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A comparison of family financial and employment impacts of fragile X syndrome, autism spectrum disorders, and intellectual disability

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ABSTRACT

This study compares the family financial and employment impacts of having a child with fragile X syndrome (FXS), autism spectrum disorder (ASD), or intellectual disabilities (ID). Data from a 2011 national survey of families of children with FXS were matched with data from the National Survey of Children with Special Health Care Needs 2009–2010 to form four analytic groups: children with FXS ($n = 189$), children with special health care needs with ASD only ($n = 185$), ID only ($n = 177$), or both ASD and ID ($n = 178$). Comparable percentages of parents of children with FXS (60%) and parents of children with both ASD and ID (52%) reported that their families experienced a financial burden as a result of the condition, both of which were higher than the percentages of parents of children with ASD only (39%) or ID only (29%). Comparable percentages of parents of children with FXS (40%) and parents of children with both ASD and ID (46%) reported quitting employment because of the condition, both of which were higher than the percentages of parents of children with ID only (25%) or ASD only (25%). In multivariate analyses controlling for co-occurring conditions and functional difficulties and stratified by age, adjusted odds ratios for the FXS group aged 12–17 years were significantly elevated for financial burden (2.73, 95% CI 1.29–5.77), quitting employment (2.58, 95% CI 1.18–5.65) and reduced hours of work (4.34, 95% CI 2.08–9.06) relative to children with ASD only. Among children aged 5–11 years, the adjusted odds ratios for the FXS group were elevated but statistically insignificant for financial burden (1.63, 95% CI 0.85–3.14) and reducing hours of work (1.34, 95% CI 0.68–2.63) relative to children with ASD only. Regardless of condition, co-occurring anxiety or seizures, limits in thinking, reasoning, or learning ability, and more irritability were significantly associated with more caregiver financial and employment impacts. Proper management of anxiety or seizures and functional difficulties of children with FXS or other developmental disabilities may be important in alleviating adverse family caregiver impacts.

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1. Introduction

Fragile X syndrome (FXS) occurs in persons with a full mutation in the *FMR1* (fragile X mental retardation 1) gene. The prevalence of FXS is estimated at 1/4000 males and 1/8000 females (Coffee et al., 2009; Peprah, 2012). FXS is characterized by cognitive and behavioral problems in affected males and, to a lesser degree, in affected females (Saul & Tarleton, 1993). FXS is the most common inherited cause of intellectual disability (ID) (Cornish, Turk, & Hagerman, 2008). FXS is one of the principal single-gene disorders associated with autism. Approximately 20% to 50% of persons with FXS meet full diagnostic criteria for autism (Moss & Howlin, 2009).

There are a variety of factors that play a role in how having a child with a disability such as FXS affects the family. These factors include characteristics of the child (e.g., age, severity of disability, extent of behavior problems), the family status (e.g., parental education, parent mental health, maternal genetic status, financial resources, social support systems, number of children with a disability), educational and employment opportunities for the child with FXS, and life events not directly associated with FXS (death of a parent, divorce, job layoff or transition). These factors inevitably interact in complex ways to shape adaptation in both positive and negative ways.

Despite the complexity of these causative influences on family adaptation, a persistent and largely unanswered question is whether families who have a child with one form of disability as a group are more or less affected by their child's particular condition than families who have a child with another form of disability. The literature on the family financial and employment impacts of caring for children with disabilities has primarily focused on autism (Cidav, Marcus, & Mandell, 2012; Kogan et al., 2008; Montes & Halterman, 2008a, 2008b) and ID (McGrath, Stransky, Cooley, & Moeschler, 2011; Schieve, Boulet, Kogan, Van Naarden-Braun, & Boyle, 2011). It has been shown that caregiver financial and employment impacts are greater in families with children with special health care needs (CSHCN) and autism compared to other CSHCN (Kogan et al., 2008). Among families of children with ID, such impacts appear to be greater among families of children with autism, cerebral palsy, hearing or vision impairment (Schieve et al., 2011). There are far fewer published studies of the impacts for families of FXS, perhaps because of the challenges in collecting needed data for rare conditions like FXS. These studies are based on convenience samples and have shown that families affected by FXS experienced a significant negative employment and financial impact (Bailey et al., 2012; Ouyang, Grosse, Raspa, & Bailey, 2010), as well as elevated rates of maternal depression, anxiety, stress, and lowered quality of life (Bailey, Sideris, Roberts, & Hatton, 2008).

Despite the documented association between FXS, ID, and autism, fine-grained analysis has revealed very different developmental, behavioral and cognitive profiles of FXS from those found in persons with idiopathic autism (Lewis et al., 2006; Moss & Howlin, 2009). Family impacts of FXS may be greater than ASD or ID alone because of the complex nature of FXS. Studying the family impact of FXS compared with ASD or ID could help put into context the needs of families affected by FXS, and inform the broader discussion on early and differentiated diagnosis, care, and services for FXS.

In addition to the major diagnoses of FXS, ID, or ASD, we also take into consideration varying functional difficulties and co-occurring conditions when investigating family caregiver impacts. To fully address the consequences of a condition, it is important to know the functional difficulties that exist, beyond receiving a clinical diagnosis (Lollar, Hartzell, & Evans, 2012). Parents caring for persons with ID consider the psychiatric or behavioral problems of their child to be an extra burden (Irazabal et al., 2012; Maes, Broekman, Dosen, & Nauts, 2003; Martorell, Gutierrez-Recacha, Irazabal, Marsa, & Garcia, 2011). The numbers of co-occurring conditions and problem behaviors such as irritability have been shown to be major contributors of family impact of FXS (Bailey et al., 2012; Ouyang et al., 2010). Identifying functional difficulties that have the greatest impact can help design appropriate management strategies and services that meet the needs of affected families.

This study aims to compare the family caregiver financial and employment impacts of having children with FXS to children with ASD and ID, ASD only, or ID only, using similar questions asked in an FXS caregiver survey and National survey of children with special health care needs (NS-CSHCN) 2009–2010. We test the hypotheses that familial caregiver economic impacts of children with FXS are similar to those of children with both ID and ASD, but greater than those with ID or ASD alone. We also investigate the role of affected children's functional limitations (learning, communication, socialization) and co-occurring conditions (depression, anxiety, and seizures) on financial and employment impacts.

2. Methods

2.1. Data source and sample

The sample of persons with FXS was a convenience sample that came from a caregiver survey administered during the year 2011 to families having a child with FXS aged 5 years or older who were enrolled in a research registry (https://www.ourfragileXworld.org). The registry hosted by RTI International, is designed to administer surveys about the nature and consequences of FXS. RTI International has partnered with two fragile X foundations, researchers, and clinicians for survey design and enrollment. Surveys from the registry have provided new knowledge about fragile X and proved that parents are a valuable source of information (Bailey, Raspa & Olmsted, 2011). Of the 508 families invited by mail to participate in the study, 350 respondents (68.9%) completed the survey. Survey respondents were parents of an individual with the full mutation or premutation of FXS, which was determined through parent-reported FXS testing results. Most (92.6%) of the respondents completed the survey online with the remainder completing the survey by phone. A detailed description of the survey can be found elsewhere (Bailey et al., 2012). We restricted the FXS sample to respondents who had a son or daughter