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ORIGINAL PAPER

# Increased Exposure to Rigid Routines can Lead to Increased Challenging Behavior Following Changes to Those Routines

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**Abstract** Several neurodevelopmental disorders are associated with preference for routine and challenging behavior following changes to routines. We examine individuals with Prader–Willi syndrome, who show elevated levels of this behavior, to better understand how previous experience of a routine can affect challenging behavior elicited by disruption to that routine. Play based challenges exposed 16 participants to routines, which were either adhered to or changed. Temper outburst behaviors, heart rate and movement were measured. As participants were exposed to routines for longer before a change (between 10 and 80 min; within participants), more temper outburst behaviors were elicited by changes. Increased emotional arousal was also elicited, which was indexed by heart rate increases not driven by movement. Further study will be important to understand whether current intervention approaches that limit exposure to changes, may benefit from the structured integration of flexibility to ensure that the opportunity for routine establishment is also limited.

**Keywords** Resistance to change · Restricted preferences · Preference for routine · Challenging behavior · Temper tantrums · Prader–Willi syndrome

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## Introduction

Individuals with neurodevelopmental disorders including autism spectrum disorder, and several genetically defined disorders such as Prader–Willi syndrome, commonly show a strong preference for routine and predictability (Kuenssberg et al. 2014; Moss et al. 2009). Importantly, this preference for predictability can manifest as challenging behavior following changes to routines or expectations (Gomot and Wicker 2012; Furniss and Biswas 2012; Richards et al. 2010; Sabaratnam et al. 2003; Woodcock et al. 2009a). In Prader–Willi syndrome, this resistance to change is particularly prevalent; and associated with temper outbursts, which have been measured in experimental settings by tracking outburst component behaviors (Oliver et al. 2009; Woodcock et al. 2011). Here, Prader–Willi syndrome is used as a model for understanding the dynamics of the association between changes to routines/expectations and specific profiles of challenging behavior, in this case temper outbursts. This work will inform a broader strategy for the development of intervention approaches targeting difficulties with change experienced by people with neurodevelopmental disorders.

The comparability of the resistance to change in people with Prader–Willi syndrome (PWS), to such behavior in individuals with other neurodevelopmental disorders is supported by research into its cognitive correlates. At a cognitive level, the preference for routine and predictability in people with PWS has been linked to a specific cognitive deficit in task set re-configuration; a component process of task switching/shifting (Woodcock et al. 2009b; Woodcock et al. 2010); which is an important aspect of executive function (Miyake et al. 2000). This relationship also appears to be present in boys with Fragile X syndrome, which has a distinct genetic aetiology (Woodcock et al. 2009b). Whilst there has been some debate on the issue (White 2013), converging evidence suggests that individuals with autism spectrum disorders also show deficits in measures of shifting (Russo et al. 2007). Importantly, performance on shifting tasks has been associated specifically with the repetitive/restricted preferences domain of autism spectrum behavior; a domain which comprises the preference for predictability (D'Cruz et al. 2013; Lopez et al. 2005). These data suggest that the preference for predictability observed across several neurodevelopmental disorders—even those with distinctly different causes and phenotypes—may be associated with the same cognitive features.

Prader–Willi Syndrome is a neurodevelopmental disorder caused by the absence of paternally derived genetic material in the q11.2–13 region of chromosome 15. There is a well characterized physical phenotype (Holm et al. 1993), alongside mild to moderate intellectual disability (Whittington et al. 2004). Temper outbursts are shown by upwards of 80 % of people with disorder (Dimitropoulos et al. 2001; Walz and Benson 2002); and a common trigger for these outbursts is change to routine or expectations (Woodcock et al. 2009a; Tunnicliffe et al. 2014). Some aspects of the phenotypic behaviors evidenced by individuals with Prader–Willi syndrome have been reported to vary across different genotypes that can cause the syndrome (e.g. Butler et al. 2002). However, both of the primary genetic sub-types appear to show similar rates of temper outbursts linked to changes to routines or expectations (Woodcock 2008).

Existing approaches that seek to address resistance to change in individuals with neurodevelopmental disorders frequently aim to increase advance planning and predictability (Mesibov and Shea 2010). The rationale behind such approaches comes from behavioral theory and involves—after having identified changes to routines/expectations as an antecedent for challenging behavior—manipulating the environment in such a way that the frequency of occurrences of antecedents for the behavior is reduced. However, these approaches often result in individuals being exposed to increased repetition of the same sequences of events i.e. routines.

In relating the increased repetition of sequences of events to cognitive theory, such repetition corresponds to infrequent, compared to frequent, required task switches. There is evidence to suggest that the nature of the cognitive demand imposed by switching is different depending on whether such switches occur frequently or infrequently. Thus, while infrequent switches place higher demands on task-set reconfiguration, more frequent switches place higher demands on task-set updating (Monsell and Mizon 2006; Nessler et al. 2012). Further discussion of the intricacies of these specific components of switching is not pertinent here. Nevertheless, these data suggest that, at least in relation to the specific switching deficit in individuals with PWS and Fragile X syndrome (Woodcock et al. 2009b), less frequent switches may place greater demands on this deficient process. This is relevant because there are data to suggest that in some individuals, disruption of a routine that the individual has experienced repeated previous exposure to can trigger challenging behavior, where disruption to a routine to which the person has been recently introduced occasions no behavioral difficulty (Woodcock et al. 2011).

If it is the case that increased exposure to routines results in increased difficulty following changes to these routines, then this would have important implications for the development of intervention strategies. It would imply that antecedent manipulation approaches, which aim to reduce the changes to expectations in people's environments, should also be sensitive to minimizing opportunities for routines to become established. The question also has important implications for potential early intervention approaches. Anecdotally, it has been reported that families of children with PWS who show little resistance to change, also appear to be those who report few opportunities for routines to become established during children's development (Woodcock et al. 2009a, 2011). Mice models have demonstrated that development in a varied environment, in which there is decreased exposure to the same stimuli and events, results in increased cognitive flexibility and reduced behavioral routines (Tanimura et al. 2008). These data suggest that increased exposure to the same sequences of events from an early age could have important potentially negative implications for later cognitive and behavioral functioning.

The primary aim of the present study was to investigate the effect of increasing length of exposure to a routine on challenging behavior following changes to that routine. Importantly, because temper outburst behavior in people with PWS was used as a model for this investigation, it was also possible to investigate the impact of such repeated routines on the physiological correlates of this behavior. Temper outbursts are often defined in relation to associated increases in emotional arousal (Potegal and Davidson 2003). Consistent with this definition, temper outbursts in individuals with PWS comprise consistent behavioral indicators of increased emotional arousal (Tunnicliffe et al. 2014; Oliver et al. 2009). Emotional arousal is associated with increased activation of the autonomic nervous system, which can be indexed by increases in heart rate (Ekman et al. 1983; Rainville et al. 2006; Fernandez et al. 2012). However, heart rate is heavily dependent on physical activity (Iellamo 2001). Thus, here both heart rate and physical activity are measured in order to index changes in emotional arousal following changes to routines to which individuals have been exposed for different lengths of time.

**Table 1** Descriptive information on participants

| | |
|---|---|
| N | 16 |
| Age range (years:months) | 9:7–47:10 |
| Mean age (SD); years:months | 25:0 (13:9) |
| N per gender: males:females | 12:4 |
| N per genetic subtype: mUPD:deletion:unknown | 6:2:8 |
| VABS adaptive behavior: range | 25–95 |
| VABS adaptive behavior: mean (SD) | 64.4 (17.92) |
| VABS daily living skills age equivalent: mean (SD) in years:months | 7:7 (3:2) |

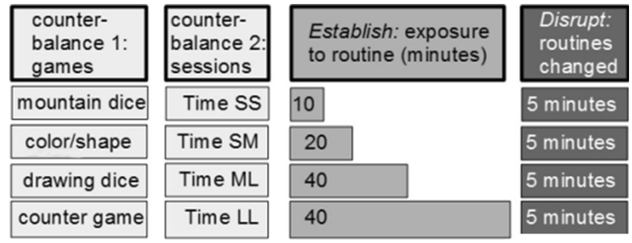


**Fig. 1** Experimental procedure. Time SS, SL, ML, LL are arbitrary labels for sessions comprising an Establish followed by a Disrupt condition, which occurred at different times

It was hypothesized that in a sample of individuals with PWS, increased exposure to a routine will be associated with increased temper outburst component behaviors and increased emotional arousal following changes to that routine.

## Method

### Participants

Ethical approval was obtained from The University of Birmingham Ethical Review Committee. All adult participants and parents of children under 16 years provided informed consent. Children under 16 also provided their informed assent. Participants were recruited from the Prader–Willi Syndrome Association in the UK (PWSA-UK) and from a group of residential homes for adults with PWS. Parents and carers were interviewed via telephone to ascertain the antecedents, component behaviors and consequences associated with the temper outbursts they observe (see *supplementary materials* for the interview schedule). Only individuals who displayed temper outbursts as a result of change to routine or expectation (though not necessarily the only trigger) were recruited. Sixteen individuals took part (Table 1). Vineland Adaptive Behavior Scales (Sparrow et al. 2005) were conducted to assess participants' adaptive behavior level to facilitate comparison with previous and future research.

### Measures

#### *Change Challenge Games*

Four table top games were designed. These games were all novel to participants (see *supplementary materials*); and allowed routines to be established during the course of play. As an illustrative example, one of the games involved choosing cards from a central pile based on rolling a die; selecting counters to discard based on the chosen card; and then discarding the card into a different pile, not to be used again during that round. Thus, one of the routines established was the separation of the already played cards from those still available, and a change to this routine was mixing of an already played card back into the pile of cards still in play.

Change challenges were presented in either Disrupt or Establish conditions. In the Establish condition, routines and/or expectations were followed as expected, thus providing participants with exposure to the corresponding routines without change. In the Disrupt condition, up to five changes (mean: 4.8; SD: 0.65) were imposed on the corresponding routines/expectations (see *supplementary materials* for a full description of these).

#### *Physiological Recordings*

Participants wore a heart rate monitor (Polar RS400; to measure heart rate) and an Actiwatch (AW4, CamNtech Ltd., Cambridge, United Kingdom), containing an accelerometer, which measures activity. The heart rate monitor was worn on a strap around the chest with a watch on the wrist, and the Actiwatch was worn on the participant's other wrist. Heart rate was recorded in average beats per minute (bpm) every second. Activity was recorded as an activity count. The accelerometer in the Actiwatch produces an electric current when movement is detected and the change in voltage is measured as an activity count. Activity counts were recorded in epochs of 10 s (extracted using Actiwatch Activity and Sleep Analysis 7, Version 7.28, CamNtech Ltd).

#### *Procedure*

The experimental design was within subjects: each participant engaged with all four activities, each presented during an Establish condition (which varied in duration across activities), and a Disrupt condition. Thus, the effect of increasing exposure to routine on response to change to a routine could be evaluated for each participant individually. The procedure is summarized in Fig. 1. Participants were assessed during 1 day at their home by a single researcher. Participants were first taught how to play each of the four games during a familiarization period (14–36 min); with matched duration of exposure to each game for any single participant. The purpose of the familiarization period was to ensure that participants understood the rules of the game. During this period, the researcher did not mention winning in order to minimize participants' focus on trying to win.

Participants then took part in Establish and Disrupt conditions in pairs, corresponding to every game. The Establish condition was always presented first; lasting either 10, 20, 40 or 80 min; and was immediately followed by the corresponding Disrupt condition. Disrupt conditions lasted at least 5 min. However, there was some variation across participants in the length of time required to explain or conduct the changes (mean duration: 7 min 23 s; SD: 2:51). Breaks were scheduled between each pair of conditions and no participants asked for breaks at any other time.

Importantly, two aspects of this procedure were fully counterbalanced across participants. Firstly, the game participants engaged in for each of the four possible durations of Establish condition; and secondly, the order with which games associated with each length of Establish condition, was presented to participants. This counterbalancing procedure minimized possible confounding effects of changes in motivation for play as the procedure progressed, and general habituation to changes being conducted by the researcher.

## Behavior Observation

Participants were filmed using a video camera whilst playing the games so that behaviors could be observed and analyzed. Behaviors of interest were temper outburst related behaviors that parents or carers had identified during the interviews (see "Participants"). Behaviors were coded in real time using ObsWin 3.2 (Martin et al. 1998) based on operationally defined categories (e.g. Oliver et al. 2009) for which inter-rater reliability; based on two researchers coding 25 % of each participant's data; demonstrated a Kappa value of at least 0.6 across 5 s time periods (Table 2).

**Table 2** Definitions and reliability of observed temper outburst behaviors

| Behavior | Operational definition | Inter-rater reliability: Kappa |
|---|---|---|
| Questioning | The participant asks the researcher a question related to the game. These could be about the rules/materials/turns. | 0.74 |
| Ignoring requests | The participant does not respond to a verbal request made by the researcher or the participant starts to verbalize about something unrelated to the request. This should be coded until a further verbal response from the researcher (either a further request or a verbalization about something unrelated to the request) or the participant stops ignoring and initiates a response. | 0.88 |
| Arguing | The participant makes verbalizations in the form of statements of disagreement, giving orders or making demands, taken from Oliver et al. (2009). | 0.85 |
| Crying | The participant shows tears or speech or non-speech vocalizations associated with crying, taken from Oliver et al. (2009). | 0.96 |
| Physical aggression | The participant responds with a deliberate act towards researcher or object involving contact that could cause harm or damage. This should also include any missed attempts at physical aggression where no contact is made. | 0.84 |
| Verbal aggression | The participant verbalizes threats or makes hurtful comments towards the researcher. This could also include any offensive language. | 0.97 |
| Gestural aggression | The participant displays a behavior that can be viewed as threatening but involves no contact with the researcher or object, for example pointing. | 0.93 |
| Picking nose | This additional behavior was coded for one participant only as this had been identified by their parents to be a temper outburst behavior. The participant engages in picking nose with fingers or tissue and includes blowing nose and includes eating any mucus from fingers or tissue. | 0.69 |

## Analyses

Analyses were based on mean percentages of time in which temper outburst behaviors were shown, mean heart rates, and mean activity counts; within the relevant conditions (i.e. Disrupt and Establish for the 10, 20, 40 and 80 min routine exposure phases respectively).

Initial inspection of the observational data revealed distributions that significantly departed from normality (Kolmogorov–Smirnov statistic up to: 0.4; with $p < .001$). Thus, non-parametric analyses were employed. Firstly, as an assessment of experimental integrity, a Wilcoxon Signed Rank test was applied (using IBM SPSS Statistics 20 software) to assess the difference in temper outburst behavior in Disrupt relative to Establish conditions. Secondly, the effect of increased length of exposure to a routine on temper outburst behavior following change to that routine was assessed using a Page's Trend test. The Page's Trend test provides a non-parametric alternative to repeated measures Analyses of Variance (ANOVA). Importantly, the approach allows a hypothesis to be tested where the order of the treatments can be predicted (i.e. that more temper outburst behavior will be demonstrated in Disrupt conditions following longer Establish conditions), but the size of the difference between each of the ordered treatments cannot be predicted (i.e. there is no reason to predict that there would be a linear effect of increasing duration of Establish conditions on the temper outburst behavior demonstrated during corresponding Disrupt conditions). The Page's Trend test therefore provides greater statistical power for the present purpose relative to alternative approaches such as the Freidman's test (Page 1963). The test was calculated manually using guidance from Meddis (1975) via the computation of $Z$ scores that provide a measure of effect size in standard deviation units.

One child was not willing to complete more than part of the Establish condition for the first game and thus was not included in the analysis. Two adults did not wish to start/complete the game associated with an 80 min Establish condition; thus these observational data were treated as missing. The missing data were dealt with in a conservative manner by taking the mean of temper outburst behaviors across all other corresponding conditions (i.e. Establish or Disrupt) for the relevant participant. Thus, the value substituted for the missing data could not strengthen the hypothesized effect if it were present (no effect on Type I errors), but dealing with the missing data in this way allowed the power of the test to be maximized (decreasing the likelihood of Type II errors).

Analyses of physiological data focused on a subset of ten participants for whom full heart rate and Actiwatch data were available. Full data were not available for six participants, either because these individuals were not comfortable with wearing the recording equipment or due to technical failure of the recording devices. Only Disrupt conditions were assessed because the relatively long duration of Establish conditions meant that they were highly subject to effects of movement, which would confound differences in heart rate linked to physiological arousal. Because the first change was not imposed immediately upon initiation of Disrupt conditions, data were averaged for each condition over only the middle 80 % of the time period of that condition. The distributions of the resulting mean activity and heart rate values did not significantly depart from normality (Kolmogorov–Smirnov $< 0.211$; $p > .200$). Thus, parametric analyses were conducted because it was necessary to assess both heart rate and activity data to inform on physiological arousal, and the assumed interplay between these two measurements meant that clear apriori directional hypotheses for

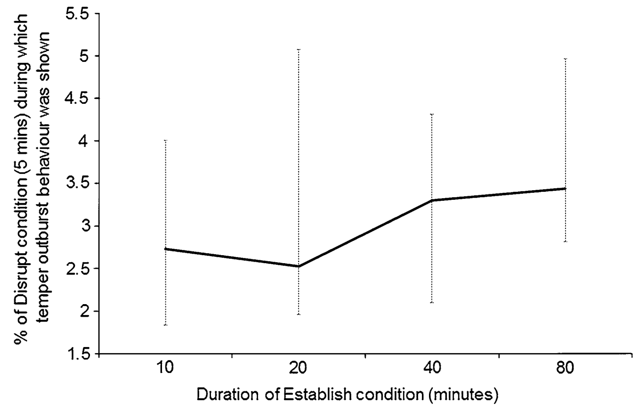


**Fig. 2** The median percentage of 5 min Disrupt conditions during which temper outburst behaviors were demonstrated. *Horizontal dashed lines* represent the interquartile range

both measures (as would be required for a Page's Trend test) could not be made. Thus, repeated measures ANOVAs with a single duration factor, comprising 10, 20, 40 and 80 min levels, were applied to assess the effect of increasing length of exposure to a routine on heart rate and physical activity following change to that routine.

## Results

### Observational Data

Supporting the experimental integrity of the present methods, the mean percentage of time during which temper outburst behaviors were presented across all games was significantly higher during Disrupt conditions relative to Establish conditions (Median Change: 3.31; median No Change: 1.43; Wilcoxon signed rank standardized value: 3.12, $p = .002$; Cliffs $d = 0.43$).

In line with our hypothesis, there was a significant main effect of increasing Establish condition duration on the percentage of time during which temper outburst behavior was demonstrated in corresponding Disrupt conditions ($L = 395$, $p = .038$; $Z = 1.79$; Fig. 2).

However, inspection of the observational data revealed high levels of individual variability (*see supplementary materials for individual participant level data*). Further exploratory analyses revealed that an important factor contributing to the individual variability was the proportion of time participants spent distracted from the game (i.e. not looking at the researcher or the game; or talking about an unrelated topic) during Disrupt conditions. When participants who evidenced higher levels of distraction (20 % or more of at least one Disrupt condition) were removed from the analysis (remaining n = 9), the main effect of duration was stronger ($L = 244$, $p = .029$, $Z = 2.19$); however this effect was not present in participants showing higher levels of distraction ($p > .90$). Overall, participants presently labelled as more distracted demonstrated more temper outburst behavior than those labelled as less distracted, but this effect only bordered significance ($p = .066$). Further, across participants, there was a significant association between increased duration of time distracted and increased total duration of temper outburst behavior (Spearman's $r = 0.52$, $p = .050$). The *supplementary materials* include additional details on these exploratory analyses.

### Physiological Data

Mean physiological measurements across relevant conditions are described in Table 3. The repeated measures ANOVA of heart rate data revealed a strong, significant main effect of duration ($F(3,27) = 3.13$, $p = .042$, $\eta_p^2 = 0.26$). As illustrated in Fig. 3, heart rate was higher in Disrupt conditions associated with longer Establish conditions, relative to that associated with the 10 min Establish condition. However, after applying a Bonferroni correction to the (one-tailed) directional paired comparisons (adjusted threshold: $p < .017$), it was only the increase in heart rate during the Disrupt condition associated with the 20 relative to the 10 min Establish condition, which attained significance ($t(9) = 3.35$, $p = .008$, $\eta^2 = 0.55$). These results are also reflected in the linear increase in heart rate across Disrupt conditions associated with increasing length of Establish conditions, which was of medium size but did not attain significance ($F(1,9) = 0.77$, $p = .404$, $\eta_p^2 = 0.078$). The quadratic main effect was strong and significant ($F(1,9) = 6.86$, $p = .028$, $\eta_p^2 = 0.43$) but this was driven by the larger increase in heart rate in the Disrupt condition following the 20 min relative to the 10 min Establish condition. To assess how far these changes in heart rate could be explained by changes in physical activity, the activity data were assessed in the same way. Here, the repeated measures ANOVA revealed no significant main effect of duration ($F(3,27) = 0.89$, $p = .46$, $\eta_p^2 = 0.090$); nor linear change in activity across Disrupt conditions (negligible linear effect; $F(1,9) = 0.006$, $p = .940$, $\eta_p^2 = 0.001$). Thus, whilst in general increased duration of Establish conditions was associated with increased heart rate in corresponding Disrupt conditions, this relationship was not linear. Changes in physical activity did not appear to drive the relationship.

**Table 3** Mean heart rate and activity counts for Disrupt conditions

| | Duration of associated Establish condition | | | |
|---|---|---|---|---|
| | 10 | 20 | 40 | 80 |
| HR (bpm) | | | | |
| Mean | 74.28 | 81.22 | 78.83 | 76.41 |
| STD | 13.51 | 16.73 | 16.38 | 13.67 |
| Activity (count) | | | | |
| Mean | 56.66 | 69.59 | 55.63 | 60.73 |
| STD | 24.29 | 27.19 | 27.88 | 17.95 |

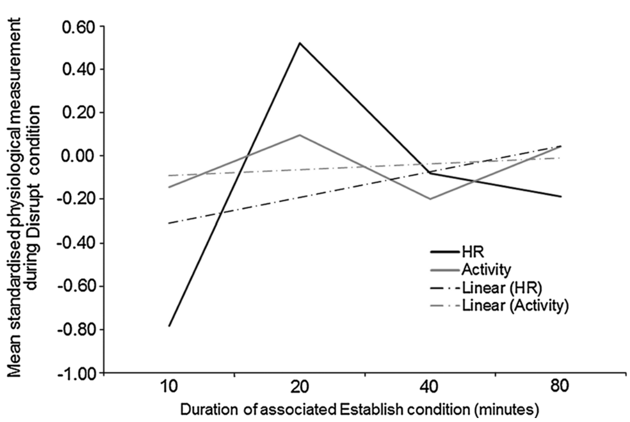


**Fig. 3** Heart rate and activity data in standardized units calculated based on the mean and standard deviation of recordings across all (Disrupt and Establish) conditions for each individual

## Discussion

Using a model of temper outburst behavior in individuals with Prader–Willi syndrome, the results provided support for the hypothesis that increasing exposure to a routine without change would be associated with increased behavioral difficulties following change to that routine. The physiological data provided additional support for this relationship because corresponding increases in heart rate were not driven by increases in physical activity; suggesting that these may have been underpinned by emotional arousal, which has been linked to temper outbursts. However, the effects of increased opportunity for the establishment of routines could not be described with a clear dose-exposure function, emphasizing the need for further research in this area. Our exploratory analyses highlighted additional factors that may impact on the relationship.

To the best of our knowledge, no previous work has directly examined the interaction between rigidity versus flexibility in individuals' environments during development and the behaviors that may be shown by these individuals following changes to routines later in life. However, an association has been demonstrated between rigidity in parent behavior during parent–child interactions and current and subsequent externalizing behavior in children (Hollenstein et al. 2004); and this work has formed the basis of several studies examining the impact of maternal depression on children's externalizing behavior (e.g. Lunkenheimer et al. 2013). In addition, obsessive–compulsive completeness traits—including a preoccupation with things being done in a particular, preferred way—are demonstrated significantly more frequently by parents of children with autism compared to parents of typically developing children; and increasing parental completeness is associated with increased resistance to change behavior in the children (Kloosterman et al. 2013). The causal direction of these relationships is unknown and parent and child behavior is likely to interact at several levels. However, taken together with the present findings, these data highlight an important need for further examination of the effects of increasing environmental rigidity during development on subsequent resistance to change and challenging behaviors following change. A cautionary note here is warranted because there are multiple reasons why increasing structure in children's environments is widely considered best practice in the context of several neurodevelopmental disorders, and the present results do not suggest any contraindication to this approach. In future, careful prospective designs, which work with the variation in standard advice proportioned to the families of individuals with different disorders or in different settings, as well as with individual differences in relevant family characteristics, are needed.

The present findings did not demonstrate a clear exposure–response relationship in the effects on challenging behavior of increasing exposure to routines. Whilst such a relationship was not specifically predicted, its characterization would have provided stronger support for a pivotal role of such exposure to routines. However, findings from several areas (e.g. the impact of environmental risks on children's externalizing behavior) also demonstrate robust associations between factors under investigation and behaviors, in the absence of exposure–response relationships (Donkin et al. 2013; Fraser et al. 2001; Liu et al. 2013; Sinha et al. 2013). Some of the complexity of these relationships is likely to arise because of the multi-level factors that influence behavior. However, also particularly relevant to the present study, is the arbitrary nature of the exposure levels that may be contrasted (e.g. Fraser et al. 2001). In the present study, different durations of exposure to routines were selected on the basis of pragmatic concerns about procedural feasibility. It is possible that there was not enough variability between each pair of durations for a dose-exposure relationship to be identified. Future studies in this area, which contrast routines that have been established over several days, weeks or even months, would be informative in this respect.

Our exploratory analyses identified a behavioral category, labeled here as *distraction*, as an additional factor that can impact the relationship between exposure to a routine and the behavioral response to changes to that routine. Distraction, as defined here, comprised times when participants were not paying active attention to the activities. The effect of increasing exposure to routines remained present, and was stronger, in participants who demonstrated little distraction. However, the effect was not present in those participants who demonstrated relatively high levels of distraction.

One possible conceptualization of distraction is that it indexes times when participants were not "on-task". On-task behavior has been closely linked to effective learning in educational settings and appears to be key to individuals benefiting from the specific features of carefully designed learning environments (Imeraj et al. 2013; Ponitz et al. 2009). Thus, it is possible that individuals who demonstrated high levels of distraction were simply less sensitive to the experimental manipulation. However, this explanation is not consistent with the finding that participants who spent more time distracted also showed more temper outburst behaviors. An alternative conceptualization of distraction is that it represented active attempts at emotion regulation by participants. Such self-distraction has been identified as a strategy, which whilst commonly shown by typical children, varies greatly across individuals in its efficacy for reducing negative emotions (Buss and Goldsmith 1998; Ekas et al. 2011). Thus, one interesting possibility is that the present participants (to differing degrees) engaged in, but were not able to successfully manage their negative emotions using, a self-distraction strategy. These data on distraction are exploratory. However, importantly, they suggest that even in a group of participants with the same genetic disorder, recruited to show a specific pattern of challenging behavior in certain environmental circumstances, differences in how environmental challenges are managed across individuals may still result in different behavioral outcomes. Such individual differences are likely to be important to consider in the design of optimal interventions.

The relationship identified between increased exposure to routines and increased physiological arousal following changes to those routines was also not clear cut, and was primarily driven by the differences in responses to changes to routines that had been established for 20 compared to 10 min. Taken together with the behavioral observation data, these data fit with a behavioral sequence model of temper outbursts. Previous research has demonstrated profiles of behaviors within a temper outburst, which progress in characteristic sequences (Oliver et al. 2009; Potegal and Davidson 2003; Green et al. 2011; Tunnicliffe et al. 2014). Interestingly, behaviors more indicative of increased emotional arousal (such as emotional vocalizations or increased salivation) often occur together either preceding or following more challenging/disruptive behaviors. Thus, it is possible that the present increase in routine establishment from 10 to 20 min effected a small difference in the response to changes (relative to when routines had been established for longer) detectable primarily in the mean heart rate data; whereas with longer exposure to routines, larger responses to changes were observed in overt temper outburst behaviors, and the effect on mean heart rate was less pronounced. One important area for future research will be to better characterize the changes in physiological arousal that occur proceeding, during and following an outburst. This would be best achieved via conjunctive measurement from multiple indices of arousal such as heart rate, galvanic skin conductance and pulse rate.

However, the possible explanation for the observed relationship between routine establishment and emotional arousal discussed above must be considered tentatively. An important limitation with the present method for indexing emotional arousal must be noted. In order to control for the effects of movement on heart rate, activity data from a separate device that comprised an accelerometer were collected. These data suggested that participants' movement was not driving the main effect of exposure to routines on heart rate. However, they did not allow the effects of movement to be removed from the heart rate data. Portable devices are now available which contain an electrocardiogram alongside an accelerometer (e.g. Koehler et al. 2011), and these would provide a purer index of emotional arousal in future related studies.

Some of the primary limitations of the present study have already been highlighted. It is also pertinent to underline the potential limitation associated with the experimental setting used for the present study. Such a setting was chosen because it was reasoned; and has been demonstrated previously (e.g. Woodcock et al. 2011); that environmental triggering events for challenging behaviors are less potent in experimental compared to natural settings. Thus, the study was designed to provide a stringent test of the present hypothesis. Using routines that were completely novel to participants before the study also allowed us to maintain the hypothesis test as stringent as possible. However, the need for studies examining longer and more realistic durations of routine establishment has already been highlighted. In addition, examination of these issues within a more natural environment may highlight other important factors, which were not evident in the experimental setting.

Finally, it is important to end with a note of caution about the generalizability of the present findings. There is fairly compelling evidence, discussed in the "Introduction", that certain similarities exist across individuals with different neurodevelopmental disorders who show challenging behaviors following changes to routines or expectations (specifically with respect to the factors that immediately impact on those behaviors). However, the present study included participants with a single disorder. Future research is necessary to assess how far the findings of the present study are pertinent to individuals with other disorders. Notwithstanding this limitation, it is hoped that the present study will promote much needed systematic research that investigates the long term, prospective impact of environmental rigidity versus flexibility on individuals with neurodevelopmental disorders who show an elevated resistance to change.

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