

Fragile X Syndrome and Family Occupations: Sertraline Treatment

Martina Dualan OTS, Elena Javier OTS, Decerie Mendoza OTS, Tracy Ye OTS, and Laura Greiss Hess PhD, OTR/L
Occupational Therapy Department, Dominican University of California

BACKGROUND

Fragile X Syndrome (FXS) is the most common inherited form of intellectual and developmental disability (IDD) and a single gene cause of autism spectrum disorder (ASD). FXS phenotype includes anxiety, decreased communication / social skills, behavioral excesses, & sensory processing challenges (NFXF, 2017). Clinical trials have shown positive responses to treatment including sertraline - Zoloft (Hess et al., 2016)

Current FXS literature is well anchored in the medical model. Many outcome measures for FXS / IDD are focused on performance skills and show little progress over time. Raw scores (RS) reflect some point increases, yet standard scores (SS) show minimal to no change. Additionally, these traditional standardized assessments fail to capture occupational performance and context.

RESEARCH QUESTION

How can semi-structured interviews reveal occupational performance changes in response to medication in a more contextually valid and sensitive manner when compared to traditional standardized outcome measures?

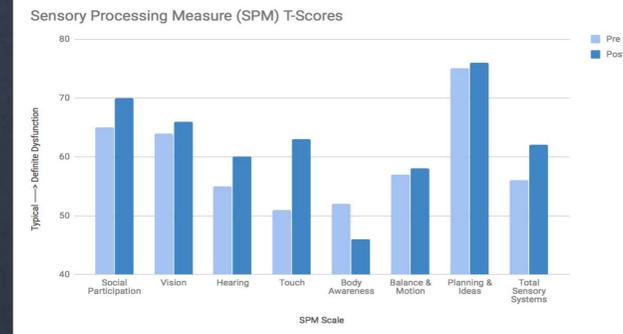
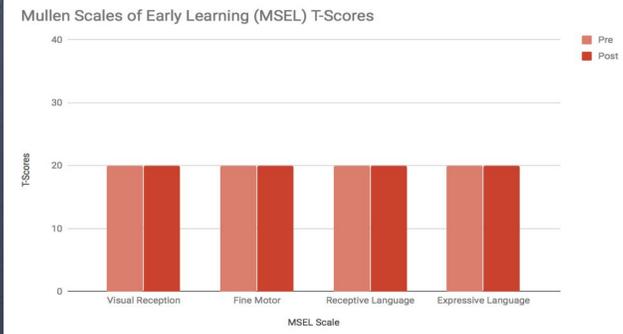
MIXED METHODS: In-Depth Case Study Analysis

Standardized assessments and interviews given at baseline and 6 months post.

Participants (N=4)	Families and children with full mutation FXS, 2-6 y/o, all received sertraline treatment
UC Davis MIND Institute Sertraline Study	(N=1, IQ = 78, N=3 on poster, mean IQ = 53.6)
Quantitative	<ul style="list-style-type: none"> Sensory Processing Measure (SPM) Mullen Scales of Early Learning (MSEL)
Standardized Assessments	<ul style="list-style-type: none"> RS examined, minimal change SS reported in results
Qualitative	Impact of FXS phenotype on family occupations
Semi-structured Interviews	<p>Constant Comparison Method (Strauss & Corbin, 1990)</p> <ul style="list-style-type: none"> - 25% of total data (8 transcripts) coded to 100% consensus across 4 researchers

RESULTS

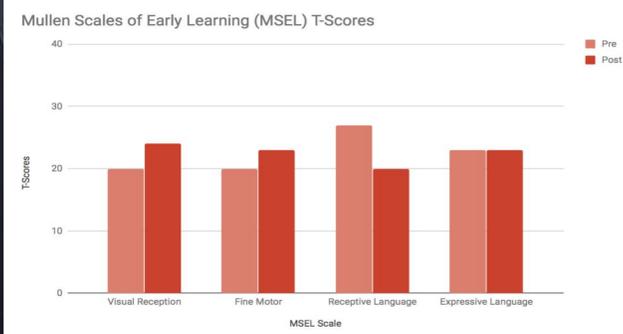
Isaac Age: 40 mo, IQ = 49



The other thing is they feel like his language is improved. It's not showing so much on the testing...he kind of went from hardly saying anything to really starting to repeat things we say and he started to say more words of his own.

Sometimes it's simple. We have rocking chairs. In their place and our place. And we just sit down and walk with him. He likes the rocking motion sometimes. If you rock with him it will calm him down.

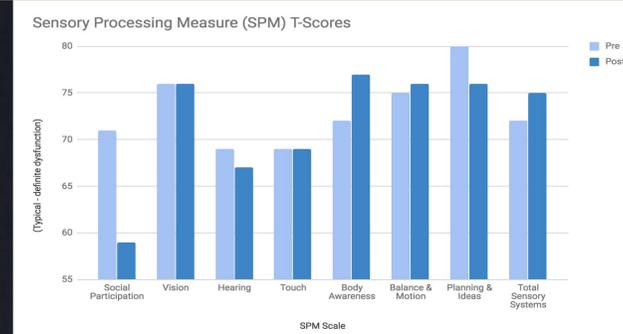
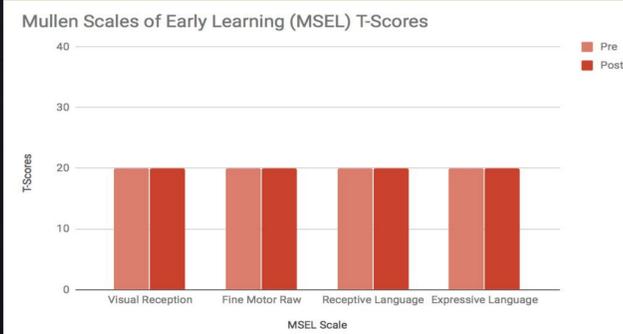
Derek Age: 41 mo, IQ = 56



Yesterday he wanted a yogurt, so he went to the refrigerator and said "eat" and I opened it up and said "what do you want?" and he grabbed his yogurt, and said "you need a spoon", then he went to the drawer. And you know, I opened the drawer for him because it's hard for him, so he can grab the spoon, and he said, "I eat."

We went out to dinner with our neighbors, and took him to a place he has never been before, and through the whole entire dinner at a restaurant, and was fine. He was completely fine. He sat and colored. So we can do more things like that.

Shiloh Age: 65 mo, IQ = 56



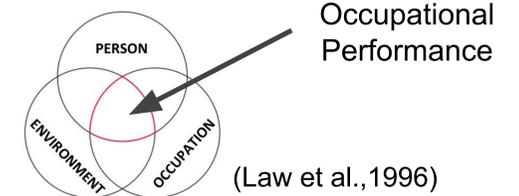
I think he is doing great, if we don't understand what he is saying, show me a picture, show me what you are talking about and he will show you.

He's very good at routine, when he's done with his goldfish crackers he'll bring me the bowl or the cup on the counter in the kitchen. We've started having him set his place at the table. Things like that, chores, he loves to help me mop.

DISCUSSION

Major themes revealed FXS phenotype impacts the family and child's occupational engagement across contexts.

- Families reported positive outcomes of sertraline
 - Improvement in daily occupational engagement and improvement in anxiety, behavior management, self-regulation and social participation.
 - Traditional standardized assessments have limited sensitivity to change for FXS.
 - Standard scores do not reflect the improvement families are reporting in their occupations and daily life.
- Interviews reveal broader and more contextually relevant changes in occupational performance in response to sertraline treatment in comparison to traditional standardized measures.



IMPLICATIONS FOR OT

A need for an occupation centered approach to serving children and families with FXS

- Look beyond assessment scores
- Examine functioning across contexts
- Include family occupations and the impact of living with FXS.
- Look at individual differences and raw score changes related to occupations.

ACKNOWLEDGEMENTS

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