

Clumsiness in fine motor tasks: evidence from the quantitative drawing evaluation of children with Down Syndrome

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Introduction

A pervasive feature of motor skill performance in participants with Down Syndrome (DS) is ‘clumsiness’, which commonly implies an ample set of movement characteristics, such as slow movements with unusual, less efficient patterns of co-ordination and high rates of failure (Galli *et al.* 2010; Rigoldi *et al.* 2011a), slower reaction times, lower muscle tone and ligament laxity (Morris *et al.* 1982; Galli *et al.* 2008). The evaluation of ‘clumsiness’ has since now taken into account gross motor tasks, mainly walking and slowness of movements seems to be one of the main, omnipresent features of DS movements. This phenomenon has been largely related to the biomechanical features of DS’s movements, such as muscular weakness and ligament laxity (Cioni *et al.* 2001; Rigoldi *et al.* 2011a). Recent literature, however, points out that clumsiness may be mainly a product of limitations at

central nervous system level, more than a product of biomechanical constraints alone (Virji-Babul & Brown 2004; Latash 2007). In this sense, biomechanical aspects are recently beginning to be linked together with the specific sensory, motor, cognitive and perceptual impairments of DS, but it remains unclear how these localised deficits impact on perceptual-motor processing and function (Virji-Babul & Brown 2004; Vimercati *et al.* 2013).

Behavioural disorders in DS include decreased attention, hyperactivity and impulsivity, which are frequently reported in DS. The exact prevalence of attention-deficit/hyperactivity disorder (ADHD) has not been clearly estimated in this population, yet a recent study (Ekstein *et al.* 2011) indicates that 43.9% of the evaluated participants with DS was affected by ADHD. The possible presence of ADHD in DS population is supported also by several studies addressing the presence of a deficit in the frontal lobes function in DS (Rowe *et al.* 2006; Kittler *et al.* 2008; Lanfranchi *et al.* 2009, 2010). Sensory deficits are present as well, such as increased risk of hearing loss and eye diseases (Ekstein *et al.* 2011). The general picture of DS thus presents itself as a very complicated picture, in which aspects of co-morbid psychiatric disorders interplay with the cognitive, motor and perceptual impairment caused by the syndrome itself. Some aspects of this picture have been more in-depth studied, such as the gross motor functions of walking and posture (Galli *et al.* 2010; Rigoldi *et al.* 2011a,b), which allowed an in-depth study of the biomechanics of movement in DS, whereas some other aspects are only recently beginning to be considered by literature. For instance, drawing ability, which has been since now almost completely omitted from the analysis, could provide important information about the cognitive state of DS. The study of drawing, in fact, allows analysing the perceptual-motor skills of the participant (in particular visual-spatial and grapho-motor abilities), together with the presence of ADHD disturbances. Simple drawing tests are commonly used for the clinical evaluation of cognitive capabilities in children with learning disabilities. Among several graphic tests that have been developed for the cognitive evaluation of children, the Denver Developmental Screening Test (DDST) (Frankenburg *et al.* 1992) is one of the most used. The test comprises a

drawing session in which the child is asked to copy the figures of a circle, a square and a cross. Copying a figure requires the child to consider the visual form (figure) as well as the neuromuscular adjustments for line control, direction, speed and pressure (Khalid *et al.* 2010), together with appropriate management of the ocular-motor co-ordination. Thus, drawing a figure can give insight on both the biomechanical control and some of the neural mechanisms underlying this control.

Graphic tests have been administered to evaluate the performance of children with a wide range of pathologies and/or difficulties, such as children at risk for school problems (Perera 2005; Bayoglu *et al.* 2007), children with developmental co-ordination disorders (Smits-Engelsman *et al.* 2003), children with autism (Sheppard *et al.* 2007, 2009) and children with learning disabilities (Galli *et al.* 2011) but, to the best of our knowledge, only one study (Clements & Barrett 1994) addressed the characterisation of drawings in children with DS, though by a qualitative visual evaluation. This study analysed the drawing performance of children and young people with DS compared with verbal-mental-age-matched children without learning difficulties. The drawing task required the participants to depict the partial occlusion of one object by another object. The drawings were given a visual score, which was then correlated with the verbal mental age of the participants. Children with DS obtained significantly lower scores than children without learning disabilities and, more interestingly, they employed different drawing strategies on individual drawing tasks. While the control group's performance correlated strongly with the verbal mental age, the performance of DS did not show the same correlation. Consequently, the authors suggested that the differences between the drawings of children with and without DS reflected a developmental difference in the underlying processes of drawing production and development rather than a delay in development.

Most of the drawing evaluation in clinical routine is nowadays still based on visual scoring systems, such as for the DDST. However, some quantitative methods have been developed and applied in research settings in the last years. In particular, a three-dimensional (3D) method for graphic gesture acquisition with the use of an optoelectronic system

was developed by Galli *et al.* (2011) to allow a quantitative, detailed description of drawings. The method was successfully applied to children with learning disability (Galli *et al.* 2011), participants with Parkinson Disease (Galli *et al.* 2012) and with dementia (De Pandis *et al.* 2010). In the present study, and based on the previous work of Galli *et al.* (2011) on the acquisition of drawings with an optoelectronic system, we analyse quantitatively the drawings of a group of children with DS and of a group of healthy, mental age-matched controls. The aim of the study was to characterise the features of fine motor skills in DS during a drawing task taken from the DDST.

Methods

Participants

Two groups of participants were enrolled for this study at the IRCCS San Raffaele Pisana, in Rome, where the acquisitions took place. The participants and their legal tutors gave their informed written consent to the study. The study was approved by the ethical committee of IRCSS San Raffaele Pisana, Tosinvest Sanità, Rome, Italy, in accordance with the ethical principles of the Declaration of Helsinki.

The DS group was composed of 23 children with DS. Their chronological mean age was 14.9 ± 4.6

years old, whereas their mean mental age, estimated from the quotient of intelligence index, was 8.1 ± 2.9 years old. The inclusion criteria for DS were right-handedness, a regular school frequency and education, no orthopaedic problems that could restrict upper limbs motion.

The control group (N) was composed of 13 children, whose mean age (9.0 ± 2.1 years old) was matched to the mental age of the DS group. Inclusion criteria for N were right-handedness, no physical or psychological dysfunction and a regular school education.

Methods

The graphic gesture was acquired with an optoelectronic system with six cameras (SMART-D BTS; Italy), at a frequency of 200 Hz, and with an integrated video system (Vixta, BTS, Italy) for video-recording. The optoelectronic system is an equipment that measures the 3D co-ordinates (X, Y, Z) of reflective markers through time. The markers were of diameter = 10 mm and were used in the configurations described here following (Fig. 1a,b).

Before every acquisition, a calibration was performed to define a global reference system frame for all the cameras and compute the extrinsic and intrinsic parameters for each camera. The calibrated volume (around $0.6 \times 0.4 \times 0.6$ m) was defined

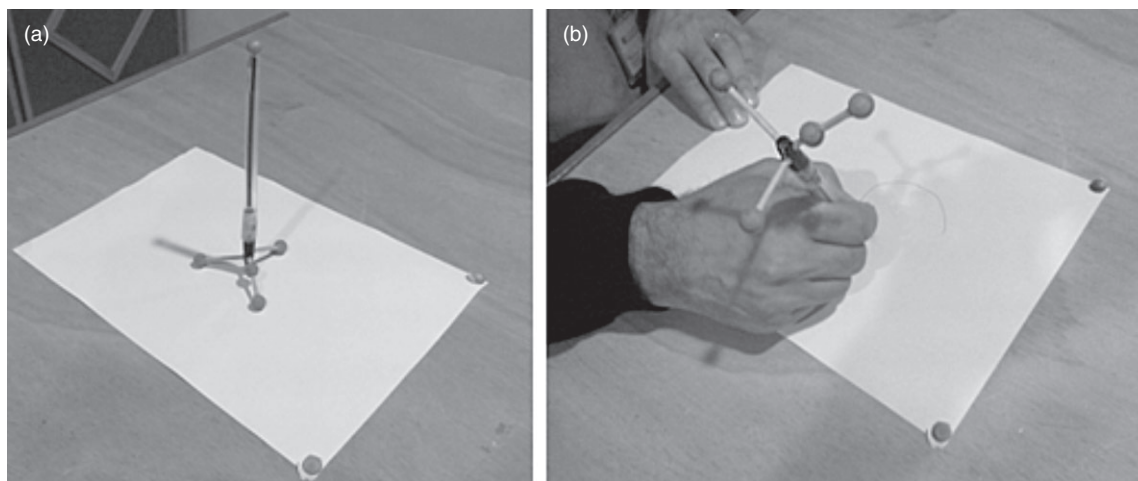


Figure 1 (a) Markers configuration for the pen during the static acquisition, (b) markers configuration for the pen and sheet during the dynamic acquisition.

considering that the volume had to include the whole motion and it had to be as small as possible, in order to obtain a high accuracy. At the end of the process, calibration was considered acceptable if the mean error on the computation of the difference between the measure and actual distance of two markers fixed on the extremities of a rigid bar at the distance of 150 mm was within 0.20 mm (standard deviation: 0.20 mm).

The children seated comfortably on an adjustable chair, in front of a desk. Their height respect to the desk was regulated to allow easy and comfortable drawing. They were given a paper sheet with a printed figure (a circle, an equilateral cross and a square) and were asked to ‘copy the illustrated figure’ with their dominant hand. The figures were presented one per time. After drawing the first figure, the child was presented with the second and then with the third. Three acquisitions (one for each drawing) were recorded for each child. Children were given a modified ink pen with markers on the cap that allowed the reconstruction of the trace drawn by the children, as described following.

Two acquisitions were necessary for each participant: a static acquisition of the markers on the pen, and a dynamic acquisition during which the participant drew. The first markers configuration, shown in Fig. 1 on the left, was used for the static acquisition, in which the participant did not take part. The pen was positioned on the table with the four markers on the cap and a marker on the tip, and the markers were acquired for five seconds, in order to calculate the position of the tip of the pen and allow the calculation of its position during the dynamic acquisition, in which the graphic test was executed by the participant. Figure 1 on the right shows the markers configuration for the drawing trials: the marker on the tip was removed, and the participant drew on the sheet. Two markers were also placed on the sheet during the dynamic acquisition. This marker configuration is a further adaptation and improvement of the method by Galli *et al.* (2011).

As shown in Fig. 2, starting from the co-ordinates of the markers on the pen cap, a system of reference (X_p , Y_p , Z_p) was defined on the pen. In this way, during the dynamic acquisition, the pen tip co-ordinates were reconstructed

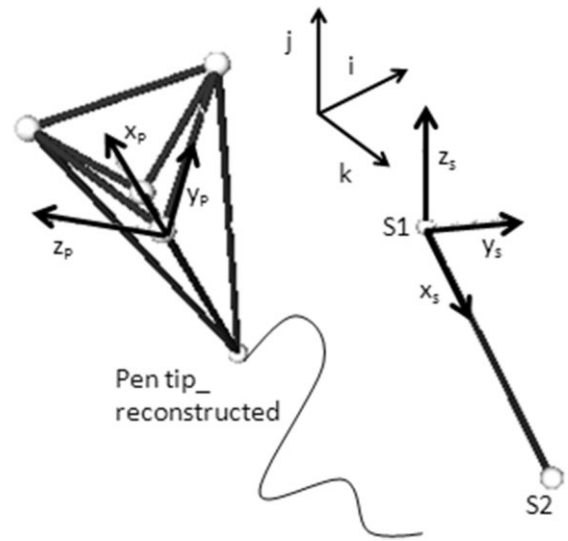


Figure 2 Pen tip reconstruction during the drawing trials and reference system on the sheet to allow rotation of the sheet during the drawing.

(Pen tip_reconstructed) and it was possible to obtain the digitalised drawing trace (i.e. the drawn figure) and the trace of the pen lifts. Another system of reference (X_s , Y_s , Z_s) was defined starting from the markers on the sheet and from the laboratory reference system (i , j , k). In this way, the sheet could be rotated by the participant during the drawing without interfering with the measurements, allowing free and natural movements of the participants.

Markers were also put on the body of the participant. Landmarks on the body were chosen in order to minimise the effect of the skin artefacts. In particular, markers were put on the head, shoulders, trunk, elbow, wrist and hand on the side of hand dominance. The protocol for markers placement is shown in Fig. 3 for a right-handed participant.

Tests and parameters

After reconstructing the 3D co-ordinates of the markers, the following parameters were computed. To characterise the position of the participant's head during the drawing, the maximum and minimum projections of the c_head marker on the table were computed and the difference between these two values was named head-table distance ($head_table_dist$) (m).

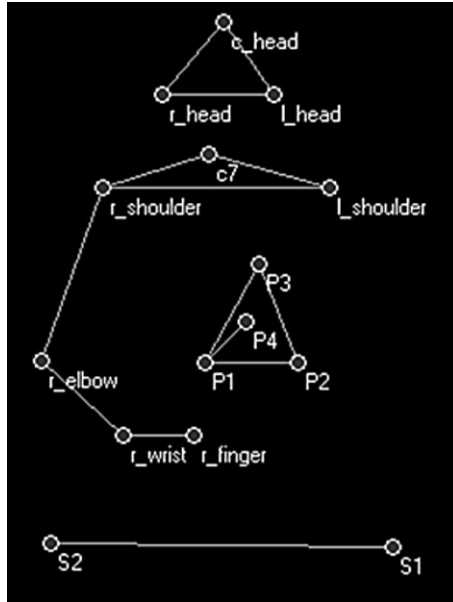


Figure 3 Protocol for markers placement; r, right; l, left; c, central.

To characterise the movement of the upper limb, the elbow angle was defined as the acute angle between the markers positioned on the shoulder, elbow and wrist. The wrist angle was defined as the acute angle between the markers positioned on the elbow, wrist and hand. The ranges of motion (ROMs) of these two angles were computed from the co-ordinates of the external markers.

To characterise the drawing traces of the different figures the following parameters were calculated.

Circle drawing

The drawing features of the circle were characterised by:

- length of the drawing track (*Length*) (m), drawing time (*Time*) (s) and drawing peak of velocity (*Max Vel*) (m/s);
- horizontal and vertical diameters lengths (*H_Dm*, *V_Dm*) (m);

Drawing accuracy was evaluated by the parameter of eccentricity:

- eccentricity (*Ecc*): $Ecc = \left| 1 - \frac{V_Dm}{H_Dm} \right|$

The more the drawn figure is close to a perfect circle, the more the parameter approaches a 0 value.

Cross drawing

The drawing features of the cross were characterised by:

- drawing time (*Time*) (s) and drawing peak of velocity (*Max Vel*) (m/s);
- length of the horizontal and vertical sides (*H_side*, *V_side*) (m);

Drawing accuracy was evaluated by the cross side error parameter, chosen to assess the tendency to draw irregular cross bars:

- cross side error (*side-ε*): $side_ε = \left| 1 - \frac{H_Side}{V_Side} \right|$

The closer the value is to 0, the more precise is the drawing, i.e. the sides have more similar lengths (equilateral cross);

Square drawing

The drawing features of the square were characterised by:

- drawing time (*Time*) (s) and drawing peak velocity (*Max Vel*) (m/s);
- length of the upper, lower, left and right sides (*S1*, *S2*, *S3*, *S4*) (m);

Drawing accuracy was evaluated by two parameters, chosen to assess the tendency to draw an irregular polygon:

- square sides error (*s - ε*) (m): $s - ε = |S1 - S2| + |S3 - S4|$

the closer the value is to 0, the more precise is the drawing, that is, the sides have more similar lengths.

- square to rectangle error (*str - ε*): $str - ε = \left| 1 - \frac{W}{H} \right|$

where *W* is the square's width, calculated as $\text{Max}(S1, S2)$ and *H* is the square's height, calculated as $\text{Max}(S3, S4)$. The closer the parameter is to 0, the closer the drawing is to a square.

These parameters were chosen to assess the tendency to draw irregular parallelepiped rather than squares.

Figure 4 illustrates some of the analysed parameters.

Statistical analysis

Data were collected for each participant and tabulated in order to compare overall results from pathological group and control group. The median,

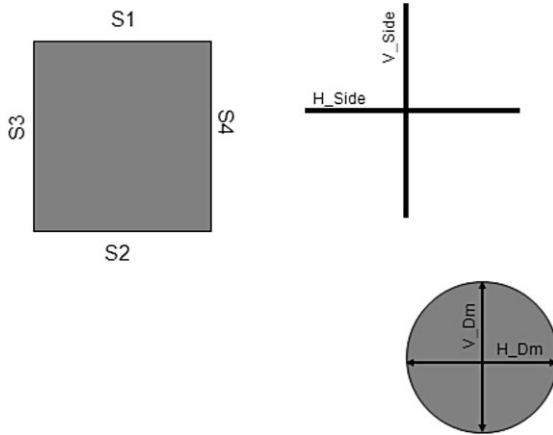


Figure 4 Representation of some of the parameters.

25° and 75° percentile values were computed for each group and parameter. The non-parametrical Mann–Whitney *U*-test was used to verify the presence of statistically significant differences between the DS and N groups. Differences were considered significant at a *P*-value < 0.05.

Results

The kinematic parameters of upper limb motion did not reveal any statistically significant difference between groups.

Table 1 presents the results for the characterisation of the drawing features and drawing accuracy in DS and N participants and for the head-table distance parameter.

For the circle, although the length of the track and the diameters' lengths were similar (similar drawing dimensions among groups) duration of the drawing was shorter for DS than for N. In agreement with this, maximum velocity was higher in DS. The eccentricity parameter was not statistically different for N compared with DS, meaning that a comparable degree of accuracy was present in the drawings of the two groups in terms of eccentricity.

For the cross, duration of the drawing was shorter in DS. The horizontal side of the cross was shorter in DS as well, whereas dimensions were comparable among groups for the vertical side. Maximum velocity was higher for DS. The accuracy parameter highlighted the presence of more inaccuracy in DS: the side-error parameter revealed in fact

Table 1 Median (25° percentile, 75° percentile) values for the drawing features and drawing accuracy parameters for the Down Syndrome (DS) and control (N) groups

Parameter	DS Median (25°, 75°)	N Median (25°, 75°)	<i>P</i> -value < 0.05
Circle drawing			
Length (m)	0.14 (0.10, 0.15)	0.15 (0.11, 0.18)	
Time (s)	2.92 (2.11, 5.70)	6.48 (5.12, 9.07)	*
Max Vel (m/s)	0.08 (0.06, 0.10)	0.05 (0.04, 0.07)	*
H_Dm (m)	0.04 (0.03, 0.05)	0.05 (0.04, 0.05)	
V_Dm (m)	0.04 (0.03, 0.05)	0.05 (0.04, 0.05)	
Ecc	1.16 (0.98, 1.24)	0.99 (0.96, 1.04)	
Head-table_ dist (m)	0.02 (0.01, 0.02)	0.03 (0.03, 0.08)	*
Cross drawing			
Time (s)	3.31 (2.07, 4.88)	6.66 (4.94, 8.35)	*
Max Vel (m/s)	0.09 (0.07, 0.18)	0.05 (0.05, 0.13)	*
H_side (m)	0.04 (0.03, 0.05)	0.06 (0.05, 0.06)	*
V_side (m)	0.06 (0.05, 0.07)	0.06 (0.05, 0.07)	
s_ε	0.26 (0.12, 0.39)	0.15 (0.07, 0.19)	*
Head-table_ dist (m)	0.01 (0.01, 0.03)	0.05 (0.03, 0.07)	*
Square drawing			
Time (s)	7.87 (4.68, 12.53)	10.01 (9.14, 11.95)	
Max Vel (m/s)	0.10 (0.08, 0.11)	0.07 (0.05, 0.07)	*
S1 (m)	0.05 (0.04, 0.06)	0.05 (0.04, 0.05)	
S2 (m)	0.05 (0.04, 0.06)	0.05 (0.05, 0.05)	
S3 (m)	0.05 (0.04, 0.06)	0.04 (0.04, 0.05)	*
S4 (m)	0.05 (0.04, 0.06)	0.04 (0.04, 0.05)	*
s_ε	0.01 (0.01, 0.02)	0.00 (0.00, 0.01)	*
str_ε	0.21 (0.11, 0.23)	0.06 (0.01, 0.11)	*
Head-table_ dist (m)	0.03 (0.02, 0.06)	0.03 (0.02, 0.06)	

* *P*-value < 0.05.

that DS's crosses were further from being equilateral crosses, with the centre of the cross being more off-centred, decentralised from the ideal position in DS respect to N.

For the square, no duration differences were found. Dimensions were similar and just some slight difference was found in the two vertical sides of the square, with DS drawing slightly longer sides. The side error was slightly higher for DS, whereas the tendency to draw rectangles instead of squares was more pronounced for DS.

The head-table distance was lower for DS in both the circle and cross drawing, whereas it was comparable for the square. Thus, the DS drew with a

closer position of the head respect to the sheet in the circle and cross drawings.

Discussion

Drawing tests are commonly administered to children with a wide range of cognitive impairments. Surprisingly, drawing tests have not been applied in literature to DS, with the exception of the work by Clements & Barrett (1994), who analysed drawings from a qualitative point of view. The aim of the study was therefore to characterise the features of fine motor skills in DS by a quantitative 3D analysis of drawing.

Literature has proven how clumsiness is a pervasive feature of DS gross movements, and how it manifests mainly as slow, less efficient movements (Galli *et al.* 2010; Rigoldi *et al.* 2011a). Traditionally, this phenomenon has been largely related to the biomechanical features of DS's movements (Cioni *et al.* 2001; Rigoldi *et al.* 2011a). On the other hand, recent literature points out that clumsiness may be mainly a product of the limitations at the central nervous system level, more than a product of biomechanical constraints alone (Virji-Babul & Brown 2004; Latash 2007). The results of the present study support this latter hypothesis. The present results, in fact, highlight shorter durations and higher peaks of velocity (with comparable drawing dimensions) in the drawings of DS compared with mental aged-matched participants without cognitive disability. An increased velocity would not be *per se* a proof of clumsiness, but it is in fact a proof of clumsiness if we consider velocity results together with accuracy results. The general accuracy in drawing, in fact, appears to be lower in DS than in controls if we take into account the cross and square drawings, whereas it is comparable in the circle drawing. The cross and square drawings, in fact, are less regular, with the square often depicted as a rectangle and the cross often depicted with a decentralised centre and uneven sides. Thus, our participants with DS tend to draw faster but with less accuracy than controls. For what concerns the circle, higher velocity peaks were found whereas accuracy seems comparable among groups. This may be attributable to the higher complexity of drawing curved lines instead of straight lines as required in the circle drawing, which caused

lower accuracy in both groups. Anyway, the percentile ranges for the two groups (DS: 0.26, N: 0.08), calculated by subtracting the 25° percentile value to the 75° percentile value, suggest the presence of a higher variability in DS, so it is possible that a significant difference could be found just by increasing the participants' number. Thus, a first comment on the results is that DS's clumsiness in fine movements such as drawing manifests itself in a different way than it does for gross movements. The fact that clumsiness acquires different features based on the kind of motor task involved (gross vs. fine motor tasks) suggests that this central characteristic of DS movement is not mainly related to muscular weakness and/or ligament laxity problems (i.e. problems at the 'effector system'), yet it is principally due to a problem at a central level, in agreement with recent studies (Virji-Babul & Brown 2004; Latash 2007). Thus, biomechanical aspects such as slowness of movement, or velocity of movement in our case, should probably be interpreted in light of the specific sensory, motor, cognitive and perceptual impairments of DS.

The fact that kinematic parameters of the upper limb did not reveal significant differences, and nevertheless drawing accuracy was lower and velocity was higher in DS, provides further evidence for the prevalence of cognitive aspects in the performance of drawing. Our results in fact show that in children with DS a psycho-motor delay, more than a biomechanical constrain, is present, which causes difficulties to represent, programme and activate correct motor sequences, manifesting motor clumsiness and lower levels of accuracy in drawings. This is in agreement with the study by Clements & Barrett (1994), who suggested that the differences between the drawings of children with and without DS reflected a developmental difference in the underlying processes of drawing production and development rather than a delay in development.

Limitations and future developments

Attentive inefficiency may have also contributed to the decreased ability of modifying the performed and the to-be-performed action of drawing. One limitation of our study is that participants were not evaluated for ADHD, so we cannot draw conclusions about the presence of ADHD in our specific

group of participants. However it is known that inattention, excessive motor hyperactivity or restlessness, and poor impulse control could lead to an impulsive drawing characterised by high speed and low accuracy, and it is known that DS has a high prevalence of attention disorders. The different posture of DS during the drawing, which led to a closer distance between their head and the sheet, may be an attempt to focus on the drawing by increasing the visual field directed on the drawing, consecutively reducing distracting factors. The analysis of drawing could be used to evaluate the presence of ADHD in DS, and future research should be addressed at evaluating and correlating the drawing performance of DS with ADHD evaluation scores. A deeper neuropsychological evaluation, together with an increase in the number of participants involved in the study, may give interesting falls out on the use of drawing tests as diagnostic tools in DS.

Conflict of interest

All authors have no conflicts of interest and no financial interest. All authors attest and affirm that the material within has not been and will not be submitted for publication elsewhere.

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