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Novel Treatment for Improvement of Sensory-Motor, Function and Emotional Behavior of a Child with Autism Spectrum Disorder: A History Case Report

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Abstract

Background: Tailored interventions. One potential novel intervention is orthotic under-garments (OUG Autism spectrum disorder (ASD) is a prevalent neuro-developmental disorder frequently presenting sensory-motor impairments. The complexity and diversity of ASD typically calls for individualized).

Aim: To explore the feasibility and effect of OUG as a supplementary therapeutic modality on motor, sensory capabilities and behavior of a child with ASD.

Participant: A five years and eight months old child with ASD that presents hypotonia, poor postural control and coordination, a significant motor delay, severe sensory modulation problems, and unusual behaviors.

Procedure: Motor, sensory and behavioral evaluations were performed pre (Pre-I) and after (Post-I) a four months intervention: Peabody Developmental Motor Scales-2 (PDMS-2), The Short Sensory Profile (SSP), therapists' reports and parental interviews served as a behavior and emotion evaluation.

Results: Motor function has improved (PDMS-2 percentile <1 Pre-I vs. Post-I 3). Sensory function has improved (Total SSP score 136 Pre-I vs. Post-I 102). Therapeutic and parental reports indicated a decrease of atypical behaviors, improvement in self-confidence and better social participation.

Conclusion: According to the current case report, the OUG was found to be useful in improving sensory-motor functions and emotional behaviors of a child with ASD. Our findings provide a possible support to the introduction of orthotic undergarment as a part of individually tailored physical therapy interventions for the child with ASD.

Keywords: Autism spectrum disorder; Sensory-motor function; Emotional behavior; Orthotic under-garment (OUG)

Introduction

Autism spectrum disorder (ASD) is a prevalent, heterogeneous neuro-developmental disorder [1,2]. About 1 in 68 children in USA has been identified with ASD according to estimates from the Center for Disease Control (CDC) [3]. Social-interaction difficulties, communication challenges and a tendency to engage in repetitive behaviors are the core symptoms of ASD. However, it is well known to be characterized by divergent of associated medical and developmental symptoms [4]. Those challenges negatively impact the children's function and participation as well as offend the quality of life of both the children and their families.

Motor difficulties in ASD have been categorized as prevalent associated symptoms [1,4,5], frequently emerging at early infancy and childhood, reaching the prevalence of 80% along the lifespan [6-9]. Recent publications suggest the possibility of regarding motor

difficulties as a part of ASD's core symptoms [10]. Sometimes it is the first suspicious sign pointing toward developmental disorder including ASD, encouraging treatment initiation [11-14].

Difficulties with Postural control (PC), coordination and motor planning. [5,14-17] are only a part of the motor challenges known to be common in ASD.

Atypical PC development has been documented in up to 92% of children with ASD [15,18]. Normal PC development is based on adequate sensory, motor and inter-systems integration [19]. Hypotonia [5], joint hyper-mobility [20] and muscle weakness are contributing factors to motor dysfunction, known to be common in ASD. Sensory modulation disorders (SMD) are presented in up to 90% of ASD children. [21,22]. In addition, abnormal PC has inverse impact on function, including motor experience and function avoidance which might reduce social engagement [15]. Thus, early intervention using appropriate therapy modalities should address PC for individuals with ASD [23].

ASD therapeutic approaches are built upon comprehensive multifaceted treatments that incorporate multiple intervention modalities [24]. In light of the motor and sensory difficulties ASD children demonstrate, do clinicians and researchers stress the significance of innovative sensory-motor interventions [23,25,26]. One such potential intervention approach is based on the use of Orthotic Undergarments (OU).

TheraTogs[™], TT[™] (Produced by TheraTogs, Inc., 305 Society Drive, Ste C-3 Telluride, CO 81435-8935 USA), are innovative OUG that were designed with intent to improve postural alignment and stability, movement skill and precision, joint stability, and prolonged muscle stretch [27]. The OUG is a live-in exo-muscular system, developed to provide a gentle and continuous pressure on soft tissue through a combination of a trunk-and-shorts system with a customized elastic external strapping system. The benefit of using OUG as an external "wear-on" system is in its extended effect assisting and supporting postural difficulties and continued deep-pressure stimulus after the conclusion of the direct hands-on intervention. There is some evidence, mostly descriptive, that OUG may be beneficial across a range of populations. Previous studies that were primarily directed toward children with cerebral palsy indicate improvements in proximal stability and mobility in gait, balance, and functional skills [27-30]. Other studies demonstrated efficacy in other populations such as Down syndrome, traumatic brain injury, and spine bifida [28]. To the best of the authors' knowledge, OUG use in ASD population has not yet been reported.

The present history case report constitutes the first documented application of OUG in ASD.

The child - IV was born at 37 weeks gestation following IVF pregnancy, birth weight of 3.050 kg, He was released home, 10 days after birth due to jaundice. IV has one younger sibling. At 9 months, IV was referred to a physical therapy (PT) consultation and intervention due to a motor delay and was diagnosed with mild hypotonia and joint hypermobility. IV acquired independent sitting by 18 months, independent standing by 22 months, and walked independently by the age of 24 months. At 13 months, a comprehensive multi-disciplinary assessment was performed due to emergence of repetitive behaviors such as hand flapping and pervasive playing as well as occasional lack of responsiveness to being called by name. Based on the assessment, IV's therapeutic program was expanded to include occupational and speech therapy. IV started to pronounce his first words at 15 months and combined words into simple sentences at 18 months. By the age of 24 months, IV was diagnosed with sensory modulation disorder (SMD) and at the age of 4 he was diagnosed with ASD, based on the Autism Diagnostic Observation Schedule (ADOS) [30]. Following the diagnosis, the educational framework was changed to meet IV's needs. Four days a week IV attended a mainstream pre-school mediated by a private special needs teaching assistant and twice a week a multi-disciplinary ASD intervention center (ASD center) operated by the Association for Children at Risk. His therapeutic intervention program consisted of weekly 1 h long individual treatment sessions in physical, occupational and art therapy, and an hour-long speech and communication group therapy. In addition, IV's parents participated in an individual dyadic session conducted by IV's art therapist.

The rationale behind using OUG for the current case study.

(1) IV's motor difficulties and avoidance of motor activities;

(2) IV's constant engagement in self-stimulation;

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(3) Difficulties IV's parents and his educational team encountered in handling him;

(4) Difficulties in social acceptance by his peers due to his uncontrolled behavior.

His high cognitive level and his expressive language abilities made IV a suitable candidate for an OUG therapeutic research study.

Methods

The study was a single case study design, aimed to evaluate the effect of OUG on the motor, sensory and emotional behavior of five years and eight months boy diagnosed with ASD. The study was approved by the IRB of Beer Yaacov Mental Health Medical Center, Nes Ziona (Approval no. 542). Publication of the details of the child and the results of the intervention was approved in writing by the parents.

Assessment tools

IV's gross motor, sensory and emotional development was evaluated before and after the intervention. In addition, a weekly behavioral follow-up was conducted by the therapeutic team before and throughout the intervention.

Peabody Developmental Motor Scales-2 (PDMS-2) is a standardized, valid and reliable measure designated to assess gross and fine motor development among children in the range of age of birth to 72 months [31]. PDMS-2 is known to be used in ASD population for clinical and research purposes [8,32]. Pdms-2 is composed of 3 quotients: motor, fine and total, which are separately standardized. In the current case report we used the Gross Motor Quotient (GMQ) which is composed of three domains: stationary (stability and equilibrium), locomotion (movement maneuvers), and object manipulation (ball skills). The GMQ was administrated by IV's PT.

The Short Sensory Profile (SSP) [33] is a standardized, valid and reliable caregiver/parent questionnaire designated to assess sensory processing among children aged 3 to 10 years. The SSP was found to be appropriate for evaluating sensory processing in children with ASD [34-36]. The SSP is a questionnaire which should be completed by the child's parents, it is consisted of seven sections: tactile sensitivity, taste/ smell sensitivity, movement sensitivity, under responsive/seeks sensation, auditory filtering, low energy/weak and visual/auditory sensitivity. The SSP was completed by IVs parents and interpreted by his occupational therapist.

Emotional behavior reports were documented based on IV's art therapist's weekly follow-up and parental interviews before, during, and after the OUG intervention. General and goal-directed behaviors were observed within the developmental setting.

Intervention procedure

The intervention included the following three stages:

IV's PT training by a certified TT[™] PT (YH-first author) following preparation of the child, his family and the educational staff;

Gradual implementation of the program at the developmental center; and

Continuous wearing the OUG throughout day and night.

Stage I: Evaluating IV's needs and deciding over the use of the OUG, educating the PT on the use of the OUG and individual short term

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trials with the child within the PT treatment room. After this process ended and a decision over the use of OUG had been made, a multidisciplinary staff meeting was conducted in order to coordinate the upcoming intervention with the professional team members. The meeting included the following:

(1) Presentation of the OUG and its evidence-based clinical benefits and side effects;

(2) Discussion over the reasons for introducing OUG therapy for IV;

(3) Defining the intervention's goals, procedure stages and assessment tools;

(4) Instructions for informing and documenting any behavior changes, especially aversive ones (to instantly identify negative reactions to the OUG use);

(5) Nominating IV's PT as the intervention case manager. Following, the OUG and intervention procedure were presented to the parents who consented to the OUG intervention by signing an informed consent.

Stage II: Adapted and detailed preparation was made possible due to IV's high cognitive level, curiosity and extremely good relationship with his PT. The PT demonstrated the importance and benefits of wearing OUG by giving IV explanations on human body systems and functions using an anatomic picture book. The PT also introduced the garment as a "power suit" invented to help children overcome their difficulties, which alleviated IV's anxiety. After three preparatory meetings, IV became very positive and enthusiastically anticipated the use of the OUG. The intervention program was added to IV's weekly

therapeutic program and was conducted continuously for four months. In the first two weeks, IV wore a complete OUG without straps for one hour a day, twice a week while he was at the ASD center. In the following two weeks, IV wore the garment at the ASD center for increasing periods of time, gradually extending wear-time to eight hours a day. In addition, one thick abdomen strap and two X-shaped body side straps were attached to the garment. The X-shaped straps were connected between the chest and pelvic components on each side in order to enhance proprioceptive input, giving IV a deeper sense of his body scheme. The abdomen strap was aimed at enhancing belly muscle activation. At that point, the PT actively involved the parents by training them to dress IV in the OUG in order to achieve full daily usage.

Stage III: The parents' involvement enabled full-day usage of the OUG. After the first month during which "wear time" was gradually increased, the parents started dressing IV in the morning and removed the garment only at night. At this point, IV insisted on sleeping with it, as if it was his "second skin." As IV was fully toilet trained, the parents needed no additional assistance in actively dressing him. The intervention continued for four months in accordance with the procedure described above.

Results

Motor function

Pre-I assessment indicated significant difficulties in fundamental age-appropriate motor skills, as describe by details in Table 1.

Domain Features	Pre-I	Post-I		
Motor functions	Preference to sedentary activities	Significant improvement in motivation to perform motor tasks		
	Extreme difficulties in maintaining PC and postural stability	Extended sitting duration on a chair or on the floor		
	Equilibrium compensatory patterns of wide base of support	Significant reduction in frequency of position changes		
	Asymmetrical posture	Longer periods of activity during physical therapy sessions		
	Significant body sway	Improved endurance, which enabled him to maintain continuous		
	Frequent position changes	activity throughout 30- minute sessions, without resting		
	Rapid tiredness after any brief motor activity			
	Low fitness and endurance			
	Low muscle tone			
	Joint hypermobility			
Sensory functions	Avoidance behavior (e.g. crawling into dark and narrow spaces)	Reduction in frequency of self-stimulation (less somato-sensory self- stimulation)		
	Self-stimulating behaviors mostly somatosensory (e.g. pressing			
	his hands deeply against his head, fixing the chair's legs over			
	his feet while sitting on the chair)			
Emotional behaviors	Low frustration threshold			
	Anxious behavior			
	Low self confidence			

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Avoidance of speaking in front of class	
Aggressive behavior	
Poor judgement	
Unusual behaviors (e.g. inappropriate laughter, pervasive speaking, moving, climbing and hanging on to an adult's body	

 Table 1: Descriptive comparison between IV's Pre-I and Post-I motor, sensory and behavioral features.

Post-I PDMS-2 GMQ total and subscales scores were increased described in Table 2, indicating an improvement of gross motor function.

GMQ subscales and total		Pre-I (68 months)		Post-I (72 months)			
	Raw score	Standard score	percentile	Raw score	Standard score	percentile	
Stationary	44	5	5	44	5	5	
Locomotion	130	5	5	152	6	9	
Object manipulation	16	3	1	34	6	9	
Total GMQ		13	<1		17	3	

 Table 2: Comparison of PDMS-2 GMQ scores Pre-I and Post-I.

IV exhibited progress on locomotion and object manipulation subscales although no change was recorded on stationary score. Post-I

observation indicated reaching the intervention's motor goals as defined in Table 3.

Domain	Goals	1
Motor functions	Improvement of postural control and stability	
	a. reducing changing positions frequency while asked to be seated	
	b. increased participation in group motor activities	√
Sensory functions	Reduction of self-stimulation	
	a. Reducing the need for holding things in hands	
	b. Reducing hiding in close places frequency	
	c. Reducing inappropriate contact with adults frequency (e.g. climbing, biting, kicking)	√
Emotional behaviors	Improvement of self-confidence and participation	
	a. Increasing participation in social activities frequency	
	b. Extending attention span in learning sessions	
	c. Increasing occasions frequency in which IV expresses himself in peer groups	√

 Table 3: Obtaining intervention motor, sensory and behavioral goals.

Sensory function: Pre-I assessment indicated a complex, mixed sensory picture of both 'seeking' and avoidance behaviors, as describe in detail in Table 1. The occupational therapist identified those difficulties as low sensory registration, based on SSP test. Post-I SSP scores were increased compared to Pre-I (136 vs. 102/190), indicating an improvement of sensory function, even though the total score classification didn't change. However, 4 out of 7 sensory function sections demonstrated an increase and positive change in

classification,	as	described	in	Table	2.	Post-I	observation	indicated
achievement of	of ir	ntervention	is se	ensory	goa	als as de	efined in Tabl	e 4.

	Pre-I (68 n	nonths)	Post-I (72 months)		
Section	Raw score	Classification	Raw score Classification		
Tactile sensitivity	21	DD	28	PD	

Taste/Smell 20 20		ТР	20	ТР
Movement sensitivity	8	DD	11	PD
Under responsive/ Seeks sensation	15	DD	23	DD
Auditory filtering	16	DD	20	PD
Low energy/Weak	6	DD	13	DD
Visual/Auditory sensitivity	16	PD	21	ТР
Total	102	DD	136	DD

Table 4: Comparison of SSP between Pre-I and Post-I.

Emotional function: Pre-I IV's main behavioral features were described in detail in Table 1. All of which limited IV's ability to participate in daily activities and maintain active interactions with peers. External proprioceptive stimulation was found to have a positive and calming effect on IV's behavior, which increased his ability to participate in social activities and increased his attention span during educational activities.

Long-term implications: Two years after initiating the current intervention, IV continues to wear the OUG on a daily basis, which might suggest his continued need for its support.

Discussion

The present case study presents the results of wearing the OUG by a five and 8 months years' old child with ASD. An assessment of the child's motor, postural, behavioral and sensory functioning suggests significant improvements in all areas after these four months of daily use of the OU garment. The results of this single case study showed that OUG was found to be a feasible intervention for a high functioning ASD child in the age of 5.5 years, with positive gains in multiple core aspects of ASD. Benefits of OU garments have been reported in several pediatric populations in the past, and the official site recommends its use for children. To the best of our knowledge, this is the first report of the effects of OUG use by a child with ASD.

Motor domain

The majority of individuals with ASD present motor difficulties [7,8]. Such difficulties might cause them to avoid physical experiences which trigger a cascade of events that leads to the adoption of a sedentary lifestyle and ultimately to severely diminished functional abilities.

IV's motor progress has improved significantly during the OUG intervention. These changes may be attributed to improved alignment and increased internal core stability, which play an important role in the development of motor skills. The effect is attributed to the OUG's effectiveness which improves body alignment and enables a more centered, focused base. These findings are similar to findings of previous researchers who reported the OUG's effectiveness in other populations [27-28].

Sensory domain

The intervention led to a significant improvement in the sensory profile that characterizes IV. Within four months, IV's sensory profile as well as his overall sensory behavior changed dramatically. Sensory difficulties have been reported in the majority of children and adults with ASD [22,37,38] and were recognized as typical symptoms of autism diagnosis [4]. In view of reliable evidence suggesting an association between sensory symptoms and severity of ASD symptoms [34,38], sensory elements are considered to be a critical area of ASD symptomatology that subsequently affects multiple areas of functioning and participation (e.g. communication, attention, emotional regulation, motor behavior, cognitive awareness) [39,40]. This OUG produced constant "correct" sensory input of proprioceptive and tactile modalities that accompany the child throughout his daily activities. The findings presented in the present article are consistent with previous findings on the positive influence of deep touch-type stimulation in children with ASD [41-43].

The authors believe that the constant use of the OUG generates more continuous, organized inputs from proprioceptive and tactile systems, thereby facilitating the integration and organization of sensory input to the CNS, and enabling the child to become more receptive to other meaningful external stimuli, such as educational interventions [44-46].

Behavioral domain

An important positive outcome of the intervention was in the behavioral domain. Challenging behaviors is one of the most significant stressors for family, caregivers and support staff these tend to become increasingly apparent with age [47]. Their consequences may include increased social isolation, restricted educational and vocational opportunities [48] and poor overall long-term outcomes [49]. It is reasonable to speculate that pressure UG usage increased IV's self-regulation, which reduced his frustration and/or irritability. This phenomenon is probably due to proprioceptive inputs (deep pressure) that previously proved to promote the production of neurotransmitters such as serotonin and dopamine, which in turn modulate the activity of the central nervous system [50]. This line of thinking should be tested in future studies.

The current findings suggest that garments, like those tested in the current study, may positively influence different core aspects of ASD. As this is the first report of the use of OUG in ASD, and the intervention represents change in IV's daily routine and habits, the educational and therapeutic team was asked to pay special attention to negative reactions, and report such behaviors in a follow-up form. No adverse effects were reported. The reports from this child's experience, as well as the experiences of children in other populations who used the OUG as a clinical intervention, have not yet revealed any negative effects of the use of OUG for children. Further studies are needed to examine this issue across a group of children with ASD.

Limitation of the intervention: generalization of the conclusions of one case study to the wider population of ASD is limited and needs to be extended to wider as well as specific ASD samples.

Future Considerations

The positive results presented, suggest that future investigation is warranted to establish evidence that support the effectiveness of the OUG for the wider population of individuals with ASD. Future research should include interventions involving larger groups of individuals with ASD fit with OUG; develop a usage protocol, with blinded accessory and specific pre-determined outcome measures. Moreover, the fact that the child in this study was able to sit calm and erect for longer durations and presented improved attunement had a positive effect on his educational achievements, yet those were not evaluated prior to the intervention and should be evaluated in future interventions.

In this study, the usage of OUG was restricted to a mere four months. Future research protocols regarding OUG should evaluate the influence of long-term OUG usage.

The participants' age is another consideration for future studies. IV's sensory difficulties drove him into a pattern of aberrant behaviors (he was unable to sit erect for long durations, avoided motor and social activities, and exhibited sensory seeking behaviors). When the intervention was initiated, IV was 5 years and 9 months old and these behaviors were already deeply established. We believe that an intervention with OUG at an earlier age might have produced better results for IV, when fixation on such behaviors would have been diminished. We therefore suggest that future interventions with OUG should be initiated with younger participants.

The adaptation of OUG for children with ASD should take into consideration the required involvement of parents/careers and the need to gain the children's full participation and co-operation in the intervention process. Due to the multiple domains (behavioral, sensory, educational and behavioral) in which significant changes occurred in this case study, collaboration with professionals in other disciplines is highly encouraged in the design, implementation, and evaluation of future interventions.

Conclusion

Based on the results of the present intervention and other clinical experiences, the OUG has shown to have significant impact on postural control and motor function of children with ASD. This type of intervention can complete and enhance direct physical therapy interventions with children with ASD. As this is a case study, generalization is limited, and further research in this area is warranted.

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