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Case Report

Effects of vagus nerve stimulation in a patient with temporal lobe epilepsy and Asperger syndrome: Case report and review of the literature

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Abstract

Seizures are a common comorbidity of autism and occur in as many as 30% of patients. This case report describes a 23-year-old man diagnosed with both Asperger syndrome and bitemporal epilepsy. The patient had behavioral regression that correlated with worsening of his intractable seizures. He subsequently underwent implantation of a vagus nerve stimulation therapy device for his refractory epilepsy. Both his seizures and his behavior were monitored for 6 months. We describe the efficacy of vagus nerve stimulation therapy in reducing seizure severity as well as improving the behavioral components of his Asperger syndrome. We also review the current literature regarding epilepsy in autistic spectrum disorders.

Keywords: Vagus nerve stimulation therapy; VNS; Asperger syndrome; Autism; Epilepsy; Seizures; Temporal lobe; Developmental delay

1. Introduction

Asperger syndrome (AS) is a complex neurobiological disorder that falls within the autistic spectrum disorders. Although patients with AS have normal intelligence and language development, they exhibit autistic-like behaviors. Their difficulties with social and communicative skills are complicated by their obsessive personalities, which often present as full-blown obsessive–compulsive disorder (OCD). With voices inclined to be flat and emotionless, these patients often display repetitive behaviors and speech that are characterized as very self-centered. Patients with AS are also very sensitive to sensory input such as sounds, smells, tastes, and touch [1].

Despite the known association of epilepsy with autism, little is known about the long-term outcomes of patients with both disorders or the effects of epilepsy on the behavioral components of autism [2]. A study by Danielsson et al. [3] showed that cognitive level and adaptive behavioral level were lower in patients with both epilepsy and autism compared with those with autism alone.

Vagus nerve stimulation (VNS Therapy, Houston, TX, USA) has been proven to reduce the frequency and severity of pharmacoresistant seizures in epileptic patients. Although the evidence is limited, preliminary studies also indicate that VNS Therapy may improve neurocognitive performance and quality of life among autistic patients receiving it [4]. To our knowledge, no observational studies of both AS and epilepsy have been reported.

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2. Case presentation

A 23-year-old man was diagnosed with both AS and pharmacoresistant temporal lobe epilepsy. The patient was admitted repeatedly to an inpatient psychiatric service for increased obsessive–compulsive behavior and agitation.

He was born after a full-term pregnancy without any complications and developed normally until age 1, when he had his first seizure, which was thought to be febrile in nature. Soon after, he began having multiple generalized tonic–clonic (GTC) seizures. He was treated with phenobarbital, which provided good seizure control until he was in the seventh grade, when he developed multiple seizure types, which were intractable. He continued to have GTC seizures, but he also developed complex partial (CP) seizures during which he would smack his lips, stare into space, and grunt. His CP seizures would often develop into secondarily generalized seizures. His EEG indicated that he had bilateral temporal lobe epilepsy.

Throughout his life, the patient has experienced some developmental delay. His social interactions were markedly impaired, as were his affective expressions. His movements were stereotyped and repetitive, and his range of interest was restricted. He was described as an average student with exceptional performance in the areas of math and spelling, and he graduated from high school. He worked in the family business as a file clerk and had held other part-time jobs including trash collector for the city. He was unable to keep jobs because of disagreements with co-workers and his obsessive behaviors.

His obsessive–compulsive tendencies, especially those associated with obtaining and collecting coins, money, cheese, and trash, worsened. At the age of 19, he was diagnosed with AS. He had a history of violence toward family members and others, especially when they tried to interfere with his rituals and obsessions.

The patient was referred to our neurology service for evaluation of his seizure medications because of excessive seizure activity, between 50 and 100 seizures per month. A review of his past anticonvulsive medications revealed that multiple medications had been ineffective. These medications included phenobarbital, carbamazepine, phenytoin, gabapentin, topiramate, levetiracetam, lamotrigine, and felbamate in various combinations. Evaluation for epilepsy surgery determined that the patient was not a suitable candidate because of the multifocal etiology of his seizures. The patient had not previously been evaluated for VNS Therapy, and he had not tried a ketogenic diet. Considering the patient's irregular and obsessive eating habits, the ketogenic diet was determined not to be a suitable option.

In January 2005, the patient was implanted with the VNS Therapy Pulse Generator and Leads. Initiation of VNS Therapy was delayed secondary to some minor postoperative medical complications. In April 2005, VNS Therapy was initiated with low baseline settings and slowly titrated upward while his seizures and behavior were monitored on a monthly basis. The output current was initially set at 0.25 mA and incrementally titrated upward monthly to 1.25 mA. The signal frequency remained 30 Hz throughout the observation period. The pulse width was set at 250 microseconds for the first 4 months and then increased to 500 microseconds in Month 5 and 750 microseconds in Month 6. The "signal on" time was initially set at 30 seconds, and, in Month 4, it was increased to 60 seconds. The "signal off" time was initially set at 5 minutes, and was titrated downward to 3 minutes in Month 2 and decreased to 1.8 minutes in Month 6.

During the 6 months after the initial programming of the device, an overall improvement was observed in this patient, who had no medication changes during this period. The number of seizures decreased by 68.8% to 25 per month in Month 5 (Fig. 1). The duration of the episodes was also reduced monthly, ending with a total decrease in average duration by 83% (Fig. 2).

We used two tools to monitor behavioral changes. The first tool was a modified Yale–Brown Obsessive Compulsive Scale (m-Y-BOCS). We took the baseline scale and added a subsection specific to this patient's symptoms. Both of his parents completed the survey before and 6 months after implantation. Comparing the pre- with postimplantation data, we observed improvement in the Abnormal Nonverbal, Social Interaction, and Emotional areas. The second tool was the Physician Quality of Life Indicators. The physician following this patient reported sustained improvement after the second month of implantation in the areas of Mood and Achievements, in which the patient improved from "poor" to "good." In the area of Memory, the patient improved from "average" to "good" in Month 6. A medication error that lasted 1 week was responsible for skewing the Month 3 data (Fig. 3).

In addition to the measured areas of improvement, the patient's family reported that he had started a new job and was functioning well. He had no further problems with anger or aggressive outbursts and he had begun dating. At an informal clinical follow-up at 12 months, the patient's family reported that his behaviors were "the best they had ever been" and that he had even been able to decrease some of his psychiatric medications.

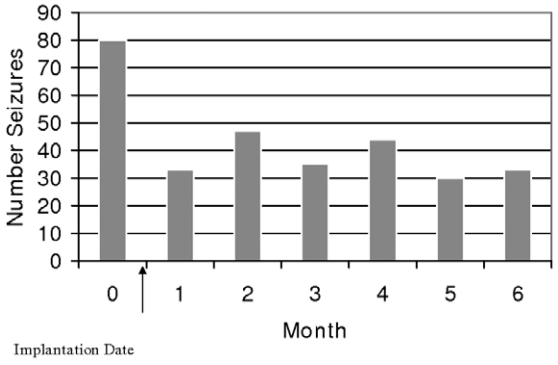

Fig. 1. Number of seizures per month after implantation of VNS device.