

THESIS

ACTIVITY PARTICIPATION AND MOTIVATION IN DOWN SYNDROME

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ABSTRACT

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This paper examines activity participation, motivational style, and daily living skills in the Down syndrome (DS) behavioral phenotype.

Children with DS ($n = 26$) and mixed-etiology intellectual disability (ID, $n = 18$) ages 5 – 22 participated. The CBCL was used to assess activity participation, the Reiss Profile MR/DD measured motivational style, and the VABS measured daily living skills.

Children with DS were more motivated by independence than children with ID ($p = .001$). In the DS group, sports participation was negatively associated with pain avoidance ($r = -.429$) and frustration avoidance ($r = -.518$) but positively associated with daily living skills ($r = .452$). Daily living skills were negatively associated with curiosity.

Though children with DS were motivated by independence, this was not necessarily related to function. Individuals with DS may need heightened tolerance for frustration and even pain to maintain motivation for physical activity. Implications for intervention are discussed.

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Introduction

Down Syndrome (DS) is a genetic disorder characterized by a range of physical abnormalities and developmental delays in motor planning skills, language, and cognitive skills (Rogers, 2005). It is the most common genetic cause of intellectual disability (Sherman, Allen, Bean, & Freeman, 2007). The prevalence of DS is 1 out of every 800 live births (12.8 per 10,000) in the United States (Canfield et al., 2006), and the rate is reported to have increased by 31.1% since 1979 (Shin, Besser, Kucik, Lu, Siffel, & Correa, 2009).

Children with DS demonstrate patterns of relative strengths and weaknesses in development, which can be characterized as the DS behavioral phenotype (Fidler, Hepburn, & Rogers, 2006). This behavioral phenotype includes relative strengths in social relatedness, mental age appropriate receptive language skills, and relative challenges (delays beyond what is expected for mental age) in motor skills and expressive language (Fidler, Hepburn, & Rogers, 2006). Hodapp (2004) hypothesized that characteristics associated with the behavioral phenotypes of individuals with genetic syndromes would directly and indirectly impact their development across time. He described one of those direct effects as being the environments and occupations an individual with a genetic syndrome chooses. Additionally, children with DS tend to display difficulty with motivational style, often quitting out of activities or using social skills to avoid challenges, and this continues to worsen as they grow older (Pitcairn & Wishart, 1994). This paper will examine the behavioral phenotype associated with DS. Specific attention will be given to particular aspects of the behavioral phenotype including socialization, adaptive behavior, motivational style, and how these factors

relate to participation in activities. By examining relationships of these factors in respect to the DS behavioral phenotype it may be possible to better understand elements related to motivation and activity engagement that could inform intervention and future research for this population.

Given the high prevalence of DS and that individuals with DS have significant needs, it is important to research this population in order to ensure that the best-possible interventions are being implemented. Early intervention, special education, and parental support are needed to help these children achieve their highest potential (Rogers, 2005).

The Down Syndrome Behavioral Phenotype

Behavioral phenotypes refer to a collection of characteristics and traits that are more likely to appear in an individual with a certain genetic disorder than in others. People with DS, while they are each of course unique, tend to display patterns of behaviors and characteristics. Evidence suggests that individuals with DS tend to demonstrate a specific behavioral phenotype (Dykens, 1995; Hodapp, 2004; Walz & Benson, 2002), which begins to emerge at an early age (Fidler, Hepburn, & Rogers, 2006). Understanding these characteristics in the DS population could be useful for intervention planning and helping to prevent functional difficulties.

It has long been understood, for example, that children with DS demonstrate deficits in the areas of expressive language (Hodapp, 2004) but that their receptive skills tend to be better (Dykens, Hodapp, & Evans, 2006; Fidler et al., 2006a). As early as age three, children with DS begin to show patterns of emerging relative strengths (visual processing and receptive language) and relative challenges (gross motor skills and expressive language) and a tendency towards sociability (Fidler et al., 2006). In

comparison to people with Prader-Willi and Williams syndrome, individuals with DS have greater visual memory and visual motor skills (Dykens et al., 2001; Hodapp et al., 2000). Compared to children with non-specific developmental delays, children with DS demonstrate deficits in motor skills (Fidler, Hepburn, Mankin, & Rogers, 2005; Fidler, Most, Booth-Laforce, & Kelly, 2008) and decelerated rates of cognitive development over time (Fidler et al., 2008; Hodapp, Evans, & Gray, 1999). Below we examine what is known about the DS behavioral phenotype in the areas of social skills, adaptive behavior, and participation in daily life.

Social skills. Social functioning is an area of particular interest within the DS behavioral phenotype. People with DS, who years ago were said to have “Prince Charming” syndrome (Dykens, 2006), have developed a stereotype of being social and friendly, and research in more recent times has upheld this stereotype. In a study looking at mothers’ perceptions of their children with DS, children were described most often as being “affectionate”, “loveable”, and “nice,” and other descriptions included “cheerful” and “fun” (Carr, 1995). Though they may be less socially interactive than their typically developing peers (Beegly, Weiss Perry, & Cicchetti, 1989; Cielisnski, Baughn, Seifer, & Contreras, 1995; Hamilton, 2005), social development emerges as a relative strength in children with DS when compared to other children with developmental disabilities (Fidler et al., 2006; Walz & Benson, 2002). They have been shown to have more frequent smiling behavior than do their peers with other developmental disabilities (Fidler, Barrett, & Most, 2005) and to have higher levels of social engagement and orientation (Fidler et al., 2008). When children with DS are compared to other children

with developmental delays, it becomes evident that sociability is a relative strength associated with this behavioral phenotype.

While sociability is a major component of the DS behavioral phenotype, it is something of a double-edged sword. Individuals with DS often use their social strengths at to escape challenging tasks. Pitcairn and Wishart (1994) found that when faced with “impossible tasks,” children with DS were more likely to use social attention-getting behavior or “party tricks” to avoid doing the task. Compared to children with developmental delays and typically developing children, children with DS displayed more negativity (i.e. facial and bodily frustration), had a lack of effective coping strategies, and were more likely to engage with the experimenter rather than seek assistance with an impossible task (Jahromi, Gulsrud, & Kasari, 2008). Similarly, children with DS when doing a puzzle, tended to look more frequently to the adult for help whereas both typically developing children and children with developmental delays looked more frequently to the adult’s puzzle for ideas (Kasari & Freeman, 2001). This is consistent with the Fidler et al., (2005) finding that children with DS elicited more help during an object retrieval task than children with developmental delays and typically developing children. This contributes to evidence that children with DS are, not only highly social, but are likely to increase social behavior to escape challenging activities rather than trying to solve problems independently. Furthermore, while strengths in social engagement and orientation emerge at an early age, children with DS display less-pronounced development of emotion regulation (Fidler et al., 2008).

Interestingly, this social behavior in individuals with DS shows mixed development over time. Fidler, Barrett, & Most (2005b), determined that children with

DS showed lower levels of anxiety and withdrawn behavior than children with mixed developmental delays but that the anxiety and withdrawn behavior increased with age for both groups. Additionally, this study points to an age-related trend towards decreased smiling and social behavior in children with Down syndrome. While young children with DS exhibited greater amounts of smiling and attention seeking behavior than did younger children with mixed developmental delays, their smiling and attention seeking behavior decreased over time. Dykens, and colleagues (2002) found that older children with Down syndrome were more withdrawn than younger children – more secretive, less talkative, and more often preferring to be alone. Sloper, Turner, Knussen, and Cunningham (1990) observed an *increase* in social contacts as children with Down syndrome got older, while Carr (2007) observed that people with DS had *fewer* relationships as they grew older and transitioned into adulthood.

Adaptive Behavior. The ability to participate in activities is supported by one's capacity for adaptive behavior. Adaptive behavior refers to the "ability to meet daily living responsibilities and respond to the needs of others, including conceptual, practical, and social skills that people need to function in their everyday lives" (Ditterline & Oakland, 2009, p. 45). Children with DS demonstrate significantly lower levels of adaptive behavior than typical peers (Coe et al., 2009), and research has indicated that adaptive behavior develops steadily early on but plateaus around middle childhood for this population (Dykens, Hodapp, & Evans, 1994). The variability of adaptive behavior in middle childhood increases, however, indicating that the plateau in adaptive development does not apply to all cases. Interestingly, Volman, Visser, & Lenvelt-Mulders (2006) reported that adaptive performance in children with DS is predicted to a

greater extent by motor ability than by mental ability. It should be noted that the assessment used to measure mental ability in this study was a single shape sorting task involving both fine motor skills and mastery motivation. Because Volman et al.'s (2006) measure of 'mental ability' may have been narrow, further evidence is needed to determine whether motor abilities is indeed a better predictor adaptive behavior than mental abilities.

Studies using the Vineland Adaptive Behavior Scales (Sparrow, Balla, & Cicchetti, 1984) have reported mixed patterns of adaptive strengths and weaknesses among individuals with DS. Fidler et al., (2006) reported that toddlers with DS had stronger adaptive socialization skills than communication and motor skills. Similarly, Dykens, Hodapp, & Evans (1994) reported children with DS as having better adaptive socialization and daily living skills relative to communication skills. Finally, Fidler et al., (2005b) found that adaptive behavior skills were significantly correlated with planning skills on battery of praxis tasks in toddlers with Down syndrome.

A descriptive study used the parent-reported Pediatric Evaluation of Disability Inventory (PEDI) to characterize the functional performance of 5-year old children with DS and found that these children demonstrate many challenges to participation in daily life (Dolva, Coster, & Lilja, 2004). Highest functioning was found in the area of mobility, and great variability was found in self-care and safety. Disability was reported in the areas of socialization and self-care. Parents reported greatest concern regarding "social interaction, language, peer play, and participation in school without being stigmatized" (p. 628). This study did not compare children with DS to other disabilities, but it highlights the importance of adaptive behavior in overall functioning and

participation. Interactions of developmental deficits over time are underpinnings to the difficulty that children with DS have with adaptive skills. A better understanding of adaptive behavior – specifically of daily living skills – within the DS behavioral phenotype may provide insight into the everyday functioning of individuals with DS.

Participation. If the behavioral phenotype for DS includes a tendency to participate in certain activities (Hodapp, 2004), it could be beneficial for therapists to use this information to guide individuals with DS and people in their support system towards activities that might be motivating. Participation in occupations is an important part of individual well-being (Hocking, 2009). A qualitative study, which investigated the experiences of individuals who had received the “Stevie Award for Outstanding Persons with Down Syndrome”, found that participants were more likely to succeed and manage challenges when they persevered in learning activities (i.e. using computers, reading new books) and were involved in extra-curricular and volunteer activities (Li, Liu, Lok, & Lee, 2006). Support from family members and teachers helped these individuals to develop talents in sports and art activities

The aforementioned qualitative study found that successful individuals with DS were those who actively engaged in occupations. However, for many individuals with DS, participation in activities is challenging. Research in the area of play, often referred to as the “occupation” of childhood, is replete with examples of how individuals with DS may experience challenges in engaging in activities. Compared to their typically developing peers, children with DS have been shown to spend less time participating in social play (Cielinski, Vaughn, Seifer, & Contreras, 1995) and symbolic play and more time in simple manipulative play (Beeghly, Perry, & Cicchetti, 1989). However, their

play behavior is similar to that of their mental age-matched peers, with both groups engaging in similar amounts of manipulative and symbolic play. Fidler et al. (2006) found that children with DS exhibit significantly more socialization during play and leisure activities than when engaged in interpersonal relationships. This might indicate that social strengths in children with DS have more to do with socializing around activity than social relationships in general.

There is a paucity of studies examining activity preferences in the DS population. The ones that do exist provide support for Hodapp's theory (2004) that phenotypes directly influence activity choice. A descriptive longitudinal study in the U.K. examining popular leisure activities among adults with DS found that about 90% engaged in listening to music and watching TV on a weekly basis, and about 80% engaged in reading and/or looking through books (Carr, 2007). A third of adults engaged in drawing and painting, the amount having decreased across time (50% of 21-year-olds engaged in drawing and painting in Carr's 1995 study). Of the participants in this study, over half participated in sports on a weekly basis including swimming, bowling, and snooker (a billiard sport similar to pool). All the sports mentioned in the article tend to have a low-intensity level and be social in nature.

A comparative study conducted by Rosner, Hodapp, Fidler, Sagun, and Dykens (2004) used the Child Behavior Checklist (CBCL) to examine both behaviors and preferred activities of children with Prader-Willi (PWS), Williams (WS), and Down syndromes. This study looked at how cognitive-behavioral profiles relate to favorite activities. Children with DS showed a higher social competence overall than the other two groups; they tended to be more involved in organizations than the PWS group, and

they behaved more positively with others than the PWS group. While children with DS had fair amount of skill in job-related activities, they scored significantly lower than the other two groups in skill and participation in non-sport activities. They tended to engage in visual-motor activities with an inclination towards arts and crafts, and they showed interest in music, particularly dancing, singing, and listening to music. In the end, this study raised questions as to *why* individuals with certain genetic syndromes tend to engage in certain activities.

Another study assessed participation levels of people with DS, PWS, and WS in specific leisure activities (Sellinger, Hodapp, & Dykens, 2006). Participants ranged in age from 2 to 54, and this study differed from that of Rosner et al. (2004) in that Sellinger and colleagues assessed *how often* participants engaged in certain activities rather than simply identifying in which activities individuals were engaging. Compared to the other two groups, individuals with DS performed coloring and drawing activities more often. They also, along with individuals with WS performed *passive* musical activities (i.e. listening to music and singing in choir) more often than did individuals with PWS; however, when the musical activity was more *active* (i.e. playing a musical instrument), people with DS participated less than those with WS. People with DS who were older were found to participate more in social activities (going out with friends and talking on the phone) but less in arts-and-crafts activities and physical activities.

Despite relative social strengths in people with DS, studies have shown mixed results regarding change in *participation* of social activities over time (Carr, 2007; Dykens et al., 2002; Sellinger, Hodapp, & Dykens, 2006; Sloper et al., 1990). Perhaps this discrepancy indicates that, while social activities continue to be desirable for

individuals with DS, opportunity for socialization and development of relationships is not always readily available. Indeed, Sloper, et al. (1990) found that familial support factors were positively correlated with the social life of children with DS. The ability to participate in social relationships is largely due to resources and availability of supervision, and a decrease in such participation may be in part due to the aging of accompanying parents (Carr, 2007). The main reasons for low social/leisure participation among adults with DS in another study were lack of available accompaniment, lack of leisure activity skill, and lack of available leisure activities (Putnam, Pueschel, & Holman, 1988). This highlights, again, the importance of building knowledge about the types of activities in which children with Down syndrome tend to participate in order to build skills and encourage participation across the lifespan.

Motivation. It has long been recognized that typically developing children possess an intrinsic motivation to explore the environment. White (1959) described a motivation towards mastery, of competently interacting with and exploring the environment in a way that naturally produces feelings of efficacy and satisfaction. Research has demonstrated that low motivation – limited curiosity and limited seeking mastery for the sake of feeling competent – tends to be an issue for children with intellectual disabilities (Bennett-Gates & Zigler, 1999; Harter & Zigler, 1974), and many studies have reported low motivation in children with DS specifically (Glenn, Dayus, Cunningham, & Horgan, 2001; Niccols, Atkinson, & Pepler, 2003). For example, Ruskin, Mundy, Kasari, and Sigman, (1994) found that children with DS, compared to typically developing children, tended to try to get out of challenging toy-related tasks and spent less time in goal-directed play with toys. Mothers of the children with DS in this

study described their children as being less persistent and less competent in object exploration. Though Gilmore, Cuskelly, Jobling & Hayes (2009) reported no difference in mastery motivation between children with DS and mental-age matched peers, it seems that children with DS tend to avoid or get out of activities that become challenging for them (Gilmore, Cuskelly, & Hayes, 2003; Kasari & Freeman, 2001; Pitcairn & Wishart, 2004). Many such children have, however, developed an effective coping mechanism which involves getting out of challenging tasks by socially engaging with whoever is directing the task (Pitcairn & Wishart, 1994; Kasari & Freeman, 2001). Though children with DS often have low intrinsic motivation for mastery, which potentially decreases persistence when activities become challenging, they may instead be motivated in other areas such as socialization.

Individuals with DS tend to be below average in skill and time spent in activities (Rosner et al., 2004), and motivational factors likely play a role. Whether this is due to inherent motivational deficits or a learned quitting behavior due to repeated failures is difficult to distinguish. Limited participation in physical activity, for example, can likely be attributed in part to factors such as hypotonia (Harris & Shea, 1991), delays in achieving motor milestones (Chen & Woolley, 1978; Jobling, 1998; Palisano et al., 200) and possibly to a tendency to develop hypothyroidism (Fidler & Daunhauer, in press), but there may be underpinnings of motivational factors as well. People with DS tend to lead sedentary lifestyles (Dykens et al., 2002; Whitt-Glover, O'Neill, & Stettler, 2006) and have a high incidence of obesity and being overweight (Cronk, Chumlea, & Roche, 1985; Rubin, Rimmer, Chicoine, Braddock, & McGuire, 1998) even when compared to other individuals with disabilities (Melville, Cooper, Morridon, Allan, Smiley, & Williamson,

2007). Adults with DS have reported that the biggest barrier to staying active is difficulty finding someone to be active with (Medlen & Peterson, 2000). Social factors may actually *promote* participation in healthy lifestyle. According to one study, factors such as friendship and social opportunities predicted healthier BMIs in individuals with DS (Fujiura, Fitzsimmons, Marks, & Chicoine, 1997). Another study found that adults with DS preferred to go bowling with others with DS rather than with people that did not have a disability (Neumayer, Smith, & Lundergren, 1993). Questions can be raised as to whether sedentary lifestyles in people with DS is due more to low intrinsic motivation in general or lack of targeting correct motivational factors, such as social motivation.

Certainly many reasons contribute to individuals with DS choosing (or not choosing) to participate in certain activities, but underlying motivation is particularly relevant to interventionists who want to encourage engagement in activity. Successfully engaged individuals with DS in the Li et al. (2006) qualitative study were said to have “positive personality traits such as motivation to learn...as well as the characteristic of perseverance” (p. 151), and support from families, teachers, and community facilities was important. The importance of motivation level is also revealed in quantitative research: mastery motivation (as assessed by persistence with goal-directed toys) in children with DS, has been found to be correlated with adaptive competence and performance of daily activities (Niccols, Atkinson, & Pepler, 2003) and to be a predictor of future academic competence (Gilmore & Cuskelly, 2009). A longitudinal study comparing three disability groups (DS, motor impairment, and developmental delay) found that children with higher mastery motivation demonstrated greater growth in mental age; however this development was considerably less prominent in children with DS (Hauser-Cram,

Warfield, Shonkoff, & Krauss, 2001). Despite deficits in mastery motivation and persistence when playing with toys (Pitcairn & Wishart, 1994; Kasari & Freeman, 2001), children with DS are still motivated by play. Ruskin, et al. (1994) found that children with DS had less mastery-motivation (i.e. length of time persisting in goal-directed play) than did typically developing children but that the quality of their play was very similar (they engaged in similar frequencies of simple exploratory play and goal directed attempts with toys). In other words, children with DS began goal-directed tasks a similar number of times (i.e. putting a shape in a shape sorter), but did not persist with this behavior for extended periods of time. They were just as engaged in play as typically developing children, but the aim of their play was different and less organized. A study that assessed 30-minute sessions of children free-playing with their mothers observed children with DS to play with similar levels of engagement, enthusiasm, and affect compared to typically developing children of the same mental age (Beeghly, Weiss Perry, & Cicchetti, 1989). This indicates that, when allowed to play as they wish rather than asked to complete challenging tasks, children with DS are not only motivated by play but also play in a similar manner as their mental age-matched peers. Naturally, children with DS are motivated by play; however it appears that their motivational style is different from the mastery-motivation that is evident in their typically developing peers.

Given the relative strengths in sociability in the DS behavioral phenotype (Fidler et al., 2006; Walz & Benson, 2002), it seems that there is a social component to their motivational profile. Fidler, Barrett, & Most (2005b) found underlying sociability in personalities of people with DS, but it was less pronounced over time which raises questions as to what other factors might play a role in their motivational profile. Fidler

(2006b) argues that the underlying verbal processing and vocabulary deficits in people with DS places them at risk to associate failure with language expression, and overtime they may try less to engage in expressive communication. This could explain, in part, why motivational problems emerge. Are there, however, alternative aspects of the DS motivational profile that can be accessed?

Wishart (1996) argues that motivational deficits in children with DS leads to cascading missed opportunities in daily activities that undermine the progress of development and that we need to find ways around their potentially challenging learning style. Fidler (2006a) indicated that researching the personality-motivational style of children with Down syndrome can aid in improving effectiveness of education and interventions. An understanding of activity choice and its relationship to motivational style in people with DS will help guide practitioners in finding activities that are meaningful and intrinsically motivating to their clients. Intervention can perhaps target areas of deficit in the behavioral phenotype such as expressive language and motor skills while capitalizing on areas of strength such as visual and social skills.

Relevance to occupational therapy. Engagement in meaningful, daily activities (or occupational participation, as termed by the field of occupational therapy) is necessary for well-being and optimal development (Hocking, 2009). Occupational therapists, as part of the important support network for many individuals with DS, play a unique roll in facilitating optimal development through participation in functional, meaningful activity. The Person-Environment-Occupation (PEO) Model from the field of occupational therapy describes the importance of finding a “fit” between these three components in order to achieve the highest possible level of occupational performance

(Law, Cooper, Strong, Stewart, Rigby, & Letts, 1996). In other words, in order to achieve the fullest level of participation, attributes of the individual (person), the type of activity (occupation), and surrounding resources and characteristics (environment) must all be taken into consideration. The model assumes that individuals are motivated and ever developing, that the environment can be supportive or constraining, and that occupations are necessary for meaningful living. The “Dynamic Performance Analysis (DPA) Framework” proposed by Polatajko, Mandich, & Martini (1999) assumes that motivation and knowledge of an activity are prerequisites for task performance. Though the motivational style of each individual is unique, the motivational style typical of the behavioral phenotype provides a good starting point. Understanding how motivational style relates to activity participation in children with DS may provide guidance for therapists in developing etiology-specific intervention approaches for this population. While infinite variables are involved, part of finding the person-environment-occupation fit and achieving optimal occupational performance involves determining what is motivating for an individual and which occupations utilize their personal motivations and abilities.

The relationship between behavioral phenotypes and intervention strategies is complicated. Basing interventions solely on phenotypical characteristics is problematic because not every individual with DS will exhibit the same makeup; however, etiology-based information is needed for families to better understand their children with special needs and to advocate for the best possible services (Hodapp, 2004). Though research into behavioral phenotypes of DS has become increasingly extensive, the application of this evidence into intervention, particularly for intervention that is focused on everyday

life and activities, is limited. In order to determine how people DS can achieve optimal occupational performance, it is important to gain an understanding of the occupations in which they tend to participate and the motivational factors that tend to play a role.

A broader understanding of a population can potentially shed light on aspects of an individual that might otherwise be overlooked. The present study will contribute to current understanding of the DS behavioral phenotype and perhaps to provide practitioners with a useful starting point for intervention planning. In order to enhance the application of the DS behavioral phenotype to practice, it would be helpful to know more about the relationship between motivational style, participation in activities, and adaptive behavior specific to daily living skills. The following questions were posed regarding participation, motivational style, and functional relevance:

1. Is the motivational style of children with DS different from that of children with mixed intellectual disabilities (ID)?
2. Is there a difference in activity participation (type of activities and how much they participate in these activities) between children with DS and children with mixed ID?
3. What is the magnitude of the relationship between level of sport and non-sport activity participation and motivational style in children with DS and mixed ID?
4. What is the magnitude of the relationship between level of sport and non-sport activity participation and daily living skills (a component of adaptive behavior) in children with DS?
5. What is the magnitude of the relationship between motivational style and performance of daily living skills in children with DS?

Methods

Participants

Participating in this study were 46 parent-child dyads: 25 children with Down syndrome (DS, *M* age 12.8 years) and 21 children with mixed-etiology intellectual disability (ID, *M* age 13.3 years). The participants were part of a larger study (see Fidler, Most, & Guiberson, 2005), and the present study is a secondary analysis of the data. Groups were equated for both mental and chronological age (see table 1).

Table 1. Participant characteristics

	DS	ID	<i>t</i> *
<i>n</i>	25	21	
<i>M</i> Chronological Age in years (<i>SD</i>)	12.8 (4.6)	13.3 (4.5)	
<i>M</i> Chronological Age in months (<i>SD</i>)	153.2 (55.4)	160 (53.5)	-.42
K-Bit Matrices (<i>SD</i>)	46.7 (9.2)	52.6 (12.9)	-1.7

*no significant differences

Hodapp and Dykens (2001) explained that in order to identify characteristics that are unique to children of a specific etiology group, it is valuable to use a group of children with intellectual disabilities as a comparison. This helps to distinguish whether characteristics pertain to a certain diagnostic group or to intellectual disability in general. In this study, the DS group includes children who had received a genetic diagnosis of trisomy 21. The mixed ID group includes children with ID of unidentifiable etiology, children who had pre-, peri-, or post-natal complications, and children with intellectual disability of other genetic or biological etiologies. The parents or legal guardians of the children were invited to participate, and more mothers participated than fathers (92% of the caregivers interacting with the DS group were mothers; 83% in the ID group were

mothers, $\chi^2 = .85$, ns). Parents of the DS group were older ($M = 47.2$ years, $SD = 6.6$) than those of the ID group ($M = 42.0$ years, $SD = 7.0$; $t = 2.48$, $p = .02$). Approval was obtained through the university institutional review board. Participants were recruited through parent support groups and school districts in a Western state.

Materials

The data for this study had already been collected and analyzed in other research (see Fidler, Most, & Guiberson, 2005), and further analyses were performed.

Kaufman Brief Intelligence Test. To measure non-verbal IQ, the matrices section of the Kaufman Brief Intelligence Test (K-BIT; Kaufman & Kaufman, 1990) was used. The K-BIT consists of vocabulary and matrices subtests and is used to measure intellectual functioning in individuals ages 4 to adult. It was designed for research or screening purposes and is widely used to assess individuals with intellectual disabilities. The DS and ID group in this study were matched for mental age according to scores on the matrices subtest of the K-BIT.

Child Behavior Checklist/4-18. The competency section of Achenbach's (1991) Child Behavior Checklist (CBCL/4-18) was used to obtain data regarding the types of activities in which children in the study typically participate and how much they participated in these activities compared to peers. The CBCL is a standardized, parent-completed checklist. The CBCL/4-18 was normed on a sample of 2,368 typically developing children aged 4 to 18 years. The mean test-retest reliability of the competence section is .87; specifically, .70 in the activity section and .92 in the social section (Achenbach, 1991). The competency portion of the CBCL reflects the number and quality of children's activities and relationships. Parents list various sport and non-sport activities in which their child participates and rate their child's performance

compared to peers. Parents provide information regarding their child's relationships with peers and siblings.

Variables from the CBCL included *types of activities* (sport and non-sport) and *level of participation* (i.e. how much children were reported to participate) in these activities. Activities which parents gave highest participation scores were used record the *types of activities* in which children participated (for any given individual, if more than one activity tied for highest score, the first activity listed was used). Sports activities were assigned to the following categories: (a) team (sports for which team play is required, e.g. basketball, baseball, volleyball), (b) individual (sports that do not require team play but are often organized into a club or regular practice, e.g. swimming, bowling, gymnastics, dance, wrestling), and (c) leisure (non-organized and typically more spontaneous sports, e.g. bike-riding, trampoline). Non-sport activities were assigned to the categories used in study conducted by Rosner et al. (2004): (a) music (e.g. singing, dancing), (b) reading (e.g. books, comics), (c) visual-motor (e.g. puzzles, workbooks, art, coloring, computer games), (d) physical activities not considered sports (e.g. walking, trampoline), (e) pretend play (e.g. dolls, action figures), and (f) focused, specific interests (e.g. collecting). Using two independent-raters, inter-rater reliability was found to be 97.2% agreement. Where discrepancies occurred, the raters discussed and came to an agreement as to the appropriate categorization. *Level of participation* was scored for sports and non-sport activities according to parent report of their child participating "above average" (1 point), "average" (2 points), or "below average" (3 points) compared to their peers in each reported activity. Scores for each activity recorded were totaled for each individual.

Reiss Profile MR/DD. According to the sensitivity theory developed by Reiss and Havercamp (1996), individuals differ in their motivational needs, and an understanding of the differing fundamental desires can predict human behavior. Based on this theory, Reiss and Havercamp (1998) developed the Reiss Profile of Fundamental Goals and Motivational Sensitivities for Persons with Mental Retardation (Reiss Profile MR/DD). This instrument is filled out by caregivers, teachers, or parents and provides a 15-factor scale analysis of underlying motives. A series of three studies were performed to develop the questions, and according to preliminary analysis, the test-retest reliability for the 15 scales ranged from .72 to .89 ($M = .81$). Lecavalier and Havercamp (2004) tested the reliability and validity of the measure using 48 individuals with mild or moderate levels of intellectual disability; they reported good internal consistency (.84), varying interrater reliability (range: 0.31-0.79, $M = 0.52$), and excellent validity (95% of the profiles were correctly identified). For the present study, the Reiss Profile MR/DD was used to determine underlying motivational style. Parents rated their child on 100 items about their child's motivational style using a five-point likert scale from strongly disagree to strongly agree. The Reiss items each correspond to one of the 15 underlying factors of motivation, including: desire for attention, helping others, social contact, curiosity, independence, order, morality, food, sexforeplay, activity, rejection avoidance, pain avoidance, anxiety avoidance, and frustration avoidance. The motivational domain scores used in this study were obtained by summing the parent responses of individual items for each domain.

Vineland Adaptive Behavior Scales. The Vineland Adaptive Behavior Scales, Interview, ed (VABS, Sparrow, Balla, & Cicchetti, 1984) is a standardized parent

interview survey that assesses socialization, communication, daily living, and motor skills. It provides norm-referenced information, and demonstrates strong test-retest reliability (domains range .88-.97) and internal consistency (Adaptive Behavior Composite ranges .89-.98; Sparrow, Balla, & Cicchetti, 1984). Standard adaptive behavior composite scores and domain scores were obtained using a sample of 3,000 individuals. For purposes of this study, which is focused on participation in daily living skills, the standard scores for the daily living skills domain were used. This domain includes skills such as eating, dressing, chores, and safety in the community.

Results

The DS group was more motivated by independence ($M = 23.14$, $SD = 5.09$) than the ID group ($M = 17.12$, $SD = 5.33$, $t_{(37)} = 3.59$, $p = .001$, $d = 1.21$, $r = .52$) with significance still being reached after the Bonferroni correction (.05/15; $ps < .003$). No significant differences were found in either the *types of activities*, nor the *level of participation* between the two groups. Table 2 provides descriptive statistics for overall reported non-sport activities. Descriptive statistics were also analyzed for highest reported sport (figure 1) and non-sport (figure 2) activities for each participant.

Table 2. Overall non-sport activities reported on CBCL

Activity Category	DS		ID	
	<i>n</i>	%	<i>n</i>	%
Visual-motor	23	38.3	23	43.4
Arts and crafts	7	10.0	8	15.0
Board games	5	8.3	0	0.0
Electronic games	4	6.7	8	15.0
Music	12	20.0	6	11.3
Reading	12	20.0	7	13.2
Pretend Play	8	13.3	10	18.9
Physical	3	5.0	3	5.7
Focused Interest	1	1.7	2	3.8
Passive TV	1	1.7	2	2.9

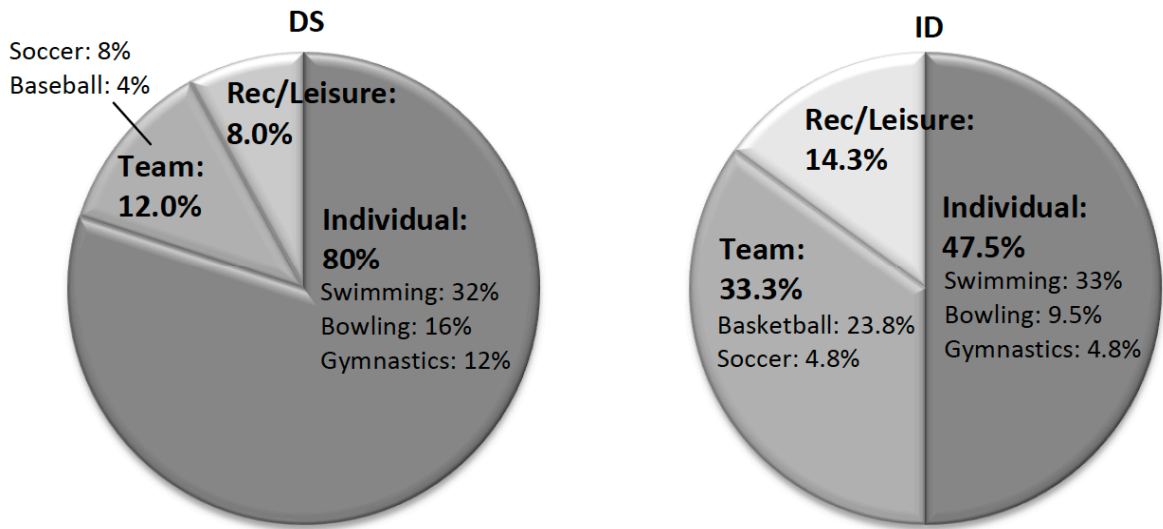


Figure 1. Sports activities in which parents reported most high participation are shown for both groups.

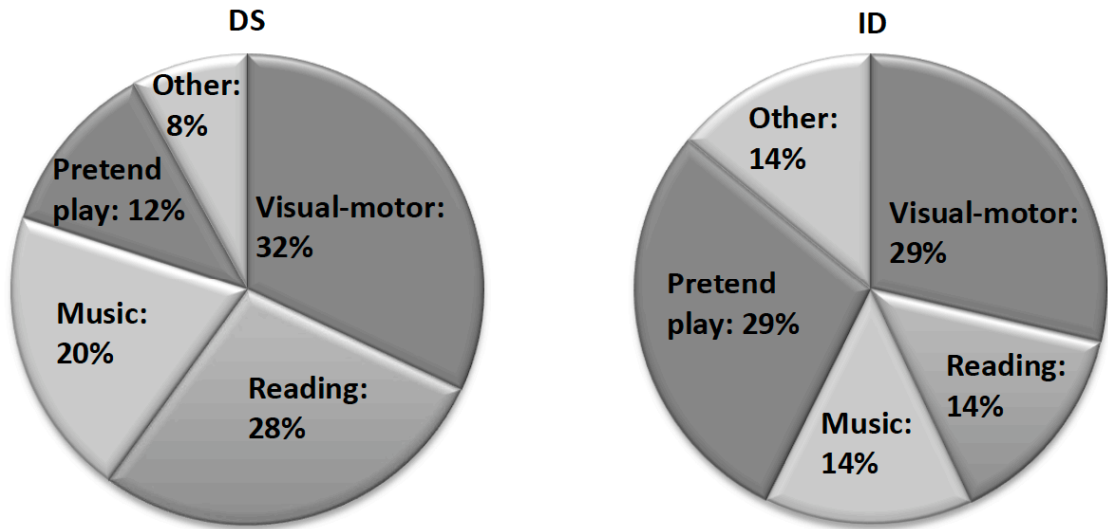


Figure 2. Non-sport activities in which parents reported most high participation are shown for both groups.

What is the magnitude of the relationship between level of sport and non-sport activity participation and motivational style in children with DS and mixed ID?

Parent-reported levels of sport participation (see table 3) on the Child Behavior Checklist (CBCL) in the DS group was negatively associated with the Reiss Profile MR/DD domains of pain avoidance ($r = -.429, p = .047$) and frustration avoidance ($r = -.518, p = .014$). Therefore, lower levels of parent-reported sport participation was associated with higher reported levels of being motivated by avoidance of both pain and frustration in the DS group. Some associations between level of sports participation and the following motivational domains approached significance in the DS group: order ($r = -.362, p = .098$), attention seeking ($r = -.311, p = .170$), anxiety avoidance ($r = -.301$), and physical activity ($r = .397, p = .067$). In the ID group, parent-reported sport participation was negatively associated with social motivation ($r = -.535, p = .040$). Therefore, lower levels of parent-reported sport participation was associated with higher reported levels of

social motivation in the ID group. For this group, the motivational domain of pain avoidance ($r = -.440, p = .101$) approached significance.

Table 3. Motivational Styles

REISS motivational factor	Sport participation		Daily living skills	
	DS	ID	DS	ID
Helping others	-.21	-.20	.29	.65**
Pain avoidance	-.43*	-.44	-.21	.29
Physical activity	.40	.24	-.10	.26
Frustration avoidance	-.52*	.05	-.20	.18
Order	-.36	.10	-.13	.13
Independence	-.04	.15	.10	.53*
Curiosity	-.14	.10	-.53*	.59*
Attention	-.31	.14	-.36	-.15
Anxiety	-.30	-.28	-.34	.49
Moral	.12	.27	-.07	.61*
Social	.08	-.54*	-.04	.19

* $p < .05$ ** $p < .01$

What is the magnitude of the relationship between level of sport and non-sport activity participation and daily living skills (a component of adaptive behavior) in children with DS?

In the DS group, sports participation as reported on the CBCL was associated with daily living skills as reported on the Vineland Adaptive Behavior Scales (VABS) ($r = .452, p = .035$); this association was not demonstrated in parent-reported sport competency ($r = .184, p = .424$). No relationships were found between non-sport levels activity participation and daily living skills.

What is the magnitude of the relationship between motivational style and performance of daily living skills in children with DS?

In the DS group, daily living skills as reported on the VABS were negatively associated with curiosity ($r = -.531, p = .016$). Therefore, better parent-reported performance of daily living skills was associated with lower levels of being motivated by curiosity. The following Reiss motivational domains approached significance with the VABS in the DS group (see table 3): attention seeking ($r = -.363, p = .127$), and anxiety avoidance ($r = -.335, p = .149$). In the ID group, daily living skills were positively associated with curiosity ($r = .592, p = .020$), helping others ($r = .650, p = .009$), independence ($r = .538, p = .047$), moral motivation ($r = .612, p = .015$), and anxiety avoidance ($r = .488, p = .065$).

Discussion

This study examines how participation, motivational style, and daily living skills relate to the Down syndrome (DS) behavioral phenotype when compared to individuals with intellectual disability (ID) of mixed etiology. Results indicated a motivational style of independence in the DS group, no difference in level of participation between the two groups, and underscored how underlying motivational style relates to participation and daily living skills. The importance of independence, participation in sports, and factors relating to daily living skills are discussed below.

Though children with Down syndrome demonstrated a motivational style comparable to their mental age matched peers with ID, the DS group was more motivated by independence than the ID group. (See table 4 for specific independence items as reported on the Reiss Profile MR/DD.) This may shed light on an important aspect of the motivational profile of individuals with DS in that there seems to be an underlying preference towards self-directed behavior and to be able to choose how tasks are

accomplished. Research has consistently demonstrated that parents of children with Down syndrome report lower stress than parents of children with other disabilities (Hodapp, Ly, Fidler, & Ricci, 2001; Kasari & Sigman 1997; Richman, et al., 2009), and this could be in part due to parents seeing their child as being more independent. On the other hand, it could be that parents of children with DS tend to see their child's "quitting out" or task-avoidant behavior (Gilmore, Cuskelly, & Hayes, 2003; Pitcairn & Wishart, 1994) as a desire to be independent or do things their own way. Knowing this "independent" tendency in children with DS might guide how parents and interventionists interact with these children. One study found that children with DS were more persistent with toys (e.g. shape sorters, puzzles) when their mothers' interaction style was *more supportive* and *less directive* (Gilmore, Cuskelly, Jobling & Hayes, 2009). Interaction style did not affect persistence level for typically developing peers of the same mental age. This indicates that, for children with DS who may have a tendency towards "doing things their own way," it is important to avoid being overly directive. Future research could examine the role that independence plays in the DS behavioral phenotype, and interventionists might consider what motivation for independence looks like for children with DS in order structure goals around helping them to achieve independence in a functional manner.

Table 4. Specific Reis Profile MR/DD items for "Independence"

Strongly prefers to make own decisions
More than most people, enjoys working independently
Strong desire to be self-reliant
Hates being dependent on others
Strong desire for autonomy
Seeks leadership roles
Personal freedom is very important to him/her

The Down syndrome group demonstrated mental age-appropriate (compared to others with ID) participation level in both sports and non-sport activities as reported by parents. Additionally, the distribution of the *types* (e.g. music, reading, visual-motor) of activities in which the two groups engaged were not significantly different overall between groups. Similarly, Rosner et al. (2004), found no difference in the level of DS participation in sports compared to children with Prader Willi (PWS) and Williams syndromes (WS). They *did* find other patterns of participation, however: children with DS demonstrated *less* skill and participation in non-sport activities, engaged more in visual motor activities than the WS group, (specifically arts and crafts), more in music than the PWS group, and more in pretend play than the WS group. The present study results do not demonstrate significant trends in participation, and this is likely due to comparison group differences between this study and Rosner et al.'s. Rosner and colleagues compared specific genetic syndromes rather than using children with mixed etiology ID.

Though non-significant, there was a trend towards participation in individual sports in the DS group compared to the ID group (See figure 1). These findings could be due to a lack of opportunity to participate in team sports. Given that the age of this sample ranges from 5 – 22 years, it is quite possible that limited team sports participation is due to team sports becoming more competitive (and less accessible for children with disabilities) as children get older. Other studies have suggested that adults with DS have greater preference to be “active” when they can do so with others (Fujiura, Fitzsimmons, Marks, & Chicoine, 1997; Medlen & Peterson, 2000), especially others with DS (Neumayer, Smith, & Lundergren, 1993); if individuals with DS are also motivated by

independence, it seems reasonable that preferred sports are those that can be done with friends while still being self-directed. Another possible reason for this trend in participation in individual sports is that parents may guide children with DS away from contact sports due to safety reasons. Though Cremers and Bol (1993) argued that, in general, children with Down syndrome do not need to be restricted from sports, the American Academy of Pediatrics had published a statement in 1984 (Schaffer, et al.) saying that all children with DS should be screened for atlantoaxial instability before participating in sports. Certainly safety should always be a concern, and appropriate precautions and adaptations should be put in place.

Though there were no group differences in participation, the level of participation in sports was related to underlying motivational profiles in the DS group. Strongest predictors for decreased sports participation were pain and frustration avoidance. Or, to look at it another way, children with DS who *participated less* in sports were rated as *more concerned* with avoiding pain and frustration. This could have interesting implications as to why individuals with DS tend to lead lifestyles of limited physical activities and are at increased risk for obesity (Dykens et al., 2002; Whitt-Glover, O'Neill, & Stettler, 2006). Perhaps motor deficits are challenging enough that children with DS need heightened tolerance for frustration and even pain to maintain motivation for physical activity. Underlying motivational profiles of pain and frustration avoidance could contribute to people with DS leading sedentary lifestyles. The negative correlation between sports participation and attention seeking behavior may indicate that if children with Down syndrome don't want to participate in a challenging sport (similar to a challenging task as in the Pitcairn & Wishart, 1994 study), they engage in attention

seeking behavior instead. Not surprisingly, perhaps, was the moderate positive correlation between participation in sports and motivation for physical activity; this indicates that parents of children with DS who report their child as *participating* in sports are also likely to see their child as being motivated by physical activity.

In the DS group (but not the ID group), parent-reported sports participation was associated with daily living skills. Other studies have reported adaptive performance to be predicted to a greater extent by motor ability than by mental ability (Volman, Visser, & Lenvelt-Mulders, 2006) and adaptive behavior skills to be significantly correlated with motor planning skills on battery of praxis tasks in toddlers with DS (Fidler et al., 2005b). Prior research has indicated a relationship between motor skills and daily living skills but has not examined whether there is a relationship between and sports or non-sports activity participation and daily living skills. Given the present findings, that there *is* an association between sports participation and daily living skills, therapists might consider building motor skills both through sport-related activities and activities of daily living. Virji-Babul, et al. (2006) argue that intervention focusing on isolated motor components (e.g. tone) and motor milestones (e.g. grasping, reaching) does not necessarily improve long-term outcomes and that a broader context of functional motor behavior should be considered.

The motivational style underlying daily living skills in the ID group was motivation for curiosity, helping others, independence, and moral motivation. This motivational profile seemed logical and even what might be expected of typical development. However, the underlying motivational style for daily living skills in the DS group was less straight-forward. Daily living skills were *negatively* associated with

curiosity. The clinical significance of this finding can be called into question, but it may be that an extreme tendency towards curiosity (e.g. strong desire to explore the environment, enjoys puzzles/mysteries, enjoys new experiences) in children with DS actually gets in the way of functional daily living skills. A marginal negative association was also found with attention seeking behavior, which is congruent to the finding by Fidler et al. (2005) that help eliciting behavior in children with Down syndrome is marginally related to poor adaptive behavior. Certainly, more information is needed in order to determine motivational styles that either promote or inhibit functional behavior.

Limitations:

One possible limitation of the present study is the use of children with mixed etiology ID as a comparison group. There was a wide variety of diagnoses within the ID group (for example some individuals with motor limitations) which may have presented functional difficulties for reasons other than having an ID. Determining appropriate comparison groups can be difficult in developmental research. Hodapp and Dykens (2001) explain that using a comparison group of mixed ID, as opposed to typically developing children of matched mental age, is helpful in identifying characteristics unique to a specific syndrome because it decreases the chance of identifying population characteristics that are due to ID in general. Other researchers, however, note the value of comparing specific diagnostic groups (e.g. DS and WS) since people with ID are, as a whole, a heterogeneous population (Burack, Iarocci, Bowler, & Mottron, 2002).

Participant characteristics presented other potential limitations as well. Though the mean age of the two groups did not differ significantly, the wide age range increased the variability of the groups. Additionally, given that the sample size was somewhat

small and was taken from a specific area (a western state) generalizability of results to the DS population as a whole should be conservative.

The measures used presented another limitation. The CBCL, though it is widely used in research on behavioral characteristics, was not specifically designed to assess the types of activities in which children participate. The Reiss Profile MR/DD has not been widely used in research, and it is difficult to determine at this point whether it provides adequate information as to the motivational profile of a population.

Future Research:

Future research might use other measures related more specifically to occupational functioning in order to examine motivation, participation, and daily living skills within the DS behavioral phenotype. For example, Pediatric Volitional Questionnaire (PVQ) is measures a child's motivation by observing how the child participates in various occupational activities (Basu et al., 2002). This might help answer questions as to how participation is related to underlying motivational profiles in children with DS. The Children's Assessment of Participation and Enjoyment & Preferences for Activities of Children (CAPE/PAC) examines how (diversity of activities, enjoyment, intensity) children ages 6-21 participate in everyday activities, the types of activities in which they participate, and the environmental and social context of these activities (King et al., 2004). It would be interesting to look further into, not only the types of activities in which children with DS participate, but also the quality and context of this participation. Future research could also describe through observation the types of activities in which children with DS engage while in their natural contexts. It would be helpful to compare DS to other populations such as typically developing children or children with other

genetic syndromes. Pediatric Evaluation of Disability Inventory - Computer Adaptive Testing (PEDI-CAT; Coster, Haley, Ni, Dumas, & Fragala-Pinkham, 2008) is a revised and updated version of the original PEDI (Haley et al., 1992) and assesses function in every day life (e.g. self-care, mobility, and social function) for infants through 21 year olds. This assessment might help pinpoint areas of strengths and challenges in performance specifically related to every day function in children with DS. Research could further examine how functional performance in every day life is related to an underlying motivational style.

It would be beneficial to examine how motivation, participation, and daily living skills may change over time within individuals with DS. For example, how might participation in sports or the motivation towards independence change over time? Patterns of motivation and participation across time within the DS behavioral phenotype might help uncover reasons for the “avoidant learning style” (Wishart, 1996) seen in this population. Additionally, research could assess what a successful or ideal trajectory of participation and motivation in DS looks like.

Future research might examine how knowledge of the DS behavioral phenotype can contribute to effective intervention planning for individuals. It would be beneficial to evaluate the effectiveness of interventions targeting or utilizing specific aspects of the DS behavioral phenotype and to determine what strategies tend to be successful. For example, can targeting areas related to the DS behavioral phenotype in early intervention prevent future problems? Future research could also examine how the findings of the present study of curiosity being negatively related to daily living skills. Does a motivation for curiosity get in the way of functional behavior in DS? Currently there is

insufficient evidence as to how curiosity fits within the DS behavioral phenotype.

Overall, more research is needed to hone in on how the DS behavioral phenotype impacts every day life.

Implications for Practice:

This study indicated an underlying motivation for independence – or perhaps a persistence to “do things my own way” – within the DS behavioral phenotype.

Practitioners might take this into consideration by planning interventions that encourage this independence to be used for functional activity while minimizing its use as ‘quitting out’ behavior. Interestingly, though independence as measured by the Reiss was found as a motivating factor for children with DS, it was not correlated with daily living skills *or* participation. This indicates that independent behavior, as operationalized in this study, is not necessarily functional in the DS behavioral phenotype. Though “independence” is generally defined as a positive characteristic, the extreme nature of “doing things my own way” or of being strong-willed within the DS behavioral phenotype seems to interfere with functional and occupational performance. Therapists might consider how the “independent” tendencies of children with DS can be organized and regulated for every day living, or how daily living skills can become fun and self-directed rather than being something that children with DS “have to do.”

Knowing that children with DS who participated *less* in sports were rated as *more concerned* with avoiding pain and frustration and that children with DS tend to try to get out of challenging tasks (Pitcairn & Wishart, 1994; Kasari & Freeman, 2001), therapists might facilitate engagement in sports activities and maximize successes in order to build confidence. They might adapt sports activities as necessary to minimize the frustration

that might be involved (e.g. playing with others of similar skill level, encouraging sports such as swimming or dance that can be less competitive and allow more independence, capitalizing on strengths in the behavioral phenotype such as using visual learning strategies). Furthermore, therapists might consider how to use the motivation towards independence as an advantage or how to prevent it from decreasing functional participation in sports. Using knowledge of an underlying motivational style of pain and frustration avoidance to shape interventions may help prevent inactive lifestyles later on. Therapists might also consider, given the association between sports participation and daily living skills, how to build motor skills in a broad context through practicing sport-related activities and activities of daily living.

Findings from this paper contribute to the understanding of the DS behavioral phenotype, and in order to best serve children with DS and their families, it is important to combine analysis of individuals with knowledge the DS behavioral phenotype. Parents reported in a study by Fidler and colleagues (2002) that occupational therapists were less likely to bring etiology-specific information for intervention planning than other members of the special education team. It would be beneficial for occupational therapists to consider characteristics of the DS behavioral phenotype when planning interventions for an individual with DS. Practitioners who understand these characteristics (e.g. visual-learning style, social relatedness, difficulty with expressive language, a tendency towards independence, pain and frustration avoidance in sports, motor difficulties) might adapt/modify environments and occupations to capitalize on strengths, prevent future problems, provide insight into why an individual is behaving in a certain way, educate families about what to expect from their child, and impact every day living and

participation in occupation. The results from this project provide a promising beginning to etiology-specific intervention for individuals with DS in the field of occupational therapy. It is hoped that this project will spark further discussion and research in this area.

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