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Kajian Kes / *Case Study*

Speech Rehabilitation of Paradoxical Vocal Fold Movement

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ABSTRAK

Pelbagai bentuk pendekatan pengendalian pergerakan lipatan vokal paradoksikal (PLVP) telah dibincangkan dalam kajian-kajian sebelum ini, tetapi adalah penting untuk mengiktirafkan sifat kompleksiti kecelaruan ini dan perlunya diagnosis yang betul bagi membolehkan pengendalian yang sewajarnya dijalankan. Dapatan penyelidikan mendapati bahawa kecelaruan ini lazimnya berlaku di kalangan wanita muda yang mempunyai sejarah masalah perubatan yang berkaitan. Artikel ini membincangkan tentang kajian kes tunggal tentang seseorang kanak-kanak lelaki berumur 11 tahun yang mempunyai kecelaruan PLVP. Keadaan PLVP ini telah didiagnosis oleh pakar Otorinolaringologi di Pusat Perubatan Universiti Malaya, Kuala Lumpur. Ujian nasendoskopi menunjukkan pergerakan lipatan vokal adalah normal dalam pernafasan senyap dan sewaktu serangan episodik. Kanak-kanak ini kemudiannya dirujuk untuk terapi pertuturan; kecelaruan PLVPnya dikendalikan khususnya menggunakan rehabilitasi pertuturan. Pengurusan kecelaruan bagi kanak-kanak lelaki ini dibincangkan daripada mula rawatan sehingga beliau didiscaj daripada rawatan.

Kata kunci: PLVP, Terapi Pertuturan, Rehabilitasi Pertuturan

ABSTRACT

Many forms of management of the Paradoxical Vocal Fold Movement (PVFM) disorder have been presented in past literature, but it is vital to recognize the complexity of the disorder and the necessity for proper diagnosis to allow for appropriate management. A review of the literature suggests that this disorder predominantly occurs in the young female, and presents with a history of associated medical conditions. A single case study of an unusual presentation of PVFM in a young eleven year old boy with PVFM is discussed in this paper. The PVFM was observed and diagnosed by the Otorhinolaryngologist at the University Malaya Medical Center (UMMC), Kuala Lumpur. Nasendoscopy revealed otherwise normal vocal fold movement in quiet breathing and during

an episodic attack. The young boy was subsequently referred for speech therapy; management of the PVFM was solely with speech rehabilitation. The management of the disorder in this young boy is discussed up to the time of discharge from therapy.

Key words: Paradoxical Vocal Fold Movement, Speech Therapy, Speech Rehabilitation

INTRODUCTION

Paradoxical vocal fold movement (PVFM) is considered a complex disorder of the laryngeal and upper respiratory tracts. It is a rare and therefore complex disorder to diagnose and manage. Owing to the rarity of its occurrence, PVFM has been labeled as "Munchausen's stridor" (Altman et al. 2000), functional airway obstruction (Appelblatt & Baker 1981), non-organic laryngeal obstruction (Patterson et al. 1974), factitious asthma (Downing et al. 1982), laryngeal spasm mimicking bronchial asthma (Chawla et al. 1984), stridor caused by vocal cord malfunction associated with emotional factors (Katlan & Ben Zvi 1985) as well as numerous other labels. Dunglison (as cited in Mathers-Schmidt 2001) has described its symptoms as early as 1842.

PVFM is defined as a disorder of the upper respiratory tract characterized by spasmodic closure of the vocal folds during the inhalation phase of respiration (Altman et al. 2000) that results in intermittent airway obstruction and stridor (Adrianopoulos et al. 2000). Kaufman (1994) further determined the severity of the disorder by stating that the extent of airway obstruction and vocal fold adduction contribute to the severity of respiratory symptoms experienced by the individual.

Respiratory distress of this nature would require immediate medical attention and care. As mentioned earlier, many individuals with PVFM are misdiagnosed as having asthma and therefore are mismanaged with bronchodilators and steroids (Newman, Mason, Schmaling 1995). In emergency situations, tracheostomies may be performed to allow ventilation (Lloyd & Jones 1995).

Additional characteristic diagnostic features of PVFM include attenuation of the inspiratory flow-volume loop during forced inspiratory-expiratory maneuver consistent with variable extra-thoracic obstruction, and adduction of true vocal folds with characteristic posterior "chinking" during the respiratory cycle demonstrated by direct visualization using fiberoptic nasendoscopy (Christopher et al. 1983).

From the literature review conducted for this study, most cases diagnosed with PVFM were found to be young females with some degree of psychological illness and mental stress (Gallivan et al. 1996), often without organic etiology (Pinho et al. 1997). The present case study reports an unusual occurrence of

PVFM in an individual who does not identify with the general profile of PVFM patients and the speech rehabilitation techniques used in the treatment of this condition.

CASE REPORT

A young eleven year old Chinese boy was referred for speech therapy after being diagnosed as having PVFM by the Otorhinolaryngologist (ORL) at the University Malaya Medical Center (UMMC), Kuala Lumpur. The young boy was accompanied by his father who was concerned that his son had frequent episodes of "noisy breathing" since the first time noticed, approximately six months prior to the ORL consult.

His condition had received several previous diagnoses and attempts at treatment at private clinics and even faith-healers, but all without success. The first medical doctor who examined him treated him with bronchodilator medication as used in patients with asthmatic conditions. When the medication failed to relieve the symptoms, the father took the young boy to another private practitioner who subjected the boy to pulmonary evaluations and examinations, and prescribed medication for pulmonary fungal infection. A third medical practitioner administered oral corticosteroids but with no avail.

The consult with the ORL at UMMC began with an interview with the father and son. The father reported that the boy would abruptly awake during the night, gasping for air and thus disrupting sleep for himself and his parents. It was also reported that he had fainted once at school, collapsed on the school field after a period of physical education, but regained consciousness within two minutes. His teachers informed the father that he was frequently tired and had difficulty concentrating in class. The teachers noted that his grades had slipped and were concerned about his forth-coming Primary 6 government examination.

On questioning the young patient, he reported that he occasionally had difficulty in breathing and felt tired after climbing two flights of stairs to his classroom. He noted that a "grunt-like noise" would occur while watching television or while playing computer games. When asked for his opinion on what he felt aggravated the symptoms, he said that stress, excessive homework and his parents' strictness / scolding increased the frequency and intensity of the "grunt-like noise".

He continued to say that when his father scolds him, he felt very scared, sad and confused and as a result had difficulty in getting restful sleep at night. He said that in the initial stages, his problem would occur only at night, but lately it occurred several times even during the day. The ORL who examined the patient referred him for various examinations to rule out pulmonary compromise and subtle cardiac problems. The results were negative, and ruled out pulmonary

and cardiac concerns. While in clinic the patient had an episode of "grunt-like" noise. The patient was immediately examined using fiber-optic nasendoscopy.

The nasendoscopic examination revealed that there was no mass obstructing the airway. The vocal folds were slightly inflamed, but no nodules, polyps, cysts or growths were noted. The patient displayed some atypical vocal fold movement in that the folds did not coordinate during the inhalation and exhalation periods of quiet respiration. During a timely episode of wheezing and dyspnea, nasendoscopy revealed that his arytenoid cartilages functioned asymmetrically and adducted inappropriately. This finding confirmed the suspicion of PVFM and he was referred for weekly speech therapy for a period of six weeks.

SPEECH REHABILITATION

FIRST CONSULTATION

The patient attended the speech therapy session accompanied by his father. He presented as a shy and reserved young boy. He constantly looked toward his father for approval signaling some lack of confidence. Perceptual assessment of his voice showed that lacked intensity but was appropriate in pitch, when he was required to sustain a prolonged /a/ to ascertain respiratory-phonatory patterns.

Prior to starting voice rehabilitation, some time was spent on increasing the young boy's confidence levels – both in speech rehabilitation as well as in himself. He was constantly assured that he had been evaluated and that his condition had been diagnosed accurately. He was also reassured that he did not require medication but could be treated successfully with speech rehabilitation that included relaxation techniques. The psychological preparation for a patient was vital for good rapport and acceptance of treatment. Formal therapy began only when this was established.

Therapy goals were established together as involvement of the patient and caregiver was felt to be important in the success of treatment and rehabilitation. The following goals were agreed upon:

1. The patient will identify the basic structures of the respiratory and phonatory systems and their respective functions in phonation.
2. The patient will maintain adequate hydration and avoid laryngeal irritants by drinking warm water throughout the day.
3. Within the 6 sessions of speech rehabilitation, the patient would be able to execute relaxed diaphragmatic breathing to reduce laryngeal and thoracic tension.
4. Voluntarily control of episodic attacks will be achieved within the 6 sessions of speech therapy.

Speech rehabilitation techniques used in the management of this patient were based on the program used by Martin et al. (1987) as well as by Pinho et al. (1997). Slight modifications were made to the techniques to suit a younger patient.

The first formal therapy session addressed self-awareness about what happens during an episode of stridor, the use of diaphragmatic breathing for relaxation and generalized muscle tension reduction. The session began with giving the parent and the patient a clear and simple explanation of how the respiratory system coordinates with the phonatory system and how they are affected by muscular tension. Simplified color charts and diagrams of the upper airway downloaded from the internet were used to complement the verbal explanations (Figs. 1 and 2).

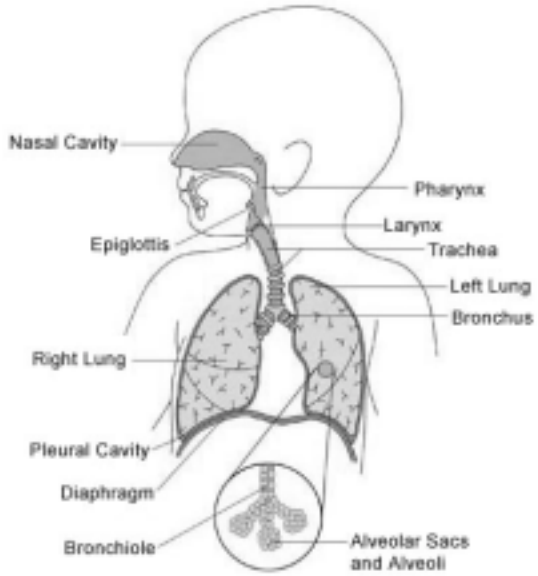

FIGURE 1. A simple diagram of the respiratory mechanism from http://uuhsc.utah.edu/healthinfo/pediatric/respiratory/lungsant.htm.

The session also reviewed the video recording of the nasendoscopy view of the patient's vocal fold function and comparison to normal physiology was made. Although young, the patient could appreciate the normal physiology of the vocal folds and was able to understand the explanation provided. This session concluded with relaxation and breathing techniques to reduce muscular tension that he was to carry-over to outside the clinic setting. The patient was then asked to write a daily log of his activities and food intake as well as of any

attacks that may occur. In order to help reduce chronic coughing or attacks, increased water intake was suggested. The patient was advised to take a half cup of warm water every half hour to maintain hydration and discourage irritation.

A letter to request for exemption from strenuous activity was issued to the school. A simple week to week addition of activity to his daily schedule was suggested to encourage gradual challenge to the respiratory system. The teacher / school was also advised on the action to be taken, should the attack occur during school-time that included claming him down, allowing free-ventilation and reassuring him that he would be alright. In order to chart his progress in speech therapy, a fancy colored chart was prepared to document his weekly performance and as a motivational tool for him to continue to improve his breathing patterns.

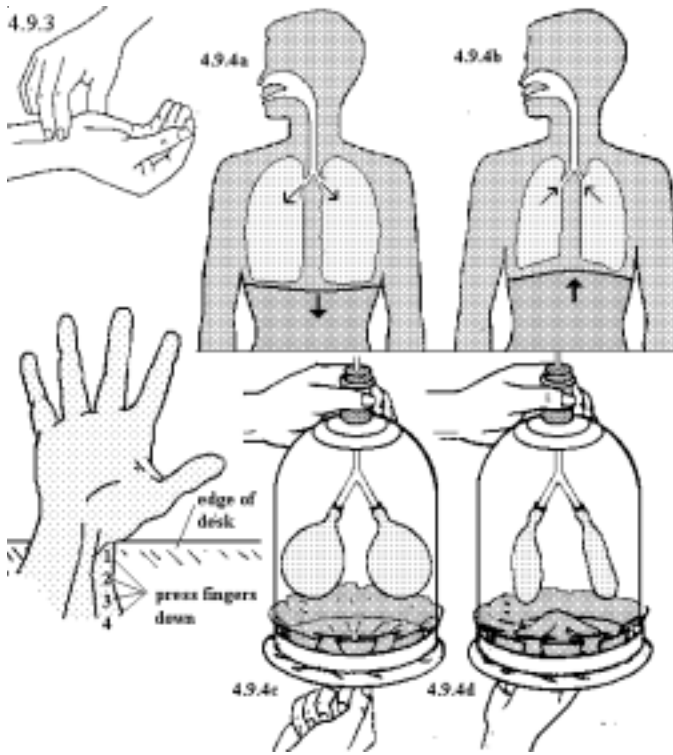

FIGURE 2. An example of respiration and diaphragmatic breathing from http://www.uq.edu.au/_school_science_lessons/BioPHYWEan.html

THERAPY SESSION 2

In the second session, the young patient was taught good breathing patterns combined with self-awareness to prepare him to voluntarily control an episodic attack. The diaphragmatic breathing technique allowed the patient to concentrate and focus on his respiration so that he could phonate without undue stress and tension on his laryngeal or thoracic mechanisms. The patient was taught how to concentrate on his respiratory patterns that required him to extend his lower abdomen on inspiration and descend the diaphragm. He was then taught that on gentle expiration, the lower abdominal muscles would support respiration.

In order to ascertain the effectiveness and understanding of this concept for relaxed respiration, the patient was taught how to adduct the vocal folds voluntarily, using the analogy of the strength of the larynx and vocal folds during the act of pushing a heavy object. This was done as negative practice.

THERAPY SESSION 3

Therapy session 3 addressed the use of "wide-open throat" breathing. The patient was taught how to execute diaphragmatic breathing while concentrating on lip seal, the tongue laying flat on the floor of mouth and the muscles of the face and jaw maintaining a relaxed state. Negative practice was carried out by teaching him how to inhale noisily while closing the vocal folds, tensing the muscles of the larynx, thorax, face and back and tilting the head posteriorly.

THERAPY SESSION 4 – 6

Therapy sessions 4 – 6 focused on increasing the patient's self-awareness of his breathing patterns, increasing his rate respiration using the relaxation techniques established in earlier sessions, without the interference of unnecessary muscular tension.

The patient was then advised to begin gentle and low-impact exercises such as walking while using relaxed breathing techniques. This was done to encourage the patient to return to a lifestyle that was as normal as possible, giving consideration to his respiratory requirements. He was advised that should he have an episodic attack, he should stop, sit down to regain his composure and breath slowly to encourage relaxation of the laryngeal musculature. It was suggested that he attempt to walk a little farther each day, progressing from a slow walk to one that was more brisk, to allow for more complex coordination of his respiration.

During each of the sessions, previously taught techniques were reviewed and corrected if necessary. He was constantly reminded that relaxed breathing would become more natural to him once he had mastered the techniques and strategies taught to him and was reassured that he was making good progress in therapy.

RESULTS

As mentioned, speech rehabilitation was conducted over a period of six weeks in addition to home practice. Nasendoscopy to view vocal fold function was carried out twice during the period; once before the third session and the second, during the final session.

The first nasendoscopy revealed that the patient's arytenoid cartilages adducted simultaneously with the true vocal folds during an episode of stridor. Additionally the right arytenoid folded over midline to cover the under lying vocal folds completely.

By the time of discharge from speech therapy, the patient was able to voluntarily produce noisy breathing and then self-correct himself. This was viewed endoscopically to present a clear picture of internal physiology. The patient was also trained to employ effective strategies to monitor, control and manage the physiological and sensory triggers that exacerbated his upper airway attacks. The patient was given a one month review appointment and then a three month follow-up appointment, prior to complete discharge. During his final appointment session, it was reported that although the attacks continued to occur, the frequency and intensity had reduced and that he was able to manage the affect effectively.

DISCUSSION

Historically, management of PVFM has used the behavioral or symptomatic speech rehabilitation approaches, (Martin et al.1987; Pinho et al.1997 and Gallivan et al. 1996), patient and family counseling (Gallivan et al. 1996; Pinho et al. 1997 and Aronson 1990), respiratory-phonatory retraining (Brugman & Simons 1998; Sataloff 1997), psychotherapy (Morrison, Rammage, Emami 1999; Martin et al. 1987; Brugman & Simons 1998), external laryngeal manipulation (Aronson 1990), laryngeal massage (Roy & Leeper 1993) and various other strategies.

Some individuals with PVFM have also been managed with reflux medication (Kaufman 1994), bio-feedback techniques (Brugman & Simons 1998) and even gas inhalation therapy (Reisner & Borish 1995). Although numerous management strategies have been employed in these studies, the current patient was managed mainly by patient education, effective counseling and motivation, respiratory re-training and relaxation techniques alone.

The initial technique of Martin et al. (1987) consists of educating the patient that he/she has a voice condition and that is important to seek therapy. The researchers also suggest that the patient whose physicians assure them that the problems are psychogenic almost automatically resist speech therapy as it is thought to be of little benefit to psychological problems. No referral was made to

the psychologist, in this patient, as it was deemed that the he was progressing satisfactorily with his sessions at speech therapy.

Pertaining to the assessment of the initially reported respiratory difficulty, Martin et al. (1987) reported that many of their patients pointed to the laryngeal region and identified it as the region of distress. The young boy in this research did not indicate such; causes of functional laryngeal or upper airway obstruction viewed as a psychogenic or somatization disorder were reported (Appelblatt & Baker 1981; Starkman & Appelblatt 1984) and as psychogenic stridor, or emotional laryngospasm (Arnold 1973).

Several authors concluded that dyspnea produced by paradoxical vocal fold motion may be increased by stressful situations (Katlan & Ben-Zvi 1985) and precipitated by physical exercises (Kivity et al. 1986) and that it is commonly accompanied by wheezing (Downing et al. 1982; Rodenstein et al. 1983; Christopher et al. 1983). Paradoxical vocal fold movement is also thought by most investigators to be a strong functional component and have reported it as a functional disorder of the larynx that mimics bronchial asthma as a manifestation of a conversion reaction stating that the patients were unable to reproduce their laryngeal motion consciously (Christopher et al. 1983). As these patients displayed symptoms of asthmatics, they were treated with a range of asthma medications, including corticosteroid and bronchodilator medication. Because of these misdiagnoses, the young patient in this study too was inadequately treated with several medications.

In addition to inappropriate inspiratory closure of the vocal folds, closing may also occur during expiration or during the entire respiratory cycle (Kivity et al. 1986). Episodic paroxysmal laryngospasm is usually recognized by a videolaryngoscopic pattern of paradoxical inspiratory adduction of the anterior two thirds of the vocal folds, a posterior, diamond-shaped, glottic chink, and attenuation of the inspiratory flow rates of the flow-volume loop, indicating partial extra-thoracic upper airway obstruction during an attack of stridor or wheezing (Altman et al. 2000). It is episodic in that it is an event or reversible series of events complete in itself, and yet forming part of a larger, uncommon, and disturbing clinical condition.

The young patient in this study showed an episodic and paroxysmal attack that presented an unusual videolaryngoscopic pattern of paradoxical inspiratory adduction showing an atypical movement of the arytenoid cartilages that folded back over the vocal folds, both at the same time or alternating with one another but not systematically. The nasendoscopy conducted for the assessment of the patient's respiration was in line with that of Kellman and Leopold (1982) who recognized that nasendoscopy was a crucial examination under normal physiological conditions as it allows much better assessment of laryngeal behavior.

Treatment of paradoxical vocal fold movement is usually multidisciplinary with the speech pathologist playing a vital role in the management process.

Patients are taught laryngeal relaxation techniques and psychological or stress-related disorders would be managed as well in cases that require the intervention of a psychologist. The combination of these two treatment modalities in concert with an ORL proves successful if the patient accepts the diagnosis (Christopher et al. 1983).

CONCLUSION

Literature on PVFM mainly presents case reports and methods of intervention that contribute to the total management of the disorder. There have been an overwhelming number of researchers who contribute to the understanding of this rare and complex disorder that is so frequently misdiagnosed and mismanaged. It is hoped that as experiences are shared in managing this condition, contributions are made to the greater understanding of its etiology, prevalence and influencing factors. So much can be learned from other professionals involved in the management of PVFM; so much still remains unknown and unexplored.

Speech pathologists who deal with voice disorders must receive in-depth training about upper and lower respiratory systems, laryngeal anatomy and physiology, and the reactions of the respiratory system to irritant substances. However, psychological factors that contribute significantly to the severity of PVFM should also be investigated and understood.

Future studies should pay careful attention to subject selection and provide in-depth investigative reports of every aspect of the individual's life that may aggravate the PVFM. Systematic intervention strategies and techniques should be employed to increase success of management with speech rehabilitation without unnecessary prolongation. Success can surely be achieved with comprehensive management of the patient as a whole by providing both physical and mental support.

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