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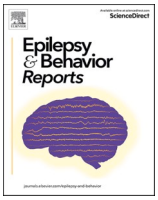

Experimental assessment of seizure-like behaviors in a girl with Rett syndrome

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ABSTRACT

Contextual events are recognized to affect seizure-like behaviors, yet there is limited research on procedures assessing contextual control. This study aimed to examine the utilization of a brief experimental precursor functional analysis within a clinical team assessment. Furthermore, the study explored if telehealth supervision could guide a parent administered replication of the functional analysis. The participants were a young female with Rett syndrome and a history of epilepsy as well as non-epileptic seizures and her mother. The functional analysis procedures consisted of the systematic alternations of contextual conditions that were hypothesized to either prevent or evoke seizure-like behaviors. The primary outcome measure was the occurrence of behavioral precursors that were identified to consequently signal subsequent seizure-like behaviors. In addition, procedure fidelity and interobserver agreement data were obtained alongside parent rating of the procedure's social validity. The clinical functional analysis clearly suggested that the seizure-like behaviors served the function of access to attention and preferred activities. A parent administered functional analysis replicated clinical functional analysis findings. The parent's fidelity to procedures was high and scores in social validity were excellent. The results show that functional analysis procedures could provide essential information in assessment of non-epileptic seizures. Strengths and limitations are discussed.

1. Introduction

Rett syndrome (RTT) is a severe neurodevelopmental disorder that almost exclusively affects females [1]. It is caused by mutations on the MECP2 gene [2] and has a prevalence of approximately 5–10 in 100 000 females [3]. The syndrome is frequently linked to severe cognitive, communicative, and motor impairments, with a primary diagnostic criterium being a period of regression [4]. Typically, after a period of developmental stagnation, there is a loss of previously acquired hand skills and communicative abilities. In this regression phase, stereotypical hand movements and episodes of screaming and crying may emerge alongside malfunctions in autonomous respiratory and vasomotor system. Between the ages of 2 and 10, the syndrome enters a more stable phase characterized by severe and multiple disabilities wherein some social and communicative behaviors may emerge. For some individuals, these challenges remain relatively stable throughout their lives, while others may encounter further declines in their motor abilities [5].

Alongside syndrome specific disabilities, epileptic seizures are a common feature in RTT patients. A register study found reports of epilepsy in approximately 68 percent of 1248 RTT patients [6]. However, there are indications of seizure-like behaviors being misinterpreted as epileptic seizures. Glaze and coworkers [7] found that among 82 individuals with RTT, 23 of 28 cases of parent reported seizures had no correlation with electroencephalogram (EEG) recordings. Moreover, when specialists examined parental reports and seizure descriptions, indications of epileptic seizures were identified in only 291 out of the 360 RTT patients who had a previously reported history of epilepsy [8]. Also, a thorough examination of 120 video-recorded seizures involving

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35 patients with various difficulties, made it evident that the non-epileptic seizures frequently bear a striking resemblance to authentic epileptic seizures [9].

Estimates of population incidence of non-epileptic seizures range from 1,4 to 4,9/100 000 persons per year with highest prevalence in young adults [10]. However, children are also affected. During 1996 to 2014 the incidence was 2,4/100 000 person years in children aged five to 17 [11]. Approximately 10 percent of affected individuals display non-epileptic seizures in comorbidity with intellectual disabilities [10].

In the absence of epileptiform activity, the occurrence of seizure-like behaviors implies other etiological considerations. Cognitive integrative models describe non-epileptic seizures as automatized manifestations of mental seizure representations, possibly with the function of regulating sympathetic activity and emotions [12]. Current conceptualizations, such as psychogenic non-epileptic seizures (PNES) and functional seizures, consider non-epileptic seizures as maladaptive coping with external or internal stressing events. Furthermore, these concepts reflect the theoretical underpinnings that emphasize the mediating role of individual vulnerability in complex interplay of inner events, such as atypical sensory processing and malfunctioning in neurological systems, that result in maladaptive coping with internal and external events [13]. Consequently, as the behavior analytic perspective emphasizes the observable contextual influence on behaviors, rather than causal or mediating events within the individual, the term seizure-like behaviors will be used throughout the current article.

Literature reviews indicate that situational events, sometimes referred to as contextual factors, within the individual's immediate environment are a common feature that might account for seizure-like behaviors, see for example Leibetseder and colleagues [14]. The influence of contextual factors on seizure-like behaviors appears to be particularly strong in individuals with intellectual disabilities [15]. For example, van Ool and associates [16] found that nursing staff reported that stressful events such as overstimulating situations, unpredicted events, and high demands triggered seizure-like behaviors in 13 of 15 patients with intellectual disabilities, and that some patients displayed seizure-like behaviors only at certain times of day or in specific places. Moreover, van Ool and colleagues [16] found that seizure-like behaviors tended to increase as epileptic seizure frequency decreased. This resembles findings in a study including 20 individuals with seizure-like behavior and epilepsy wherein two individuals with intellectual disabilities along with early-onset epilepsy developed seizure-like behaviors as a means of securing attention from caregivers or exerting control over various aspects of their immediate social environment [17].

Although the importance of contextual factors is recognized, there is a shortage of research investigating the impact of the individual's immediate context on seizure-like behaviors. Nevertheless, nearly five decades ago, Iwata and Lorentzson [18] found that increasing daily activities in conjunction with differential reinforcement of other behavior and withholding attention upon seizure onset, resulted in a decrease of weekly occurrences of seizure-like behaviors from over 10 occurrences to less than one in an adult male with ID. More recently, seizure-like behaviors in an adult male with intellectual disability were hypothesized to serve various functions, including obtaining access to food, and eliciting caregiver attention. In certain situations, these behaviors also enabled the avoidance of physical exercise. Following the implementation of function-based behavioral interventions, which included verbal antecedent cues and differential reinforcement procedures, the number of days without seizure-like behaviors increased from a mean of 1.75 per week to 4.75 [19].

DeLeon and colleagues [20] employed a procedure known as functional analysis to explore the influence of contextual factors on seizure-like behaviors in a male with ID. The results of the functional analysis informed the programming and implementation of a treatment package of positive reinforcement strategies that effectively reduced seizure-like behaviors, as well as additional problem behaviors.

Functional analysis of behavior is experimental in nature and consists of systematic and repeated presentations of contextual stimuli (i.e., independent variables) to determine which stimuli may lead to the target behavior (i.e., dependent variable). The procedure was originally developed by Iwata and colleagues [21] who demonstrated diverse situational functions of self-injurious behaviors in nine children and adolescents, all of whom exhibited some level of developmental delay. Since then, a substantial amount of research has proven functional analysis procedures effective in identifying functional relationships between contextual factors and behavioral topographies, in several populations and settings, and various adaptations of functional analysis have evolved towards less intrusive practices [22]. Brief functional analysis entails fewer and shorter sessions [23] and when observational data is clear and unambiguous, a single test condition can be interchanged with a control condition in a single function analysis [24]. Also, in recent years, numerous studies have demonstrated the efficiency of telehealth supervised parent administered functional analysis [25]. Another adaptation is to identify less severe behaviors that precede and signal target behavior occurrence and to utilize such antecedent behaviors as proxies for the target behavior in precursor functional analysis [23].

In busy clinical settings, adapted functional analysis procedures may provide sufficient data to inform intervention plans that target specific contextual factors to reduce seizure-like behaviors and ultimately increase the individual's quality of life. Therefore, this study was designed to examine whether a brief functional analysis of precursor behaviors could contribute to the understanding of seizure-like behaviors in a young female with Rett syndrome. In addition, the study explored the feasibility of telehealth technology supervision of a parent administered brief functional analysis.

2. Materials and methods

2.1. Participant and case description

A mother – child dyad was recruited as participants. The child was a six-year-old female diagnosed with Rett Syndrome and profound intellectual disability. Her communicative repertoire was limited to eye-gaze and generalized expressions of liking or disliking via body language, facial displays, and vocalizations. The young female's mother was recruited to administer an additional functional analysis. Both parents were informants on the project's social validity. None of the participants had any previous experience of behavior analytic procedures.

During the young female's first years of life two distinct patterns of seizures emerged. One pattern of minor and relatively frequent episodes of absence, stumbling and sudden head droppings. A second pattern constituted more prominent and long-lasting seizures characterized by a forward leaning body posture, high tension in legs and hip, tremor and jerks in lower limbs that seemed to cause great discomfort and pain. Several EEG exams indicated epileptiform activity but only limited correspondence with overt seizure behaviors. A combination of anti-seizure medications (i.e., Levitiracetam, Valproic acid, and Lamotrigine in dosages 61 mg, 29,5 mg, and 4 mg per kilo bodyweight respectively) eventually decreased episodes of absence, stumbling and head droppings. However, the second pattern of seizure-like behaviors remained at a relatively steady rate of approximately four episodes daily.

The clinical assessment team that consisted of a neurologist, an orthopedist and a physiotherapist ruled out epilepsy and concluded that the seizure-like sequences culminated in tensor fascia latae (i.e., a thigh muscle) sliding over greater trochanter (i.e., a protrusion located at the top of the thigh bone) causing intense vibrations that sometimes produced a popping sound. A potentially painful phenomenon known as snapping hip. In addition, a parent interview using a Swedish version of The Functional Assessment Interview form [26] alongside direct observations indicated that the seizure-like behavior sequence served two functional categories, (a) escape from potential unpleasant situations and (b) access to parent attention and preferred activity. Furthermore,

the assessments revealed a sequence of specific behaviors that seemed to precede the seizure-like behaviors. These precursor behaviors were treated as proxies for seizure-like behavior in subsequent functional analysis.

2.2. Measures

The main outcome measures were occurrence and latency of precursor behaviors operationalized as the young female displaying a distinct facial expression of scowling with down turned mouth and initiating a forward leaning body posture most commonly in parent's general direction. To minimize risk for potential discomfort the predictive value of the precursor behaviors was not further analyzed.

Procedure fidelity for the parent administered functional analysis was measured by scoring parent conducting procedure activities as either yes or no.

Inter-observer data was obtained by two independent raters that reviewed video recordings and scored occurrence and timing of precursor behaviors for clinical, and parent administered functional analysis and the procedural fidelity of the parent administered functional analysis.

Social validity data concerning importance of selected target behavior, procedure acceptability, and satisfaction of outcomes were collected from both parents via ratings of agreement to six statements on a Likert scale.

2.3. Brief functional analysis

Escape function was observed in physical exams and due to the young female's substantial need for assistance, it was not feasible to separate the contextual events of attention and preferred activities. Consequently, we established a single test condition where there was no access to activities or tangible items and the mother being present but providing minimal or no social attention. In contrast, the control condition involved the mother providing full access to an iPad with a highly preferred animated movie and offering frequent social attention. The functional analysis was conducted at the clinic, in a room with minimal competing stimuli. The procedure was completed in one session, beginning and ending with prolonged control conditions. Due to miscalculation of response latency the initial programmed 30 s condition duration was replaced by ad hoc decisions of when to end control condition and present test condition. After a period of non-occurrence of precursor behaviors in the control condition (duration from 35 s to 2 min and 25 s), the test condition was presented. Whenever precursor behavior occurred, the test condition interval was terminated, and the control condition was reinstated. A parent administered replication of the brief functional analysis was conducted in the family home, in a room with few competing stimuli and included the same conditions as described above. To establish a more robust experimental methodology, the control condition and test conditions were programmed to last for 3 min. However, the test condition was terminated immediately if the precursor behavior occurred. Prior to the in-home functional analysis, the mother underwent telehealth administered behavior skills training, targeting skills such as identifying precursors and alternating test and control condition intervals according to the functional analysis design. The training consisted of two video sessions and included verbal and written instructions and roleplay with feedback. Behavior skills training was provided by a behavior analyst that was blind to the outcome of previous functional analysis.

2.4. Data analysis

A non-parametric test, Tau-U [27] calculated in an online calculator was employed to control for unwanted phase trends in interval durations in control and test conditions respectively (https://singlecaseresearch.org/calculators/tau-u). Inter-rater reliability was computed as percentage of agreement and Cohens' Kappa by the formula k = (Pr(a) − Pr(e))/(1 − Pr(e)) in Excel where Pr(a) and Pr(e) are probabilities based on observations and based on chance, respectively.

3. Results

As displayed in Fig. 1, precursor behavior in the clinical brief function analysis consistently occurred in test condition intervals and were consistently absent in all control condition intervals. In the parent administered functional analysis the response pattern resembled that of the clinical functional analysis in that occurrence and non-occurrence of precursor behavior was dependent upon the condition.

Table 1 exhibits interval durations for clinical and parent administered functional analysis. Statistical analysis revealed no problematic within condition patterns, there were no significant trends in control condition interval durations (column 1 and 3 of Table 1) in neither clinical functional analysis (Tau = .16; p = .53; CI 90 % −.60 < >.27) nor parent administered functional analysis (Tau = .2; p = .62; CI 90 % −.47 < >.87). Analysis of test condition interval durations (column 2 and 4 of Table 1) found no significant trends in clinical functional analysis (Tau = .14; p = .62; CI 90 % −.61 < >.33) or in parent administered functional analysis (Tau = .33; p.49; CI 90 % −1 < >.47).

Parent ratings of social validity were consistent and indicated high relevance of target behavior, procedures being acceptable and producing satisfactory outcomes. The overall percentage of inter-rater agreement was substantial with a mean of 93.3 % (range 89 %–94 %) with Cohens' Kappa inter-rater reliability of k = .88 and k = .78 for clinical and parent administered functional analysis, respectively. Procedural fidelity of parent administered functional analysis was 92 % with an inter-rater reliability of k = .78.

4. Discussion and conclusion

The initial functional interview and observations identified possible contextual factors evoking a seizure-like behavioral sequence that consequently preceded the snapping hip. Within this study the contextual factors were refined and assessed in a brief functional precursor analysis that clearly demonstrated experimental control over precursor behaviors, leading to the conclusion that the seizure-like behaviors served the functions of gaining access to preferred activities and attention. In addition, the brief functional analysis data proved reliable when replicated in a parent administered functional analysis.

Seizure-like behaviors are recognized to be influenced by contextual factors, and in that sense they do not differ from other problematic behaviors. Contextual factors can relatively easily be manipulated and rearranged to explore potential eliciting and/or reinforcing effect on behaviors. For this purpose, an experimental functional analysis is considered the gold standard. Consequently, whenever seizures show no corresponding epileptiform activity in EEG exams, behavior analytic expertise should be consulted to provide behavior assessments and functional analytic procedures in further clinical assessment.

Seizure-like behaviors occurred at a rate of approximately four episodes per day. However, when tentative contextual factors and possible precursor behaviors were identified the rate could be increased through repeated and relatively rapid manipulation of contextual factors, hence making the behaviors subject to meaningful measures and analysis. Refining and applying relevant independent variables are a central feature of functional analysis and through comprehensive preceding behavioral assessments, functional analysis procedures can be designed to investigate contextual influence on problematic behaviors that occur at high rates, as well as behaviors occurring only a few times per week or month.

The functional analysis procedures employed in this study were conducted for less than 30 min each. Besides being effective, the limited assessment time, and the utilization of precursors as proxies for seizure-like behavior, minimized the risk of the individual experiencing

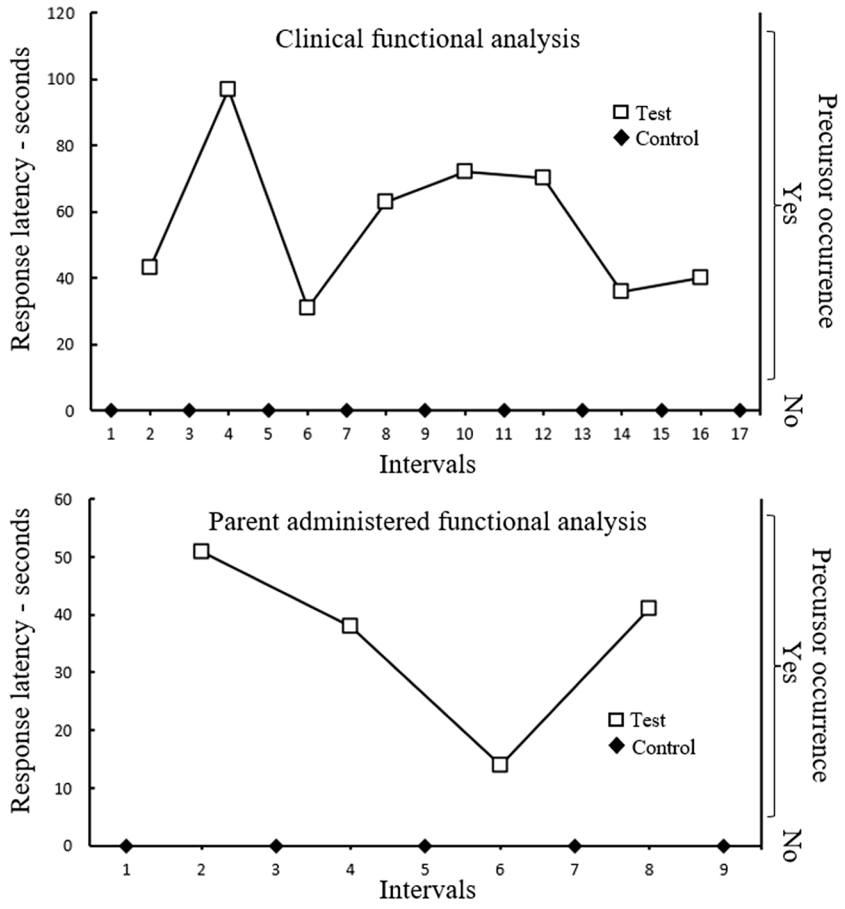

Fig. 1. The left y-axis shows Response Latency of Precursor Behavior for both Test and Control Conditions. The right y-axis exhibits Prcence and Absence of Precursor Behavior for both Conditions. Top graph exhibits data from Clinical Functional Analysis and the bottom graph shows data from Parent Administred Functional Analysis.

Table 1
Interval duration for clinical, and parent administered functional analysis.

Clinical functional analysis		Parent administered functional analysis	
Condition	min:sec.	Condition	min:sec.
Control	02:17	Control	03:20
Test	00:43	Test	00:51
Control	01:13	Control	03:53
Test	01:37	Test	00:38
Control	00:51	Control	03:19
Test	00:31	Test	00:14
Control	00:39	Control	03:20
Test	01:03	Test	00:41
Control	00:45	Control	03:43
Test	01:12		
Control	00:35		
Test	01:10		
Control	00:40		
Test	00:36		
Control	00:49		
Test	00:40		
Control	02:25		

discomfort. Also, after considering ethical aspects of the functional analysis procedure we concluded that withholding access to attention in test condition resembled mild versions of everyday interaction sequences (e.g., delay of social attention when parent being busy talking on the phone or preparing dinner). At the same time, the immediate attention along with access to tangibles provided at occurrence of precursor behavior constituted an increased access to reinforcing stimuli. Furthermore, the functional analysis data directly informed antecedent strategies that, according to anecdotal parent reports, resulted in an immediate decrease of seizure-like behaviors to a near zero level.

In addition, this study, as well as previous research, indicate that behavior analytic expertise can utilize telehealth technologies to provide caregivers with sufficient skills to conduct functional analysis procedures in home settings. Obviously, this adds further accessibility to specialist care.

4.1. *Strengths and limitations*

Functional analysis is the gold standard in assessing problematic behaviors and at the same time, the embedded replications of condition alternation (i.e., ABABA…) within functional analysis procedures is experimental in nature. Hence, the procedures yielded robust data that corresponded with clinical priorities. Another strength was the twofold payoff from the parent administered brief functional analysis. This procedure replicated the clinical findings and simultaneously provided support for making specialist services available via telehealth technologies.

There are some limitations to this study. In the clinical functional analysis four of the eight test condition intervals (A), were surrounded by control condition intervals (B) with shorter durations (for details, see column 1 and 2 of Table 1), leaving room for time to be a causal factor. However, precursor occurrence was fully dependent on the manipulation of the independent variable in the clinical functional analysis, and we argue that sufficient control was established with multiple withdrawal sequences (i.e., ABA ABA ABABA). Furthermore, a systematic assessment of the precursor behavior's predictive value would have added to the validity of the functional analysis outcome. Also, effects of

function-based interventions are typically used to further evaluate the data obtained in functional analysis and such data would have added valuable information. Finally, the generalizability of findings in this study is limited.

4.2. Implications for future research

Future research should further explore the inclusion of in-clinic as well as caregiver administered functional analysis procedures in assessments of seizure-like behaviors. Also, a next step could be to evaluate validity of the functional analysis through research on subsequent interventions informed by the specific functional analysis data. Moreover, future research should explore generalization of functional analytic procedures to other rare genetic syndromes and other complex problematic behavior topographies.

4.3. Compliance with ethical standards

Informed consent was collected from the participants and informed consent from the young female was obtained by-proxy through her legal guardians (i.e., both parents). All procedures involving human participants were in accordance with the ethical standards of The Swedish Ethical Review Authority (ethical approval reference number 2020-05867) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

4.4. Potential conflict of interest

First author's work hours were funded by The Swedish National Center for Rett syndrome and related disorders. A public, non-profit, competence and research center within the Swedish health care system. Besides the statement above, none of the authors have any conflict of interest to disclose.

CRediT authorship contribution statement

Magnus Starbrink: Writing – original draft, Visualization, Project administration, Methodology, Investigation, Formal analysis, Conceptualization. **Svein Eikeseth:** Writing – review & editing, Writing – original draft, Supervision, Methodology. **Sigmund Eldevik:** Writing – review & editing, Supervision, Methodology. **Johanna Edervall:** Writing – original draft, Validation, Project administration.

Declaration of competing interest

The authors declare the following financial interests/personal relationships which may be considered as potential competing interests: First author 's work hours were funded by The Swedish National Center for Rett syndrome and related disorders. A public, non-profit, competence and research center within the Swedish health care system. Besides the statement above, none of the authors have any conflict of interest to disclose.

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