

CASE REPORT

Multidisciplinary Approach To An Adolescent With Vocal Cord Dysfunction: Case Report

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ÖZET

Vokal Kord Disfonksiyonu (VKD), ses tellerinin uygun olmayan şekilde kapanmasına neden olarak gırtlak üzerinde duyulabilir hırıltıya ve stridora yol açar. VCD'nin etiyolojisi net değildir, ancak psikojenik faktörlerin de kritik rol oynayabileceği karmaşık ve heterojen bir bozukluk olarak görülmüştür. Bu vakada VCD, astım ve gastroözofageal reflü hastalığı olan 14 yaşında adölesan hasta sunulmaktadır. Tıbbi tedaviye ve konuşma terapisine dirençli olan olguda yapılan psikiyatrik değerlendirme, somatik semptom bozukluğu ile uyumlu çeşitli dinamikleri ortaya çıkarmıştır. Bilişsel Davranışçı Terapi (CBT) odaklı 5 seanslık müdahale, semptomların başarıyla giderilmesini sağlayarak psikiyatrik yaklaşımların VCD'nin optimal tedavisinde kolaylaştırıcı bir rol oynayabileceğine işaret etmektedir.

Anahtar kelimeler: Bilişsel davranışçı terapi, somatik belirtiler, vokal kord disfonksiyonu

ABSTRACT

Vocal Cord Dysfunction (VCD) causes improper adduction of vocal cords resulting in audible wheezing and stridor over the larynx. The etiology of VCD is not clear, however, it has been viewed as a complex and heterogeneous disorder in which psychogenic factors might also have a critical role. We present the 14-years-old female with VCD, asthma, and gastroesophageal reflux disease. Her condition was resistant to medical treatment and speech therapy. A psychiatric evaluation revealed that several dynamics consistent with somatic symptom disorder. Cognitive Behavioral Therapy (CBT) oriented 5 sessions of intervention resulted in symptom relief successfully. Child and adolescent psychiatrists can play a facilitating role in the optimal treatment of VCD regarding the multifactorial aspects of the condition.

Keywords: Cognitive behavioural therapy, somatic symptoms, vocal cord dysfunction

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INTRODUCTION

Vocal Cord Dysfunction (VCD) causes adduction of the vocal cords generally in inhalation and sometimes in exhalation producing audible wheezing and stridor over the larynx (1). Patients may also experience shortness of breath and breathy dysphonia mimicking asthma (2,3) Previously, VCD has been described in many names such as Munchausen stridor (Patterson et al., 1974), psychogenic stridor, paradoxical vocal cord motion, episodic paroxysmal laryngospasm, emotional laryngeal wheezing or factitious asthma mostly referring to its psychological origin.

The etiology of VCD is not clear yet, however it has been commonly viewed as a complex and heterogeneous disorder (4). Pathophysiologically proposed subtypes of VCD are somatic (psychogenic) VCD, spontaneous VCD, irritant VCD, and exerciseinduced laryngeal obstruction (EILO) (5). Patients with VCD were diagnosed with many psychiatric conditions including conversion disorder, depression, anxiety disorder, factitious disorder, family conflict, personality disorder, and post-traumatic stress disorder (6) The problems associated with VCD other than psychogenic cause include gastroesophageal reflux disease, laryngopharyngeal reflux, upper airway inflammation due to allergies, sinusitis, or recurrent viral infections, strenuous exercise (7) exposure to irritant fumes (8) and environmental allergens and/or pollutants.

Regarding the psychogenic origin of VCD, a multidisciplinary approach including psychiatric intervention is important for the

successful treatment of the condition (9). Although cognitive-behavioral therapy (CBT) is poorly studied in children and adolescents with VCD, some reports support the effectiveness of short-term CBT (10). We present a juvenile with VCD poorly responsive to speech therapy and medical treatment responding well to additional 5 sessions of CBT.

CASE REPORT

C. is 14 years old female referred to the child and adolescent psychiatry clinic by a pediatric pulmonologist. She had a long history of asthma for 7 years and also had a diagnosis of gastroesophageal reflux disease (GERD). 7 months prior to the referral to a child and adolescent psychiatry outpatient clinic, she had had a severe attack of dyspnea accounted for asthma and respiratory infection. She had been on several medications including corticosteroids due to her resistant condition and got various treatments including botox injection after the diagnosis of VCD. She was on medical treatment with inhaled corticosteroids, long-acting beta-agonist, and proton pump inhibitors for her GERD and asthma. She hospitalized for treatment lasting 45 days. Nonetheless, the condition poorly responded to medical interventions and speech therapy. Although difficult to differentiate, predominantly VCD accounted for the patient's symptoms, and asthma and GERD symptoms were under control with medications.

In the psychiatric interview, she presented with breathy dysphonia, intermittent wheezing, dyspnea, fatigue, and exercise

limitation. She complained of a little bit of discomfort about the noisy breathing due to being noticed by others. She had no history of psychiatric disorders such as anxiety. depression, trauma, etc. After a short period of time starting the school, she had an attack that described as a severe episode of dyspnea and dizziness. After the episode, her school life was disrupted, and couldn't attend school regularly. She was very successful at school and has always been appreciated by her teachers due to her high academic achievements and obedience. She had no family history of psychiatric illness. She was physically active before the symptoms began. In addition, the familiy reported that she occasionally witnessed to her sick grandfather who confined to bed reportedly receiving lots of interest from the family.

During sessions, her mom was a little bit depressive and very anxious about her child's condition. Her speech was usually melodramatic and emotionally intense as well. She was preoccupied with her daughter's symptoms and showing self-sacrificing behaviors. Th mother was seemed mostly convinced by the possible medical causes.

In her mental state examination and general observation, her appearance was cushingoid and obese (>97p). She had whistly, short breathy noises sometimes interfering with her speech, otherwise, the breathy dysphonia was mostly during resting. Her attitude and speech were quite puerile and immature. Her mood was too positive and affection was inappropriate. For example, she didn't show any affective

discomfort about her school attendance and her hospitalization. She could state no idea about the possible causes about the condition. Her mental capacity was quite good however, insight was partial.

She did not meet the criteria of any psychiatric disorder in a semi-structured interview applied via Schedule for Affective Disorders and Schizophrenia for School-Age Children Present Version-DSM-5 (K-SADS-P-DSM-5). Conversion disorder was not also diagnosed according to DSM-5 because of underlying VCD. However, many features of the case suggested the diagnosis as described in Figure 1.

PSYCHIATRIC INTERVENTION

We spared the first visit for a detailed psychiatric assessment and taking the patient's history. The characteristics of the case presented in Figure 1, were in line with psychosomatic disorders. We shared the psychiatric formulation of the condition with the patient and the family. CBT could be the choice of treatment since catastrophizing thoughts related to the condition were standing out for both the patient and the mother. Also, behavioral accommodation possibly leading to secondary gain and avoidant behaviors were the prominent and would-be target of behavioral intervention.



A total of 5 sessions of 50 minutes were carried out by a CBT trainee, resident child and adolescent psychiatrist under supervision. The content of the first session was primarily psychoeducation and providing relaxation techniques such as breathing exercises and progressive muscle relaxation in stressful situations. The psychoeducation included the clarifications about the possible psychogenic origin of the VCD, the possibility of different

medical explanations and results of repetitive medical investigations, psychiatric overlapping of the VCD with conversion disorder, and the possible causes and treatment of the condition. We also introduced the CBT model to the patient and the family to help them relate thoughts about the condition and safety-seeking / avoidant behaviors in particular. 43

In the second session, we focused on unveiling the patient's assumptions and automatic thoughts. Although the patient was not as anxious as her mom, she was experiencing expectational anxiety of having an attack of breathlessness. We defined cognitive assumptions such as "if I exercise, I will be dyspneic and have an attack" or "if I go to school, I will be helpless" and automatic thoughts such as "I am going to faint" when she experiences dizziness. We made a brief CBT formulation shown in Figure 2 and, worked on the effect of safety/avoidant behaviors like withdrawal from physical activities and unnecessary emergency visits through Socratic questioning, although the emotional component was not potent as might be expected in conversion disorders. Then we designed behavioral experiments to test assumptions starting from the least anxiety-provoking situations.

Since family dynamics played an important role in the case, we reserved the 3rd session for parents. We worked on the value and the effects of a set of maintaining factors belonging to the family such as mother's excessive worry and comforting behaviors, symptoms, possible attention over the secondary gains especially over the sick role, overinvolvement, accommodation, and selfsacrificing behaviors mainly via psychoeducation. The family and the mother in

particular actively participated in the process and followed the formulation which in turn led them to be assured about underlying psychiatric dynamics. We planned the first 3 sessions weekly, then in order not to be a part of repetitive medical visits we agreed upon sessions with 1-month intervals. In the 4th and 5th sessions, we mainly went on testing assumptions over safety/avoidant behaviors and designed gradual exposure to avoidant situations. We also aimed for improving problem-solving skills and adaptive coping strategies in stressful situations. She was already able to be physically more active, less concerned about attacks, and more independent of the mother. After 5 sessions and about 3 months after referral, she came as free of complaints; able to speak fluently without being interrupted by breathy dysphonia and audible wheezing. We agreed upon the termination of the therapy in order not to sustain the sick role after the improvement. We had video recordings of before and after but were not allowed to share as supplementary material by the family. During the therapy process, the patient went on to attend speech therapy and took asthma and GERD medications.

After 6 and 9 months from the discharge, we made 2 telephone calls. Her mother reported that she had no complaints or attacks and going on to take regular asthma and GERD medications for prophylaxis.



DISCUSSION

We present a 14-years-old female with VCD, asthma, and GER disease. Although all medical conditions might be an important etiological consideration, psychogenic causes may also play a vital role while understanding the multifactorial aspects of the VCD. Psychiatric intervention and CBT, in particular, facilitated the successful treatment of the case.

Literature suggests that patients with vocal cord dysfunction show a pattern consistent with conversion disorder (11) Conversion disorder may result in along with several problems like school absenteeism, poor school performance, and being viewed as sick parents (12), unnecessary medical by investigations and treatments risking children for iatrogenic harm (3). The characteristic of the case had a considerable amount of attribution for conversion and somatic symptom disorders. Brugman et al. (1994) studied 37 children with VCD from 4-19 years old, both inpatients and Their characteristics outpatients. were identified and they found that %68 of the sample were female, 81% were Caucasian, 29 of the patients had comorbid asthma and only 8 had pure VCD. Competitive youngs like high academic achievers and athletes were found at a high percentage, 11% of the sample showed evidence of sexual abuse, and %70 of the patients had dysfunctional families as well (13). Pediatric aged patients with VCD may have comorbid asthma up to %40 (3) as it is in our case. The main characteristics of our case shown in Figure 1 are mostly comparable with the previous literature.

To date, there is still no clearly standardized evaluation and treatment guidelines for children and adolescents with VCD. Several therapeutic approaches such as biofeedback, hypnotherapy personal construct therapy, and patient education were reported to be beneficial in VCD patients especially as an adjunct to speech therapy and treatment of comorbid conditions such as GERD (14). Nevertheless, there is no randomized controlled study exploring the effects of psychotherapy. In a study done with 59 children and adolescents, Maturo et al. (2011) reported that 63% of patients responded well to speech therapy as first-line treatment. A total of 30.5 % of patients had underlying psychiatric conditions. Interestingly, patients whose symptoms were symptomatic at rest had more psychiatric conditions and responded excellently within 3 months to psychiatric interventions. The authors suggested that symptoms at rest could be a harbinger of psychiatric conditions and treatment needs should be conceptualized personally (15). Another study evaluating the effects of 4 sessions of cognitive-behavioral intervention in children and adolescents showed that therapy was related to symptom-specific reduction and improvement in individual control and coping ability associated with symptoms (10). In our case, the patient was also symptomatic at rest, poorly responded to only speech therapy, and responded very well to psychiatric intervention. Further studies are warranted to validate the effectiveness of CBT.

This study has many limitations. First of all, although our treatment plan had a robust base, it wasn't fully structured and we couldn't

be able to document the healing process. Then, we still don't know the long-term effects of the intervention. Along with these, one of the most important considerations might be which components of the treatment were responsible for the outcome has remained unclear.

CONCLUSION

In this case, the medical conditions and psychogenic factors were likely overlapping and the treatment regimen included a multidisciplinary approach. The complex nature of the problem suggests that VCD might not be managed by a single health care provider and instead requires a team of health professionals (1). We suggest a detailed psychiatric evaluation for juveniles with VCD and CBT in suitable cases.

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Disclosure Statement

The authors have no conflicts of interest to declare in connection with this paper.

Ethical Standards and Informed Consent

The authors of this article guarantee that all procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2000. Informed consent was obtained both from the patient and his mother for being included in the study.

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