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TITLE OF CASE

Effect of vibrostimulatory wearable technology on stereotyped behaviour in a child with autism and intellectual disability

SUMMARY

The aim of the work has been to report on the effects of vibrostimulation, administered through wearable technology, on stereotyped behaviour of a 6-year-old child with autism, intellectual disability and severe behaviour in the "stereotypic behavior" subscale of the Restricted and Repetitive Behavior Revised Scale (RBS-R). He received vibrostimulation (210 Hz, 2.8µm), with a continuous pattern of vibration: 3 vibrations of 700ms, each separated by a rest period of 500ms and a pause of 8,000ms. Vibration was delivered bilaterally by two devices, repeating the vibration pattern for 3 minutes. The measures were repeated 4 times alternately, with the device turned off and on. The outcome measure was frequency of stereotyed behaviour, which was evaluated for 3 minutes with and without vibrostimulation. The results and observations, over three minutes of stimulation, showed the disappearance of stereotyped movements during vibrostimulation and better precision in intentional hand movements. Subjectively, the child enjoyed vibrostimulation.

BACKGROUND

Stereotyped movements are repetitive motor behaviours that appear impulsive and non-purposive.[1] The 5th edition of the Diagnostic and Statistical Manual of Mental Disorders DSM-V, [2] also included, as diagnostic criteria for Autism Spectrum Disorders (ASD), the presence of restrictive and repetitive behaviour patterns (RRB), including stereotyped or repetitive movements, use of objects or speech. Moreover, stereotypies follow a unique repertoire specific to each individual and occur in varying temporal patterns, either transient or persistent, and could be a common manifestation of some disorders, such as autism spectrum disorders or intellectual disability. [3-5] Despite their high frequency and great diagnostic importance within ASD, RRBs pose many uncertainties due to their wide spectrum and complexity. [6 7]

As for treatments, when very intense, various behaviour modification techniques stand out, [8-10] occasionally using sensory stimuli -visual signals, beeping, vibration- as distractors. [11 12] However, after a systematic review of 60 articles, Lanovaz[13] suggests a shift towards other behaviours, which are not always appropriate. Other interventions have tested the influence of some sensory stimulation modalities in reducing stereotyped behaviours. [14 15] Similarly, Bressel *et al*. [16] studied the effects of vibrostimulation, administered through vibration platforms, on stereotyped behaviours, finding short-term positive results in reducing some types of stereotyped behaviours in children with autism.

Vibration stimulation, or vibrostimulation (VS), is a haptic stimulation that provides tactile and proprioceptive sensations [17] and allows the perception of physical objects and the external world. The frequency of the vibration used as a stimulus determines tactile sensations, because inputs from multiple mechanoreceptor types (Pacinian and Meissner corpuscles, Merkel's disks , and Ruffini endings) are integrated in the cortex. Human skin is found to be most sensitive to vibrations at frequencies between 150 and 300 Hz, where the Pacinian channels are predominantly activated.[18] Although the neural mechanisms underlying high frequency vibrotactile perception are still relatively unknown, some research has found that vibrostimulation at high frequencies (100 - 400 Hz) mainly stimulates the secondary or associative somatosensory cortex SII, while vibrations under 100 Hz stimulate the primary somatosensory cortex SI. [19-23] However, Kim et al[24] showed contradictory findings in the frequency of 150

Hz. On the other hand, some recent literature[25-27] reported that response inhibition processes could be more efficient when tactile stimuli are processed via SII, compared to SI somatosensory areas.

There are also conflicting results on somatosensory perception thresholds in the population with ASD and intellectual disability. Buyuktaskin *et al.* [28] report that in ASD, the perceptual threshold is elevated, requiring a more intense stimulus than in neurotypical populations. Moreover, there has been an increasing amount of literature correlating stereotyped behaviour in ASD with sensory processing disorders.[29-30] Wolf et al[31], identified that repetitive behaviours (including stereotyped behaviour) and unusual sensory response patterns co-occur and share common brain-behaviour relationships.

Considering that the occurrence of some stereotyped behaviours in ASD, could represent differences in the processing of somatosensory information, it was hypothesised that proprioceptive stimulation by high-frecuency vibration, using wearable technology, could decrease the frequency of stereotyped behaviours. For this reason, the objective of this case has been to report on the effects of vibrostimulation, administered through wearable technology, on stereotyped behaviour of one child with autism and intellectual disability.

CASE PRESENTATION

The case of one child of 6 years old, with autism, intellectual disability and severe stereotyped behaviour is presented.

Therapists at the clinical center attended by the patient report that it is difficult to do activities with him because he spends part of his time performing stereotyped movements that interfere with the activity program. For example, when the patient sits in front of a table to carry out an activity, he usually presents stereotyped behaviours such as body rocking, legs swimming and babbling. The therapists and special teachers refer that subjectively it seems that he recreates himself in these movements. He shows no signs of discomfort when performing them. They indicate that they can be interrupted by a reference person without the patient displaying disruptive behaviour or an expression that indicates disgust, but they reappear shortly after. They also consider that these behaviours interfere with their learning processes and their adaptive behaviour.

INVESTIGATIONS

The patient had a confirmed diagnosis of autism spectrum disorder (ASD). This assessment was conducted by an independent psychiatrist according to DSM-V[2] and showed persistent deficits in social communication and social interaction across multiple contexts. The child had deficits in social-emotional reciprocity and failed to initiate/respond social interactions: He was non-verbal and showed poorly nonverbal communicative behaviours in social interaction developing, deficits to understand relationships, interest in peers and lack of imaginative play. He had stereotyped motor movements (legs swinging, when seated) and body rocking. Also non-propositive bubbling was observed. He had insistence on sameness, ritualized patterns (getting on and off the same swing over and over again) and restricted interests. He had hyporeactivity to sensory input (sounds) and apparent indiference to pain/temperature. These symptoms have appeared in the early developmental period and cause clinically significant impairment in social, occupational, or other important areas of current functioning. The Autism Diagnostic Interview-Revised (ADI-R)[32] and Autism Diagnostic Observation Schedule (ADOS) [33] scores were ADI-R Total (M=33.7) and ADOS Total score (M=19.8).

The presence of severe behaviour in the first items of the "stereotypic behaviour" subscale of the Restricted and Repetitive Behavior Revised Scale (RBS-R)[34] was determined as Severe.

Apart from ASD features, he had also fulfilled DSM-5 criteria for Intellectual Disabilities (ID). He showed deficits in both intellectual and adaptive functioning. These symptoms appeared in the early developmental period and cause clinically significant impairment in conceptual, social and practical domains. Binet Standord Scale[35] Fifth Edition showed SB5 non verbal IQ<40. Specific measures showed widespread very low scores in all dimensions. He showed deficits in adaptive functioning in activities of daily life across multiple environments, such as home, school, work, and community. Vineland scale[36] was used to measure the severity of adapative functioning. It indicated an equivalent age of 1 year, 9 months, Total Score: 30,5 and a Social Quotient (SQ): 26, Range: Severe.

DIFFERENTIAL DIAGNOSIS

A differential diagnosis with ADHD was made. According to DSM-V and child development stage, the patient did not meet the criteria in either of the two dimensions of ADHD. In addition, parents and teachers completed the 18 criteria for ADHD from the DSM-5, obtaining the same results.

The neurological exploration excluded epilepsy, spasticity, chorea, ballism, tics, athetosis, dystonia and myoclonia. Psychiatric diagnosis of mental illness was excluded. The patient was not under pharmacological treatment.

TREATMENT

For the design of the wearable vibrostimulatory device, an open source smartwatch was used, specifically the Sony Smartwatch 3. The built-in vibrator was also tested to ensure that it met the required technical specifications (210 Hz and 0.517G=2.8µm amplitude).The beta version of a specific app was created using the Android Studio software. When the app is activated, the watch's vibrator vibrates with the set vibration pattern (Figure 1). The watches are hermetically-sealed to prevent liquids seeping through (e.g. saliva).

As an initial step, the psychologists conducted a functional behavioural analysis to identify the stereotypical behaviours under study. This was followed by a desensitisation phase to familiarise the patient with the smartwatch and the way in which it vibrated. The patient was trained to remove the watch easily. It was decided that if the patient ever took off his watches or made the intention of taking them off and was unable to do so, he would be helped to remove them. In addition, it was established that, prior to the test, patient had to agree to wear two watches (one on each wrist) for at least three minutes, with and without vibration (three minutes in each case). The turning on of the clock (ON condition), is the condition in which the device provides vibrostimulation (VS).

The test was conducted in the clinical centre attended by the child. It was repeated 4 times in two consecutive weeks (Tuesdays and Thursdays). During the test, the child would sit in front of a table and a psychologist would sit next to him. Once seated, the watches would be put on and the test would begin. The psychologist would alternatively suggest making two-piece puzzles of 6 animals (imitating the onomatopoeic sounds) and playing stacking blocks. The psychologist would not touch the child. If the stereotyped movements arose, they would not be interrupted.

Due to the severity of the patient's symptoms, a time duration of 6 minutes was defined for the activity to be carried out (3 minutes with VS and 3 minutes without VS). Days one and three: first 3 minutes with VS (ON condition), next 3 minutes without VS (OFF condition). Days two and four: first 3 minutes without VS (OFF condition), next 3 minutes with VS(ON condition). If the patient could not remain seated for the complete duration, he was allowed to stand up.

The dependent variable of this test was the percentage of time engaged in the identified stereotyped behaviours rated as severe on the RBS scale and measured every 30 seconds during the test. The independent variable being the presence or absence of VS (ON/OFF condition). The tests were videotaped and analysed by two psychologists of the team. The reliability of the inter-observer observations was determined by the percentage of agreement in relation to the total measurements (agreement and disagreement).

Following other studies in the population with ASD and/or ID, a vibrostimulation pattern was defined, repeated every 8,000ms, consisting of 3 vibrations of 700ms, each separated by a rest period of 500ms. [37- 39] Vibration would be delivered bilaterally for 3 minutes by two smartwatches, placed on both wrists, repeating the vibration pattern shown in Figure 1 uninterruptedly. Both smartwatches would vibrate synchronously.

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Figure 1. Vibration pattern.

At the end of the test, observers would collect qualitative observations on activity performance, paying particular attention to the patient's facial expressions and behaviours as an indicator of their acceptance or non-acceptance of the VS. Declaration of Helsinki was approved by the ethics committee of the institution conducting the study, according Helsinki principles. The guardians of the participant provided informed consent.

OUTCOME AND FOLLOW-UP

The results are shown in Figure 2 and Table 1. The inter-observer reliability showed agreement between observers was 96.7% (Mean), 3,5 (s.d).

In the 4 repetitions, during OFF condition, the patient wore the watches all the time. The presence of stereotyped behaviours are shown in Figure 2 (left column) and Table 1. The means and s.d. Mean(s.d.) of the percentages of time, measured over 3 minutes, were 28.01(1.58), in babbling; 64.10(4.52), in legs swinging and 16.09(1.53) in body rocking. The patient collaborated in doing the puzzles, and playing with the stacking blocks. In addition, the patient tried to fit the pieces of the puzzles with verbal help, although he did not succeed. However, the patient managed to stack some blocks. Subjectively, no expression or behaviour showing displeasure/discomfort was observed. The patient did not get up from the table. During the 4 repetitions of the test, there were 3 moments in which the patient hit the screen of the watch and then put the watch to his ear, seeming to search for the vibration.

During the VS-phase (ON condition), and on all 4 occasions, the disappearance of all stereotyped behaviours (babbling, legs swinging and body rocking) was observed, both present in the OFF condition. On the 4 occasions, at the beginning of the VS there were stereotyped behaviours and their disappearance occurred between the first 2 and 3 seconds, from the VS. This implies the presence of 2-3 seconds of stereotyped behaviours during the first measurement (made at 30 seconds). This fact is not visually perceived in the graphs (right column) of Figure 2, because the average percentage of time of stereotyped behaviours (measured in the first 30 seconds) ranges between 0.07-0.08% (see Table 1). The patient agreed to perform the activities proposed by the psychologist (puzzles and blocks). During the VS condition, upon the psychologist's demonstration, he repeated the onomatopoeic sound of a rooster (chil said “ki-ki-kiiii”, psycologist said “kikiriki”) and a dog (child said “wao”, psycologist said “guau”), what was reproduced in the different repetitions of the test. It was also observed that the child made his attempts to fit the pieces more slowly than without VS. Although the patient needed verbal instructions, on several occasions, he was able to fit some pieces with slower and more precise hand movements. It was also observed that the stacking of the blocks was done in a slower and more precise way. On all the

occasions when the VS began, the child brought the clocks closer to the ears and mouth, seeming to seek to recognize/perceive the VS in those areas. He did not get up from the chair in either phase. Subjectively, the child enjoyed vibrostimulation.

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Table 1. Mean (standard deviation) of the percentages of time spent on stereotyped behaviours in ON/OFF conditions. Mean(s.d.) values were obtained from the 4 measurements in the different time intervals (measures 1 to 4).

Observation Time		**Babbling Mean (s.d.)**		**Legs swinging Mean(s.d.)**		**Body rocking Mean(s.d.)**	
(minutes)	(seconds)	OFF Cond.	ON Cond.	OFF Cond.	ON Cond.	OFF Cond.	ON Cond.
	30	29.74(12.12)	0.08 (0.00)	65.39(3.86)	0.07 (0.00)	18.35(2.41)	0.07 (0.00)
1	60	25.28(13.98)	0.00 (0.00)	61.03(0.67)	0.00 (0.00)	14.35(4.45)	0.00 (0.00)
	90	28.15(13.23)	0.00 (0.00)	56.99(0.34)	0.00 (0.00)	16.23(1.60)	0.00 (0.00)
2	120	28.28(9.57)	0.00 (0.00)	64.83(9.38)	0.00 (0.00)	14.85(2.40)	0.00 (0.00)
	150	27.36(11.06)	0.00 (0.00)	66.36(7.35)	0.00 (0.00)	17.35(3.33)	0.00 (0.00)
3	180	29.25(11.78)	0.00 (0.00)	69.99(8.70)	0.00 (0.00)	15.43(4.96)	0.00 (0.00)
Mean(s.d) along 3 minutes		**28.01(1.58)**		**64.10(4.52)**		**16.09(1.53)**	

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Figure 2. Effect of vibrostimulation on stereotyped behaviours (Mean value of 4 measurements in red). Left: Results OFF condition (wearing the device without VS). Right: Results ON condition (wearing the device with VS). Measurements 1 and 3: results of 3 min. OFF condition/3 min. ON condition(VS). Measurements 2 and 4: results of 3 min. ON (VS) condition/3 min. OFF condition.

DISCUSSION

In all the measurements carried out, a disappearance of stereotyped behaviours was observed during VS. The fact that a possible response to vibrostimulation has been observed in the case presented opens the door to further research in this area. Althought the effect of the VS has only been studied in a brief period of time (3 minutes), during these short periods of time, some improvements have also been observed in the control of hand movements and the imitation of onomatopoeic sounds (vocalizations). These findings could also suggest implications for learning. The greater control in the movement of the hands could be explained as a secondary effect of the disappearance of movements such as legs swinging and/or body rocking, because when these movements appear, the rest of the body also moves a little (hands included). Picking up an object in these conditions implies picking it up from a body that is in motion, despite the patient being seated. Additionally, the findings could suggest that the presence of babbling was preventing intentional vocalizations.

On the other hand, the observed findings would not imply causal effects. Moreover, future studies are needed to address in more depth the etiology of stereotypies and their relationship with neurodevelopmental disorders. Similarly, more research studies would contribute to clarifying the mechanisms by which stereotyped behaviours can disappear or decrease under vibrostimulation conditions and answer questions such as the permanence of the VS effect for longer periods of time, or what would be the most appropriate vibration pattern and what consequences could VS condition have on the patient if it were applied as a therapy.

There are many studies that apply VS for the rehabilitation of various motor functions in the presence of neurological impairment[40] or chronic stroke,[41] even with wearable technology. However, far too little attention has been paid to neurodevelopmental disorders.

Other studies have used similar technology to reduce anxiety and self-regulation problems, [42,43] with limited results, and one study proposes the use of smartwatches to monitor stereotyped movements, but not to reduce them[44]. Regarding other experiences on vibrostimulation, Bressel *et al.* [16] observed the reduction (not disappearance) of some types of stereotypies in children with autism through the use of vibration platforms during a case study. In the same line, some contributions study how the oscillatory movement produced on the body by physical activity reduces stereotyped behaviours in people with autism. [45,46] However, in both cases (vibration platforms and physical exercise), the type of vibratory stimulus is different from the one used in this study, as vibration platforms and physical exercise provide stimuli at low frequencies but with much higher amplitudes, so the results are not comparable with those of this work. The parameterization of the vibrations of the device used in this study (frequency, amplitude, vibration pattern and duration of the stimulus), together with the fact that the device is easily replicable, will facilitate the comparison of future studies and even the comparison of long-term results, in the case of therapies.

In conclusion, although in this case report the VS has been shown to be correlated with the disappearance of stereotyped behaviours in a very short periods of time, further experimental research is needed to confirm this correlation and evaluate effect saturation, VS patterns, frequency and intensity of vibrations and including the impact of the device (shape, weight, texture) on the results. Therefore, despite the methodological limitations of case reports, the present study could be a starting point for further progress in the reduction of stereotyped behaviours through the use of wearable vibrostimulatory technology.

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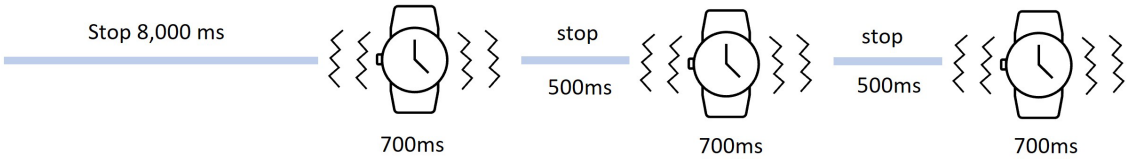

Figure 1. Vibration pattern.

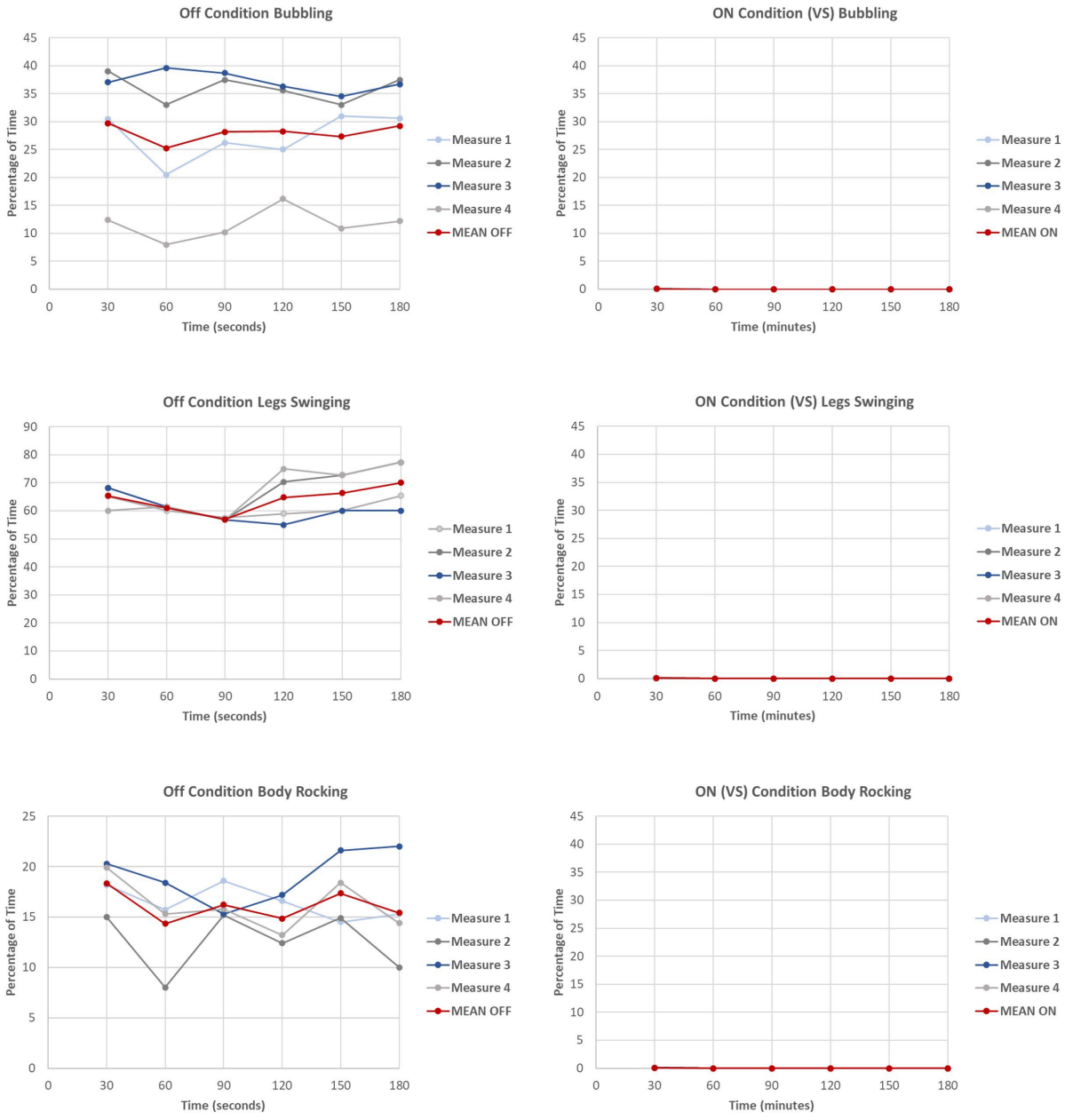

Figure 2. Effect of vibrostimulation on stereotyped behaviours (Mean value of 4 measurements in red). Left: Results OFF condition (wearing the device without VS). Right: Results ON condition (wearing the device with VS). Measurements 1 and 3: results of 3 min. OFF condition/3 min. ON condition(VS). Measurements 2 and 4: results of 3 min. ON (VS) condition/3 min. OFF condition.