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RUNNING HEAD: PROBLEM BEHAVIOR IN FRAGILE X AND SMITH-MAGENIS
SYNDROME

An Indirect Examination of the Function of Problem Behavior Associated with Fragile X
Syndrome and Smith-Magenis Syndrome

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Abstract

Fragile X syndrome (FXS) and Smith-Magenis syndrome (SMS) are associated with a number of specific topographies of problem behavior. Very few studies have examined the function served by problem behavior in these groups. Using the Questions About Behavioral Function scale (Matson & Vollmer, 1995) the current study examined group differences in the function of problem behavior displayed by children with FXS and SMS, in comparison to a control group of children with non-specific intellectual and developmental disabilities. Between-group analyses showed children with SMS were more likely to display problem behavior related to physical discomfort. Both within- and between-group analyses showed children with FXS were less likely to display attention-maintained problem behavior. These findings hold implications for the assessment, treatment and prevention of problem behavior associated with both FXS and SMS.

Key words: functional assessment, problem behavior, fragile X syndrome, Smith-Magenis syndrome.

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An Indirect Examination of the Function of Problem Behavior Associated with Fragile X Syndrome and Smith-Magenis Syndrome

Approximately 10% of children with severe intellectual and developmental disabilities (IDD) display problem behavior, such as self-injurious behavior (SIB), aggression or property destruction (Kiernan & Kiernan, 1994). Children who display problem behavior are likely to be disadvantaged across a number of indices of quality of life (e.g., Borthwick-Duffy, 1994; Rusch, Hall, & Griffin, 1986). Such behaviors may also be a considerable source of stress for families and caregivers (Hastings, 2002).

Genetic variables appear to be an important risk factor for the development of problem behavior in people with IDD (May et al., 2009). Indeed, problem behavior is considered to be phenotypic of a number of single gene disorder syndromes associated with IDD. Two syndromes where such gene-behavior associations have been widely investigated are fragile X syndrome (FXS) and Smith-Magenis syndrome (SMS).

FXS is the most common inherited cause of IDD, occurring in 1:3,600 males and 1:8,000 females in the general population (Turner, Webb, Wake, & Robinson, 1996). The genetic locus of FXS lies in a mutation on a single gene on the X chromosome known as the FMR1 gene (Verkerk et al., 1991). This mutation, which consists of an amplification of CGG repeats, leads to hypermethylation of the promoter region of the FMR1 gene resulting in the reduced production of the Fragile X Mental Retardation Protein (FMRP), a protein that plays an important role in regulating brain development (Gothelf et al., 2008). Indeed, FXS appears to be associated with the heightened prevalence of both aggression (Einfeld, Hall, & Levy, 1991) and SIB (Symons, Clark, Hatton, Skinner, & Bailey, 2003). SIB occurs in over 50% of boys with

FXS and is particularly likely to involve hand-biting, although other topographies, such as head-hitting and skin picking also occur at elevated rates (Symons, Clark, Hatton, Skinner, & Bailey, 2003). Certain stereotypical behaviors, such as hand-flapping, also appear to occur at unusually high rates in FXS (Meerenstein et al., 1996).

SMS has an estimated prevalence of 1/25,000, with an equal distribution between the genders (Greenberg et al., 1991). SMS is caused by an interstitial deletion of chromosome 17p11.2, although recent research suggests that haploinsufficiency of the *RAI1* gene is the primary genetic cause of the syndrome (Edelman et al., 2007; Slager et al., 2003). Problem behaviors appear to form a relatively prominent feature of SMS (Clarke & Boer, 1998; Dykens & Smith, 1998). In comparison to other groups, SMS is associated with relatively high levels of aggression, as well as a range of stereotypical behaviors (Dykens, Finucane, & Gayley, 1997; Dykens & Smith, 1998). Estimates of the prevalence of SIB in SMS vary between 67-96% (Dykens & Smith, 1998; Finucane, Dirrigl, & Simon, 2001; Greenberg et al., 1996; Martin, Wolters, & Smith, 2006). Some topographies of SIB, such as onychotillomania (pulling out finger- and toenails) and polyembolokomania (insertion of objects into body orifices) appear to be relatively unique to the syndrome (e.g., Finucane, Dirrigl, & Simon, 2001).

Research on FXS and SMS to date has predominantly examined the form of problem behavior rather than its function. Functional assessment methodologies aim to identify those variables that evoke and maintain problem behavior. Problem behavior displayed by people with IDD has been repeatedly demonstrated to serve an operant function with such behaviors being commonly maintained by socially- and non-socially mediated forms of positive and negative reinforcement (e.g., Hanley, Iwata, & McCord, 2003). These relations appear to influence problem behavior even in cases where those behaviors are recognised as being phenotypic of a

particular syndrome (e.g., Hall, Oliver, & Murphy, 2001; Sloneem, Arron, Hall, & Oliver, 2009; O'Reilly, Lacey, & Lancioni, 2000). It seems unlikely, therefore, that genes have an effect on problem behavior independent of environmental influence (Langthorne & McGill, 2008).

It has been suggested that developmental changes associated with certain genetic syndromes may influence the occurrence of problem behavior by altering the reinforcing value of some of the consequences that commonly maintain such behaviors (Kennedy, Caruso, & Thompson, 2001; Langthorne & McGill, 2008; Oliver, 1993). If, in some cases, genetic events provide some of the 'motivation' for problem behavior then differences (both between- and within-syndrome) in the function served by problem behavior across certain syndrome groups should be expected.

There is some preliminary evidence to indicate that people with FXS and SMS may differ in the probability of displaying problem behaviors that serve certain functions. It appears that individuals with FXS may be less likely to display problem behavior that is maintained by the provision of social attention than would typically be expected and more likely to be maintained by the removal of aversive stimuli, and/or the provision of tangibles (Hall, DeBernadis, & Reiss, 2006; Roberts, Weisenfeld, Hatton, Heath, & Kaufmann, 2007; Symons, Clark, Hatton, Skinner, & Bailey, 2003; Woodcock, Oliver, & Humphreys, 2009). Symons et al for example, using a modified version of the Functional Assessment Interview (FAI; O'Neil, Horner, Albin, Storey, & Sprague, 1990) reported that only 3% of children with FXS displayed attention-maintained SIB. In comparison, 65-87% were reported to display SIB in response to task demands and changes in routine. Others have noted the apparent high levels of social escape and avoidance behaviors associated with FXS (Hall et al., 2006; Roberts et al., 2007).

In contrast several clinical reports and studies of SMS (e.g., Bass & Speak, 2005; Dykens & Smith, 1998; Smith, Dykens, & Greenberg, 1998) have noted the apparently high level of ‘attention seeking’ behaviors associated with the syndrome. A recent study by Taylor and Oliver (2008) involving descriptive functional assessment methods reported that for four out of the five children with SMS in their study, problem behavior was more likely to occur following periods of low adult attention or following reduced levels of demands and was likely to lead to an increase in attention or demands for those same children. Such evidence indicates that attention may hold different reinforcing properties for children with SMS than for other groups, such as children with FXS.

The current study aimed to further this line of research by examining problem behavior displayed by children with FXS and SMS, in comparison to one another and to a control group of children with non-specific IDD. This is the first study of which we are aware to have examined between-syndrome differences in the function of problem behavior. The Questions About Behavioral Function scale (QABF; Matson & Vollmer, 1995) was used to provide a more robust indirect measure of behavioral function.

Method

Participants

Participants were individuals with IDD aged 5-21 years who were reported to display at least one of the following topographical classes of problem behavior: self-injury, aggression or property destruction. Participants belonged to one of three etiological groups: FXS, SMS, or non-specific IDD. There were 34 participants with FXS, 25 with SMS and 30 with a non-specific IDD.

All participants from the FXS and non-specific IDD groups were recruited via relevant parental support groups based in the United Kingdom. Participants from the SMS group were recruited via a combination of the following methods; via parental support groups (N= 5), via regional genetics testing centres (N= 7) and via word of mouth and advertisement in relevant publications (N= 13).

All participants with FXS and SMS had a confirmed genetic diagnosis and evidence of this was requested by the researchers. In cases where this was not forthcoming, parents were asked to provide consent for the researchers to request this information from the child's regional genetic testing centre or paediatrician.

Procedure and Measures

The study received multi-site ethical approval from the National Health Service and from the Tizard Centre Ethics Committee. All participants were sent information about the study and were asked to return a completed consent form indicating a convenient time for the interviews to be conducted and a completed copy of the Aberrant Behavior Checklist- Community Version (Aman, Burrow, & Wolford, 1995). All remaining measures were conducted with parents/caregivers over the telephone. Parents were sent paper copies of all questionnaires in advance and were prompted to have all measures to hand when completing the telephone interview.

All interviews began with the researcher establishing whether this was a convenient time for the interview. If necessary, interviews were rearranged for an alternative time. The Vineland Adaptive Behavior Scale-Screener version (Sparrow, 2000) and the QABF (Matson & Vollmer, 1995) were completed as part of the interview.

The QABF was completed for each general category of problem behavior (aggression, self-injury, and property destruction) the child was reported to display. The QABF provides summary statistics across five functional categories ('attention', 'escape', 'tangible', 'physical discomfort' and 'self-stimulatory'). Participants were asked to rate the extent to which each item applied to their child's problem behavior using a 4-point scale. Total scores for each subscale were then calculated. The QABF's five subscales have been confirmed via factor analysis and the scale has good reliability (Paclawskyj, Matson, Rush, Smalls, & Vollmer, 2000) and predictive and convergent validity (Matson, Bamburg, Cherry, & Paclawskyj, 1999; Paclawskyj, Matson, Rush, Smalls, & Vollmer, 2001). The QABF has been used to measure behavioral function in a number of other large N studies (e.g., Didden, Korzilius, & Curfs, 2007). To prevent individual variation in QABF scale scores being masked by the aggregation of individual scores, the data used in the current study were analyzed categorically.

Various approaches to the categorical analysis of the QABF have been used (see Matson & Vollmer, 1995; Matson & Boisjoli, 2007; Paclawskyj, Matson, Rush, Smalls, & Vollmer, 2001). As many participants were reported to display behaviors that were potentially multi-functional, a relatively high cut-off score of 10 was selected as it is the lowest total scale score requiring that a minimum of 4 items be endorsed.

Statistics

Participant age and severity of problem behavior were analyzed using one-way ANOVAs at a significance level of 0.05. Differences in the gender of the groups was examined using a chi square test at a significance level of 0.05. As the data for the Vineland Adaptive Behavior Scales were not normally distributed a series of non-parametric Kruskal-Wallis tests were used to examine group differences across all sub-domains. To reduce the

probability of making a Type I error by multiple comparisons, $\alpha = 0.05$ was divided by the number of comparisons made, resulting in a significance level of 0.012. Henceforth where the same process has been used to account for multiple comparisons, this shall be referred to as the Bonferroni adjustment. Mann Whitney tests were used to examine post hoc between-group differences on the Vineland using a significance level of 0.016 (Bonferroni adjustment). Between-group differences in the proportion of participants who displayed problem behavior that served single compared to multiple functions were analyzed using a series of chi-square tests at a significance level of 0.012 (Bonferroni adjustment). Within-group differences in the proportion of participants with FXS who met criteria for attention-maintained problem behavior in comparison to other functions were examined using a series of Cochran Q tests at a significance level of 0.016 (Bonerroni adjustment). Within-group pairwise differences were then examined using a series of McNemar tests at a significance level of 0.012 (Bonferroni adjustment). Between group differences in the proportion of participants who met criteria for specific functions on the QABF were analyzed using a series of chi-square tests at a significance level of 0.003 (Bonferroni adjustment).

Results

Table 1 shows participant characteristics for each group. Participants in all three groups were matched for age ($F [2, 85] = 1.195; p > 0.05$), severity of overall problem behavior ($F [2, 85] = 1.065; p > 0.05$) as measured by the Aberrant Behavior Checklist (Aman, Singh, Stewart, & Field, 1985). There were significantly more females in the SMS group than the other two groups ($\chi^2 (2) = 11.505; p < 0.05$). The groups were matched for global levels of adaptive behavior as measured by the screener version of the Vineland Adaptive Behavior Scales (Sparrow, 2000) (χ^2

[2] = 8.05; $p > 0.012$), as well as the Daily Living Skills sub domain: $\chi^2(2) = 5.727$; $p > 0.012$; and the Socialization sub domain: $\chi^2(2) = 1.205$; $p > 0.012$. There were significant between group differences for the Communication sub domain of the Vineland: $\chi^2(2) = 12.99$; $p < 0.012$. Pairwise comparisons showed scores for the SMS group were significantly greater than both the FXS group ($U = 203$; $p < 0.016$), and the mixed etiology group ($U = 157.5$; $p < 0.016$). Differences between children with FXS and the mixed etiology group were not significant ($U = 403$; $p > 0.016$).

A total of 2.9% of participants in the FXS group, 4.0% of those in the SMS group and 20.0% in the non-specific IDD group were reported to present with one topographical class of problem behavior, 51.4%, 17.1% and 36.7% in each respective group presented with two topographical classes of problem behavior and 45.7%, 72.0% and 43.3% in each group presented with three topographical classes of problem behavior.

[Insert Table 1 about here]

Table 2 shows the percentage of participants in each group who presented with behaviors that served a single versus multiple function across each topographical class of problem behavior, as well as those participants in each group for whom no function was identified. There were no significant between-group differences in the proportion meeting criteria for behavioral function for self-injury or aggression, there were, however, significant differences for property destruction ($\chi^2[2] = 19.0$; $p < 0.012$).

[Insert Table 2 about here]

Figure 1 shows the percentage of participants in each group who met criteria for each subscale of the QABF across each topographical class of problem behavior.

[Insert Figure 1 about here]

Examination of the within-group data for participants with FXS suggests the proportion of participants meeting criteria for the attention subscale of the QABF was lower than for other scales. For self-injury, 6.7% of the group were identified as displaying attention-maintained behavior, 43.3% as tangible-maintained, 40.0% as escape-maintained, 30.0% as discomfort-related and 23.3% as self-stimulatory. For aggression, 6.2% were identified as displaying attention-maintained behavior, 46.9% as tangible-maintained, 59.4% as escape-maintained, 21.9% as discomfort-related and 3.2% as self-stimulatory. For property destruction, 8.7% were identified as displaying attention-maintained behavior, 26.1% as tangible-maintained, 30.4% as escape-maintained, 4.3% as discomfort-related and 26.1% as self-stimulatory. These differences were significant for self-injury ($Q [4] = 16.43, p < 0.016$) and aggression ($Q [4] = 43.59, p < 0.016$); however there were no significant differences for property destruction ($Q [4] = 7.89, p > 0.016$). Pairwise comparisons were conducted to test the hypothesis that the proportion of participants meeting criteria for attention-maintained problem behavior would be less than other functions. For self-injury significant differences were found between the attention and tangible subscales ($p = 0.003$) and there was a non-significant trend in the same direction for the attention and demand subscales ($p = 0.013$). For aggression significant differences were found between the attention and tangible subscales ($p = 0.000$) and between the attention and demand subscales ($p = 0.000$).

The between-group data suggest differences in the proportion of participants in each group meeting criteria for attention-maintained behaviors. Less participants in the FXS group met criteria for attention-maintained behavior than in the SMS and non-specific IDD group for self-injury (6.7% Vs. 43.5% Vs. 33.3% respectively), for aggression (6.2% Vs. 62.5% Vs. 32.1%

respectively) and for property destruction (8.7% Vs. 75.0% Vs. 40.0% respectively). Significant differences were found between the groups on the attention subscale for aggression, $\chi^2(2) = 20.3$; $p < 0.003$, and property destruction $\chi^2(2) = 19.7$; $p < 0.003$. There was also a non-significant trend in the same direction for self-injury $\chi^2(2) = 10.2$; $p = 0.006$.

Examination of the between-group data also suggests differences in the proportion of participants in each group meeting criteria for physical discomfort-related behaviors. More participants in the SMS group met criteria for physical discomfort-related behavior than in the FXS and non-specific IDD group for self-injury (52.2% Vs. 30.0% Vs. 29.2% respectively), for aggression (70.8% Vs. 21.9% Vs. 28.6% respectively) and for property destruction (60.0% Vs. 4.3% Vs. 20.0% respectively). There were significant between-group differences on the physical discomfort subscale for aggression, $\chi^2(2) = 15.6$; $p < 0.003$, and property destruction, $\chi^2(2) = 17.2$; $p < 0.003$.

Discussion

The current study found statistically significant between-group differences in the function served by problem behavior in FXS and SMS. Participants with FXS were significantly less likely to be reported as displaying attention-maintained problem behavior than either comparison group. Participants with SMS were significantly more likely than participants in the comparison groups to be reported as displaying problem behavior related to physical discomfort.

Examination of the within-syndrome data for participants with FXS suggested that a smaller proportion of individuals with FXS displayed attention-maintained problem behavior than other socially mediated functions of problem behavior (e.g., tangible- or escape-maintained) for self-injury and aggression.

The results of the current study suggest that children with FXS may be less likely to display attention-maintained problem behavior than escape- or tangible-maintained problem behaviors. These findings are consistent with previous studies to have examined behavioral function in FXS using measures derived from the FAI (Symons, Clark, Hatton, Skinner, & Bailey, 2003; Woodcock, Oliver, & Humphreys, 2009) and extend that work through the use of a more robust measure of behavioral function. The findings of the current study are also consistent with studies to have reported relatively high levels of social escape behaviors in FXS when in contexts characterised by high social- or performance-related demands (e.g., Hall, DeBernadis, & Reiss, 2006; Roberts, Weisenfeld, Hatton, Heath, & Kaufmann, 2007). It remains relatively unclear whether attention serves to function as an aversive stimulus or simply a less effective type of reinforcement than would be typically expected for many children with FXS. The use of a measure of behavioral function that included an escape from attention subscale in future research may help to determine the relative role of social attention in problem behavior in this group.

The mechanisms that underpin some of the relations described above for children with FXS have yet to be identified. However it has been postulated that the social escape behaviors that are characteristic of FXS (such as gaze avoidance) may result from the abnormal functioning of the limbic-hypothalamic-pituitary-adrenal (L-HPA) axis, a major part of the neuroendocrine system which plays an important role in modulating the human stress response (Cohen, 1995; Hessel et al., 2002). Indeed, recent studies have suggested an association between levels of cortisol (a marker of L-HPA functioning) and gaze avoidance (e.g., Hall, DeBernadis, & Reiss, 2006). Further work is needed to examine the influence of such variables on the function of problem behavior in people with FXS.

The current study found that in comparison to other groups, children with SMS were more likely to display problem behaviors related to physical discomfort than either comparison group. Anecdotally, parents frequently cited sleep disruption as being related to their child's problem behavior; however, it is possible that other health conditions associated with the syndrome, such as peripheral neuropathy, may have exerted an influence. In addition, children with SMS had significantly higher scores on the attention subscale of the QABF than children with FXS.

Chronic sleep disturbance is considered to be characteristic of SMS (Finucane, Dirrigl, & Simon, 2001; Greenberg et al., 1996). People with SMS appear to have a relatively specific form of sleep disturbance, suffering from frequent night-waking and excessive daytime sleepiness, which is thought to be related to an inversion in the circadian melatonin cycle (De Leersnyder, de Blois, Claustrat et al., 2001). Given such high levels of sleep disturbance then it is perhaps not surprising that children with SMS were reported to display high levels of problem behavior related to physical discomfort. Studies involving people with SMS that have targeted sleep disturbance for intervention have noted concomitant improvements in problem behavior (De Leersnyder, de Blois, Vekemans et al., 2001). Treatment of such physiological conditions would appear to represent an obvious first step for clinicians working with individuals with SMS.

Existing reports of the SMS behavioral phenotype (e.g., Dykens & Smith, 1998; Smith, Dykens, & Greenberg, 1998) and the findings of other studies to have examined behavioral function in this group (e.g., Taylor & Oliver, 2008) suggest that attention may be a particularly potent type of reinforcement for individuals with SMS. The findings from the current study partially support these findings. For example, children with SMS were more likely to display attention-maintained problem behaviors than children with FXS. However, there were minimal

within-group differences in reported behavioral function for children with SMS. This suggests that there may not be a specific relationship between SMS and behaviors maintained by social contact. Instead children with SMS may be likely to display problem behaviors that serve multiple functions, one of which may be to access social attention.

The findings of the current study hold a number of applied implications for the prevention of problem behavior associated with FXS and SMS. Directed efforts towards the prevention of problem behavior in children with genetic syndromes, such as FXS or SMS, are appropriate given that these children are at a heightened risk of developing particular forms of problem behavior. Knowledge of the probable functions that problem behaviors are likely to serve in young children with specific genetic conditions will aid these efforts. Based on the findings of the current study children with FXS could be taught alternative means of requesting preferred tangibles or to request a break from tasks before problem behaviors become established in the child's behavioral repertoire. For children with SMS the current findings suggest that the treatment of any health conditions should be done at as early a stage as is possible. It would seem important that additional efforts are focused on teaching children with SMS mands that serve multiple functions, one of which is to access attention. The success of any such strategy would be dependent on caregiver responsiveness to such requests.

The findings of the current study also have implications for the assessment and treatment of problem behavior in individuals where such behaviors are already established. Although no substitute for conducting a thorough functional assessment, it would be of benefit for clinicians to be aware that certain individuals are especially likely to display problem behavior that serve specific functions. For example, it would be useful to be aware that problem behaviors displayed

by an individual with SMS may be influenced by physical health conditions, as the role of such variables can be easily overlooked (see Langthorne, McGill & O'Reilly, 2007).

There are a number of limitations in the current study. First, the measure of behavioral function was indirect and it may be that the responses of parents/caregivers do not correspond to the actual contingencies that influence the behavior of their child (e.g., Vollmer & Smith, 1996). For example, neither the respondent nor the investigators were blind to the diagnostic status of each child. It is possible that respondents' prior knowledge of their child's syndrome may have influenced the answers given to items on the QABF. Whilst the use of a highly structured checklist aimed to mitigate against this, it is also possible that demand characteristics introduced some bias in responses to the QABF. Second, the inclusion of several different topographies within each general category of problem behavior, may have led to behaviors that formed separate response classes being aggregated together. Whilst completing the QABF for each individual topography would have allowed for a more sensitive analysis, the differential pattern of results across each behavioral function suggests that the methods adopted in the current study were sufficient to determine general differences in the probability of certain functions being endorsed. Finally, the diagnostic status of the non-specified IDD group was not controlled. The group was selected to ensure that control participants had comparative levels of problem behavior to those in the FXS and SMS groups. This raises the possibility that participants in this group may have been more likely to have an alternative diagnosis, such as an Autistic Spectrum Disorder. As such, it is possible that members of the control group systematically differed in the function served by problem behaviors from what would typically be expected in the population of people with intellectual and developmental disability per se (see O'Reilly et al 2010, for

example). Such limitations aside, the findings of the current study are promising and should stimulate further research on the influence of genetic events on behavioral function.

Further research is required, which adopts experimental functional analysis methods in order to provide a more rigorous examination of behavioral function than was possible in the current study. Future studies should also ensure that investigators are blind to the nature of the child's syndrome in order to protect against potential demand characteristics.

The current study provides preliminary evidence for between and within-syndrome differences in the function of problem behavior displayed by children with FXS and SMS. This is the first study to the authors' knowledge to have noted between-syndrome differences in the function of problem behavior. The use of the QABF as a measure of behavioral function also extends the existing literature on FXS and SMS. The results of the current study holds important applied implications and suggests that further systematic exploration of such relations both within and across different syndromes associated with IDD may be merited.

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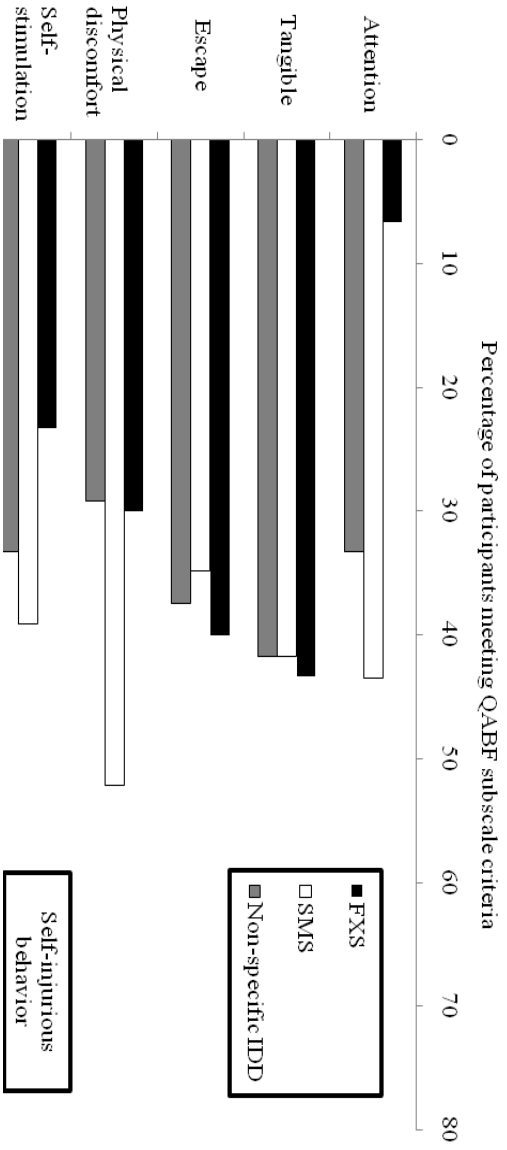
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figure 1

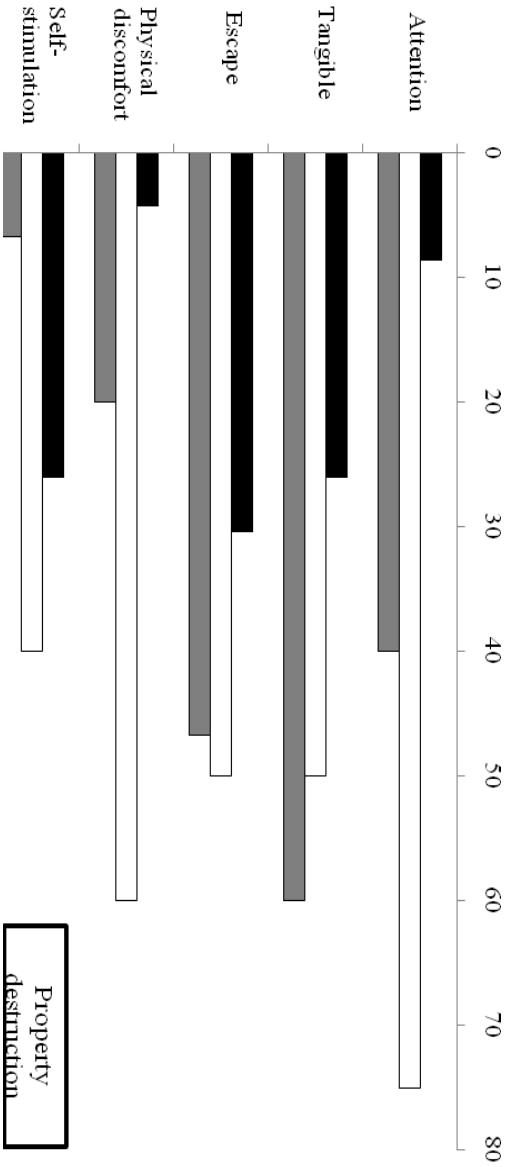
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Figure 1. Percentage of participants with FXS, SMS and non-specific IDD who met criteria for each subscale of the QABF by topographical class.



Self-injurious behavior

QABF subscale



Aggression

Property destruction

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Table 1

Participant Characteristics

	FXS (N=34)	SMS (N=25)	Non-specific IDD (N=30)
<hr/>			
Chronological Age (months).			
Mean (SD)	133.5 (36.3)	138.0 (46.6)	121.3 (34.7)
Gender (%)			
Male	91.4	52.0	80.0
Female	8.6	48.0	20.0
Vineland Age Equivalent Scores (months)			
Overall Mean (SD)	46.9 (21.2)	53.9 (19.8)	42.3 (23.6)
Communication (SD)	50.6 (30.7)	68.1 (27.8)	40.4 (28.2)
Daily Living Skills (SD)	42.1 (26.2)	50.9 (19.6)	42.8 (30.4)
Socialization (SD)	48.0 (17.8)	49.2 (16.8)	43.8 (18.7)

Aberrant Behavior Checklist (total scores)

Overall Means (SD)	65.9 (35.3)	73.3 (28.0)	77.0 (28.9)
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Number of participants with:

1 form of problem behavior (% of group)	1 (2.9)	1 (4.0)	6 (20.0)
2 forms of problem behavior (% of group)	18 (51.4)	6 (17.1)	11 (36.7)
3 forms of problem behavior (% of group)	16 (45.7)	18 (72.0)	13 (43.3)

Table 2

Number of Participants Meeting Criteria for Behavioral Function

Topography	FXS	SMS	Non-specific IDD	χ^2
	N	N	N	
Self-injury				5.5
not meeting criteria	11 (36.7%)	2 (8.7%)	6 (25.0%)	
with single function	7 (23.3%)	7 (30.4%)	6 (25.0%)	
with 2 or more functions	12 (40.0%)	14 (60.9%)	12 (50.0%)	
Aggression				8.0
not meeting criteria	8 (30.8%)	1 (4.5%)	2 (9.1%)	
with single function	5 (19.2%)	4 (18.2%)	6 (27.3%)	
with 2 or more functions	13 (50.0%)	17 (77.3%)	14 (63.6%)	
Property destruction				19.0*
not meeting criteria	7 (36.8%)	0 (0.0%)	1 (7.7%)	
with single function	5 (26.3%)	2 (11.1%)	7 (53.8%)	
with 2 or more functions	7 (36.8%)	16 (88.9%)	5 (38.5%)	

*sig at p<0.012

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