**Title:** What Skills are Lost When Children with Fragile X Syndrome Experience Declines in Adaptive Behavior?

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**Introduction:** We previously reported that more than half of a longitudinal sample of 55 children with fragile X syndrome (FXS) showed some raw score declines on the Vineland Adaptive Behavior Scales (VABS) (Hahn, et al. 2015). The VABS is made up of four domains: communication, social, daily living skills and motor. This raises two obvious questions. First, what specific skills are being lost in each domain? Second, what parent and child variables are associated with these declines? The answers to these questions are fundamental to understanding the nature of these declines and identifying potential mechanisms to eliminate them and their impact on child functioning.

**Method:** These data are part of an ongoing longitudinal study of 55 mothers and their child with FXS (44 males and 11 females) that began when the children were age 2 on average. Here we present data for 52 children who were assessed at age of 9 or 10 (mean age 9.5 years, SD=.31). At each home visit, the mother reported on her child’s ability to demonstrate each skill using the VABS to indicate 0=no never, 1=sometimes or partially, 2=Yes, usually for each item/skill. Mothers also completed the Child Behavior Checklist (CBCL) and the Parent Stress Index (PSI). The focus of this analysis is at the VABS item level, allowing us to determine what specific skills had declined for each child from previous observations. For this analysis, child performance on each item was categorized as declining or maintaining. The definition of a “declining item” was one in which the child’s score at age 9-10 was less than at least one of their previous scores. The definition of a maintaining item was one in which the child scored greater than 0 at age 9-10 and this score was greater than or equal to all previous scores at earlier points in time.

**Result:** We focused our analysis on items in each domain that showed declines for 8 or more individuals at the 9-10 age period. There were 14 items in the VABS social domain that showed declines across 8 or more children—more than all other domains combined. Further analyses indicated that children who declined on the social domain had higher scores on “withdrawn/depressed” and “anxious/depressed” subscales of the CBCL, and higher ratings of parent dysfunctional interaction and total stress from the PSI. Interestingly, ratings of maternal responsivity were higher for this group.

**Discussion:** The picture these data paint for children with FXS who showed declines is one of social withdrawal by the child. The skills that have been “lost” according to the VABS include items such as “shares toys”, “has a preferred friend,” “has a group of friends”, etc. The loss of these skills is associated with increased ratings of child depression and dysfunctional parent child interaction as compared with children who maintain these skills. Data on maternal responsivity for mothers of these children suggests that they may have recognized these declines in their child and increased their efforts to be responsive to the child to enhance development.


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