Measuring intellectual ability in cerebral palsy: the comparison of three tests and their neuroimaging correlates.

Júlia Ballester-Plané[a,b], Olga Laporta-Hoyos[a,b], Alfons Macaya[c], Pilar Póo[d], Mar Meléndez-Plumed[e], Élida Vázquez[f], Ignacio Delgado[f], Leire Zubiaurre-Elorza[g], Ana Narberhaus[a], Esther Toro-Tamargo[e], Maria Eugenia Russi[d], Violeta Tenorio[h], Dolors Segarra[a], Roser Pueyo[a,b,*]

[a] Departament de Psicologia Clínica i Psicobiologia, Universitat de Barcelona, Barcelona, Spain
[b] Institut de Neurociències, Universitat de Barcelona, Barcelona, Spain
[c] Grup de Recerca en Neurologia Pediàtrica, Vall d'Hebron Institut de Recerca, Universitat Autònoma de Barcelona, Barcelona, Spain
[d] Servei de Neurologia, Hospital Universitari Sant Joan de Déu, Barcelona, Spain
[e] Servei de Rehabilitació i Medicina Física, Hospital Universitari Vall d'Hebron, Barcelona, Spain
[f] Servei de Radiologia Pediàtrica, Hospital Universitari Vall d'Hebron, Barcelona, Spain
[g] Departamento de Fundamentos y Métodos de la Psicología, Facultad de Psicología y Educación, Universidad de Deusto, Bilbo-Bizkaia, Spain
[h] Servei de Neonatologia, Hospital Clínic de Barcelona, Universitat de Barcelona, Barcelona, Spain

Abstract

Standard intelligence scales require both verbal and manipulative responses, making it difficult to use in cerebral palsy and leading to underestimate their actual performance. This study aims to compare three intelligence tests suitable for the heterogeneity of cerebral palsy in order to identify which one(s) could be more appropriate to use. Forty-four subjects with bilateral dyskinetic cerebral palsy (26 male, mean age 23 years) conducted the Raven's Coloured Progressive Matrices (RCPM), the Peabody Picture Vocabulary Test-3rd (PPVT-III) and the Wechsler Nonverbal Scale of Ability (WNV). Furthermore, a comprehensive neuropsychological battery and magnetic resonance imaging were assessed. The results show that PPVT-III gives limited information on cognitive performance and brain correlates, getting lower intelligence quotient scores. The WNV provides similar outcomes as RCPM, but cases with severe motor impairment were unable to perform it. Finally, the RCPM gives more comprehensive information on cognitive performance, comprising not only visual but also verbal functions. It is also sensitive to the structural state of the brain, being related to basal ganglia, thalamus and white matter areas such as superior longitudinal fasciculus. So, the RCPM may be considered a standardized easy-to-administer tool with great potential in both clinical and research fields of bilateral cerebral palsy.

What this paper adds?

There is a need to find a brief and easily applied psychometric intelligence test that minimizes the interference of motor and communicative impairments experienced by people with cerebral palsy. To our knowledge, this is the first study that aims to compare several intelligence tests in order to determine which one(s) may be more appropriate to use in this population. For this purpose, our study not only includes two widely used tests in the field of cerebral palsy (Raven's Coloured Progressive Matrices and Peabody Picture Vocabulary Test-3rd) but also one which is a newly-adapted version of the Wechsler's scales, not yet applied to this group (Wechsler Nonverbal Scale of Ability).

Furthermore, this is one of the few studies in cerebral palsy that aims to investigate the relationship between intelligence tests and an extensive neuropsychological battery, which includes measures of attention, language, visuoperceptual abilities, memory and executive functions. Finally, it also aims to investigate the relationship between intelligence scores and measures of brain volume. To date, the intelligence-brain relationship in people with cerebral palsy has been mainly studied in a qualitative manner. Using quantitative techniques for analysing magnetic resonance images, such as voxel-based morphometry, allow us to be more precise and to detect relationships that could not be perceived otherwise.

1. Introduction

Cerebral palsy (CP) is one of the most common causes of physical disability in early childhood and this disabling condition persists throughout the life of the affected children (Krageloh-Mann & Cans, 2009). The term 'cerebral palsy' encompasses a group of permanent disorders of movement and/or posture and of motor function due to non-progressive disturbances in the developing or immature brain, and in which accompanying impairments are becoming increasingly important (Rosenbaum et al., 2007; Surveillance of Cerebral Palsy in Europe, 2000). The Surveillance of Cerebral Palsy in Europe (SCPE) collaborative group recommends collecting information on at least four associated impairments, including the intellectual function (Christine et al., 2007). In this direction, the International Classification of Functioning, Disability and Health (ICF) considers the intellectual function as one of the essential ICF Core Sets for children and youth with CP (Schiariti, Selb, Cieza, & O'Donnell, 2014). Moreover, intellectual functioning is taken into account to gain access to different medical, educational and social resources.

It has been reported that between 34% and 64% of people with CP have an intellectual disability (intelligence quotient, IQ<70) (Novak, Hines, Goldsmith, & Barclay, 2012). However, these data were based on different methods such as 1) estimates from clinical observation, from cognitive description provided by parents or from the type of education; 2) application of developmental scales; or 3) through formal IQ assessments based on psychometric tests. The lack of an established objective assessment for people with CP may lead to over- or underestimate their real intellectual ability. Brief and easily applied psychometric tests can provide a solution to this problem, since most intelligence scales usually involve long administration times and both verbal and manipulative responses, which hinders its application in people with CP. The use of inaccurate tools can lead to confuse the inability to respond to a task with a low intellectual performance, with the diagnostic consequences derived therefrom (Braden & Elliott, 2003; Sabbadini, Bonanni, Carlesimo, & Caltagirone, 2001). As some authors highlight, there is a need to identify which psychometric measures are more desirable in order to obtain standardized assessments for this population (Sabbadini et al., 2001; Schiariti et al., 2014; Sherwell et al., 2014). Moreover, it should be noted that most studies focus only on children, being still scarce the attention to intellectual performance in adults with CP. As some authors point, it is important to adopt a lifelong perspective on the disorder (Colver, Fairhurst, & Pharoah, 2014; Haak, Lenski, Hidecker, Li, & Paneth, 2009).

1.1. Intellectual ability assessments used in cerebral palsy

A recent systematic review has examined the most used IQ assessments for children and young adults with CP and concluded that all identified assessments are potentially suitable for cases with minimal motor involvement (GMFCS I-III and MACS I), but not for those cases with greater motor involvement (GMFCS IV-V or MACS II-V) or with communication impairments (Yin Foo, Guppy, & Johnston, 2013). However, some studies have shown that the typical and widely used Wechsler's intelligence scales not only cannot be carried out by severe cases (GMFCS IV-V, MACS IV-V or quadriplegia), but also that milder cases have problems completing them (Sherwell et al., 2014; Sigurdardottir et al., 2008). Specifically, in the study of Sherwell et al. (2014) 25% and 17% of GMFCS and MACS II and 43% and 47% of GMFCS and MACS III failed to complete the test. The use of shorter batteries, such as Stanford-Binet Intelligence Scales or Leiter

International Performance Scale, increases the percentage of subjects who complete them, but still remains around a 20% who cannot (El-Tallawy et al., 2014; Majnemer et al., 2013; Majnemer, Shevell, Law, Poulin, & Rosenbaum, 2012). Intelligence assessments composed of one single task, or a smaller number of subtests, using non-verbal assessments with minimal motor requirements suitably accommodate the heterogeneous range of impairments experienced by this population. This could provide a standardized tool that would assess all subjects objectively and equally, minimizing the interference of motor and communicative impairments, which could be used both in clinical practice and in research. The most widely used tests in the field of CP that meet these criteria are the Raven's Coloured Progressive Matrices (RCPM) and the Peabody Picture Vocabulary Test (PPVT).

The RCPM aims to measure the ability to deduct relationships (Raven, Court, & Seisdedos Cubero, 2001) and its use is recommended for people with physical disabilities, aphasia, deafness or CP (Strauss, Spreen, & Sherman, 2006). The PPVT is an old and very commonly used standardized test of receptive vocabulary (Strauss et al., 2006) considered as a screening test of intellectual functioning (Dunn & Dunn, 1997). Because of its non-verbal aspect it is appropriate for those cases with severe expressive language impairment (Strauss et al., 2006) and it has been widely used in CP (Yin Foo et al., 2013). Recently, the Wechsler Nonverbal Scale of Ability (WNV) (Wechsler & Naglieri, 2006) has been created with the aim of measuring general intellectual ability but eliminating or minimizing the verbal content. While WNV has been tested in special groups, such as language disorders, hearing difficulties or intellectual disability, there is still no study that has applied it to people with CP. In addition, to date few studies have compared the RCPM and the PPVT in people with CP and very few indications exist of which one may be more suitable. Only three studies have provided correlations between both tests, with a result of medium (r=.41) (Nicholson, 1970; Peeters, Verhoeven, van Balkom, & de Moor, 2008) to large effect (r=.734) (Pueyo, Junqué, Vendrell, Narberhaus, & Segarra, 2008). However, these observed relationships do not automatically imply that there is good concordance between these two methods (Bland & Altman, 1986). Therefore, more studies comparing the IQ scores of these different intelligence tests are needed in an attempt to know how they score, how they classify subjects' cognitive ability and if they could be interchangeable or not.

1.2. Specific cognitive functions and intellectual ability

General intelligence describes the strong common core that specific cognitive tests share and is responsible for much of their predictive validity (Deary, Penke, & Johnson, 2010). RCPM has been associated with perceptual tasks (Courchesne, Meilleur, Poulin-Lord, Dawson, & Soulières, 2015; Cronin-Golomb & Braun, 1997), language (Baldo, Bunge, Wilson, & Dronkers, 2010; Goharpey, Crewther, & Crewther, 2013) and attentional measures (Blake, McKinney, Treece, Lee, & Lincoln, 2002; Maeshima et al., 2002) in some neurological conditions. The PPVT has been linked to several language tests and to some executive functions in healthy and clinical subjects (Strauss et al., 2006). The only study to date that has applied the WNV has found relationship with several attentional measures in healthy children (Hurford et al., 2014). However, the relationship between IQ measures and specific cognitive functions has been poorly studied in CP, being executive functions and long-term memory undervalued. To identify which easy-to-administer test provide as much information as possible on the different cognitive functions would satisfy this need that emerges both in clinical settings and in the research field of CP.

1.3. Neuroimage and intellectual ability

General intellectual ability has also been considered as a reflection of the state and functionality of the brain. Many studies have analysed the relationship between total brain volume and intelligence measures. With the advance of magnetic resonance imaging (MRI) techniques it has become possible to extend this study to different brain tissues and specific brain regions. In healthy subjects, higher IQ scores have been associated with larger total, grey and white matter volumes, specifically in dorsolateral prefrontal cortex, anterior cingulate cortex, parietal, temporal and occipital lobes (Deary et al., 2010; Jung & Haier, 2007). In lesion mapping studies, areas of the frontal, parietal and temporal lobes have also been associated with intellectual performance (Baldo et al., 2010; Gläscher et al., 2009). Recently, an increasing amount of evidence shows that subcortical structures such as basal ganglia play a relevant role in intelligence (Burgaleta et al., 2014; Rhein et al., 2014; Sandman et al., 2014). These structures, together with the thalamus, are important in CP. Bilateral lesions of the thalamus and basal ganglia have been reported and associated with the severity of cognitive and motor impairment in this population (Kate Himmelmann & Uvebrant, 2011; Krägeloh-Mann et al., 2002). To date, there are still few studies that have tested the intelligence-brain relationship in people with CP. Most of them have focused on the qualitative connection between the pattern and extent of brain injury and the frequency of intellectual disability (Fedrizzi et al., 1996; Kate Himmelmann & Uvebrant, 2011; Riva et al., 2012; Schatz, Craft, Koby, & Park, 1997). To find out the relationship between these three tests and different quantitative measures of brain volume in CP could indicate whether they are sensitive to the state of the brain, which areas are involved in and if there are differences between them or not.

1.4. Research objective

In conclusion, although the SCPE and the ICF Core Sets for CP highlight ‘what’ to measure, they do not address ‘how’ and poor consensus exists on the best tool to assess intellectual function in CP. Therefore, the aim of this study is to compare three intelligence tests suitable for the heterogeneous range of people with CP, including cases with severe motor impairment, in order to determine which may be more appropriate to use. In this sense, we will include two widely used single-task tests in CP (RCPM and PPVT) and a third one which is a newly-adapted version of the Wechsler’s scales (WNV), not yet applied to this group. Initially, we will compare the IQ measures obtained with the three tests and their degree of concordance in classifying the subjects’ cognitive ability, and then we will study the relationship between these measures and both the performance in specific cognitive functions and brain volume data. Having such comprehensive information about these tests could indicate which one(s) may be more appropriate to use in this population.

2. Methods

2.1. Sample

This study is part of a larger work examining cognitive performance in people with dyskinetic CP. The study was approved by the University of Barcelona’s (CBUB) Institutional Ethics Committee, Institutional Review Board (IRB 00003099, assurance number: FWA00004225; http://www.ub.edu/recerca/comissiobioetica.htm). The research was conducted in accordance with the Helsinki Declaration. Participants were mainly recruited (83%) from the Hospital Vall d’Hebron and the Hospital Sant Joan de Déu in Barcelona, Spain. The sample was completed with some patients from previous studies (4%) and from other clinical services for people with CP (13%), in which some of the subjects from the hospital were also attended. The inclusion criteria were: 1) clinical diagnosis of dyskinetic CP with bilateral motor involvement; 2) being able to understand instructions assessed by means of the receptive part of the Screening Test of Spanish Grammar (Toronto, 1973); 3) age between 6 and 60 years. Exclusion criteria were: 1) identified hearing abnormalities or severe visual difficulties that precluded neuropsychological assessment; 2) lack of an intelligible 'yes'/'no' response system in any communicative modality (e.g. vocalizations, movement of head, facial miming or gestures with other parts of the body). Forty-seven subjects met the inclusion and exclusion criteria but three of them dropped out the study. The final sample consisted of forty-four subjects (sixteen children and twenty-eight adults). Perinatal information was obtained from medical reports and complemented with information provided by participants or their relatives, if needed. According to this information, the main perinatal antecedents were: signs of perinatal asphyxia in 34 cases (based on medical reports and/or MRI pattern compatible with hypoxic-ischemic injury), signs of vascular events in 1 case, kernicterus in 2 cases, congenital brain malformations in 2 cases and signs of infection in 1 case. In 4 subjects the perinatal antecedents were unknown. Motor function impairment was assessed following the recommendations of the SCPE: Gross Motor Function Classification System (GMFCS) for the lower limbs function (Palisano, Rosenbaum, Bartlett, & Livingston, 2008) and Bimanual Fine Motor Function (BFMF) (Elvrum et al., 2014) and the Manual Ability Classification System (MACS)(Eliasson et al., 2006) for the upper limbs. Daily communication abilities were assessed by means of the Communication Function Classification System (CFCS) (Hidecker et al., 2011). An informed consent was obtained from all participants or their parents. Demographic and clinical data of the sample is shown in table 1. Demographic and clinical data separated by age can be found as online supplementary material.

2.2. Neuropsychological assessment

As recommended by Sabbadini et al. (2001) subjects were encouraged to answer in an autonomous way. Therefore, the subjects were allowed to use the response technique that best fitted to their degree of disability and the communication devices they normally used. In this manner, subjects answered the tests orally and/or pointing (with finger or hand, an adapted pointer on the head or fixing the gaze if the original distribution of multiple choices allowed it). To access the computer tests, a mouse/joystick (controlled by hand or with the chin) and two switches (pressed by hand, cheek or head) were used. In those cases in which autonomous response was not possible, the examiner indicated the various response alternatives while asking the patient if it was his/her choice (Sabbadini et al., 2001). These subjects answered "yes" or "no" by means of vocalizations, movement of head, facial miming or gestures with other parts of the body, as they usually did.

2.2.1. Intelligence measures

Three different measures of intelligence were used (RCPM, PPVT-III and WNV). The RCPM test consists of 36 items, grouped into three sets of 12 items of increasing difficulty within each set. The problems are printed with a bright background colour, which makes the test more appealing to attract the patient's attention. Each item contains a pattern problem with one part removed and the participant has to choose which of the six alternatives completes the pattern. Each correct answer is scored one point, until the maximum score of 36. The raw scores were converted into IQ scores using normative data of the test for children and normative data provided by Measso et al. (1993) for adults. For adolescents, we used a linear interpolation to estimate which mean and standard deviation needed to be used to calculate IQ scores.

The PPVT-III adaptation for Spanish population consists of 192 items sorted by level of difficulty and grouped into sets of 12 items. Each element consists of 4 illustrations in black and white and the task of the examinee is to select the image that best represents the meaning of the word provided orally by the examiner. For each correct answer one point is scored. Raw scores are converted into IQ scores using normative data provided by the test.

The WNV provides a brief version composed of two subtests: Matrices and Spatial Span. The subtest of Matrices is similar to the RCPM. It contains 41 items of increasing difficulty in which the subject must discover the logic of a pattern problem and select the portion that has been removed among 4 or 5 different options. The subtest of Spatial Span comprises two tasks: the direct order condition, in which the examiner points out some cubes and examinee must indicate the same cubes in the same order, and inverse order condition, in which the examinee must indicate the cubes in the reverse order. In both conditions the length of the sequences is gradually increased. The Spatial Span subtest was adapted in different ways for those cases that were not able to point the finger, either by using the hand, an adapted pointer on the head or fixing the gaze. These two subtests allow obtaining an IQ score using normative data provided by the test.

Mean and standard deviation for each intelligence test is shown in table 1. Intelligence measures separated by age can be found as online supplementary material. For all the IQ assessments, two cut-off points were established: one standard deviation below the mean (IQ<85), including those subjects with low-average functioning, borderline intellectual functioning and intellectual disability, and two standard deviations below the mean (IQ<70), including only subjects with intellectual disability.

2.2.2. *Specific cognitive assessment*

Participants completed a broad neuropsychological assessment. The selected tests are extensively used in neuropsychological assessment and most of them allow a nonverbal response. Cognitive measures were grouped into 5 cognitive domains: attention, language, visuoperception, memory and executive functions, grouping those tests that measure different aspects of the same cognitive function (Lezak, 2012; Strauss et al., 2006).

Attention was assessed using the Digit Span subtest of the Wechsler Intelligence Scale for Children-4th edition or Wechsler Adult Intelligence Scale-3rd edition (Wechsler, 1997, 2003) and Spatial Span subtest of WNV (Wechsler & Naglieri, 2006). In patients with a non-verbal communication, response to Digit Span subtest was given pointing to written numbers placed in front of them. Subjects were required to point out using their usual systems to enable them to answer as fast as possible. Every time the numbers were dictated, the written numbers were removed from the visual field of the subject to prevent that the response was based on visual rather than on verbal component. To examine language functions and to assess comprehension, we administered the receptive part of the Screening Test of Spanish Grammar (Toronto, 1973) and Peabody Picture Vocabulary Test-3rd edition (Dunn & Dunn, 1997). Benton's Facial Recognition Test and Benton's Judgment of Line Orientation Test were used to assess visuoperceptual abilities (Benton, 1994). For memory assessment we selected the Pattern Recognition Memory subtest of the Cambridge Neuropsychological Test Automated Battery (CANTAB), which is a visual recognition memory test that provides a measure of short and long term memory (Cambridge Cognition, 2006). Finally, executive functions were assessed with three tests. The 64-item computerized version of the Wisconsin Card Sorting Test (WCST) was used as a measure of cognitive flexibility (Kongs, S. K., Thompson L. L., Iverson, G. L., & Heaton, 2000). Perseverative errors score was used. To assess planning and problem solving ability, Stockings of Cambridge subtest of CANTAB was applied (Cambridge Cognition, 2006). The Balloon Analogue Risk Task youth version (BART-Y) was used as a measure of risk taking behaviour (Lejuez et al., 2007).

In all tests, participants' raw scores were normalized to *z* scores using the normative data provided by each test, except for the delayed recall of the Pattern Recognition Memory (Roque, Teixeira, Zachi, & Ventura, 2011). Composite *z* scores for each participant in the five cognitive domains were calculated by averaging the *z* scores of all tests within that domain [table 2].

2.3. Neuroimaging

2.3.1. MRI acquisition

MRI images were performed on a Siemens Magnetom TRIO 3,0 Tesla scanner (Erlangen, Germany) from the Hospital Universitari Vall d'Hebron (Barcelona, Spain) at the same time as neuropsychological assessments were made. High-resolution three-dimensional T1-weighted images were acquired in the sagittal plane with a MPRAGE sequence (TR/TE 1900/2.46 ms; TI 900 ms; flip angle 9º; 320 x 307 matrix and voxel size 0.7 mm x 0.7 mm x 1 mm). In order to minimize movement during MRI, a muscle relaxant was administered in twelve patients and four were sedated. Good quality images were obtained in thirty-three cases, but in the statistical analysis we only included those thirty who had completed the three intelligence tests.

2.3.2. Image processing

All images were processed with Statistical Parametric Mapping 8 software (SPM8; http://www.fil.ion.ucl.ac.uk/spm/software/spm8/) using the voxel-based morphometry toolbox version 8 (VBM8; http://dbm.neuro.uni-jena.de/vbm/). Image processing included the following steps: 1) Antero-posterior commissure alignment and image reorienting of all images; 2) Tissue probability maps were generated from our own sample using the Template-O-Matic toolbox (TOM8; https://irc.cchmc.org/software/tom.php) in order to improve the segmentation and normalization precision (Wilke, Holland, Altaye, & Gaser, 2008); 3) The images were segmented into grey matter, white matter and cerebrospinal fluid segments. Total intracranial volume was computed as the sum of these three cerebral tissues; 4) Tissue segments were normalized to the tissue probability maps created before using an affine transformation and it was used to create customized DARTEL templates in MNI space (Ashburner, 2007); 5) The affine-registered grey and white matter segments were warped to this average template using the high-dimensional DARTEL approach and non-linear modulated to ensure relative differences in regional volumes correcting for individual brain size; 6) Data quality was checked in order to identify possible artefacts and failed segmentation or normalization; 7) Finally, maps were smoothed with an 8 x 8 x 8 mm^3 full-width-half-maximum filter (FWHM) kernel.

2.4. Statistical analysis

All statistical analysis were computed using IBM SPSS Statistics version 22. The significance level was set at p-value less than .05. In order to compare means for the three IQ measures Friedman's ANOVA for repeated measures was applied. Post hoc tests were conducted using Wilcoxon's signed-rank test. For this analysis, a Bonferroni correction was applied and level of significance was set at .0167. To determine the degree of agreement in classifying the subjects' cognitive ability a Cohen's Kappa (κ) was run establishing two different cut-off points: IQ<85 (including low-average functioning, borderline intellectual functioning and intellectual disability) and IQ<70 (including only intellectual disability). Guidelines from Landis & Koch (1977) were taken into account for the interpretation of the results. A multiple linear regression model (stepwise method) was performed to identify which specific cognitive functions contributed most to explain different IQ scores. To avoid overlapping of some tests, the attention domain included only the Digit Span subtest when WNV was analysed, and the language domain included only the Screening Test of Spanish Grammar when PPVT-III was analysed. Assumptions of normality, independence and homoscedasticity of residuals and multicollinearity were checked. Cook's distance was used to check for influential data points. For neuroimaging analysis, the relationship between the IQ measures and global brain volumes (total, grey and whiter matter) was analysed by Pearson's correlation coefficient, correcting for total intracranial volume measures and controlling for the effect of age and gender. Voxel-based morphometry (VBM) method (Ashburner & Friston, 2000) was used to analyse which specific brain regions were related to the intelligence measures. For this purpose, a multiple regression analysis was conducted including age and gender as covariates in each statistical design and only clusters larger than 50 voxels were considered. For the VBM analysis focused in basal ganglia and thalamus, a region-of-interest was applied by means of the WFU PickAtlas (version 3.0) (http://fmri.wfubmc.edu/software/pickatlas) and only clusters larger than 20 voxels were considered. For statistical purposes, we used a threshold corrected at the family-wise error level (p<.05).

3. Results

All the subjects completed the RCPM and PPVT-III whilst seven subjects failed to complete the WNV, mainly due to their inability to perform the Spatial Span subtest because of motor impairment. Comparing IQ measures, results showed a significant difference in the mean scores of the three tests ($\chi^2_F(2)=28.86$, $p<.001$). As shown in table 1, the highest mean IQ score is observed with RCPM, closely followed by WNV, and the lowest mean IQ score is provided by PPVT-III. Pairwise comparisons showed a medium effect size between RCPM and the other two tests (WNV, z=-3.313, p=.001, r=-.31; PPVT-III, z=-4.687, p<.001, r=-.44) and a small effect size among WNV and PPVT-III (z=-2.845, p=.004, r=-.27).

The degree of agreement in classifying the subjects' cognitive ability changed depending on the cut-off established. When the cut-off was set at IQ<85 there was an almost perfect agreement between RCPM and WNV (κ=.83, p<.001) and a substantial agreement between PPVT-III and the other two tests (RCPM, κ=.61, p<.001; WNV, κ=.69, p<.001). When the cut-off point was set in IQ<70 the degree of agreement between tests declined. The highest agreement was observed again between RCPM and WNV, which was moderate (κ=.41, p=.002). Between the PPVT-III and the other two tests the degree of agreement observed was fair (RCPM, κ=.21, p=.025; WNV, κ=.34, p=.021). Figure 1 shows the percentage of cases included in each group according to the IQ level obtained with each test.

Regression analysis was performed to identify which of these specific cognitive functions contributed most to explain different IQ scores. As shown in table 3, the best predictors of RCPM

IQ scores were attention, visuoperception and language (model 3); for PPVT-III IQ scores the best predictor was visuoperception (model 1); and for WNV IQ scores were visuoperception and attention (model 2). All predictor variables had a positive effect on IQ scores.

Regarding the neuroimaging analysis, the relationship between IQ measures and global brain volumes was tested first. Neither age nor gender shows relation to IQ measures. Age shows a negative correlation with grey matter volume ($r=-.835$, $p<.001$) and a positive correlation with white matter volume ($r=.650$, $p<.001$), and gender shows an effect on total intracranial volume ($r=.631$, $p<.001$). Therefore, a partial correlation was made controlling for the effect of these two variables. As shown in table 4, all three tests correlated positively with total intracranial volume and white matter volume, and negatively with grey matter volume. Furthermore, we analysed which specific brain regions were related to intelligence measures. VBM analysis showed only significant results in white matter areas with RCPM and WNV [table 5, figure 2]. Specifically, IQ measures obtained with RCPM were positively correlated with left superior longitudinal fasciculus, extending to corticospinal tract and superior corona radiata; and areas in the right hemisphere, including retrolenticular part of the internal capsule, cingulum, superior corona radiata and superior longitudinal fasciculus. In relation to WNV, significant positive correlations were observed in right hemisphere including retrolenticular part of the internal capsule, extending also to the posterior limb, and cingulum; and in left superior longitudinal fasciculus, extending to superior corona radiata and white matter in the pre-central and post-central gyrus. Regarding PPVT-III, the results did not reach statistical significance although uncorrected results ($p<.001$) showed a significant cluster including the left superior longitudinal fasciculus.

Correlation analyses between grey matter areas and IQ measures did not reach statistical significance at the corrected level. However, when the statistical maps were displayed uncorrected for multiple comparisons ($p<.001$) and based on peak level some areas were observed as being relevant. Specifically, positive correlations were found with both RCPM and WNV in the left grey matter of the pre-central and post-central gyrus, being larger in case of RCPM. Negative correlations were observed between RCPM and right temporal fusiform cortex; WNV and right temporal fusiform cortex and subcallosal cortex; and between PPVT and the temporooccipital part of the left middle temporal gyrus.

Finally, VBM analysis focused in basal ganglia and thalamus showed significant positive correlations with RCPM and WNV [table 6, figure 3]. Specifically, both tests were related to thalamus and putamen. Moreover, RCPM was also related to caudate nuclei. The results of the PPVT-III did not reach statistical significance.

4. Discussion

All subjects included in the study, even severe cases, were able to complete the two single-task tests, RCPM and PPVT-III. This suggests that both tools are suitable regardless of motor severity and verbal communication impairments. Although still a 16% of cases cannot complete the Spatial Span subtest of the WNV, particularly cases with greater motor impairment, this percentage is lower than those observed with other versions of Wechsler's scales or other intelligence tests (Sherwell et al., 2014; Sigurdardottir & Vik, 2011).

Our results show that there are differences between the three IQ scores. The highest mean IQ score is observed with RCPM, closely followed by WNV. Our results coincide with the longitudinal study of Smits et al. (2011) in which the RCPM IQ scores of children with dyskinetic CP ranged from 83 to 93. The PPVT-III shows the lowest mean IQ score. This result is also consistent with previous findings in which all subjects with bilateral dyskinetic CP had an impaired receptive vocabulary measured with the PPVT-R (Pueyo, Junque, Vendrell, Narberhaus, & Segarra, 2009), and the two studies which found an average verbal IQ of 63 and 73 in children with dyskinetic CP (Sigurdardottir & Vik, 2011; Sigurdardottir et al., 2008). Such differences observed between PPVT-III and the other two tests (RCPM and WNV) may be related to the Cattell-Horn concepts of fluid and crystallized intelligence (McGrew, 2009). Fluid intelligence is defined as the ability to solve abstract problems in novel situations, while crystallized ability represents knowledge and skills acquired through education and experience. The Raven's progressive matrices are regarded as a quintessential measure of fluid intelligence, while vocabulary tests, like PPVT-III, are considered good measures of crystallized intelligence (Strauss et al., 2006). The WNV, which includes a matrix reasoning task, can be considered a measure with a high load of fluid intelligence. This could explain the greater degree of agreement between RCPM and WNV, as two measures of the same factor, and the large differences observed between RCPM and PPVT-III, as representative measures of these two different factors involved in general intelligence.

The significantly low mean score of the PPVT-III may be due to the fact that this test evaluates the knowledge acquired through either formal education or informal everyday experiences, and people with CP, in many cases, require special education and have a lower level of social involvement than their peers (Imms, 2008). As for education, in healthy subjects crystallized intelligence has shown a strong association with educational attainment (Kaufman, Kaufman, Liu, & Johnson, 2009). In CP, Ito and colleagues found that after attending elementary school verbal IQ increased (Ito, Araki, Tanaka, Tasaki, & Cho, 1997). Regarding informal everyday experiences, it has been shown in people with intellectual disability that life experiences exert a significant effect on the crystallized component of intelligence (measured with the PPVT) but not on measures of fluid intelligence, such as RCPM (Facon, Bollengier, & Grubar, 1993; Facon, Facon-Bollengier, & Grubar, 2002; Facon & Facon-Bollengier, 1999). In the same line of reasoning, motor difficulties that inhibit speech ability in CP affect the frequency and the quality of communicative acts: people with CP tend to adopt a respondent role, take fewer turns in conversation or produce many yes/no answers and seldom ask questions (Pirila et al., 2007; Reed & Warner-Rogers, 2008). These speech and communicative impairments can consequently lead to a poorer vocabulary comprehension or a delay in their acquisition (Bishop, Brown, & Robson, 1990; Pueyo et al., 2013). Taken together, this data show that the low scores obtained with the PPVT-III in people with CP may reflect a limited access to different learning experiences and specifically communicative experiences that promote learning vocabulary, rather than providing an accurate estimate of their overall cognitive performance. The fact of using a vocabulary test to assess general cognitive performance in a population with speech and communicative impairments may result in an underestimate of their intellectual abilities.

The present results also show that the level of agreement between tests in classifying the subjects' cognitive ability changes depending on the cut-off established, being almost perfect or substantial in classifying subjects as IQ below 85 and moderate to fair in classifying them as IQ below 70. This indicates that there are some cases where the performance is impaired (less than 70) in one test and it is categorized as low-average or borderline level in another (from 70 to 84). Again, the highest degree of agreement is observed between RCPM and WNV, being lower with PPVT-III. So, the three tests have shown a good agreement identifying those subjects who did not have a completely average intellectual performance. Thus, all three could be used as an exclusion or classification criteria on scientific studies, or to identify those who will require some specialized intervention at clinical level. However, when the objective is to establish a diagnosis of intellectual disability we have to be careful because the results show that the three tests are not interchangeable. Moreover, it is important to keep in mind that intellectual disability is characterized by low intellectual functioning as well as problems in adaptive behaviour, so clinical decisions should not rely on single IQ assessments but should also consider adaptive functioning.

Regarding the relationship between IQ measures and performance in specific cognitive functions, regression analysis show that the degree of contribution of these cognitive functions differs between tests. Regarding PPVT-III, the best predictor of IQ scores is visuoperception. It was expected that language was the most significant cognitive function in the PPVT-III since this is a test that measures vocabulary. In this case, the language domain is only formed by the receptive part of the Screening Test of Spanish Grammar, which may be insufficient to assess language. This poor representation of language domain could be the reason why the visuoperceptual skills become more relevant. Moreover, the amount of variation that is accounted for by the model is the lowest compared to the other IQ tests (R^2=.437), remaining unexplained more than 56% of the variation (table 3). Once again, RCPM and WNV exhibit similar results and both are related to attention and visuoperception functions. This was expected as both tests have been previously associated with attentional measures (Blake et al., 2002; Hurford et al., 2014; Maeshima et al., 2002) and the involvement of visuoperceptual functions in both tasks has been well established (Lezak, 2004; Strauss et al., 2006). Furthermore, the results show that RCPM is additionally linked to language performance. This fits with other studies that have found evidences that language has a critical role for the non-verbal reasoning measured with RCPM. They concluded that RCPM can be considered as a measure of non-verbal, but also as a measure of verbal functions (Baldo et al., 2010; Goharpey et al., 2013; Pueyo et al., 2008). Regarding executive functions, they were not significantly associated with measures of intelligence in any model of the regression analysis. This lack of results may be due to the fact that not all components of executive functions have been equally associated with intelligence (Au et al., 2015; Toplak, Sorge, Benoit, West, & Stanovich, 2010). There are also no significant results as to memory. We were expecting an association between memory and IQ scores of WNV because it includes a task with a remarkable component of short-term and working memory. However, memory domain included measures of long term visual memory and these differences in the type of memory may explain the lack of results. The fact that regression models do not include executive functions and memory as significant factors means that these variables do not add additional independent information to the statistical models.

At this point, it is important to consider the fact that the sample comprises only subjects with dyskinetic CP, which generally differ from spastic in functional profiles and neurological substrates. Dyskinetic CP has been associated with severe forms (Himmelmann et al., 2009), leading to a greater number of cases with bilateral involvement. Given that this study only includes people with bilateral CP, the present results cannot be generalized to milder cases, such as unilateral CP, who suffer less and milder impairments and can also perform more comprehensive intelligence assessments. However, the results might be applicable in cases with more severe motor disturbances, such as bilateral forms, which also manifest difficulties answering typical intelligence scales and who have shown similar alterations in the overall cognitive performance (Hagberg & Olow, 1975; Krageloh-Mann et al., 1993; Miller & Rosenfeld, 1952; Pueyo, Junque, & Vendrell, 2003; Sigurdardottir et al., 2008). In addition, the application of new neuroimaging techniques has allowed to observe that bilateral CP forms show a similar neural substrate, with the involvement of both deep grey matter (basal ganglia and thalamus) and white matter (Krägeloh-Mann et al., 2002; Yoshida et al., 2011). On the other hand, the lower frequency and also the great severity of dyskinetic CP may explain why this type is less researched and why the particular neuropsychological features and needs of individuals with dyskinetic CP are less known. The few cognitive studies that have found differences between the two types of CP certainly point to a better visuospatial performance in individuals with dyskinetic CP (Dorman, 1987; Kozeis et al., 2007; Miller & Rosenfeld, 1952; Pueyo et al., 2003, 2009; Stiers et al., 2002). However, these studies included very few subjects with dyskinetic CP which could be the reason why in the recent systematic review by Ego et al. (2015) visuoperceptual impairments have not been significantly related to any CP type. If further studies with larger sample sizes representing all types of CP proved such differences, it would be expected to play out differently in performance to the detriment of those subjects with poorer visuoperceptual skills. Nevertheless, the use of the test that our results suggest more appropriate, RCPM, may be adequate for those subjects with visuoperceptual disturbances since only one third of the items are predominantly visuospatial (Lezak, 2012; Strauss et al., 2006b). In the rest, the task shifts from pattern completion to reasoning by analogy. These differences in the nature of the items suggest that RCPM assess a complex of cognitive skills that go beyond visuoperceptual skills. Moreover, it has been observed that modifying the item presentation format of RCPM (enlarging considerably the stimuli) not involve a significant change in the performance of a group of children with CP (Warschausky et al., 2011). Finally, it is noteworthy that RCPM has shown similar results to the current ones in a sample mainly formed by bilateral spastic CP (Pueyo et al., 2008b), which strengthens the idea that these findings might be applicable to all bilateral forms of CP.

Neuroimaging analysis showed that all tests correlated positively with total intracranial volume and white matter volume, as expected, but negatively with grey matter volume. These results may be linked to the dynamic relationship between intelligence and brain development which experiences regionally specific, age-dependent variations. In healthy subjects, grey matter volume increases at early ages followed by sustained loss starting around puberty, and white matter volume tend to increase in a more linear and less variant manner across regions and ages. The maturational process of grey matter loss begins first in dorsal parietal cortices, particularly the primary sensorimotor areas, and then spreads over the frontal, parietal, occipital, and finally the temporal cortex (Gogtay & Giedd, 2004). Intelligence is related to these dynamic properties of cortical maturation, as predominantly negative correlations between IQ and cortical thickness in the early healthy childhood progress to positive correlations as brain regions mature through adolescence and young adulthood (Shaw et al., 2006). As IQ has been found negatively related to grey matter volume in our sample, our results could be interpreted as an indication of a disturbance in cortical organization and/or retardation in the maturational processes of some cortical grey matter areas. Specifically, VBM grey matter results, though uncorrected, have shown positive correlations in frontal and parietal areas, which mature earlier, and negative correlations in some areas of the cortex in the temporal lobe, which shows a characteristic late maturation pattern. Other studies in pathological samples have also described negative correlations between temporal areas (fusiform gyrus and posterior temporal cortex) and intellectual performance (Baglio et al., 2014; Isaacs et al., 2004). The wide age range of our sample may also diminish the possibility of finding other significant areas in which age-associated differences could be relevant. The lack of a control group makes it difficult to test this hypothesis, although this also goes beyond the scope of our study. In the same manner, with a control group could be tested if the presence of compensatory white matter mechanisms might be counterbalancing these grey matter dysfunctions.

Regarding VBM analysis of white matter, significant results were only observed with RCPM and WNV IQ measures. Both tests show a positive relationship with right parts of the internal capsule and cingulum, and left parts of the superior longitudinal fasciculus and superior corona radiata. Beyond the similarities, the RCPM presents a relationship with larger areas and bilateral superior regions such as longitudinal fasciculus and corona radiata. Recent studies using diffusion tensor imaging (DTI) have found that the extent of brain damage in dyskinetic CP tends to be more diffuse than previously thought, not only including deep grey matter but also white matter regions like internal capsule, corticospinal tract, posterior thalamic radiation, cingulum or superior longitudinal fasciculus (Yoshida et al., 2011). Most of these areas coincide with our findings indicating that these tests, and specifically the RCPM, are sensitive to the state of white matter areas typically affected in CP. Our results also match with the recent study of Rai et al. (2013) which finds correlations between integrity of different white matter fibers including parts of the internal capsule, corona radiata and posterior areas, with measures of intelligence in children with CP. Another recent study in CP suggest that network-level structural disruptions, with specific vulnerability of long range fiber tracts, may contribute to the neurocognitive impairments observed in this population (Englander et al., 2013). On the other hand, current research in the field of intelligence has emphasized the crucial role of white matter tracts underlying parieto-frontal association cortices, such as the superior longitudinal fasciculus (Jung & Haier, 2007). Accordingly, our results also suggest that the state of white matter may have an important role in intellectual performance in people with CP.

Finally, VBM analysis focused in basal ganglia and thalamus showed that RCPM and WNV IQ measures were positively correlated to thalamus and putamen. RCPM was also related to caudate nuclei. First, these results indicate that these tests are also susceptible to the main neurological impairment suffered by these subjects: damage to basal ganglia and thalamus (Bax, Tydeman, & Flodmark, 2006; Kate Himmelmann & Uvebrant, 2011; Robinson et al., 2009). Secondly, it is remarkable that RCPM is also associated with the caudate nuclei, which is highly connected to the prefrontal cortex and is considered to be involved in solving novel, abstract problems; whereas the WNV is more associated with thalamus, which has been linked to visuospatial processing and spatial working memory (Burgaleta et al., 2014; Sandman et al., 2014). Moreover, our results are consistent with those of Burgaleta et al. (2014) who found that fluid intelligence was associated with the right basal ganglia and thalamus in a sample of healthy subjects.

Regarding PPVT-III, previous studies have found positive correlations with brain volume in healthy subjects, specifically in frontal and temporal lobes (Wells et al., 2009), and with the integrity of underlying white matter fibers (Mullen et al., 2011). Although uncorrected for multiple comparisons, our results are consistent with these findings, identifying relationship with frontal and temporal structures. The lack of results of the PPVT-III with basal ganglia and thalamus agrees with studies that found that verbal aspects were not correlated with basal ganglia volumes (Burgaleta et al., 2014; Rhein et al., 2014).

4.1. Limitations

In addition to the limitation already mentioned in the discussion regarding the generalizability of the present results to the whole CP, the small sample size as well as the wide age range may also limit the statistical power of these findings. However, it is worth mentioning that there are no significant differences between children and adults on demographic, clinical and intelligence data (see online supplementary material). Differences were only observed in communication (CFCS) in the subsample included in neuroimaging study. Although the present study does not allow drawing specific conclusions about whether the tools compared work equally well for children and adults, the fact that there are no significant differences between the groups does allow extend the present results for all ages tested.

Secondly, the PPVT-III includes some limitations. One is that it sets a floor effect on the lower limit of IQ, with a minimum of 55, losing part of discriminative information between moderate and severe cases. This feature of the test may have influenced the results related to the performance on cognitive functions and neuroimaging data. When the new PPVT version becomes available for non-English speaking countries, it may offer a solution to this problem as it includes a greater range of standard scores and expands low levels of mental ability. Moreover, if further studies are performed using the new version, language domain should be assessed in a more comprehensive way than performed in this work. The other drawback is that there is evidence that PPVT tends to underestimate the cognitive performance in people with low incomes and/or minority groups. The lack of studies in Spanish population does not provide clear evidence of this limitation, but the same authors of the Spanish adaptation already reported lower IQ scores in those people living in rural areas, with low educational level and who attended public schools. These important constraints, together with the results of the present study, do not encourage using the PPVT-III as a proxy for intellect, especially in populations with speech and communicative impairments.

Third, most intelligence tests are validated with typically developing subjects, which raises doubts about whether the tests can be used similarly in atypical populations. However, several studies have shown that RCPM can be used with confidence when comparing people with and without intellectual disability (Facon, Magis, Nuchadee, & De Boeck, 2011; Facon & Nuchadee, 2010; Goharpey et al., 2013). Finally, the application of more precise techniques of analysis of different brain tissues, such as cortical thickness or DTI, could provide more specific information on brain-intelligence relationship in people with CP.

4.2. Conclusion

The findings of the present study bring some light on the decision of which test is more suitable when assessing general intellectual ability in people with bilateral CP. The results show that PPVT-III, even though it is a widely used test in CP, provides limited information on cognitive performance and brain status of this population, which is more likely to have trouble learning vocabulary due to communication difficulties, getting markedly lower IQ scores than the other two tests. The WNV, a test not yet applied in the field of CP, provides similar although reduced outcomes as RCPM, with the disadvantage that subjects with severe motor impairment have difficulties in performing one of its subtests, which requires some minor motor skills. Finally, the results suggest that RCPM is a tool that can provide comprehensive information on the cognitive performance of people with bilateral dyskinetic CP, comprising not only visual but also verbal functions. It is also sensitive to the structural state of the brain, including basal ganglia, thalamus and white matter areas such as superior longitudinal fasciculus. Moreover, the fact that this test is a single task with minimal motor and communicative requirements, suitably accommodate the heterogeneous range of impairments experienced by CP, even cases with severe motor impairment. All this makes the RCPM a standardized easy-to-administer tool with great potential both in clinical practice and in the field of research of bilateral CP.

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