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Case Reports

Primum non nocere: Endotracheal Intubation for Prolonged Functional/ Dissociative Seizures in an Adolescent

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Abstract

Functional/dissociative seizures (FDS) are one of the most common manifestations of functional neurological disorder (FND) in both adult and pediatric populations. FND and FDS result in significant healthcare utilization and costs, often due to excessive medical interventions and workup. Previously regarded as a diagnosis of exclusion that required extensive testing to identify, FND and FDS are now more accurately understood as "rule-in" diagnoses, based on the presence of characteristic positive clinical findings. Early identification and evidence-based therapies can facilitate patient recovery. However, patient outcomes continue to be negatively affected by clinicians' limited familiarity and comfort with this diagnosis. We describe the case of an adolescent with FDS who was intubated and admitted to the intensive care unit for presumed convulsive status epilepticus, and who has since had resolution of FDS after treatment by an interdisciplinary team. Our goal is to highlight the need for increased training of healthcare personnel in the recognition and management of this important clinical entity to facilitate early diagnosis, improve clinical outcomes, and potentially decrease healthcare utilization.

BACKGROUND

Functional neurological disorder (FND) has been defined as "a neuropsychiatric disorder involving aberrant changes within and between neuron-glial networks as well as complex interactions between brain, mind, body, and context".[1] The pathophysiology of FND is complex, involving hyperactivity of the limbic system and impaired sensory integration in terms of predictive coding and bottom-up processing of sensory inputs.[2] Functional/dissociative seizures (FDS), also known as psychogenic non-epileptic seizures, are a common type of FND affecting adults and children.[3] The annual incidence of FDS in children ranges between 7.4 and 18.3 per 100,000.[4-6] However, this is likely an underestimation.[7] As awareness of FDS increases, these numbers may continue to grow. In non-adult populations, the incidence of FDS is highest among individuals between 11 and 19 years of age.[5,6] Environmental factors can play a role in its development. For instance, increased incidence of FND was reported in relation to the COVID-19 pandemic and its impact on mental health.[8,9]

Non-epileptic events account for approximately 15-30% of pediatric admissions to epilepsy monitoring units. Among non-epileptic events, FDS are the most common type, particularly in older children and adolescents.[10,11] FND and FDS are associated with significant healthcare utilization and costs.[12] Approximately 20% of pediatric patients with FDS are readmitted or re-present to the emergency department within 30 days.[13] Although there are limited data for pediatric populations, FDS in adults are associated with increased morbidity and mortality.[14] Individuals with FDS can be misdiagnosed as having convulsive status epilepticus, leading to intubation and other iatrogenic complications.[15-18]

Despite its clinical relevance and considerable healthcare utilization, clinicians often show limitations when it comes to identifying and communicating a diagnosis of FND or FDS to pediatric patients and their caregivers.[19] Moreover, research surrounding the optimal management of pediatric FND and FDS remains limited. To the best of our knowledge, only one randomized controlled trial on the management of FDS in children has been published.[20,21] To illustrate some of the challenges in the clinical diagnosis and management of FDS, we describe the case of an adolescent who was intubated and admitted to the intensive care unit (ICU) for presumed convulsive status epilepticus. Our goal is to highlight the need for

increased training of clinicians, particularly first-line providers and hospitalists, in the recognition and treatment of FND.

CASE PRESENTATION

A 14-year-old gender-nonbinary individual had an event concerning for seizure while undergoing a scheduled outpatient electroencephalogram (EEG) at a community hospital. This EEG was ordered for the evaluation of multiple episodes of impaired awareness that had been occurring over the two preceding weeks. During hyperventilation, a common EEG activation procedure, the patient became dizzy, was unresponsive to verbal stimuli, and then had an event described as asynchronous and vigorous body jerking while keeping their eyes forcefully closed. Real-time review of the EEG by a neurologist was not available. EEG recording was terminated, and the patient was transferred to the emergency department, where multiple intravenous doses of lorazepam and levetiracetam were administered. Over the next several minutes, the patient had multiple seizure-like events without returning to neurological baseline. The patient was intubated for presumed convulsive status epilepticus. Vital signs, basic toxic-metabolic workup, non-contrast head CT, and electrocardiogram were all normal. This excluded differential diagnoses such as cardiac arrhythmias and intracranial space-occupying lesions. The presentation was not consistent with processes such as syncope, migraine, tics, or panic attacks

The patient's past medical history included Lyme disease 3 years prior resulting in residual facial palsy; inpatient psychiatric hospitalization followed by partial hospitalization for depression, anxiety, and suicidal ideation two years prior; and concussion 6 months prior. While the patient was bullied in middle school, they had recently started 9th grade. They had a new group of friends and participated in different extracurricular activities. They followed regularly with a therapist and with a psychiatrist. Home medications included sertraline, guanfacine, hydroxyzine, and melatonin. The patient had no recent systemic illnesses, and there was no family history of neurological disease.

After intubation, the events stopped. The patient was transferred to the ICU. Shortly after arrival, and upon discontinuation of anesthetics, they were able to follow commands and were extubated to room air. They continued to have numerous episodes consisting of stiffening and asynchronous shaking of both upper and lower extremities, pelvic thrusting, and forceful eye closure. Shaking was suppressible when painful stimuli were applied. These events, which typically lasted for periods of 10 minutes or longer, were captured on continuous video-EEG, which confirmed the diagnosis of documented FDS.[22] The EEG was normal, with no focal or epileptiform abnormalities. Child neurology and psychiatry were consulted. The patient and their family were receptive to the diagnosis of FDS. Prior to discharge, FDS had decreased in frequency significantly. The patient was discharged to a medicine/psychiatry partial hospitalization program. During this admission, they continued to show FDS with variable semiologies including upper and lower extremity twitching, eyelid fluttering, dissociation, unresponsiveness/staring, and falling to the ground. They also exhibited episodic motor and vocal functional tics. Throughout their partial hospitalization, they received physical and occupational therapy, as well as individual, group, and family psychotherapy multiple times per week. They practiced identifying triggers and warning signs for FND. In addition, they worked on thinking and behavioral strategies including relaxation and calming strategies, adaptive emotional expression, cognitive behavioral therapy, and dialectical behavior therapy-based techniques to rewire overloaded and misdirected neural pathways that led to FND symptoms. Decompressing and redirecting these neural pathways through the aforementioned methods were the primary goals of treatment, leading to a reduction in FND symptoms overall.[23] Since discharge from the partial program, they have gained a full understanding of their diagnosis, have been following regularly with their neurologist, psychiatrist, and therapist, and have been seizure-free for over two years.

DISCUSSION

Unfortunately, the case that we describe is neither unique nor exceptional. Although healthcare professionals from a variety of fields (pediatrics, emergency medicine, hospital medicine, neurology, mental health, rehabilitation, among others) encounter patients with FDS with relative frequency, their recognition and management remain a challenge and, as exemplified here, some patients may undergo intubation and ICU admission.[15-17]

This case illustrates key interventions that can facilitate timely and accurate diagnosis of FND and FDS. These include: adopting a "rule-in" model by using positive clinical signs, educating clinicians, implementing clinical pathways, and improving communication with patients. The first one relates to the diagnosis of FDS. Prior to the publication of the Diagnostic and Statistical Manual of Mental Disorders 5th Edition (DSM-5) in 2013, FND was a diagnosis of exclusion that could be made if all the available neurological tests were normal and there was a link to a psychological stressor. After publication of the DSM-5, FND became a "rule-in" diagnosis, based on the presence of typical semiology and/or findings on physical examination.[24] Our patient's semiology exhibited numerous features that were consistent with FDS, which unfortunately went unrecognized by first-line providers (EEG technologist, nurses, emergency physicians). These included forceful eye closure, pelvic thrusting, asynchronous limb movements, suppressibil-

ity when external stimuli were applied, long duration (>3 minutes), and fluctuating (waxing/waning) course. Other "rule in" features, although not present in our patient, include ictal crying, preserved awareness during bilateral motor activity, and absence of postictal confusion.[24] Moreover, the patient's events were precipitated by a standard EEG activation procedure.[25] As previously noted, FDS may be more common in adolescents than in other age groups.[4-6] Transgender/gender diverse individuals[26-28] and people with psychiatric comorbidities[25,29] may show increased prevalence of FND and FDS.

It has been described that the ability to accurately diagnose FDS varies by specialty and level of training. One study that used videos of teenagers and adults with seizure-like events found that 88% of epileptologists and 73% of general neurologists correctly differentiated FDS from epileptic seizures, whereas only 44% of emergency physicians and 54% of adult internists were able to make the correct diagnosis.[30] Although the shift in the way FND is diagnosed took place a decade ago, most first line providers still do not feel comfortable with the diagnosis and initial management of FDS. Since many patients with FDS are first evaluated by non-neurologists, education efforts targeting internists and hospitalists are critical.

Potentially, optimization of clinical processes and pathways could have prevented intubation and ICU admission in this patient. For instance, if the clinician who requested the initial outpatient EEG for the evaluation of episodes of impaired awareness had indicated in the referral order that there was a suspicion for FDS, the EEG technologist's approach could have been different when dealing with an acute clinical event of this kind. Real-time review of the EEG recording by a qualified EEG reader, which was not available in this case, could have shown that the episode was consistent with FDS, potentially avoiding the need for transfer to the emergency department, administration of antiseizure medications, and endotracheal intubation. These medical interventions are not benign and can be traumatizing with lasting psychological and/or physical impacts for patients and families. The International League Against Epilepsy has published consensus guidelines for FDS in children.[25] Clinicians taking care of these patients should become familiar with these guidelines.

FDS are occasionally misdiagnosed as status epilepticus. On the other hand, misdiagnosis of status epilepticus as FDS is considered to be rare but can occur, especially in cases presenting with atypical semiologies or in patients with coexistence of both epileptic seizures and FDS.[31, 32] Multimodal approaches that integrate careful assessments of seizure semiology and timely access to EEG can minimize misdiagnosis.

Effective strategies to deliver the diagnosis of functional/dissociative seizures (or FND in general) center on clarity, validation, and partnership. The diagnosis should be introduced when FND is high in the differential rather than after completion of all neurodiagnostic testing. The clinician should explain the positive clinical signs that allow them to make the diagnosis of FND in a given patient. For FDS in particular, the clinician should point out signs like eye closure, pelvic thrusting, disorganized movements during the episode, prolonged duration of the event, etc. This allows patients to understand that their symptoms are authentic and disabling, developing trust in the clinician and the diagnosis. After delivering the diagnosis, it is key to emphasize that FND/FDS is treatable and that complete remission can occur. The clinician should explain that future reevaluation may be needed as comorbidities or new symptoms may occasionally appear. The diagnosis should be framed within a biopsychosocial context so that patients and families can appreciate the multifactorial nature of this condition and understand the need for a tailored, multidisciplinary management plan.[24,33,34]

Over the past decade, several studies have been published regarding the management of FDS in children. Most of these studies used multidisciplinary clinics that provide confirmation of the diagnosis and psychological, physical, and occupational therapy for the management of FDS.[25] The framework of these programs is the biopsychosocial approach to the treatment of FND. Complete resolution of symptoms has been reported in up to 63 to 95% of pediatric patients included in these types of programs,[35] as was the case for our patient. For FDS, cognitive behavioral therapy and the use of other psychological techniques are key. These focus on patient education, identification of triggers, development of skills for detection and control of functional seizures. Plans should be tailored to each individual patient. Techniques such as attention redirection and promoting adaptive coping strategies are emphasized.[33,34,36]

Increased training of clinicians, including hospitalists, in the recognition and treatment of FND and FDS is critical to reduce potentially harmful, unnecessary, and costly medical interventions.[18] Early diagnosis and treatment with multidisciplinary programs increase the likelihood of favorable clinical outcomes. Nevertheless, there is still a paucity of high-quality research evaluating different approaches for the management of FND and FDS in children.[25]

Disclosures/Conflicts of Interest

None

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