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Challenging behavior in adults with epilepsy and intellectual disability: An analysis of epilepsy characteristics

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ABSTRACT

Purpose: The study aimed to describe the frequency and severity of self-injurious, stereotyped, and aggressive/destructive behavior in adults with both epilepsy and intellectual disability (ID) who reside at a tertiary epilepsy center and to investigate the associations between challenging behavior and epilepsy and ID characteristics.
Method: The frequency and severity of self-injurious, (motoric) stereotyped, and aggressive/destructive behavior among 189 patients was assessed using the Behavior Problem Inventory. Comparisons were made with an adult reference population with ID, based on gender, to determine whether the behavior was clinically deviant. Epilepsy characteristics, including age at onset, epilepsy type, seizure types, seizure frequency, and use of antiepileptic drugs (AEDs), were retrieved from patient files. The level of ID was classified using the Diagnostic and Statistical Manual of Mental Disorders — Fifth Edition (DSM-5) and an ID domain discrepancy was allocated if there was a substantial difference between two domains of adaptive behavior within a subject.
Results: Self-injurious behavior was present in 35% of subjects, stereotyped behavior in 60%, and aggressive/destructive behavior in 63%. The behavior exceeded clinical norms in 7%, 18%, and 12%, respectively. Aggression was the behavior evaluated most often as being problematic, despite its reported frequency being the lowest. When adjusting for level of ID and use of psychotropic medication, logistic regression analyses showed that self-injurious behavior was significantly associated with a lower number of AEDs (odds ratio (OR) = 0.4); that stereotyped behavior was significantly associated with a higher number of seizure types (OR = 1.4) and a lower number of AEDs (OR = 0.4); and that aggression was significantly associated with the presence of an ID domain discrepancy (OR = 3.1).
Conclusion: Challenging behavior is a serious issue among adults with epilepsy and ID. Although some of the epilepsy and ID characteristics seemed to contribute independently to these types of challenging behavior, the effects of epilepsy-related characteristics are modest when compared with ID.

1. Introduction

Challenging behavior is a serious concern among people with epilepsy and intellectual disability (ID) [1]. It is defined by Emerson [2] as "culturally abnormal behavior(s) of such an intensity, frequency or duration that the physical safety of the person or others is likely to be placed in serious jeopardy, or behavior likely to seriously limit use of, or result in the person being denied access to, ordinary community facilities". Various types of challenging behavior are encountered in the daily care for this population, such as aggression, self-injury, noncompliance, hyperactivity, and stereotyped mannerisms. These behaviors can result in a fear of harm or actual injury to the person or to others and might have adverse consequences for the individual's development and opportunities for community integration [3].

The prevalence of challenging behavior among people with ID was studied in multiple large population studies, which resulted in point prevalence rates between 10 and 22.5% [4–7]. The prevalence in those with both epilepsy and ID is less well-documented. Two systematic review studies on challenging behavior in this population concluded that people with epilepsy and ID did not clearly exhibit more challenging behavior when compared with those without epilepsy [8, 9] although the results were inconclusive. More specifically, having epilepsy was not associated with aggression, behavioral disturbances, social impairments,

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or challenging behavior in people with ID [10–13]. McGrother et al. [14], however, found higher rates of being uncooperative, seeking attention, and disturbing others at night in people with epilepsy and ID compared with those without epilepsy, after adjusting for gender, age, and level of intellectual understanding. Studies comparing ID populations with and without epilepsy might oversimplify the association between epilepsy and challenging behavior, considering epilepsy is a very heterogeneous disorder with variability in localization, syndromes, etiology, seizure types and frequency, and treatment strategies often including polypharmacy in people with ID.

The literature on the impact of specific epilepsy-related characteristics on challenging behavior in people with ID is scarce. Espie et al. [15] explored associations between epilepsy factors as well as nonepilepsy concerns and challenging behavior and psychiatric symptoms. They concluded that psychiatric symptoms were most strongly related to epilepsy characteristics, such as seizure frequency and severity, whereas behavioral outcomes were most strongly predicted by nonepilepsy concerns, including sensory, intellectual, and motor impairments, as well as adverse effects of drugs [15]. Other studies on behavioral changes associated with antiepileptic drugs (AEDs) show that effects vary among different AEDs, with positive as well as negative effects, although high-quality evidence in people with ID is lacking [1].

The aim of the study was to describe the frequency and severity of self-injurious, stereotyped, and aggressive/destructive behavior in adults with both epilepsy and ID who reside at a tertiary epilepsy center and to investigate the associations between challenging behavior and epilepsy and ID characteristics.

2. Method

2.1. Study design and participants

This study had a cross-sectional design and was part of the TRIANGLE study (The Relation between epilepsy, ID, And Neuropsychiatric comorbidities in a Group of patients in Long-term care for Epilepsy), which was conducted within the tertiary care facility of Kempenhaeghe, the Netherlands. The TRIANGLE study is approved by the medical-ethical committee of Kempenhaeghe (No. 15.01), and the medical-ethical committee of Erasmus University Medical Center concluded that the rules laid down in the Medical Research Involving Human Subjects Act do not apply to this study (MEC-2016-408). The inclusion criteria were as follows: 1) age ≥ 18 years, 2) diagnosis of epilepsy according to the clinical definition by Fisher et al. [16], 3) diagnosis of ID or current adaptive functioning at the level of ID as evaluated by the individual's psychologist, and 4) currently living at the residential care facilities of Kempenhaeghe for at least 1 year. The consent was provided by individuals themselves if they were capacitated, by their legal guardian in case individuals did not have the capacity, or by both the individual and their legal guardian if the individual was capacitated but also had a legal guardian.

2.2. Instruments and procedure

Data on challenging behavior, epilepsy characteristics, and ID were collected using the multiple methods listed below.

2.2.1. Challenging behavior

To assess challenging behavior, the Dutch version of the Behavioral Problem Inventory-01 (BPI) [3] was completed by a professional caregiver who had been familiar with the subject for at least 1 year. The BPI consists of three subscales: self-injurious behavior (SIB; 15 items), stereotyped behavior (25 items), and aggressive/destructive behavior (11 items). Self-injurious behavior was defined as behavior that may cause damage to the person's body and that occurred repeatedly in an unvarying manner (e.g., head-hitting); stereotyped behavior included peculiar or inappropriate voluntary acts that occurred repetitively and habitually (e.g., rocking); and aggressive/destructive behavior referred to deliberate, abusive attacks against others or objects (e.g., hitting others). For each item, the caregiver was asked to evaluate the frequency and severity of the particular behavior in a subject during the past two months. Frequency was rated on a five-point scale (never, monthly, weekly, daily, or hourly) and – if the item occurred at least monthly – the severity was rated on a three-point scale (slight problem, moderate problem, or severe problem). The sum of items yields a continuous (nonstandardized) frequency and severity score per subscale, with a higher score representing more frequent or severe challenging behavior. In addition, whether the scores were clinically deviant was examined by comparing the subject's score with clinical norms of an international population with ID (USA, UK, the Netherlands, and Romania) [17]. A score was considered clinically deviant if it exceeded the mean score plus 1.5 standard deviation (>93rd percentile) of the adult group with the corresponding gender.

The BPI is found to have good psychometric properties [3, 18]. Reliability analyses in this study showed Cronbach's alpha values ranging from 0.684–0.858 (internal consistency) and split-half reliability values of 0.557–0.864. The BPI was also found to have good factor and criterion validity [3].

2.2.2. Epilepsy characteristics

Epilepsy characteristics, including age at onset, epilepsy type and etiology, seizure type, number of seizures (including nocturnal seizures) in the past year, and the use of AEDs, were retrieved from the subject's medical records. With respect to all aspects of epilepsy, the patients are regularly followed up by a neurologist specialized in epilepsy. Seizures were recorded by the direct support staff and relied therefore on direct or secondary observations. Nonepileptic events, such as psychogenic nonepileptic seizures, were excluded. Non-EEG seizure-detection systems were used to detect nocturnal seizures if applicable. The epilepsy type was classified according to the most recent classification system by the International League Against Epilepsy (ILAE) [19].

2.2.3. ID

Regarding the ID, there were two variables of interest: overall level of ID and ID domain discrepancy. The level of ID was based on the three domains of adaptive deficits as described in the Diagnostic and Statistical Manual of Mental Disorders — Fifth Edition (DSM-5) [20]: the conceptual, social, and practical domains. Each domain was assessed separately. The conceptual domain was assessed using a psychological test in combination with an expert opinion by the subject's psychologist. The psychological test applied was either a 4-subtest version of the Wechsler Adult Intelligence Scale — Fourth Edition (WAIS-IV) [21], in case of expected mild to (high-) moderate level of ID (n = 79), or the Peabody Picture Vocabulary Test — Third Edition (PPVT-III) [22], in case of (low-) moderate to severe level of ID (n = 57). The remaining 53 subjects were classified by expert opinion of the subject's psychologist. The WAIS-IV short form was validated among people with neurological disorders and impaired intellectual functioning [23]. The PPVT-III is a measure of receptive vocabulary and is considered a valid screening tool for global cognitive functioning [24, 25]. The social and practical domains were assessed using the corresponding Daily Living Skills and Socialization subscales of the Vineland-II Expanded Interview Form [26] (Dutch translation by Dijkxhoorn and Verhaar [27]), which were completed on all subjects.

The results for each domain were converted into a classification of mild, moderate, severe, or profound deficits. Internationally used cutoff points, described by the Diagnostic and Statistical Manual of Mental Disorders — Fourth Edition (DSM-IV), the International Statistical Classification of Diseases — tenth edition (ICD-10), and Vineland II, were applied, all using cutoff points of 70–50/55 for mild deficits, 50/55–35/40 for moderate deficits, 35/40–20/25 for severe deficits, and below 20/25 for profound deficits [26, 28, 29]. The lower-end values were applied. An ID profile was considered to be discrepant when there was a

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