Impact of somatosensory orthoses on behavioral and postural control in individuals with autism and severe proprioceptive dysfunction: an open retrospective exploratory study

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Introduction

Background:

Compression garments (CG) are an adjuvant treatment for hypermobility spectrum disorder (HSD) including Ehlers-Danlos syndrome hypermobility type. Their action is likely to be related to a better proprioceptive control.
Patients with ASD show higher rates poor motor coordination probably due to sensorial abnormalities. For some of them, Severe proprioceptive dysfunction (SPD) may underlie both motor problems and some challenging behaviors.
HSD and ASD often comorbid in severe cases. For these reasons CG could be an convenient way to improve a pattern of challenging behaviors assigned to SPD

Objective:

We focused on a population of inpatients with severe autism and challenging behaviors resisting to a multi disciplinary management in a Parisian neurobehavioral unit (USIDATU).
We aim to explore the use of CG in individuals with severe autism and severe proprioceptive dysfunction (SPD) including HSD on postural control and challenging behaviors.
This exploratory case study is a first step for a larger scale feasibility study provide that will define clinical saliency assigned to a better outcome

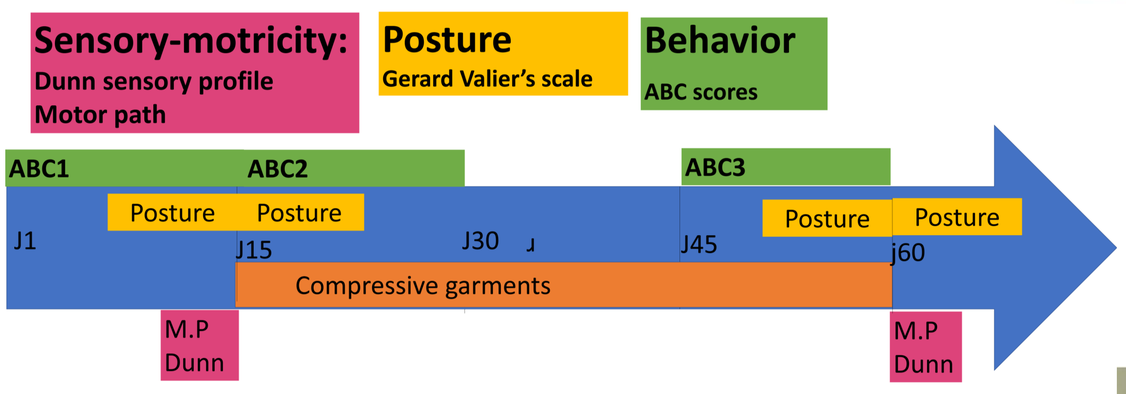
Table 1: Clinical characteristics at admission of the participants (N=14)

Socio-demographics	
Males: N (%)	13 (93)
Age (years): mean (±SD) [range]	18,2 (±5,5) [8,2 – 30,0]
SES: N (%), low/middle/high	4 (29) / 3 (21) / 7 (50)
Hospitalization	
Duration (months.): mean (±SD) [range]	10,8 (±5,5) [0,1 – 21,5]
Comorbidities: N (%)	
Syndromic ASD	7 (50)
Epilepsy	6 (43)
Catatonia	6 (43)
Obesity	3 (21)
OSA	1 (7)
Osteoporosis	1 (7)
GERD	3 (21)
Œsophagitis, duodenitis, gastritis	11 (79)
Chronic constipation	12 (86)
ENT and maxillofacial infections	4 (29)
Parasitic infection	3 (21)
Cardiopathy	2 (14)
Anaemia	3 (21)
Pruritic skin diseases	2 (14)
Iatrogeny	3 (21)
At baseline	
Patients receiving medication: N (%)	13 (93)
Poly-medication: N (%)	10 (71)
Equivalent chlorpromazine (mg) per patient receiving medication: mean (±SD) [range]	664 (± 665) [67 – 1750]
History of exception treatment: N (%)	2 (14)
Autism history	
ADI-R: 4–5 years., mean (±SD)*	
Social impairment score	26,1 (±2,8)
Communication score	9,6 (±2,1)
Repetitive interests score	7,7 (±2,4)
Developmental score	4,7 (±1,8)
Clinical characteristics	
Main reason for referral	Self-injurious behavior (N=7) Catatonia (N=2) Agitation (N=2) Destructive behavior (N=2) Hetero-aggression (N=1)
Language	Fluent (N=0),Few words (N=7)Nonverbal (N=7)
ABC, mean (±SD) [range]	61 (±23) [21 – 109]
CARS, mean (±SD) [range]	38 (±5) [30 – 51]
Vineland Adaptive Behavior Scales**	
Developmental age: years.	2,7 (±1,1) [1,8 – 5]

Table 2: Systematic search of Ehlers-Danlos Syndrome and retained diagnosis based on *Malfait et al., 2017* criteria

	Hypermobile EDS (hEDS) or Hypermobility Spectrum Disorder (HSD) criteria																
	Generalised joint hypermobility	Soft skin	Hyperextensive skin	Stretch marks	Piezogenic papules	Multiple abdominal hernias	Atrophic scars	Prolapse	Dental crowding, high-arched palate	Arachnodactyly	Arm span-to-height ratio ≥1.05	Mitral valve prolapse	Aortic root dilation	Family history	Muscular pain, joint dislocation	Differential diagnosis excluded	**hEDS**	**HSD**
P1	1	1	0	0	0	0	0	0	0	0	1	0	0	0	?	1	N	**Y**
P2	1	0	1	1	0	0	0	0	1	1	1	0	0	0	1	1	**Y**	N
P3	1	0	1	1	0	0	1	0	1	1	1	0	0	1	?	1	**Y**	N
P4	1	1	1	0	0	0	0	0	0	1	1	0	0	0	1	1	N	**Y**
P5	1	1	1	0	0	0	0	0	1	1	1	0	0	1	0	1	**Y**	N
P6	1	1	1	0	0	0	0	0	0	0	0	1	0	0	1	1	N	**Y**
P7	1	0	1	0	0	0	1	0	1	1	1	0	0	0	1	1	**Y**	N
P8	0	0	0	1	0	0	0	0	0	0	0	0	0	0	?	0	N	N
P9	0	1	0	1	0	0	0	0	0	0	0	0	0	0	0	0	N	N
P10	0	0	0	0	0	0	0	0	0	0	0	0	0	0	?	0	N	N
P11	0	0	0	0	0	0	0	0	1	1	0	0	0	0	?	0	N	N
P12	0	0	0	0	0	0	0	0	0	0	0	0	0	0	?	0	N	N
P13	0	0	0	0	0	0	0	0	0	0	0	0	0	0	?	0	N	N
P14	0	0	0	0	0	0	0	0	0	0	0	0	0	0	?	0	N	N

Figure 4 description of the Protocol



6 weeks of portage. 1h/day minimum.

Methods:

Recruitment

We retrospectively describe 14 patients with autism and SPD, hospitalized for severe challenging behaviors. Symptoms were resistant to an intensive and multidisciplinary management, including medication, treatment of organic comorbidity and behavioral restructuring (Table 1).
Proprioceptive abnormalities were assessed by trained physiotherapists based on a list of 19 symptoms.
All patients were screened for HSD and Elher Danlos syndrome (Table 2).

Procedure

Each patient received a compression garment orthose (Figure 1) to wear at least 1 hour/per day for 6 weeks. We assessed participants at baseline, 2 weeks and 6 weeks for challenging behaviors with the Aberrant Behavior Checklist (ABC), sensory integration with the Dunn questionnaire, postural sway (figure 2) and gross motor performance through a self-designed motricity path that includes ten workshops (figure 3).

Figure 1: Compressive garments designed for the pilot study

Figure 2: Postural measurement (dorsal and profile) of an autistic patient with catatonic symptoms, with or without compression, according to G. Vallier's classification.

Figure 3: Motricity path used for assessing gross motor control.

Results:

Protocol achievement

14 patients achieved the 6 weeks protocol (7 during the hospitalization, 7 others 6 weeks after discharge). 4 participants presented positive criteria for hEDS according to the initial expert examination. 3 others had generalized joint hypermobility (GJH)
The medication was stable and CG were overall well tolerated. Only one side effect was noted: a transient hand swelling in a child who slept with the compressive garment.
The protocol allowed a great variability in the duration for which the CG was worn, but it exceeded 4 h/day for 13 patients.

Motricity and posture

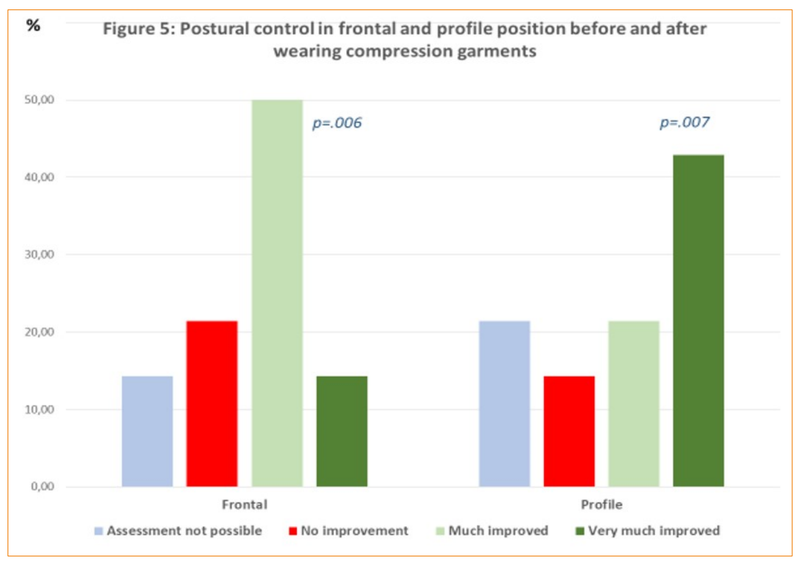
We used non parametric Wilcoxon paired sign rank test for analyze.
Postural control in dorsal and profile position significantly improved before and after wearing compression garments (p=.006 and .007, respectively, see Figure 5).
Motor performance was also significantly improved (see Table 3).
We found no significant change in Dunn sensory scores.
A comorbid HSD was not associated with a better outcome.

Behaviors

For ABC scores, we used the Friedman test and found a significant effect on most the total score, p=.004, irritability, p=.007, hyperactivity, p=.001, lethargy, p=.001, at 2 weeks, that persisted at 6 weeks (see Table 3 and figure 6).
Surprisingly there was significant difference on the stereotypies score despite the clinical impression of a reduction of self injurious behaviors for most patients.

Table 3: Clinical changes for outcome variables between baseline and T-1 (after CG) (N=14)

Variable	Baseline	T-1 (after CG)	Delta	Effect size	p*
ABC-SCORES					
ABC-irritability	21.64(9.52)	16.46(9.03)	-5.18(7.78)	-0.65	.028
ABC-lethargy	12.79(8.14)	7.57(6.89)	-5.21(4.25)	-1.19	.002
ABC-stereotypies	7.64(3.77)	6.71(3.09)	-0.93(2.5)	-0.36	NS
ABC-hyperactivity	17.36(12.59)	11.61(8.85)	-5.75(7.97)	-0.7	.003
ABC-inappropriate speech	1.79(3.38)	1.68(3.21)	-0.11(1.76)	-0.06	NS
ABC-total	61.21(23.44)	44.04(20.19)	-17.18(20.19)	-0.83	.008
DUNN QUESTIONNAIRE					
	Non significative				
MOTRICITY PATH					
Successful items	23.73(18.45)	29.91(20.43)	6.18(7.31)	0.81	.025
Emerging items	12.91(9.88)	11.82(9.9)	-1.09(6.5)	-0.16	.442
Failed items	13.27(12.19)	8.18(8.87)	-5.09(9.16)	-0.53	.074
POSTURAL CONTROL					
Frontal (mediolateral)	NA	NA	0.92(0.67)	1.37**	.006
Profile (anteroposterior)	NA	NA	1.36(0.81)	1.69**	.007



Conclusion:

Compression garments appear to be a promising adjuvant treatment for behavioral, postural and gross motor impairments in individuals with autism and SPD.
In our clinical sample of complex patients, we were unable to conclusively determine whether the CG modulates a latent pain disturbance through a "gate effect" simply by providing a relaxing feeling of comfort, or actively substitutes for some sensorial disturbances, and may even facilitate sensory integration.
Exploring the complex relationship between HSD, ASD through SPD and motor control may open new therapeutic perspectives for a subgroup of patients that will need to be more closely circumcised in future (figure /)

Figure 7 Autistic manifestations expression in a hyperlaxis body