Review

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An Interesting Model for Functional Neurological Disorders: Integrative Approach to Psychogenic Dysphonia

Fonksiyonel Nörolojik Bozukluklar İçin İlginç Bir Model: Psikojenik Disfoniye Bütüncül Yaklaşım

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ABSTRACT

Functional neurological disorder or conversion disorder, is a mental disorder that manifests itself with neurological symptoms that cannot be explained by a neurological condition or medical condition. Psychogenic Dysphonia is a symptom type of functional neurological disorders. With DSM-5, the diagnostic criteria of conversion disorder have changed and as a result, it is imperative to develop a new perspective on the diagnosis and treatment of conversion disorder. Also recently, functional neurological disease definition has been used instead of the definition of conversion disorder. Thus, an integrative approach was required in the diagnosis, evaluation and treatment of this disorder. Studies on functional neurological disorders have uncovered psychogenic dysphonies. The present review aimed the analyzing the literature to illuminate diagnosis, evaluation and treatment approaches in the patients with psychogenic dysphonies. Psychogenic dysphonia is very interesting in terms of its formation with emotional distress as well as its response to a functional intervention such as standard circular laryngeal therapy. Therefore, psychogenic dysphonia is an interesting model for the diagnosis, evaluation and treatment of functional neurological disorders. As psychogenic dysphonia symptoms and psychological factors mutually affect each other, the combination of voice therapy and psychotherapy for symptoms seem to be the gold standard treatment for now.

Keywords: Psychogenic dysphonia, functional neurological disorder, conversion disorder

ÖZ

Fonksiyonel nörolojik bozukluk ya da konversiyon bozukluğu, nörolojik veya tıbbi bir hastalıkla açıklanamayan nörolojik belirtilerle kendini gösteren bir ruhsal bozukluktur. Psikojenik disfoni ise fonksiyonel nörolojik bozuklukların bir belirti tipidir. DSM-5 ile birlikte, konversiyon bozukluğu tanı kriterleri değişti ve bunun sonucu olarak konversiyon bozukluğu tanı ve tedavisine yeni bir bakış açısı geliştirmek şart oldu. Ayrıca, son zamanlarda konversiyon bozukluğu tanımı yerine fonksiyonel nörolojik bozukluk tanımlaması kullanılmaya başlandı. Böylece bu bozukluğun tanı, değerlendirme ve tedavisinde bütüncül yaklaşımlar gerekliliği doğdu. Fonksiyonel nörolojik hastalıklarda yapılan çalışmalar çoğunlukla psikojenik disfonileri içermemektedir. Bu derlemede psikojenik disfonili hastaların tanı, değerlendirme ve tedavisine yaklaşımları literatür ışığında tartışmak amaçlamıştır. Psikojenik disfoniler, oluşumunda emosyenel stresin rolüne rağmen standart sirkümlaringeal terapiye yanıt vermesi açısından ilginçtir. Bu nedenle foksiyonel nörolojik bozuklukların tanı, değerlendirme ve tedavisinde ilginç bir modeldir. Psikojenik disfoni semptomları ve psikolojik faktörler karşılıklı olarak birbirini etkilediği için ses terapisi ve psikoterapi kombinasyonu en iyi tedavi gibi görünmektedir.

Anahtar sözcükler: Psikojenik disfoni, fonksiyonel nörolojik bozukluk, konversiyon bozukluğu

Introduction

Functional neurological disorder (FND) or conversion disorder is characterized by neurological symptoms that are unexplained by neurological conditions or other classical medical conditions. FND was previously referred to as conversion disorder but together with The Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5), it was called FND (APA 2013). In DSM-5, this disorder is included under the heading of somatic symptom and related disorders. However, it has some differences from other somatic symptom disorders. Symptoms in FND are incompatible with any recognized neurological condition (APA 2013). DSM-5 emphasizes that physician must provide evidence that physiological function is preserved despite presence of neurological symptoms. DSM-5 diagnostic criteria also include the requirement that clinical signs and symptoms are not fully compatible with a neurological disease and general

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Introduction

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Disability in functional neurological disorders are almost similar to structural diseases. More than 50% can not recover within 8 months (Stone et al. 2003). In patients with unexplained, atypical neurological disease symptoms, clinicians should be vigilant for FND.

Studies have shown that 78% -93% of FND patients are women (Tomasson et al. 1991, Deka et al. 2007). More than 2/3 of these patients have comorbid psychiatric diagnosis (Singh and Lee 1997, Carson et al. 2000).

Psychogenic dysphonia (PD), is a vocal cord dysfunction that is not a structural or neurological laryngeal disorder. Usually, patients with PD are observed to have an acute onset, and most of them describe stress related problems (Andresson and Schalén 1998, Baker 2002). Psychogenic aphonia is a type of conversion disorder in which the ability to make sounds disappear. In psychogenic mutism, the patient does not make an effort to talk. Approximately 5% of psychogenic dysphonies are cases of psychogenic mutism (Andresson and Schalén 1998, Baker 2002). In psychogenic aphonia, the use of the volunteer sound is affected and there is no articulation. But vegetative vocal functions such as coughing, laughing, sighing, or crying are generally not affected (Van Houtte et al. 2011, Davids et al. 2012). PD was under the motor symptom or deficit type of conversion disorder in DSM 4 (as aphonia). However, in DSM-5 it took place under speech symptom type as dysphonia (APA 2013).

Emotional distress can produce pseudoneurological symptoms and this has been known for many years. But neurobiological signs have recently been investigated for FND (Becker et al. 2013). Neurobiology of FND is not fully illuminated. But volumetric imaging and functional studies of the brain, physiological testing have started to provide evidence (O'Neal and Baslet 2018).

The hypothesis, which argues that the FND is caused by the disruption of the network in the brain, is actually related to the emotional process. In animal models and human imaging studies, high and uncontrolled stress exposure was associated with severe neuronal response in the prefrontal cortex and amygdala (Arnsten 2015). Stress-related disorders have been associated with emotional disregulation and reduced control of the prefrontal cortex over the amygdala (Etkin and Wager 2007, Stark et al. 2015). The amygdala is associated with emotional perception and the prefrontal cortex is associated with emotional regulation. In addition, amygdala-prefrontal cortex cycle is the core interaction in the emotional process network (Etkin et al. 2011).

Although we have more data on functional neurological diseases, there is limited information about the diagnosis and treatment approaches in psychogenic dysphonia. Therefore, integrative approaches to psychogenic dysphonia, which is an interesting model for FND, are discussed.

Integrative Approaches in Diagnosis and Evaluation

Diagnosing FND is based on the incompatibility of neurological and other symptoms with organic disease (Stone et al. 2005). But in the integrative approach, the diagnosis is made after examining the psychological factors together. Factors predisposing, precipitating, and perpetuating to FND should be determined (Baslet et al. 2016). Today, there is a consensus that it is not sufficient to exclude organic diseases for the diagnosis of FND, and that psychiatric evaluation with a multidisciplinary approach is also essential (Espay et al. 2018, Baker et al. 2021).

Misdiagnosis rate of FND is about 4% (Stone et al. 2005). Tezcaner et al (2019), have reported that the time from onset of symptoms to diagnosis can continue to 7-90 days in the patients with PD. In the literature, some clinical features that distinguish patients with functional voice disorders from organic diseases during examination have been described by other disciplines (Chung et al. 2018). For example, acute onset, variable symptom severity during different speech examination activities, suggestibility, symptom improvement with distraction, potential for rapid reversibility or improvement of symptoms, paradoxical increased muscle contraction with sound fatigue, denial or indifference to abnormal speech/voice are among these features. Especially, it is frequently emphasized that there is a sudden onset in psychogenic voice disorders (Chung et al. 2018). Sometimes PD is accompanied by other functional voice disorders (Andresson and Schalén 1998, Baker 2002). Usually, PD starts acutely and there are stress-related problems in the past (Andresson and Schalén 1998).

Diagnosis of PD is difficult and detailed organic examination is required for diagnosis and differential diagnosis (Tezcaner et al. 2019). But it is not enough to exclude organic diseases for diagnosis. For example, videolaryngoscop can not be of differentiating psychogenic dysphonia from other functional dysphonias. In psychogenic dysphonia, there may be similar findings with other functional dysphonias (such as failure

of the vocal folds to adduct, paradoxical movements of the vocal folds, vocal fold bowing, ventricular band) (Baker 2002). Some studies indicate that patients with PD have normal contraction and relaxation of laryngeal muscles during coughs and breathing, and pathological supraglottal contraction during phoning (Spengler et. 2017). However, disease history, course of disease, response to voice therapy and fluctuations through noncommunicative voices are important for diagnosis (Tezcaner et al. 2019). Muscle tension dysphonia, hyperfunctional voice disorders, presbylarynx, vocal fold atrophy should be considered for the differential diagnosis of PD (Van Houtte et al. 2011, Davids et al. 2012).

Diagnosis should be made after the patient is evaluated by both otolaryngologist and psychiatrist. Eventually, diagnosis should be made with an integrated multidisciplinary approach (O'Neal and Baslet 2018, Tezcaner et al. 2019). An empathic, supportive approach is essential in the patients with PD. The patient should be interviewed carefully and precisely. The message that there is no underlying cause is wrong. Because, this message may discourage the patient from treatment. Feigning or malingering discourses are dangerous. It should be emphasized that the symptoms are unconscious.

Evaluation of the patient's personality traits also helps to determine the treatment framework. In a recent study on patients with dysphonia, including patients with functional dysphonia, differences in introversion, neuroticism and tendency for activating the behavioral inhibition system were reported in these patients (Josep et al. 2021). Sensitivity characteristics of the patient such as avoidance tendencies, alexithymia and somatoform tendencies for FND should be defined (Beghi 2015, Baslet et al. 2016). Because these are actually the main framework when making a psychotherapy plan.

Long-term follow-up is important in these patients as different functional neurological symptoms may develop later. Explaining that the patient will be followed up for a long time gives the message that the symptoms are important and real. During this follow-up, the possibility of concomitant organic pathology should not be overlooked.

Integrative Approaches in Treatment

Recent studies on FND focus on physiological factors as well as psychological factors. Changing diagnostic criteria for FND in DSM-5, emphasizing functional concept and integrating etiological factors recently requires integration in treatment (APA 2013). Despite the psychological etiology in PD the response to standard circular laryngeal manual therapy is good (Roy 2003). This proves the need for an integrative approach in psychogenic dysphonies like other FNDs.

In the treatment of psychogenic dysphonia, it is aimed to restructure the effect of the brain on the vocal cords. PD treatment actually starts with explaining the diagnosis to the patient and accepting the treatment plan by the patient (O'Neal and Baslet 2018). The relationship between psychosocial stress factor and dysphonia symptom should be explained to the patient. The patient may not understand the importance of treatment if the connection between her/his symptoms and psychological state is not explained.

Once the diagnosis is clear, the patient and family should be informed. Even accurate information initiates the therapeutic effect (Stone 2014). It should be noted that FND is common and curable. The family should listen and take care of the patient at other times, not when the symptom appears. When explaining the secondary gain to the patient, it should be stated that the gain in the short term turns into loss in the long term. The relationship between brain, nerve, vocal cord trio in psychogenic dysphonia should be explained. It should be explained that this relationship can be disrupted unknowingly due to stress. The treatment should be described as the retraining of the brain.

Psychotherapy

Cognitive-behavioral therapy (CBT) refers to a series of attempts that share the basic antecedent that mental disorders and psychological distress are maintained by cognitive factors (Hofmann et al. 2012). CBT effect was demonstrated by randomized pilot studies in the treatment of psychogenic nonepileptic seizures, a FND subtype (LaFrance et al. 2014, Goldstein et al. 2010). CBT can be recommended especially for those with episodic symptoms.

CBT includes education about FND and the stress response cycle, attempts for the patient to manage stress and develop new behavioral patterns and helps patients identify and change unhelpful thoughts. It is important to identify and replace thought patterns that strengthen patients' symptoms and do not help (O'Neal and Baslet 2018). Daily vocal exercises such as blowing, whistling, and spelling can be given to the patient. The selfguided CBT booklet has been shown to reduce FND symptoms. CBT treatment workbooks can be recommended to help these patients for understand the diagnosis and develop the necessary skills to overcome FND symptoms (Williams et al. 2011).

Psychotherapy should be explained to the patient how it will help the patient to improve her/his symptoms. This approach can change the patient's 2 false perceptions. First, the patient does not see psychotherapy as meaningless. Second, the patient does not see psychotherapy as a magic wand. The goal of psychotherapy is to change the brain's way of processing information, to reduce the tendency to express stress with physical symptoms, and to develop new patterns of behavior by breaking the unconscious pattern that causes this symptom. Step approach is important in psychotherapy. This approach consists of keeping a diary of the patient's symptoms, performing a chain analysis of the trigger factors and giving strategies to save the patient from pervasive symptoms.

Voice Therapy

The difficulty of talking of the patient with psychogenic aphonia / dysphonia increases functional hoarseness. So it pulls the patient into a vicious circle (O'Neal and Baslet 2018). Actually, the goal of voice therapy is to break this vicious circle. In the past, some researchers have suggested voice therapy as an alternative therapy in patients who do not respond to psychotherapeutic interventions (Kolbrunner et al. 2010). In a more recent study 93.1% of patients with PD have been shown to respond to voice therapy. However, in the same study, the rate of relapse was found to be high in those who received only voice therapy (Tezcaner et al. 2019).

In fact, there are goals similar to psychotherapy in voice therapy. Voice therapy is similar to the behavioral component of cognitive behavioral therapy. The goal in voice therapy is to create a more adaptive voice. In the voice therapy primarily natural voice is created by using noncommunicative voices like clearing the throat, coughing, gargling sounds and laughing. After switching to natural voice, there is gradually transition to speech production. Usually, voice therapy is scheduled in two to four sessions and each session consists of 40 minutes (Tezcaner et al. 2019).

Although voice therapy is effective in PD there is limited knowledge about the long-term success of voice therapy. In a study conducted in patients with psychogenic voice disorder the risk of renewal only in those who received voice therapy was higher than those who received voice therapy together with psychotherapy (Tezcaner et al. 2019). Combination of voice therapy and psychotherapy is the most frequently suggested method for PD treatment (Elias et al. 1989, Maniecka-Aleksandrowicz et al. 2006).

Antidepressant Treatment

Like all FNDs in PD, the risk of komorbid anxiety disorders, depressive disorders, post-traumatic stress disorder and hypochondriasis is high (Hoge et al. 2007, Waller and Scheidt 2004). There are no studies other than a few uncontrolled studies on antidepressant or anxiolytic efficacy in patients with FND. Only no randomized placebo-controlled research has demonstrated efficacy of some antidepressant for FND (Voon and Lang 2005). Therefore, if there is a comorbid diagnosis, antidepressant can be started. The antidepressant effect in PD patients without a comorbid psychiatric diagnosis is an issue that needs to be questioned. Eventually, antidepressants should not be initiated in a case without a comorbid diagnosis in terms of rational drug use.

Other Treatment

The effect of hypnosis on FND has been demonstrated in randomized controlled trials (Moene et al. 2002). Treatment of psychogenic movement disorder which is FNB with short term psychodynamic psychotherapy is found effective a randomized clinical trial (Kompoliti et al. 2014). Other uncontrolled studies in FNDs include mindfulness based psychotherapy (Baslet et al. 2015), inpatient programs (Kuyk et al. 2008), psychoeducational interventions (Zaroff et al. 2004), and transcranial magnetic stimulation over the motor cortex (Chastan and Parain 2010).

Conclusion

Despite the importance of psychological factors in PD etiology, the fact that it responds to a functional intervention and at the same time decreases renewal with psychotherapy supports the importance of integrative approach in these patients. On the one hand, this makes PD an interesting model to show the link between chronic stress, emotional dysregulation and functional neurological loss. Like all FNDs, an integrated multidisciplinary approach to treatment is essential in PD. Mental health professionals should be in collaboration with otolaryngologist, both during the differential diagnosis and during the treatment phase. In order to prevent unintended diagnosis and treatment interventions, the diagnosis should be shared with otolaryngologist when it is clear. Approaching the problem only psychologically or only functionally makes diagnosis and treatment difficult. Consequently dysphonia symptom and psychological factors affect each other mutually. Integrated therapy using psychotherapy and voice therapy together is the gold standard approach for PD therapy.

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