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The comparison of expressed emotion of parents of individuals with fragile X syndrome to other intellectual disabilities

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Abstract

Background: Parenting children and young adults with intellectual disabilities, including individuals with fragile X syndrome and Down syndrome, is challenging, joyful, and complicated. Exploring how parents talk about their children, and the quality of the parent/child relationship can provide insight into the home environment and interactional patterns of the family.

Method: Expressed emotion (EE) is a measurement of a family's emotional climate based on a parent or caregiver's report of warmth, emotional overinvolvement, hostility, and criticism. The purpose of this study was to describe EE for a sample of parents of individuals with intellectual disabilities and to determine any differences in EE amongst individuals within subgroups. Based on previous research about fragile X syndrome and family systems, we hypothesized that there would be significant differences between the disability groups (higher EE in families with children/young adults with fragile X syndrome).

Results: Results showed relatively high proportions of EE across groups of individuals with intellectual disabilities, however, there were no significant differences between the subgroups. Null findings suggest that differences in EE may not relate directly to a child's specific genetic condition. Rather, increased EE in caregiver populations may simply reflect well-documented stressors related to stigma, caregiver burden, and limited community supports. Critical statements were infrequent, however, over half of the participants reported dissatisfaction with their situation, and many were categorized as having emotional overinvolvement, as measured by frequent statements of intense worry and self-sacrifice.

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Jeanine Coleman and Talia Thompson contributed equally to this work.

AUTHOR CONTRIBUTIONS

Jeanine Coleman is the first author of this article due to the formation of the study and a majority of data collection and data analysis. Jeanine Coleman and Talia Thompson contributed equally in writing of the article. Karen Riley and Korrie Allen contributed additional writing and editing to this article and the remaining authors reviewed the article and provided feedback.

Conclusion: Findings point to potential utility in family-level interventions focused on providing structured caregiver therapy to manage excessive worry and grief related to a diagnosis of intellectual disability, and respite care to encourage caregiver independence and pursuit of personal care.

Keywords

expressed emotion; 5 min speech sample; fragile X syndrome; intellectual disability

1 | INTRODUCTION

Parents of children and young adults with intellectual disabilities, such as fragile X syndrome and Down syndrome, are not often asked about their relationship with their child or how their child's disability affects their own well-being, their relationships, and the overall family system (Laghezza et al., 2010). While more researchers are studying the connection between parental mental health (i.e., depression and anxiety) and their children's disabilities, research is still limited to many with significant intellectual disabilities, particularly low incidence disabilities, such as fragile X syndrome (Scherer et al., 2019; Schuiringa et al., 2017). Parents are asked, most often, how their child is functioning, what are their strengths, and what are their needs. Yet, exploring *how* parents talk about their children with intellectual disabilities, and the quality of the parent/child relationship can provide insight into the home environment and interactional patterns of the family. The bioecological systems theory of human development posits that growth and skill acquisition are highly impacted by the quality of reciprocal interactions between children and their immediate caregivers (micro-system) as well as broader community supports (exo-system) and cultural and societal attitudes (macro-system) (Bronfenbrenner & Ceci, 1994). Similarly, family systems theory views the family as an emotional unit, where members are emotionally interconnected and influence each other's behaviours (Bowen, 1978). As such, family systems are highly impacted when a child has an impairing condition such as intellectual disability, and the parent/child dyad is paramount to understand and support.

Expressed emotion (EE) is a measurement of a family's emotional climate based on a parent or caregiver's report of warmth, emotional overinvolvement, hostility, and criticism (Calam & Peters, 2006; McCarty & Weisz, 2002). High EE may be an indication of a disruptive or complicated emotional relationship between the parent and the child and has been associated with poor adherence to intervention plans and maladaptive behaviours for the child (Weston et al., 2017). Studies show that the relationship between child behaviour and parent EE is bidirectional (Greenberg et al., 2006). For example, longitudinal studies have shown high levels of parental criticism can predict the development of problem behaviours over time (Baker et al., 2011) while child functioning can impact parent warmth and criticism (Hickey et al., 2020).

Thus, EE provides a novel and nuanced way to examine the challenges and joys of parenting a child with intellectual disability and may be useful in developing valid and feasible family-systems level interventions to improve child outcomes. Further, as prior studies have documented differences in parenting experiences amongst parents of those with

known genetic causes of intellectual disability (Sterling & Warren, 2018), an examination of differences in EE between groups, including fragile X syndrome and Down syndrome, and idiopathic or unknown causes of intellectual disability could inform etiologically specific caregiver supports.

1.1 | Parenting children with intellectual disabilities

Overall, parents of individuals with intellectual disabilities face unique challenges shown to increase stress, which can subsequently impact child outcomes as well as overall family wellbeing (Peer & Hillman, 2014; Woodman et al., 2015). Stressors related to parenting a child with intellectual disabilities can include increased demands for intense caregiving well into adulthood, comorbid behavioural challenges, and social isolation or lack of support (Hart & Neil, 2021; Heller et al., 2021; Hodapp et al., 2019; Sheldon et al., 2020). However, despite ample literature documenting increased stress, there is also evidence for positive parenting experiences of parents of children with intellectual disabilities, such as close and loving relationships with the child and a sense of increased meaning and purpose in life (Green, 2007; Hastings & Taunt, 2002; Thompson et al., 2020). Furthermore, parenting experiences can vary widely within different populations of intellectual disabilities based on parent coping strategies and demographic factors (Poehlmann et al., 2005), comorbid diagnoses (De Clercq et al., 2022), and genetic causes (Sterling & Warren, 2018).

Fragile X syndrome is the most common *inherited* cause of intellectual disability and is associated with a complex and challenging behavioural phenotype; including high rates of comorbid autism spectrum disorder, hyperactivity, aggression, and anxiety (Salcedo-Arellano et al., 2020). Parents of children with fragile X syndrome describe a high emotional burden of caregiving due to frequent medical appointments, child-induced injury to the parent, financial challenges, and the need for constant parent supervision for most waking hours (Bailey et al., 2012). Sterling and Warren (2018) compared communication practices between parents of children with fragile X and Down syndromes and found that parents of children with fragile X utilised more redirection and zaps (comments restricting behaviour) in their communication with their children, highlighting important differences in dyadic interaction patterns between the two genetic conditions. Further, heritability patterns of fragile X syndrome mean that in any given family unit, the mother or father are also genetically impacted, either with the fragile X full mutation or the premutation. Individuals with the fragile X premutation have increased (55–200) CGG repeats of the fragile X gene (FMR1) (Hessl et al., 2011) and are at increased risk for a variety of their own medical and psychological complications that might impact their parenting experience, including fragile X associated neuropsychiatric disorders (FXAND), such as anxiety, depression, attention deficit/hyperactivity disorder, and learning disabilities (Kour Sodhi & Hagerman, 2021; Hagerman et al., 2018). Despite these risk factors, many families of children with fragile X syndrome demonstrate great resiliency, reporting a high quality of life and satisfaction with parenting (Raspa et al., 2014; Wheeler et al., 2008).

Studies have documented a ‘Down syndrome advantage’, in which children with Down syndrome are reportedly easier to rear than those with other genetic causes of intellectual disabilities (Hodapp et al., 2019). Research shows that families of children with Down

syndrome report higher levels of well-being than families of children with other causes of intellectual disabilities (Fidler et al., 2010), however, critics of the 'Down syndrome advantage' call attention to troubles with the metrics used to assess family levels of wellbeing, the importance of analysing parent age and support systems when assessing well-being, and the necessity to consider the ages of the children at the time of assessment (Glidden et al., 2014). Overall, across genetic conditions, parents of children with intellectual disabilities who demonstrate more adaptive coping strategies for stress (e.g., positive reappraisal of the circumstances) report higher levels of well-being (Abbeduto et al., 2004; Rooke & Pereira-Silva, 2016; van der Veek et al., 2009).

1.2 | Measuring expressed emotion

The Five-Minute Speech Sample (FMSS, Magaña-Amato, 2015) is a concise way of measuring EE which is coded through a non-scripted parent monologue that lasts for exactly 5 min. Within those 5 min, statements spoken by the parents can reveal the loving relationships, challenges, and sacrifices that parenting a child or young adult with disabilities brings to families. Parent monologues are analysed for both content and tone, specifically noting instances of warmth, critical comments, emotional displays (e.g., crying; laughing), self-sacrifice, statements of attitude, excessive detail, statements about relationship status, and positive remarks. Like many social-emotional measures, a higher score is more concerning or problematic.

Several studies have documented how constructs measured by the FMSS (e.g., high EE, criticism, emotional overinvolvement, warmth, positive and negative remarks) correlate with direct observational data from parent–child interactions in a variety of clinical populations (Weston et al., 2017), including children with attention deficit/hyperactivity disorder (Daley et al., 2003; Marshall et al., 1990; Psychogiou et al., 2008) and youth with internalising and externalising problems (McCarty et al., 2004; Scott et al., 2011). While these findings suggest the FMSS is a very brief, direct interview that provides researchers and clinicians with rich insight to the unique relationships between parents and their children, more research is needed to specifically examine the validity and utility of the FMSS for families of children with intellectual disabilities. Coleman (2010) found no significant correlations between EE from the FMSS and behavioural questionnaires completed by mothers of children with fragile X syndrome, suggesting there may be differences in the relationship between EE and child outcomes amongst different populations of individuals with intellectual disabilities, as measured by behavioural questionnaires.

1.3 | Expressed emotion in families impacted by intellectual disabilities

Research indicates a sizable proportion (~40%) of parents of children with intellectual disabilities exhibit high EE, highlighting the family system as a potential target for intervention (Thompson et al., 2018). However, research on EE in parents of children with intellectual disabilities caused by specific genetic conditions such as fragile X syndrome and Down syndrome is limited. Preliminary studies indicate parents of children with fragile X syndrome have increased EE, with particularly high rates of EOI, characterised by statements of intense expressions of love for their children, parental self-sacrifice, and worry about their children's future (Coleman, 2010; Coleman & Riley, 2014). Furthermore,

for children with fragile X syndrome, parent EE profiles of increased warmth coupled with decreased criticism are associated with better behavioural outcomes (Greenberg et al., 2012). Research on EE in parents of children with Down syndrome is more limited but initial findings suggest a pattern of high parent warmth (De Clercq et al., 2022). Beck et al. (2004) found lower EE than expected in a sample of parents of children with ID and hypothesized that the high proportion of children with Down syndrome in their sample may have skewed their results due to the 'Down syndrome advantage' within intellectual disability populations. Further, Cregenzán-Royo et al. (2018) found significantly increased EE in parents of children with fragile X syndrome compared to those of children with Down syndrome, however, the authors cautioned interpretation of their results as they used a survey methodology rather than direct interviewing with the FMSS to capture EE data. Therefore, ample opportunity remains to describe parent EE within and between different etiological groups of children with intellectual disabilities.

1.4 | Present study

The purpose of the present study was to describe EE for a sample of parents of individuals with intellectual disabilities and to determine any differences in EE by aetiology. We aimed to understand if the presence or absence of high EE is related to the child's genetic cause for intellectual disability; or is high EE a universal phenomenon across different populations of individuals with intellectual disabilities? Based on previous research demonstrating the significant challenges in families impacted by fragile X syndrome related to parent premutation status, child cognitive capacity, parent and child emotional and behavioural regulation, and overall social competence, we hypothesized higher EE in parents of children with fragile X syndrome than Down syndrome, or other/idiopathic causes of intellectual disabilities. The following research questions guided the study:

1. What are the EE profiles of parents of individuals with intellectual disabilities, including subgroups of fragile X syndrome, Down syndrome, and other/idiopathic causes, as measured by the FMSS?
    - How many parents in the pooled sample as well as each subgroup show high EE?
    - What are the frequencies of each EE subcategory (CRIT, EOI) across subgroups?
    - What are the initial statements, relationships, criticism, dissatisfaction, and EOI, as measured by the FMSS, in a sample of parents of individuals with intellectual disabilities?
2. What are the differences in EE and EE subcategories amongst subgroups of intellectual disabilities, including those with known genetic aetiology (e.g., fragile X syndrome and Down syndrome) and those with other/idiopathic causes of intellectual disabilities?

2 | MATERIALS AND METHODS

2.1 | Participants and procedures

Families were recruited from another, multi-site study validating a novel cognitive assessment for individuals with intellectual disabilities. Participation was voluntary and did not impact enrollment for the validation study. All participants provided informed consent and the study was approved by the University of Denver Institutional Review Board. Participants were included in the study if they were parents or caregivers of individuals ages 6–25 years with a diagnosed intellectual disability, and they spoke English. Parents completed demographic surveys and the FMSS while the individuals with intellectual disabilities participated in standardised testing (including IQ testing) for the validation study in a separate room.

The participants in the study ($N = 152$), were parents or caregivers of individuals with intellectual disabilities, including fragile X syndrome, Down syndrome, and other/idiopathic causes (see Table 1). All caregivers completing the FMSS were involved in the individuals' daily lives and knew the individuals very well. The majority of respondents were mothers (85%) and most individuals with intellectual disabilities were male (63%) and identified as white (72%). Most participants (84%) were independent parent respondents, meaning that only one parent of the family completed the FMSS on one child, and 12 families in the sample had two parents completing the FMSS on the same child ($n = 24$; 16%). The families with two parents who completed there FMSS were identified with the participant's specific identification number and an identifier with either 'mother' or 'father'. The intellectual disability subgroups were statistically similar for proportions of mothers and fathers completing the FMSS, child age group (middle childhood, adolescent, young adult), ethnicity, and gender. There were significant differences in terms of caregiver education; the group of individuals with other/idiopathic causes of intellectual disabilities had significantly more caregivers with an advanced or graduate degree. However, for all groups, education levels were relatively high with majority earning at least a college degree. The group of individuals with other/idiopathic causes of intellectual disabilities also had significantly higher FSIQ scores, as measured by the SB5 (Table 1). All comparison statistics were adjusted for child IQ and parent education to account for these group differences.

3 | MEASURES

3.1 | Five minute speech sample (FMSS)

The FMSS is a standardised procedure, which includes video/audio recording a parent or close relative talking about their child for exactly 5 min (Magána-Amato, 2015). The FMSS has been shown to be a valid and reliable predictor of child outcomes (Hooley & Parker, 2006) and shows good interrater reliability [*kappa* = .35–.82] and concurrent validity ([r = .58–.74]; Moore & Kuipers, 1999). Members of the research team provided standardised instructions to parents asking them to speak for 5 min about their child and their relationship to the child, including anything they felt described how they got along with their child. Recordings were transcribed verbatim and then coded and scored across five categories: (1) initial statement (positive, neutral, or negative), (2) relationship (overall rating of positive,

neutral, or negative; includes instances of warmth), (3) criticisms (frequency count), (4) dissatisfaction (present or absent), and (5) emotional overinvolvement (present, absent, borderline; includes self-sacrificing, extreme worry, over-protection). The FMSS measures overall levels of EE (high vs. low) which are comprised of subcategories: high critical (CRIT), borderline critical (b/CRIT), high emotional overinvolvement (EOI), and borderline emotional overinvolvement (b/EOI). There are nine potential EE subcategory profiles under the two overall levels of EE (High: CRIT, EOI, CRIT + EOI, CRIT + b/EOI, EOI + b/CRIT; Low: Low (absence of CRIT or EOI), b/CRIT, b/EOI, c/CRIT + b/EOI).

Intercoder reliability involved an approach by Syed and Nelson (2015) that involves both interrater reliability and consensus building in cases lacking agreement. Adhering to this process, the first author independently coded all the transcripts according to the criteria outlined in the FMSS manual (Magaña-Amato, 2015). The second author then independently coded 20% of the transcripts and intercoder agreement was calculated. Both coders had previously completed a 5-day, live teleconference training seminar with the creator of the FMSS coding system and passed intercoder agreement tests with each other and with the FMSS developer. Agreement on the dual coded transcripts was above 80%. In accordance with the process, any differences were discussed until consensus was met. Codes were then totaled and classified to determine final FMSS EE categories and subcategories (Figure 1).

3.2 | Demographic survey

Parents completed a brief survey on parent and child age, education, and ethnicity as well as other diagnoses of the child.

3.3 | Stanford Binet, 5th edition (SB5)

IQ was measured through the SB5, a standardised, individually administered test that has been validated as a gold standard measure for individuals with intellectual disabilities (Roid & Miller, 2003). Tests on the SB5 include fluid reasoning, knowledge, quantitative reasoning, visual–spatial processing, and working memory. SB5 scores are normed to have a mean of 100 and standard deviation of 15. SB5 scores were used in this study to control for differences in EE by child cognitive capacity.

3.4 | Data analysis

Descriptive statistics (frequency and proportions) were used to characterise the sample demographics, EE status (high vs. low), EE subcategories and combinations (CRIT, b/CRIT, EOI, b/EOI), and specific scoring categories of initial statement, relationship/warmth, criticisms, dissatisfaction, and emotional overinvolvement. For the pooled sample, we used a *T*-test to assess mean FSIQ differences between Low and High EE, and Chi-square to examine differences in EE by parent gender and between families with one versus two parent responders. *T*-tests, Chi-square, and Fisher's exact tests were used to compare the subgroups of individuals with intellectual disabilities across demographic variables and EE. Analyses were intended to be descriptive and hypothesis-generating, therefore no adjustments were made for multiple comparisons. Logistic regression models were used to control for the effects of socioeconomic status (SES; as measured by parent education), child

age, and child IQ, and all statistics reported are adjusted for these variables. The statistical package SPSS-28 was used to run all analyses and statistical significance was set at $p < .05$.

4 | RESULTS

In the pooled sample, approximately 44% of caregiver participants showed high EE on the FMSS, with 48% from the group of individuals with fragile X syndrome, 40% from the group of individuals with Down syndrome, and 46% from the group of individuals with other/idiopathic causes of intellectual disabilities (Table 2 and Figure 2). Mothers were significantly more likely to show high EE than fathers/male caregivers, X^2 (1, $N = 152$) = 7.83, $p = .006$, although results should be interpreted with caution as there were few fathers/male caregivers ($N = 23$) enrolled in the study. FSIQ did not differ significantly between Low EE ($M = 48.38 \pm 10.90$) and High EE ($M = 47.71 \pm 9.71$) groups, $t(133) = -3.7$, $p = .712$. Results showed no significant differences in EE between families where one or two parents completed the FMSS X^2 (1, $N = 152$) = .50, $p = .511$.

In contrast with our hypothesis, there were no statistical differences in EE amongst the subgroups, X^2 (2, $N = 152$) = .95, $p = .623$ (Table 2, Figure 2). Additionally, there were no significant differences in EE subcategories amongst subgroups, X^2 (8, $N = 152$) = 5.9, $p = .988$ (Table 3). The most frequent EE subcategory was b/EOI (n = 30), but it was statistically proportional to the other subgroups.

In terms of general FMSS monologue content, most parents began their FMSS with a positive (47%) or neutral (51%) statement, such as ‘My daughter is a smart and funny person’ or ‘My son is 13 years old and has fragile X syndrome’. Parents described relationships with their children as mostly positive (50%) or neutral (41%), with less than 10% being negative. There were very few critical statements ($n = 18$; <12%). Although, many parents expressed dissatisfaction (52%) with the situation that their family is in (not dissatisfaction with their child), indicating very challenging parenting times. Within the whole sample, more parents made statements that were coded as b/EOI or EOI, rather than CRIT (high critical). Furthermore, self-sacrificing/overprotective behaviour was the highest coded subcategory within EOI (present 16% of the time and borderline 17% of the time).

5 | DISCUSSION

The current study is the first to examine differences in EE between parents of children with idiopathic intellectual disabilities and intellectual disabilities caused by fragile X syndrome or Down syndrome. Based on previous research about increased caregiver burden and family stressors in fragile X syndrome, we hypothesized that there would be significant differences between the disability groups (higher EE in parents of children with fragile X syndrome). Results showed relatively high proportions of EE across the subgroups (>40% for all three subgroups), however, there were no significant differences between the subgroups. These findings support an emergent theory of understanding how parents of children with different intellectual disabilities talk about their children and express their emotions about their overall relationship with their child. This evolving model illustrates that regardless of the actual diagnosis and or the aetiology of the child’s intellectual disability, parents’ expressed

emotion is relatively similar, suggesting universal experiences for parents raising children with intellectual disabilities.

Previous literature suggested that given the phenotypic patterns of parents with the fragile X premutation (Hagerman et al., 2018; Hessl et al., 2011) and that managing the stressors associated with a fragile X syndrome diagnosis is challenging for the whole family (Weber et al., 2019), parents of children with fragile X syndrome might be more emotionally charged and have higher EE. Furthermore, studies have shown parents of children with Down syndrome might present with a different and more positive pattern than their counterparts with children with fragile X syndrome or other/idiopathic causes (Beck et al., 2004; Fidler et al., 2010). Therefore, it was important to determine whether there are differences between the aetiology or diagnosis in relation to EE and how parents are supported based on their child's disability. Results from this study did not support our hypothesis and indicate that there are no significant differences between intellectual disabilities subcategories in relation to EE.

Nonetheless, our descriptive results from the pooled sample are directly in line with multiple other studies that have shown approximately 40% of parents of children with intellectual disabilities show high EE (Thompson et al., 2018). Our null findings suggest that differences in EE may not relate directly to a child's specific genetic condition. Rather, increased EE in intellectual disability caregiver populations may reflect well-documented environmental stressors related to stigma, caregiver burden, and limited community supports (Green, 2003; Hodapp et al., 2019; Mitter et al., 2019). Additionally, prior research has shown child age might impact FMSS results (Vostanis et al., 1994), in which case our study design might not have properly accounted for age, although we did adjust all comparisons for child age group. Another explanation could be methodological limitations with the FMSS in the fragile X syndrome subgroup. The FMSS was originally based on the Camberwell Family Interview (CFI), developed for research with individuals with mental illness (Brown, et al., 1972; Vaughan & Leff, 1976). Fragile X syndrome is a unique and highly involved genetic condition, impacting both children and caregivers. As seen in autism research (Benson et al., 2011; Daley & Benson, 2008) and with preschool populations (Daley et al., 2003), the coding criteria for the FMSS may need to be adapted to accurately measure EE for parents of those with intellectual disabilities caused by different genetic conditions. For example, an adapted measure for fragile X syndrome may need to lower the threshold for criteria for emotional over-involvement.

While there were no group differences by subgroup, our study identified differences in EE between mothers and fathers, indicating that mothers of children with intellectual disabilities may be at particularly high risk and may need more support to balance their EE. Prior research has already documented higher caregiver burden (Shearn & Todd, 2000), poorer health related quality of life (Bourke et al., 2008), and higher parenting stress (Woodman et al., 2015) in mothers of children with disabilities. Our research extends these trends to the construct of EE, suggesting that the parenting experience is challenging, especially for mothers of children with intellectual disabilities. We speculate that ongoing societal pressures for mothers to carry higher caregiving loads than fathers may disproportionately impact a mother's ability to connect and relate to her child with intellectual disability.

However, limitations in our sample size for fathers preclude us from generalising these findings to the entire population, and further research is needed to better understand differences in EE for mothers and fathers.

Despite relatively high EE in the total sample, most of the parents expressed much love and joy from their children. Initial statements were often neutral or positive, and parents described warm relationships and made few critical comments about their child. These findings align with studies documenting the resilience of some families of children with ID (McConnell et al., 2014) and that many families report their child with intellectual disabilities benefits the entire family system (Green, 2007; Hastings & Taunt, 2002; Thompson et al., 2020). Despite these positive descriptions, higher numbers of EOI and b/EOI across the sample suggest that while parents in our sample may not be overly critical of their children, they may suffer from intense worry and self-sacrifice related to the child's disability. Furthermore, over half of our participants described dissatisfaction that comes with a diagnosis, although they did not wish to change anything specific about their child. These combined findings point to potential utility in family systems-level interventions focused on providing structured caregiver therapy to manage excessive worrying and grief related to the intellectual disability diagnosis (Hastings & Beck, 2004) and respite care to encourage caregiver independence and pursuit of personal care (Strunk, 2010).

5.1 | Limitations

One limitation of the study is that our sample did not include an equal number of fathers and mothers. It is important to include more fathers in future studies so that we can look deeper at the differences between genders and potential heteronormative parenting styles. Additionally, this sample did not include families with children below 6 years of age. Most families that participated had children in the adolescent and young adult age ranges, therefore limiting the age ranges for analysis. We also did not collect data on fragile X premutation status for parents of individuals with fragile X syndrome. This limited our ability to analyse any impact premutation carrier status might have on EE in the fragile X syndrome subgroup. Lastly, the sample had limited ethnic and racial diversity. It is well known that most families that participate in research identify as White with middle or high-incomes, and this phenomenon is reflected in this study.

5.2 | Clinical implications

The concept of EE is central to the need to focus interventions not just on the child but the entire family system, especially those struggling with their children's challenging behaviours, limited self-care skills, and other everyday stressors. Ensuring parents are supported and their stresses and concerns are addressed will help ensure alignment between the home and the therapeutic environment to achieve maximum benefit for children with fragile X syndrome and other intellectual disabilities. The FMSS provides a novel way of looking at the parent–child dyad and could provide critical information to guide future intervention in relation to balancing EE for new parents with young children with fragile X syndrome and different intellectual disabilities. The intervention may be parent mediated interventions to support the parents' balance of EE in relation to cognitive and behavioural interventions for their children with intellectual disabilities. We know that high EE is

predictive of poorer outcomes for children (Calam & Peters, 2006). High EE can interfere with the effectiveness of the treatment or therapies that children receive. Interestingly, given the amount of evidence documenting the challenges that mothers of children with intellectual disabilities face, there is very limited psychological research exploring treatment options or how best to support this community. This is a significant gap in the literature and more efforts to understand this unique group of individuals are needed. This data would suggest that mental health support and counselling is warranted regardless of the diagnostic label or the aetiology of the intellectual disability. This would also suggest that understanding stress related to having and raising a child with intellectual disability should be included in preservice training for psychologists and counsellors. Parents who can maintain the balance of EE in relation to the stressors and challenges of parenting a child with intellectual disability may see that maximum benefit from therapeutic interventions can be achieved.

5.3 | Future directions

The next step in this research is to examine the relationship of EE to child characteristics (e.g., adaptive development, behaviours, cognition) both within and across the subgroups of intellectual disabilities. Further investigation of EE in parents of younger children with intellectual disabilities will also broaden our understanding across the lifespan. It would be valuable to determine the differences between EE for parents with young children (under the age of 5 years) with and without developmental delays and disabilities to understand the impact of age on parents' expressed emotion. Previous research indicated that parents were high EE if their children were younger—especially EOI (Coleman & Riley, 2014). We have started to explore the differences between mothers' and fathers' expressed emotion, but we need more fathers to participate in this research to fully understand the differences. To address potential interventions for families with young children with disabilities and their ability to balance their EE, a next step is to examine the impact of counselling and support services for families. Lastly, it would be valuable to explore the positive impact that individuals with intellectual disabilities have on their families and communities. Much of the research with individuals with disabilities focuses on the negative aspects, such as what the individuals cannot do, or the challenges associated with caretaking. Therefore, it is important to highlight the positive experiences and contributions individuals with intellectual disabilities bring to their communities to combat ableist rhetoric and add to the research on thriving and self-determination.

6 | CONCLUSION

This study showed relatively high EE in parents of children with intellectual disabilities, with no significant differences between the subgroups (fragile X syndrome, Down syndrome, other/idiopathic). Mothers showed higher EE across the subgroups and may require additional supports to manage EE. Despite high EE, many parents described positive relationships and few criticisms, however, emotional overinvolvement was more common. In all, mental health support is necessary across intellectual disability diagnoses, but the need for etiologically specific intervention may not be a requirement. Essentially, this reveals that parents of children with intellectual disabilities with varying diagnoses and etiologies

are not statistically different from one another, however, they do present patterns that are different than those of parents of neurotypical children, which supports the recommendation that these parents receive ongoing mental health support.

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CONFLICT OF INTEREST

The authors declare that the research was conducted in absences of any commercial or financial relationships that could be construed as a potential conflict of interest. There are no conflicts of interest for the following authors, J. Coleman, T. Thompson, K. Riley, K. Allen, C. Mickalak, R. Shields. E. Berry-Kravis has received funding from Acadia, Alcobra, AMO, Asuragen, Avexis, Biogen, BioMarin, Cydan, Erydel, Fulcrum, GeneTx, GW, Healx, Ionis, Jaguar, Lumos, Marinus, Neuren, Neurogene, Neurotrope, Novartis, Orphazyme/Kempharm, Ovid, PTC Therapeutics, Retrophin, Roche, Seaside Therapeutics, Taysha, Tetra, Ultragenyx, Yamo, Zynerba, Moment Biosciences, and Vtesse/Sucampo/Mallinckrodt Pharmaceuticals, to consult on trial design or run clinical or lab validation trials in genetic neurodevelopmental or neurodegenerative disorders, all of which is directed to RUMC in support of rare disease programs; Dr Berry-Kravis receives no personal funds and RUMC has no relevant financial interest in any of the commercial entities listed; D. Hessl: Dr. Hessl has received funding from the following, all of which is directed to UC Davis, in support of fragile X treatment programs, and he receives no personal funds and has no relevant financial interest in any of the commercial entities listed: Autifony, Ovid, Tetra, Healx, and Zynerba pharmaceutical companies to consult on outcome measures and clinical trial design.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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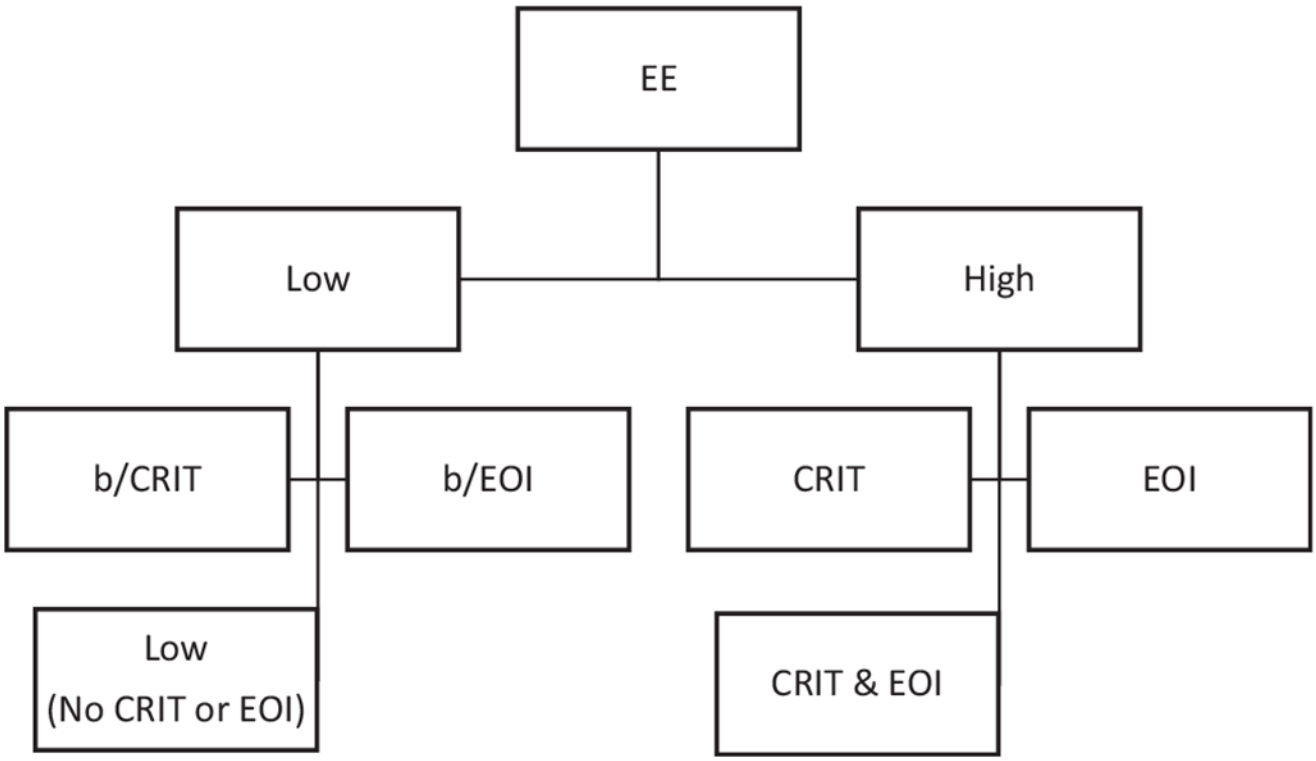

FIGURE 1.
FMSS scoring structure. As described in Magaña-Amato, A. B., 2015. EE, expressed emotion; b/, borderline; CRIT, critical; EOI, emotionally overinvolved

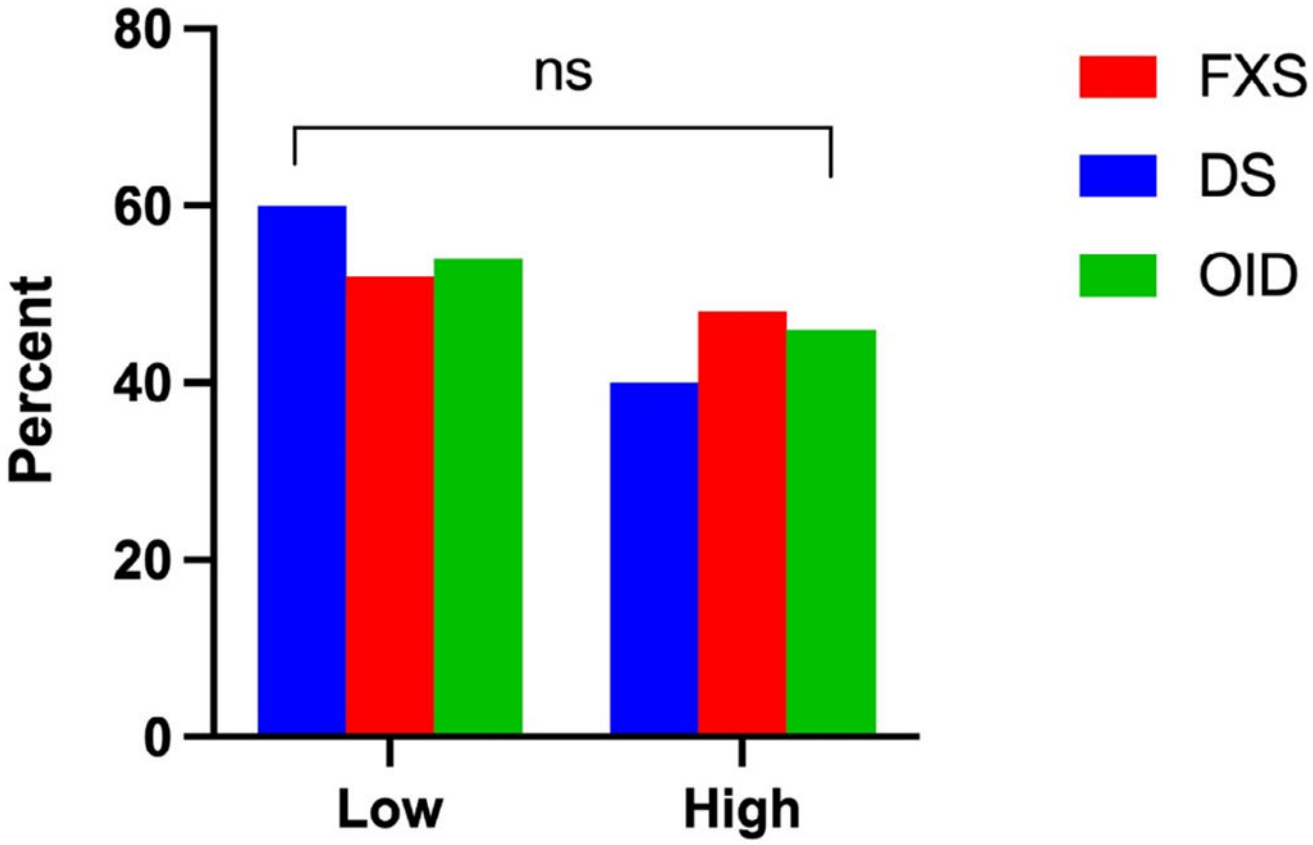

FIGURE 2.
Comparison of expressed emotion scores by diagnostic group

TABLE 1

Demographic composition of sample

	Total sample *n* = 152	**FXS *n* = 46**	**DS *n* = 65**	**OID *n* = 41**	**Sig**
Caregiver completing the FMSS					.809
Mother	129 (84.9)	39 (84.8)	54 (83.1)	36 (87.8)	
Father/stepfather/grandfather	23 (15.1)	7 (15.2)	11 (16.9)	5 (12.2)	
Caregiver education					**.004**
High school or GED	4 (2.6)	1 (2.2)	1 (1.7)	2 (4.9)	
Some college	42 (27.6)	16 (34.8)	18 (27.7)	8 (19.5)	
College graduate	69 (45.4)	26 (56.5)	31 (47.7)	12 (29.3)	
Graduate degree	35 (23)	3 (6.5)	15 (23.1)	17 (41.5)	
Did not report	2 (1.3)	0	0	2 (4.9)	
Child gender					.081
Male	96 (63.2)	35 (76.1)	36 (55.4)	25 (61)	
Female	56 (36.8)	11 (23.9)	29 (44.6)	16 (39)	
Child age					.500
Middle childhood (6–11 years)	46 (30.3)	17 (37)	18 (27.7)	11 (26.8)	
Adolescent (12–18 years)	54 (35.5)	12 (26.1)	24 (36.9)	18 (43.9)	
Young adult (19 years and above)	52 (34.2)	17 (37)	23 (35.4)	12 (29.3)	
Child ethnicity					.052
Black or African American	14 (9.2)	6 (13)	4 (6.2)	4 (9.8)	
Mexican or Mexican American	3 (2)	0	0	3 (7.3)	
Other Latinx	11 (7.2)	2 (4.3)	5 (7.7)	4 (9.8)	
Indigenous or American Indian	4 (2.6)	1 (2.2)	2 (3.1)	1 (2.4)	
Asian	3 (2)	0	1 (1.5)	2 (4.9)	
White	108 (71.1)	37 (80.4)	50 (76.9)	21 (51.2)	
2 or more	2 (1.3)	0	0	2 (4.9)	
Other	5 (3.3)	0	3 (4.6)	2 (4.9)	
Did not report	2 (1.3)	0	0	2 (4.9)	

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	Total sample *n* = 152	**FXS *n* = 46**	**DS *n* = 65**	**OID *n* = 41**	**Sig**
Child IQ	48.09 ± 10.37	46.29 ± 8.66	44.75 ± 6.37	56.81 ± 13.50	**<.001**

Note: Statistics reported: *N* (%), *M* ± SD. Bold type indicates significant findings, $p < .05$.

TABLE 2

Comparisons of expressed emotion by diagnostic group

Emotional expression overall	Total $N = 152$	FXS $N = 46$	DS $N = 65$	OID $N = 41$	Chi-square tests of independence
Low	85 (55.9)	24 (52.2)	39 (60)	22 (53.7)	X^2 (2) = .95
High	67 (44.1)	22 (47.8)	26 (40)	19 (46.3)	p = .623 n = 152

Note: Statistics reported: N(%), significance set at $p < .05$.

Abbreviations: DS, Down syndrome; FXS, fragile X; OID, other/idiopathic ID.

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TABLE 3

Comparison of expressed emotion subcategories by diagnostic group

Emotional expression subcategories	Total *N* = 152	FXS *N* = 46	DS *N* = 65	OID *N* = 41	Fisher's exact tests of independence
Low (absence of CRIT or EOI)	19 (12.5)	4 (8.7)	9 (13.8)	6 (14.6)	X^2 (16) = 5.99
High CRIT	7 (4.6)	1 (2.2)	4 (6.2)	2 (4.9)	p = .988 n = 152
High EOI	26 (17.1)	9 (19.6)	10 (15.4)	7 (17.1)	
b/CRIT (low)	15 (9.9)	4 (8.7)	7 (10.8)	4 (9.8)	
b/EOI (low)	30 (19.7)	9 (19.6)	15 (23.1)	6 (14.6)	
CRIT & EOI (high)	11 (7.2)	5 (10.9)	4 (6.2)	2 (4.9)	
b/CRIT & b/EOI (low)	21 (13.8)	7 (15.2)	8 (12.3)	6 (14.6)	
CRIT & b/EOI (high)	8 (5.3)	3 (6.5)	3 (4.6)	2 (4.9)	
b/CRIT & EOI (high)	15 (9.9)	4 (8.7)	5 (7.7)	6 (14.6)	

Note: Statistics reported: *N*(%), significance set at $p < .05$.

Abbreviations: b/, borderline; CRIT, critical; DS, Down syndrome; EOI, emotionally overinvolved; FXS, fragile X; OID, other/idiopathic ID.