

# The impact of parenting on fragile X syndrome



**Dr Steven F Warren**, University Distinguished Professor of Speech-Language-Hearing: Sciences and Disorders and Investigator within the Schiefelbusch Institute of Life Span Studies at the University of Kansas, has a keen interest in the developmental delays of children with severe intellectual disabilities. Much of his research with his colleague **Professor Nancy Brady** has focused on early communication and language development. In particular, it has analysed the effects of various intervention strategies, with the aim to assess and treat individuals with minimal verbal skills associated with Down syndrome, autism, and fragile X syndrome (FXS).

Over the past decade, Professor Steven Warren and his colleague Professor Nancy Brady have dedicated their research to the behaviour development of children with fragile X syndrome (FXS). To date, their findings indicate that this genetic disorder is not just intrinsically biological, but rather, there are significant environmental variables, such as parent responsivity, that have a substantial impact on the way children with FXS develop.

Dr Warren's recent work is part of a larger investigation of Family Adaptation to FXS – supported by NICHD and representing a collaboration of investigators at the University of North Carolina and the University of Wisconsin. In his latest work, he shifts its scope from looking at the organic development of communication and development in children with FXS. Instead, he broadens the scope to include external forces, namely the impact of parenting. Collaborating with Dr Nancy Brady, a colleague from the Institute for Life Span Studies, Dr Warren has specifically been studying the role of maternal responsivity, a key element of parent-child interaction. Thus far, the findings suggest that there are several fundamentally important variables shaping child development and that FXS is not just intrinsically biological. Rather, it is a condition attributable to the dynamic interaction of biology, behaviour and environment.

## WHAT IS FRAGILE X SYNDROME?

Fragile X syndrome (FXS) occurs due to the expansion of the CGG triplet repeat within the FMR-1 gene on the X chromosome. Full mutation of the FMR-1 gene prevents the production of a protein vital to neurological functioning. FXS is the most common inherited genetic condition that causes intellectual disabilities, behavioural and learning challenges, and mild physical impairment. While FXS occurs in both genders, males are affected more frequently, and with greater severity, as they possess just one X chromosome. FXS is also the most common genetic cause of autism which affects 30 to 35% of males with FXS – it is estimated that up to 70% of them have autism spectrum disorder (ASD).

## MATERNAL RESPONSIVITY IS KEY

Drs Warren and Brady define maternal responsivity as a "healthy, growth-producing relationship characterised by warmth, nurturance, and stability as well as specific behaviours, such as contingent positive responses to child initiations". Highly responsive parents or caregivers create an environment that facilitates and promotes many aspects of child development, including language development. High levels of contingent maternal responsivity to a child's communication attempts result in better language and vocabulary skills, while low levels and/or excessive negative responses have been associated with poor language growth.

**Collaborating with Dr Nancy Brady, Dr Warren has specifically been studying the role of maternal responsivity, a key element of parent-child interaction, within fragile X syndrome patients**



Within the framework of their research, Dr Warren and Dr Brady started with the notion that parenting is known to have effects on child development, and that over time, maternal responsivity may impact how a child develops socially, linguistically, emotionally and cognitively. Given that parenting is among the most consistent elements of a child's environment from birth to adolescence, they focused on the extent to which it influences the development of children with FXS.

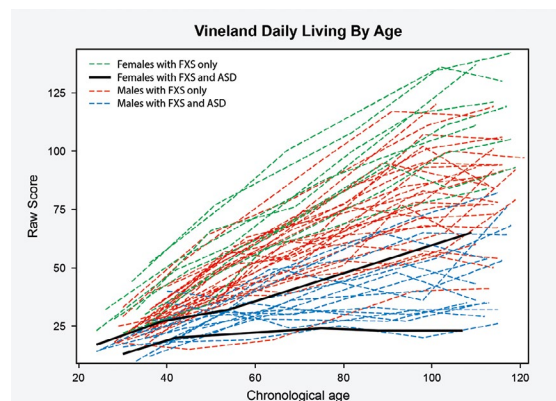
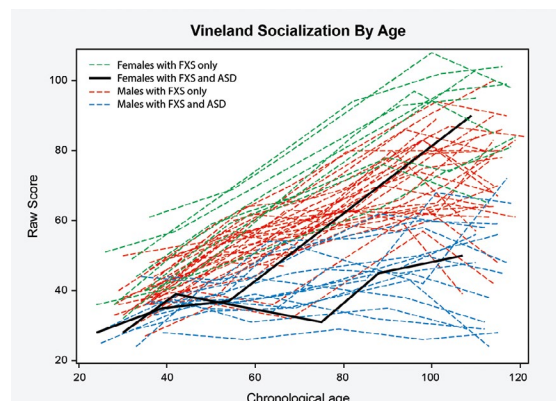
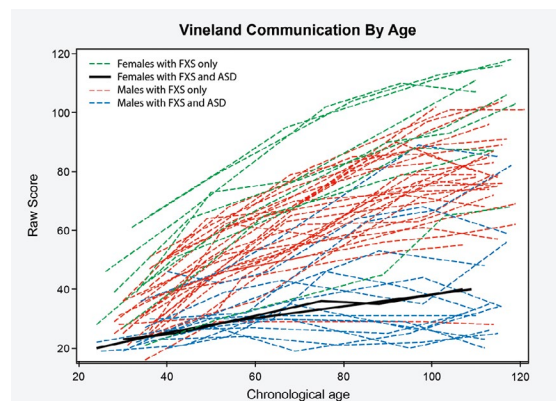
## THE STUDY

In 2002, Dr Warren and Dr Brady began investigating the effects of maternal responsivity on the development of children with FXS. Taking a "natural history" approach, they looked at several variables that influence the type and quality of parenting and the mother-child relationship, including autism status, gender, severity of child behaviour problems, and maternal mental health. Their test group consisted of 55 mother-child dyads from 28 different US states. The children were two years of age on average at the start of the study. All 55 mothers were FXS carriers – three of whom had the full mutation. 33% of the 55 children had comorbid autism – 16 males and two females.

Each family was visited for three to four hours every 18–20 months in their home. Mother-child interactions were videotaped during these visits in a variety of daily situations, i.e., during book reading or making snacks. The video data was then coded for performance measures of child communication behaviours, parent responsivity and behaviour management. A primary focus was on the number and type of maternal responses that were contingent on child initiations, including verbal expansions and directives. In addition, they rated more qualitative variables such as the maternal tone, warmth, and flexibility of commenting and expanding on the child's responses.

## THE OUTCOME

The overall aim of the study was to determine whether sustained maternal responsivity affects measures of language, cognitive, social, emotional and adaptive development. What Dr Warren and Dr Brady discovered was that some effects of parental responsiveness are quite significant even when controlling the child's developmental level, presence of autism and gender. Maternal responsivity, actually accounted

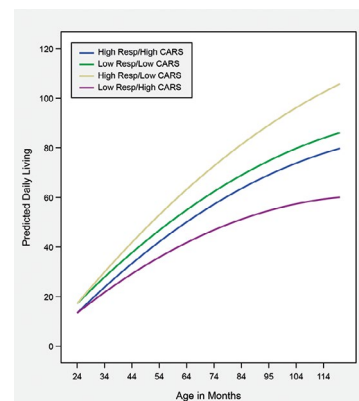
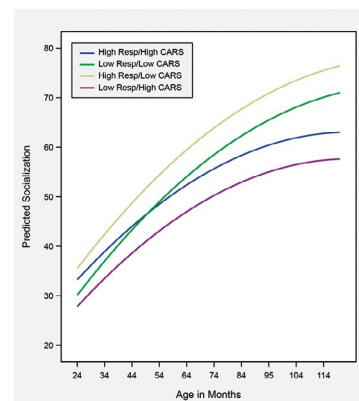
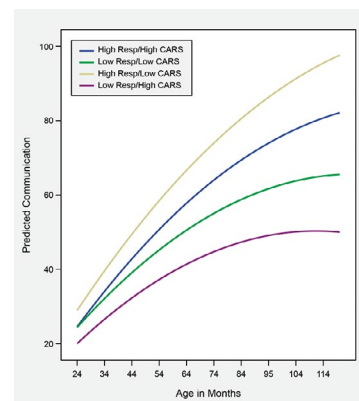


The graphs on the left show growth in Vineland measures of communication, socialisation, and daily living skills across early and middle childhood. The different line colours reflect females with FXS only, females with FXS and ASD, males with FXS only and males with FXS and ASD. Graphs on the right show average growth for these measures separated into four subgroups: children with highly responsive mothers and low scores on the CARS (a measure of autism symptoms) are indicated by the top line labelled "High Resp/Low CARS"; children with low parent responsivity and high CARS scores are represented by the bottom line.

for over 40% of the variance in receptive and productive vocabulary development in children up to the age of nine.

Dr Warren and Dr Brady then expanded their analyses to the adaptive behaviour development of the children with FXS. Adaptive behaviour refers to the social and practical competencies of an individual to meet the demands of everyday living. The Vineland Adaptive Behavior Scales specifically measure personal and social skills in five areas: communication, daily

living skills, socialisation, motor skills, and maladaptation. The results indicated that there was a significant decline in adaptive behaviour for over half of the sample in middle childhood. However, children who experienced highly responsive parenting over early and middle childhood either showed no decline in skills or only very modest declines from early childhood to middle childhood. High levels of sustained maternal responsivity significantly abated regression in communication, socialisation, and daily living. Thus, high levels of



# Q&A

## How has your research been received by the general public?

The reaction has been very positive. Many people inherently believe that parenting plays an important role in development and that we, as humans, aren't simply products of our genetic heritage. In the case of FXS, our study allows people to see what parenting is influencing the child's development, and what is not.

## How do behavioural patterns in parent-child relationships that involve a child with intellectual disabilities differ from relationships where there is none?

These patterns vary a great deal just as they do with parenting typically developing children. In general, the more a child with a disability initiates, the more parents and others in the child's environment can respond and incidentally help teach the child about the world around them. Low rates of child initiation, which are often observed in children with intellectual disabilities may generate a more directive than responsive style in some parents, especially if the child has problem behaviours.

## In what ways will the results of your research advance clinical treatment of individuals with FXS?

Our findings may influence efforts to enhance the parenting skills in family members both early in a child's development as well as later on. Our research suggests that sustained responsive parenting over time is likely to generate better long term outcomes than early intervention alone.

## Why did you choose to look at mother-child interactions specifically, as opposed to father-child?

We choose mother-child interactions as opposed to father-child because we believed that on average mothers would have somewhat more interaction with the child across development. We also chose mothers because they are carriers of the FXS disorder. Thus we are interested in the extent to which their carrier status might influence their behaviour over time as well. Virtually all the mothers in this study have contributed valuable insights into what it is like to parent a child with FXS. In many respects, they have been amazing partners in this research.

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# Detail

## RESEARCH OBJECTIVES

Professor Warren's recent research focuses on understanding the developmental delays associated with a chromosomal disorder called fragile X syndrome (FXS). His latest work, done in collaboration with Dr Nancy Brady, investigates the effects of parenting on the development and behaviour of adolescents with FXS.

## FUNDING

- Eunice Kennedy Shriver National Institute of Child Health and Human Development (NICHD)
- National Institutes of Health (NIH)

## COLLABORATORS

Nancy Brady, PhD (University of Kansas); Kandace Fleming, PhD (University of Kansas); Leann Smith, PhD (University of Wisconsin); Elizabeth Berry-Kravis (RUSH University Medical Center, Chicago); Life Span Institute

## BIO

Dr Steven Warren has over 40 years' experience studying the development of children with developmental delays and has published over 185 papers. His collaboration with Dr Nancy Brady, Associate Professor of Speech-Language-Hearing, draws on her substantial research experience of the development of communication in early childhood and in individuals with developmental disabilities.

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contingent maternal responsivity appear to both enhance development and act as a brake on later declines in development experienced by some children with FXS.

## MOVING FORWARD

Dr Warren and Dr Brady are now following this same sample of 55 families as the children move through adolescence to adulthood with support by an additional five years of funds provided by the National Institutes of Health (NIH R01HD084563). Their aim is to examine mother-child

relationship patterns and trajectories from early childhood through adolescence, taking into consideration several fundamentally important variables and domains. They are convinced that understanding the effects of parenting on the development and behaviour of children and adolescents with FXS has implications for educational planning and intervention, as well as clinical treatment of individuals with FXS and related genetic disorders.