Title: Role of Occupational and Physical Therapy on Motor Skills in Infants with Down Syndrome and Fragile X Syndrome

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Introduction: Fragile X syndrome (FXS) and Down syndrome (DS) are both neurogenetic disorders associated with intellectual disability (ID) and developmental delays (Dykens, Hodapp, & Finucane, 2000; Hagerman & Hagerman, 2002). Infants with either of these conditions are at high risk for acquiring their fine and gross motor skills at a slower rate than that of a typically developing infant (Bailey et al., 2001; Hinton et al., 2013; Periera et al., 2013). Due to these early motor delays, coupled with timing of early diagnosis, infants with FXS and DS often receive occupational therapy (OT) and/or physical therapy (PT). Each of these address early sensory motor impairments and functional performance; however, the substantial dearth of behavioral phenotyping during early development in both FXS and DS may translate to a lack of targeted intervention specifically addressing the unique needs of each disorder. Furthermore, the impact of therapy type and dosage on early motor skills in these neurogenetic conditions is largely unsubstantiated. As such, the current study aims to investigate the rates of each type of therapy within FXS and DS, examine differences in motor abilities of infants with FXS and DS, and determine whether there are systematic differences in motor abilities as a function of receiving early intervention services within each group.

Method: Participants included a total of 46 children assessed at 12-months of age: 33 FXS (chronological age $M = 12.90$ months, $SD = 1.04$ months) and 13 DS (chronological age $M = 12.42$ months, $SD = 0.76$ months). Parents reported whether children were receiving therapy, which type, and the amount of time spent in therapy each week (in minutes). The Vineland Adaptive Behavior Scales, Second Addition (Vineland-II; Sparrow et al., 2005) interview was conducted with participant’s parents. Children’s motor skills were also assessed using the Mullen Scales of Early Learning (Mullen, 1995). Raw scores from the gross and fine motor subdomains of the Vineland-II and from the gross and fine motor scales of the Mullen were used in analyses. We used descriptive analyses to determine the proportion of participants in each group – FXS or DS – that were receiving PT or OT. We then used non-parametric group comparisons, due to discrepant sample sizes across groups, to test group differences on fine and gross motor functioning across direct assessment (MSEL) and parent report (VABS-II). Finally, we used non-parametric group comparisons to determine whether groups differed on amount of therapy each received, as well as to determine, within each group, whether those receiving therapy were significantly different on motor skills from those not receiving therapy.

Results: Descriptive analyses demonstrated that 42.42% of the FXS group were in OT services, whereas 39.39% were in PT. In the DS group, 46.5% were in OT services, whereas 76.92% were in PT services. Groups significantly differed on fine and gross motor domains across the MSEL and VABS-II (all $p's < .015$); however, there were no significant differences in the amount of therapy – PT or OT – between children with FXS and those with DS. Finally, we examined whether within each group, there were systematic differences in motor skills in those receiving OT or PT services. Interestingly, there were no significant differences in fine or gross motor on the MSEL or VABS-II between children with FXS receiving OT and those not, and this finding was also consistent for those receiving PT and those not (all $p$-values > .05). Similarly, infants with DS receiving OT did not significantly differ on any measure of fine or gross motor from those not receiving OT, and this was also consistent across infants with DS who received PT and those that did not (all $p$-values > .05).

Discussion: Findings indicate relatively high rates of infants with FXS and DS receiving early intervention services. Although infants with FXS demonstrated significantly higher gross and fine motor skills than infants with DS, this could not be accounted for by higher amounts of early intervention, as groups were equivalent on time spent in therapy. Furthermore, within each group, there were no differences in motor abilities as a function of receiving OT or PT services. The preliminary nature of this study and modest sample sizes require these findings to be considered with caution. However, it is quite notable that despite relatively high amounts of weekly therapy, these intervention efforts did not manifest in better motor functioning in any domain in FXS or DS. It is possible that early intervention may prevent those receiving services from falling behind in motor development relative to their peers not receiving services. Alternatively, it may also be the case that current early intervention approaches are insufficient in addressing the unique needs that infants and toddlers with FXS or DS may experience as a result of their...
neurogenetic phenotype. Future work is necessary to fully delineate the efficacy of early intervention in addressing delayed development in FXS and DS.

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