoulos, E. T. (1986). Relationship between intraoral air pressure and vocal intensity in children and adults. *Journal of Speech and Hearing Research*, 29, 71-74.
- Stemple, J. C. (1993). Voice therapy: Clinical studies. St. Louis, MO: Mosby-Year Book.
- Wilson, D. K. (1987). Voice problems in children (3rd ed.).
 Baltimore, MD: Williams & Wilkins.
- Zajac, D. J., Farkas, Z., Dindzans, L. J., & Stool, S. E. (1993).
 Aerodynamic and laryngographic assessment of pediatric vocal function. *Pediatric Pulmonology*, 15, 44-51.
- Zajac, D. J., & Linville, R. N. (1989). Voice perturbations of children with perceived nasality and hoarseness. *Cleft Palate Journal*, 26, 226-232.
- Zalzal, G. H., Loomis, S. R., Derkay, C. S., Murray, S. L., & Thomsen, J. (1991). Vocal quality of decannulated children following laryngeal reconstruction. *Laryngoscope*, 101, 425-429.
- Zalzal, G. H., Loomis, S. R., & Fischer, M. (1993). Laryngeal reconstruction in children: Assessment of vocal quality. Archives of Otolaryngology Head and Neck Surgery, 119, 504-507.

Received December 1, 1993 Accepted August 25, 1994

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