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## ORIGINAL ARTICLE

# Developmental coordination disorder subtypes in children: An unsupervised clustering

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## Abstract

**Aim:** To identify subtypes of developmental coordination disorder (DCD) in children.

**Method:** Children with DCD diagnosed through comprehensive evaluation at Robert-Debré Children's University Hospital (Paris, France) were consecutively enrolled from February 2017 to March 2020. We performed an unsupervised hierarchical clustering based on principal component analysis using a large set of variables encompassing cognitive, motor, and visuospatial scores (Wechsler Intelligence Scale for Children, Fifth Edition; Developmental Neuropsychological Assessment, Second Edition; Movement Assessment Battery for Children, Second Edition).

**Results:** One hundred and sixty-four children with DCD were enrolled (median age 10 years 3 months; male:female ratio 5.56:1). We identified distinct subgroups with mixed visuospatial and gestural disorders, or with pure gestural disorders that predominantly impaired either speed or precision. Associated neurodevelopmental disorders, such as attention-deficit/hyperactivity disorder, did not influence the results of the clustering. Importantly, we identified a subgroup of children with marked visuospatial impairment with the lowest scores in almost all of the evaluated domains, and the poorest school performance.

**Interpretation:** The classification of DCD into distinct subgroups could be indicative of prognosis and provide critical information to guide patient management, taking into account the child's neuropsychological profile. Beyond this clinical interest, our findings also provide a relevant framework with homogeneous subgroups of patients for research on the pathogenesis of DCD.

Developmental coordination disorder (DCD) is defined by difficulties in acquiring age-appropriate motor skills, with consequences on school performance and activities of daily living. Children with DCD present various

combinations of slowness, motor imprecision, and/or alteration of visuospatial processing.<sup>1,2</sup> DCD is a heterogeneous disorder with various subtypes of motor dysfunction and disability.

**Abbreviations:** ADHD, attention-deficit/hyperactivity disorder; DCD, developmental coordination disorder; MABC-2, Movement Assessment Battery for Children, Second Edition; NDD, neurodevelopmental disorder; NEPSY-II, Developmental Neuropsychological Assessment, Second Edition; PCA, principal component analysis; WISC-V, Wechsler Intelligence Scale for Children, Fifth Edition.

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Attempts have been made to classify children with DCD using hierarchical clustering (Table S1), but findings are inconsistent and may not adequately reflect the clinical diversity of DCD.<sup>3</sup> Methodological reasons likely to account for these limitations include (1) relatively small sample sizes ( $n < 100$ );<sup>4–11</sup> (2) sample not restricted to children with DCD;<sup>6</sup> (3) absence of detailed evaluation of visuospatial<sup>6,7,9</sup> or fine motor functions;<sup>6</sup> or (4) failure to consider the influence of developmental comorbidities such as attention-deficit/hyperactivity disorder (ADHD).<sup>4–8,11,12</sup>

We performed unsupervised hierarchical clustering on a large sample of children with DCD using a set of evaluation measures based on current recommendations.<sup>13</sup> We paid special attention to visuospatial<sup>2,14</sup> and fine motor skills as these are critical parameters correlated with daily life and academic achievement.<sup>15,16</sup> On the basis of deep phenotyping, we hypothesized that clusters would emerge in an unsupervised manner, particularly around visual-spatial processing and fine motor skills, and that these clusters would correlate with school difficulties.

## METHOD

### Participants

We retrospectively analysed the medical records of consecutive children aged between 7 years and 16 years 11 months diagnosed with DCD at the Robert-Debré Children's University Hospital (Paris) between February 2017 and March 2020. Children were referred because of concerns about motor skills. DCD was diagnosed according to the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) criteria,<sup>17</sup> on the basis of clinical examination, assessment of problems in daily life (parent assessment using the Developmental Coordination Disorder Questionnaire and Movement Assessment Battery for Children, Second Edition [MABC-2] checklist), and the results of a comprehensive neuropsychological test battery, with a focus on visuospatial and fine motor skills. The presence of an associated neurodevelopmental disorder (NDD) was not an exclusion criterion. ADHD symptoms were screened according to DSM-5 criteria, and attention tests were added when appropriate. Comorbid NDDs were diagnosed according to DSM-5 criteria, including speech and language evaluation (reading disability), clinical and neuropsychological evaluation (ADHD), or multidisciplinary assessments (autism spectrum disorder) when appropriate.

The study was approved by the Robert-Debré Hospital Ethics Committee and the French National Data Protection Agency (Commission Nationale de l'Informatique et des Libertés). Parents provided written informed consent.

### Data collection

Clinical data were obtained from medical records. The Developmental Coordination Disorder Questionnaire and

### What this paper adds

- Unsupervised hierarchical clustering identified four subgroups of children with developmental coordination disorder.
- Two subgroups had combined visuospatial/gestural difficulties, and two had pure gestural disorders.
- Severe visuospatial impairment was associated with poor performance in most domains including school.
- Difficulties in the gestural-only clusters were predominantly either gestural precision or speed.

MABC-2 checklist were completed by the parents. All the children had received a detailed neurological examination with a focus on motor aspects. Brain magnetic resonance imaging (MRI) was available if there was a history of perinatal injury, head circumference less or more than 2 standard deviations (SD), or specific skin or ophthalmological abnormalities. Neuropsychological data included three sets of variables: (1) psychometric assessment by the Wechsler Intelligence Scale for Children, Fifth Edition (WISC-V); (2) motor skill, manual dexterity, visuospatial, and graphomotor tests by Developmental Neuropsychological Assessment, Second Edition (NEPSY-II) subtests, the Movement Assessment Battery for Children, Second Edition (MABC-2), the Rey-Osterrieth Complex Figure Test for children aged 9 years or more or Developmental Test of Visual Perception, Second Edition, spatial relation subtest for those younger than 9 years, and the Concise Evaluation Scale for Children's Handwriting (the French criterion standard test for dysgraphia); and (3) attention tests by the Test of Everyday Attention for Children and NEPSY-II subtests when clinically appropriate. WISC-V-defined strength (or weakness) was retained for a child when the difference between one of its subtest-scaled scores and the average of all its subtest-scaled scores was more than the 90th centile (or < 10th) of the distribution of this difference in the standardization population. We finally selected 36 non-redundant quantitative age-standardized neuropsychological variables from our routine set.

All but two variables (NEPSY-II route finding and visuomotor precision accuracy) were converted to ordinal scale variables) were collected with standard deviation (population-based age-related calibration data provided with each neuropsychological test).

A stratification based on functional domains is provided in Table 1. Following DCD recommendations,<sup>13</sup> we considered the psychometrics from the WISC-V associated with visuospatial and fine motor variables (a + b) to be the reference set of variables for the study (27 of the 36 variables). Sets limited to psychometric variables (a) or including attentional variables (a + b + c) were considered only for sensitivity analyses. Seventeen categorical developmental and clinical covariables were recorded for association analyses (Appendix S1).

**TABLE 1** Quantitative neuropsychological variable stratification for statistical analysis.**Quantitative neuropsychological variables = age-standardized scores ( $n = 36$ )****Variables 'a': psychometrics from the Wechsler Composite Intelligence Scale, Fifth Edition ( $n = 10$ )**

Similarities; vocabulary; matrix reasoning; figure weights; visual puzzles; block design; digit span; picture span; coding; symbol search

**Variables 'b': complementary 'fine motor and visuospatial' subtests ( $n = 17$ )**

|   |  |
|---|--|
| Fine motor subtests                                     | Visuospatial and constructive subtests               |
| Imitating hand positions (NEPSY-II)                     | Local design copying (NEPSY-II)                      |
| Fingertip tapping, preferred hand (NEPSY-II)            | Global design copying (NEPSY-II)                     |
| Fingertip tapping, other hand (NEPSY-II)                | Arrows (NEPSY-II)                                    |
| Visuomotor precision, speed (NEPSY-II)                  | Route finding (NEPSY-II)                             |
| Visuomotor precision, accuracy (NEPSY-II)               | Block construction (NEPSY-II)                        |
| Trail (MABC-2)  | Spatial relations (DTVP-2) for children <9 years old |
| Manual dexterity preferred hand and other hand (MABC-2) | ROCF $\geq 9$ years old                              |
| Bi-manual coordination (MABC-2)                         |  |
| Motor design copying (NEPSY-II)                         |  |
| BHK speed   |  |
| BHK quality   |  |

**Variables 'c': complementary 'attention and executive tests' ( $n = 9$ )**

|  |                                 |
|--|---------------------------------|
| TEA-Ch: selective attention: sky search (number of correct targets)            | NEPSY-II: auditory attention    |
| TEA-Ch: selective attention: sky search (time per target)                      | NEPSY-II: response set          |
| TEA-Ch: sustained attention, auditory task: score                              | NEPSY-II: inhibition naming     |
| TEA-Ch: divided attention: sky search dual task (combine sky search and score) | NEPSY-II: inhibition-inhibition |
|  | NEPSY-II: inhibition switching  |

Abbreviations: BHK, Concise Evaluation Scale for Children's Handwriting; DTVP-2, Developmental Test of Visual Perception, Second Edition; MABC-2, Movement Assessment Battery for Children, Second Edition; NEPSY-II, Developmental Neuropsychological Assessment, Second Edition; ROCF, Rey-Osterreith Complex Figure Test for Children; TEA-Ch, Test of Everyday Attention for Children.

## Statistical analysis

Analyses were performed in R (R Foundation for Statistical Computing, Vienna, Austria) with a 5% alpha risk corrected for multiple comparison by the false discovery rate (Benjamini-Hochberg correction) when appropriate.

## Principal component analysis and patient clustering: set-up and consistency analysis

First, we performed a principal component analysis (PCA)<sup>18</sup> based on the reference set of variables followed by a Ward's unsupervised hierarchical clustering<sup>19</sup> with the Duda-Hart stopping rule (pseudo- $F$  and pseudo- $t^2$  statistics to determine the optimal number of clusters) to cluster patients on the basis of this reference PCA. Unit variance scaling of the variables was used in the PCA.

As a consistency analysis for both the number and the limits of clusters, we confronted the PCA-based clustering with one driven by Gaussian mixture modelling<sup>20</sup> (see Appendix S1 for clustering details). As a sensitivity analysis of the set of variables, to determine whether removing or adding variables would change the classification, we also performed PCA-based clustering on the psychometric variables alone (a) and on an extended set of attentional variables in addition to the reference set (a + b + c). For the sensitivity analysis, we included all the patients and imputed data that could not be obtained (test not suitable for the age range) using the multivariate imputation approach with chained equations<sup>21</sup> (see Appendix S1). The clusterings were compared with the Jaccard index and Sørensen-Dice coefficient.

## Component and cluster analysis: association with neuropsychological and clinical variables

Components were considered of interest if they explained more than 3.7% of the total variance (equipartition between the 27 reference variables) or were significantly associated with a retained patient cluster. Variables were reported as notably contributing to a component when the 3.7% threshold was exceeded, either with positive or negative correlation: the clinical meaning of the component was more specific when a low number of variables exceeded that threshold. To describe clinical correlates, 17 categorical variables were used as supplementary variables in the PCA (Appendix S1): not included in the PCA calculation, their factor scores were obtained by positioning into the PCA space and projection onto the principal components. Associations between components or variables and PCA-based clusters were assessed with a V-test: association with a variable was considered of interest for a posteriori analysis of power over 80% and medium effect size (for instance the 20% having the largest Cohen's  $d$ ).

## Disentangling visuospatial treatment and elementary reasoning skills

The first component of the reference PCA was highly associated with visuospatial treatment and elementary reasoning (Figure 1), and was the main axis segregating clusters. To better understand the staggering of the clusters along this axis, we computed the mean of four elementary reasoning variables that did not (or mildly) depend on visuospatial treatment, 'M1' (WISC-V similarities, vocabulary, matrix reasoning, and figure weights) and

the mean of five visuospatial treatment variables, 'M2' (WISC-V block design, WISC-V visual puzzles, NEPSY-II block construction, NEPSY-II global design copying, and NEPSY-II arrows), and compared their distribution between the clusters.

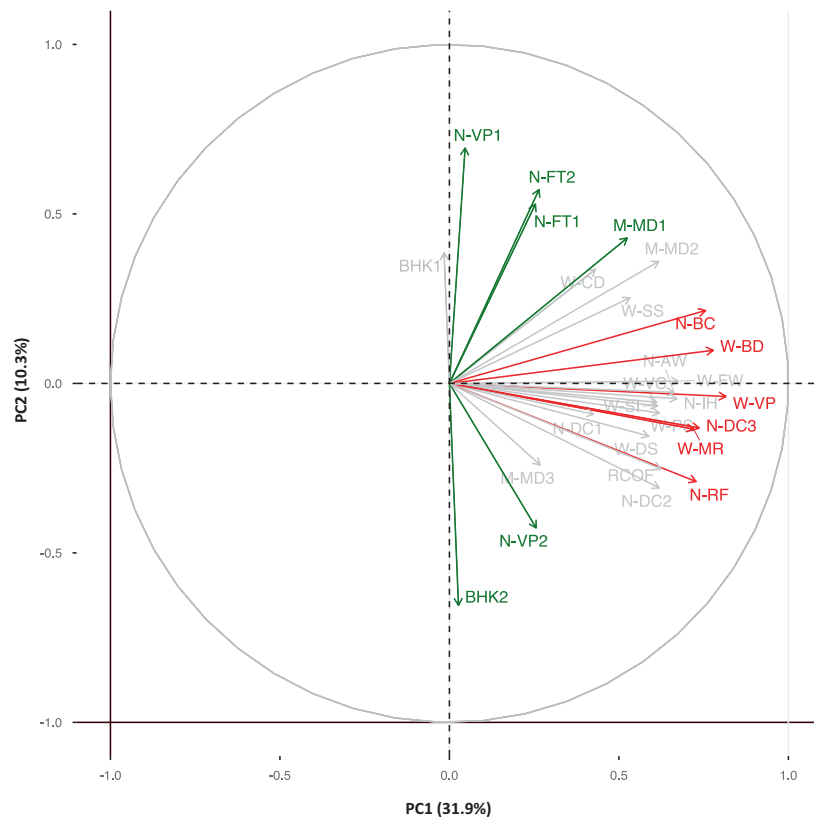
## RESULTS

### General characteristics of the patients

Among the 211 children referred to our centre because of concerns about their motor skills, 46 did not fulfil the diagnostic criteria for DCD (Table S2 for details) and one

declined to participate. We finally enrolled 164 children with DCD (Table 2). Male and left-handed children were over-represented. Twenty-six per cent of the children had a perinatal history without clinical evidence of cerebral palsy; 25% had minor non-specific brain MRI abnormalities, such as enlarged Virchow–Robin spaces or mild white-matter abnormalities (Table S3). The children frequently had developmental motor subsigns: walking acquisition over 15 months (40%), orofacial impairment (33%), and synkinesis (100%). Associated NDDs, mostly ADHD, were frequently observed (51%). None of the children had intellectual disability.

All the children had problems in daily life ascertained by at least one parent-completed questionnaire



**FIGURE 1** The principal component analysis (PCA) correlation circle. PCA simplifies the complexity of a data set with a high number of variables by providing new composite variables (components) that account for a high amount of the total variance. These uncorrelated components define directions of high variability in the data. The variables are illustrated as vectors pointing away from the origin; direction of vectors is determined by component loadings. The angle between the vectors approximates the correlation between the variables. A small angle indicates the variables are positively correlated, an angle of  $90^\circ$  indicates the variables are not correlated, and an angle close to  $180^\circ$  indicates the variables are negatively correlated. The length of the line and its closeness to the circle indicate how well the variable is represented in the two components. The six variables in red and the six in green largely contribute to principal component 1 and 2 (PC1 and PC2) respectively. Along the PC1 axis, patients showed a major increase in visuospatial skills and level of fluid reasoning. PC2 almost exclusively reflected gestural skills with significant loading of numerous variables related to speed or precision. Patients showed greater speed but less precision of gesture along the PC2 axis. BHK 1, BHK speed; BHK 2, BHK quality; M-MD 1, MABC2 manual dexterity 1 (preferred hand and other hand); M-MD 2, MABC2 manual dexterity 2 (bi-manual coordination); M-MD 3, MABC2 manual dexterity 3 (trail); N-IH, NEPSY-II imitating hand positions; N-FT 1, NEPSY-II fingertip tapping (preferred hand); N-FT 2, NEPSY-II fingertip tapping (other hand); N-VP 1, NEPSY-II visuomotor precision (speed); N-VP 2, NEPSY-II visuomotor precision (accuracy); N-AW, NEPSY-II arrows; N-BC, NEPSY-II block construction; N-RF, NEPSY-II route finding; N-DC 1, NEPSY-II design copying (motor); N-DC 2, NEPSY-II design copying (local); N-DC 3, NEPSY-II design copying (global); ROCF, Rey–Osterrieth Complex Figure test; W-SI, WISC-V similarities; W-VC, WISC-V vocabulary; W-MR, WISC-V matrix reasoning; W-FW, WISC-V figure weights; W-VP, WISC-V visual puzzles; W-BD, WISC-V block design; W-DS, WISC-V digit span; W-PS, WISC-V picture span; W-CD, WISC-V coding; W-SS, WISC-V symbol search. Abbreviations: BHK, Concise Evaluation Scale for Children's Handwriting; DTVP-2, Developmental Test of Visual Perception, Second Edition; MABC-2, Movement Assessment Battery for Children, Second Edition; MRI, magnetic resonance imaging; NEPSY-II, Developmental Neuropsychological Assessment, Second Edition; ROCF, Rey–Osterrieth Complex Figure Test for Children; WISC-V, Wechsler Intelligence Scale for Children, Fifth Edition.

**TABLE 2** Characteristics of the 164 patients.

| Characteristics  | n (%)                              |
|--|------------------------------------|
| <b>Demographics</b>  |                                    |
| Socioprofessional category of the mother <sup>a</sup> = 1        | 7 (4.3)                            |
| Socioprofessional category of the mother <sup>a</sup> = 2        | 31 (18.9)                          |
| Socioprofessional category of the mother <sup>a</sup> = 3        | 66 (40.2)                          |
| Socioprofessional category of the mother <sup>a</sup> = 4        | 36 (21.9)                          |
| Socioprofessional category of the mother <sup>a</sup> = 5        | 24 (14.6)                          |
| Age, years:months <sup>b</sup>                                   | 10:3 (9:0–11:2)                    |
| Sex ratio (male:female)  | 5.56:1                             |
| <b>Clinical characteristics and investigations</b>               |                                    |
| Perinatal history without clinical evidence of cerebral palsy    | 42 (26.2)                          |
| Language acquisition delay <sup>c</sup>                          | 27 (16.4)                          |
| Left-handed  | 28 (17.1)                          |
| Minor non-specific brain MRI abnormalities                       | 42/68 (61.8)                       |
| <b>Subtle developmental motor subsigns</b>                       |                                    |
| Orofacial impairment <sup>d</sup>                                | 54 (32.9)                          |
| Walking acquisition <15 m/15–17 m/≥18 m                          | 98 (59.7)/39 (23.8)/27 (16.5)      |
| Synkinesis   | 164 (100)                          |
| <b>Associated disorders</b>                                      |                                    |
| Attention-deficit/hyperactivity disorder                         | 84 (51.2)                          |
| Autism spectrum disorder   | 26 (15.8)                          |
| Reading disability   | 42 (25.6)                          |
| Speech language disorder   | 8 (4.9)                            |
| Tics <sup>e</sup>  | 18 (11.0)                          |
| Sleep disturbances <sup>f</sup>                                  | 49 (29.9)                          |
| Ophthalmological history <sup>g</sup>                            | 80 (48.8)                          |
| <b>School curriculum and professional orientation</b>            |                                    |
| School assistant/repeating a year in primary school <sup>h</sup> | 61 (37.2)/22 (13.4)                |
| Positive DCDQ/positive MABC-2 checklist                          | 157 (95.7)/142 (94.0) <sup>i</sup> |
| <b>WISC-V index mean (SD)/range (minimum–maximum)</b>            |                                    |
| Verbal Comprehension Index                                       | 115.45 (15.23)/71 (84–155)         |
| Visual Spatial Index   | 94.22 (13.93)/71 (67–138)          |
| Fluid Reasoning Index  | 106.83 (13.36)/64 (76–140)         |
| Working Memory Index   | 100.8 (13.16)/71 (67–138)          |
| Processing Speed Index   | 80.37 (12.45)/82 (53–135)          |
| <b>WISC-V defined strength/weakness</b>                          |                                    |
| Verbal Comprehension Index                                       | 110 (67.0)/1 (0.6)                 |
| Visual Spatial Index   | 4 (2.4)/40 (24.4)                  |
| Fluid Reasoning Index  | 55 (33.5)/0                        |

**TABLE 2** (Continued)

| Characteristics   | n (%)                    |
|---|--------------------------|
| Working Memory Index                                      | 16 (9.7)/9 (5.5)         |
| Processing Speed Index                                    | 1 (0.6)/117 (71.3)       |
| <b>Handwriting impairment in the population</b>           |                          |
| BHK < -1.3 SD: speed/quality/both in speed and in quality | 85 (52)/129 (79)/61 (37) |

Abbreviations: BHK, Concise Evaluation Scale for Children's Handwriting; DCDQ, Developmental Coordination Disorder Questionnaire; MABC-2, Movement Assessment Battery for Children, Second Edition; MRI, magnetic resonance imaging; SD, standard deviation; WISC-V, Wechsler Intelligence Scale for Children, Fifth Edition.

<sup>a</sup>Classified from 1 to 5, according to the Institut National de la Statistique et des Etudes Economiques classification of 2016 (1: craftsmen, shopkeepers, company managers; 2: executives and higher intellectual professions; 3: intermediate profession; 4: employees; 5: workers).

<sup>b</sup>Age is expressed as median and interquartile range.

<sup>c</sup>First canonical sentences after 3 years of age.

<sup>d</sup>Following clinical evaluation and language assessment by a speech therapist.

<sup>e</sup>If present for more than 1 year, without a break of more than 3 consecutive months.

<sup>f</sup>Restless leg syndrome ( $n = 2$ ), obstructive sleep apnoea syndrome ( $n = 2$ ), bedtime resistance ( $n = 40$ ), parasomnia ( $n = 4$ ).

<sup>g</sup>Following ophthalmological examination (see Table S3).

<sup>h</sup>Among children aged 11 years or older.

<sup>i</sup>Over 151 children.

(Developmental Coordination Disorder Questionnaire or MABC-2 or both). A significant discrepancy between the Verbal Comprehension Index and Processing Speed Index or Visual Spatial Index was present in 81% and 54% of the children respectively. A combination of Verbal Comprehension Index as a strong point (67%) and Processing Speed Index as a weak point (71%) was found in 51% of the children. A reduced Processing Speed Index mainly accounted for poor performance at the coding subtest. The MABC-2 global score was no more than the 15th centile in 93.3% of the children. Weak visuospatial performance – defined as at least half of the eight visuospatial subscores (WISC-V: visual puzzles, block design; NEPSY-II: local design copying, global design copying, arrows, block construction, route finding; Rey–Osterreith Complex Figure Test for Children or Developmental Test of Visual Perception, Second Edition: spatial relations) less than  $-1$  SD was observed in 41% of the children. One hundred and thirteen children underwent attention testing by Test of Everyday Attention for Children and NEPSY-II because of ADHD symptoms.

### PCA with the reference set of variables

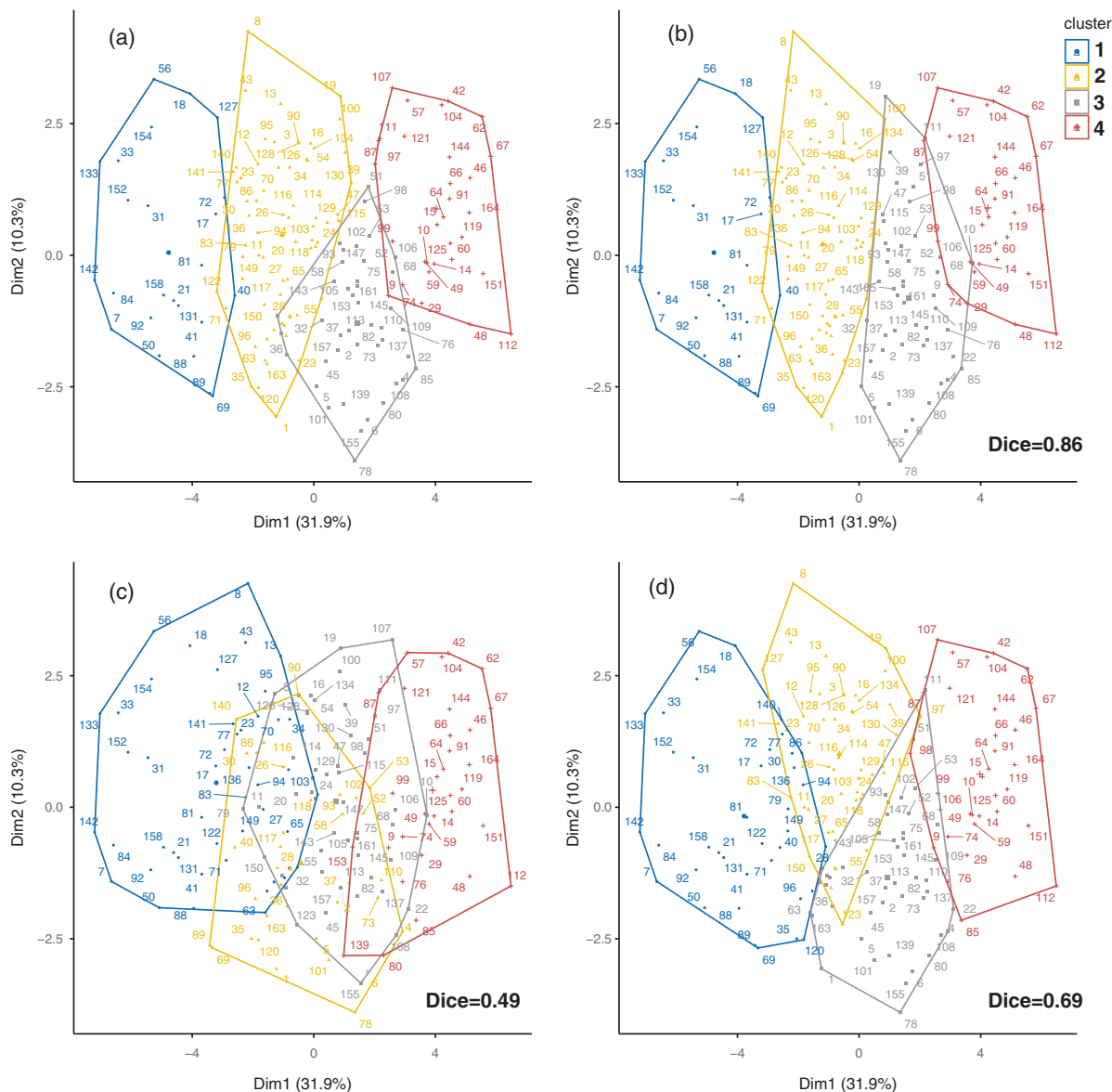
The PCA resulted in five components of interest (Table S4): the first two principal components (PC1 and PC2) accounted for 42% of the interindividual variance, and the five for 61% (Figure 1). About 60% of the variables (16 out of 27) contributed to PC1 (32% of the variance), indicating association with multiple skills. Among the six largest contributions to PC1, five were visuospatial subtests (WISC-V visual puzzles, WISC-V block design, NEPSY-II block construction, NEPSY-II global

design copying, NEPSY-II route finding) and one was a non-verbal inductive reasoning subtest (WISC-V matrix reasoning), with much higher loadings than those of gestural speed and precision subtests or verbal reasoning. Ten variables contributed to PC2 (10% of the variance). PC2 almost exclusively reflected gestural skills with significant loading of numerous variables related to speed (NEPSY-II visuomotor precision, speed, NEPSY-II fingertip tapping, other hand, NEPSY-II fingertip tapping, preferred hand, MABC-2 manual dexterity preferred hand and other hand) or precision (BHK quality,

NEPSY-II visuomotor precision, quality). Patients showed greater speed but less precision of gesture along the PC2 axis. We considered the PC1–PC2 plane as the reference space for analyses.

### Data-driven patient clustering

Unsupervised PCA-based hierarchical clustering with the reference set of variables (Figure 2a) resulted in a four-cluster



**FIGURE 2** Unsupervised hierarchical clustering of 164 children with developmental coordination disorder (DCD): based on psychometric, motor, and visuospatial variables. (a) Cluster 1 and cluster 2: DCD with mixed visuospatial and gestural impairment (severe visuospatial impairment in cluster 1, weakness in visuospatial skills in cluster 2). Cluster 3 and cluster 4: DCD with pure gestural impairment. Marked slowness in cluster 3, with better motor precision than other clusters. Children in cluster 4 are quicker and less precise than those in cluster 3. (b) Second unsupervised stratification strategy with a Gaussian mixture model on the same reference set of variables: only 7% of the children ( $n = 12$ ) changed class; these changes all occurred at the border of two clusters (2 to 3 and 3 to 4). (c) Principal component analysis (PCA) with only ‘a’ psychometric variables: the low Sørensen–Dice coefficient of 0.46 is due to 49% of the children changing cluster ( $n = 81$ ), all between adjacent clusters (54% from cluster 2 to cluster 3 or from cluster 3 to cluster 2, 37% from cluster 2 or 3 to cluster 1 or 4). (d) Adding attentional variables to the reference set (PCA with 38 variables ‘a + b + c’). High Sørensen–Dice overlap coefficient of 0.69, with only 29 children (17%) changing cluster, all between adjacent clusters. Among these 29 children who changed cluster, 22 belong to cluster 2 (15 changed from cluster 2 to cluster 1).

optimum based on PC1–5. All the clusters were strongly associated with PC1, accounting for their staggering along this axis of maximum variance, and three with PC2 segregating clusters 3 and 4. There were 24 patients in cluster 1, 67 in cluster 2, 42 in cluster 3, and 31 in cluster 4. The alternative unsupervised clustering strategy based on a Gaussian mixture model on the same reference set of variables consistently resulted in a four-cluster optimum, with very good overlapping with the PCA-based clustering (Sørensen–Dice coefficient = 0.86) (Figure 2b). Restricting PCA-based clustering to the ‘a’ set of variables (10 WISC-V subtests) resulted in a roughly similar four-cluster structure, but with poor cluster overlap (Figure 2c). Conversely, the overall classification of children with DCD was less sensitive to the addition of the 11 attentional variables to the reference set of variables (‘a + b + c’), with changes restricted to patients near the cluster boundaries (Sørensen–Dice = 0.69) (Figure 2d).

### Correlations between clusters and variables

#### PCA axis and quantitative neuropsychological variables

Children from cluster 1 showed the poorest achievement in almost all the evaluated domains compared with other clusters, with a main deficit in visuospatial skills (Table 3). Children from cluster 2 showed greater achievements than those from cluster 1 and poorer ones than those from clusters 3 or 4, with a weakness in visuospatial skills. These children were quicker in gestural ability but less precise than those from cluster 3. Children from cluster 3 had pure gestural impairment, without significant visuospatial impairment. They had a marked slowness but with a greater fine motor precision compared with other clusters, and fair reasoning abilities. Children from cluster 4 also had pure gestural impairment with the best reasoning and visuospatial skills compared with the three other clusters. They were similar to the patients from cluster 3 but quicker and less precise in their gestural performance.

All the clusters were highly correlated to PC1, with levels of visuospatial skills and fluid reasoning increasing from clusters 1 to 4. The difference between mean elementary reasoning performance and mean visuospatial treatment performance was always significant and roughly similar: around 4 standard points better for reasoning (Figure 3).

#### Developmental and clinical qualitative variables

Sex, laterality, socioprofessional category (maternal occupation), delayed oral language acquisition, and most associated disorders (NDDs, tics, sleep disturbances, ophthalmological disorders) were evenly distributed among the four clusters, except for reading disability (less frequent in cluster 1). Late walking acquisition (>15 months), perinatal history, and orofacial impairment were significantly linked to cluster 1, as was

the need for a school assistant and repeating a school year. On the contrary, the absence of perinatal history was associated with cluster 2, the absence of orofacial impairment and no need for a school assistant with cluster 3, and the absence of imaging abnormalities with cluster 4 (Tables 3 and S5).

## DISCUSSION

We propose a data-driven classification of four DCD subtypes distinguishing mixed visuospatial and gestural disorders (clusters 1 and 2) from pure gestural disorders predominantly affecting either gestural speed (cluster 3) or precision (cluster 4). Elementary reasoning and visuospatial treatment abilities remained strongly linked, so that children with marked visuospatial impairment had the poorest achievements in almost all the evaluated domains (cluster 1), but always with a differential in favour of reasoning abilities. These findings are relevant in clinical practice as they may help refine the diagnosis into discrete subtypes, formulate a prognosis, and guide a management strategy. They could also provide a relevant framework with homogeneous subgroups of patients for research about DCD pathogenesis.

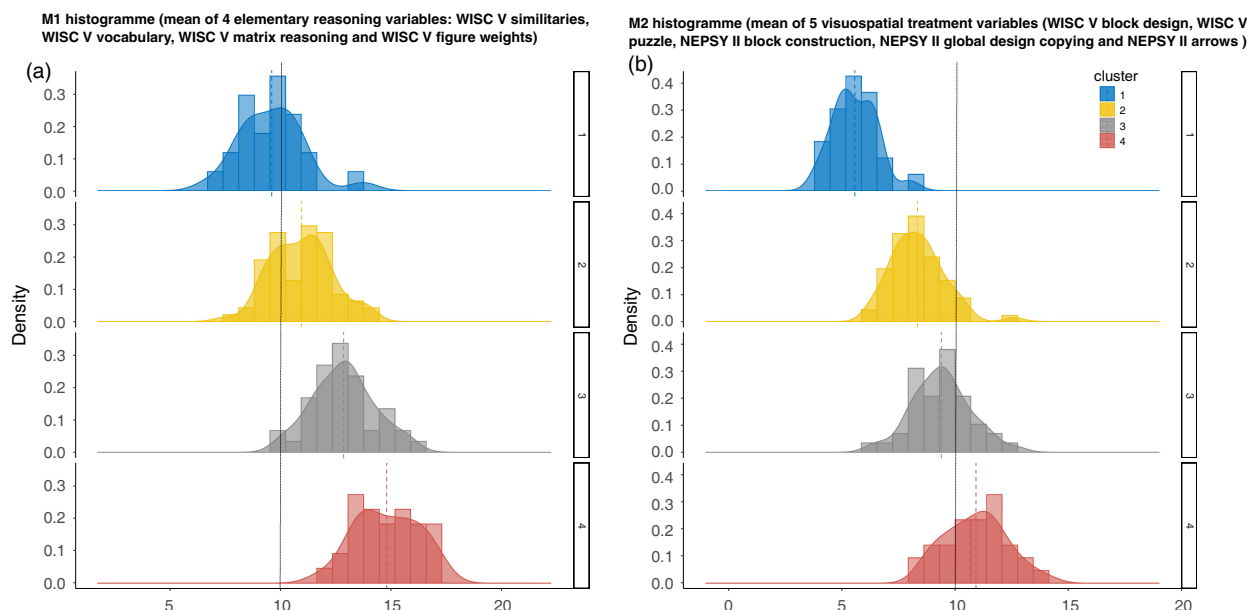
The strengths of our study are (1) the large sample size; (2) the use of rigorous criteria for DCD diagnosis and comprehensive clinical characterization; (3) the use of multiple standardized evaluation tools including questionnaires and tests; (4) the focus on visuospatial and fine motor skills that are critical for DCD pathogenesis and determining daily life disability; and (5) the consistency of the findings when the same reference set of variables was analysed with two different unsupervised stratification strategies. Some limitations deserve to be mentioned. We chose not to exclude patients with associated NDDs such as ADHD. This is disputable as associated NDDs are present in most children with DCD and can be seen as part of the DCD clinical spectrum.<sup>1</sup> We took care to characterize the NDDs and carefully considered their potential distribution over the clusters. We included patients with a perinatal history without evidence of cerebral palsy as they represent a significant proportion of unselected children with DCD.<sup>22</sup> Despite the large number of patients, our sample might not be fully representative of a random population of children with DCD owing to possible referral bias. First, male children were largely over-represented (85%), possibly reflecting that females are less likely to be referred, as demonstrated in other NDDs such as ADHD.<sup>23</sup> Second, none of the children had mild intellectual disability or motor skills deficits not better explained by intellectual developmental disorder (diagnostic criteria from DSM-5).<sup>17</sup> Similarly, children with an associated speech/language disorder were under-represented in our population.<sup>24</sup> This recruitment bias was related to the local organization of care with dedicated referral centres for developmental motor problems (our team), intellectual disability, and speech/language disorders. Despite these possible recruitment biases, the general characteristics of our patients were consistent with those of previous series.<sup>25</sup>



**TABLE 3** Contributing components, main variables contributing for each cluster, and motor and visuospatial subtest results in each cluster.

|  | Cluster 1 <i>n</i> = 24  | Cluster 2 <i>n</i> = 67   | Cluster 3 <i>n</i> = 42   | Cluster 4 <i>n</i> = 31   |
|--|--|---|---|---|
| Contributing components in descending order of importance    | 1, 5, 3  | 1, 3, 2, 5  | 2, 4, 1   | 1, 2, 4   |
| Main <sup>a</sup> variables contributing (Cohen's <i>d</i> ) | NEPSY-II arrows (1.54)<br>NEPSY-II route finding (1.5)<br>NEPSY-II block construction (1.4)<br>NEPSY-II global design copying (1.4)<br>WISC-V visual puzzles (1.29)<br>WISC-V block (1.24) | WISC-V similarities (0.44)<br>WISC-V vocabulary (0.4)<br>WISC-V picture span (0.36)<br>WISC-V visual puzzles (0.34)<br>WISC-V matrix (0.32)<br>WISC-V figure weights (0.31) | NEPSY-II visuomotor precision speed (0.74)<br>NEPSY-II route finding (0.63)<br>BHK speed (0.6)<br>NEPSY-II visuomotor precision accuracy (0.61)<br>BHK quality (0.57)<br>NEPSY-II local design copying (0.58) | WISC-V block (1.23)<br>WISC-V picture span (1.22)<br>WISC-V matrix (1.06)<br>WISC-V similarities (1.07)<br>WISC-V visual puzzles (1.03)<br>WISC-V figure weights (1.08) |
| Supplementary categorical variables                          | Perinatal history (0.48)<br>Orofacial impairment (0.52)<br>School assistant (0.61)<br>Repeating a school year (0.36)<br>Walking acquisition (0.54)<br>Reading disability (0.40)            | Perinatal history (0.22)<br>School assistant (0.33)   | Orofacial impairment (0.3)<br>School assistant (0.33)   | Absence of MRI abnormalities (0.33)   |
| Complementary subtests: results <sup>b</sup>                 |  |   |   |   |
| Speed  | NEPSY-II VP 1<br>BHK Speed   | 8.76 (2.42)<br>-1.08 (1.02)   | 5.92 (1.78)<br>-1.88 (0.98)   | 9.17 (2.66)<br>-0.87 (1)  |
| Speed and quality  | NEPSY-II FT 1<br>NEPSY-II FT 2   | 6.5 (3.08)<br>6.04 (2.66)   | 7.32 (2.03)<br>7.2 (2.52)   | 8.52 (2.02)<br>8.56 (2.28)  |
| Quality  | MABC-2 MD 1<br>MABC-2 MD 2<br>NEPSY-II VP 2  | 3.92 (3.27)<br>2.29 (2.07)<br>3.58 (0.89)   | 6.17 (2.82)<br>5.52 (2.8)<br>4.61 (0.62)  | 9.32 (2.69)<br>7.74 (2.49)<br>4.057 (1.27)  |
| Visuospatial   | NEPSY-II DC 1<br>MABC-2 MD 3<br>BHK quality<br>NEPSY-II DC 2<br>NEPSY-II DC 3<br>NEPSY-II Arrows<br>NEPSY-II RF<br>NEPSY-II BC<br>DTVP-2 or ROCF   | 4.67 (2.3)<br>2.17 (1.76)<br>-2.33 (1.53)<br>2.67 (1.97)<br>3.42 (2.12)<br>5.33 (3.17)<br>1.97 (0.87)<br>3.13 (1.57)<br>-4.17 (3.39)  | 8.08 (2.13)<br>5.12 (3.44)<br>-1.64 (1.33)<br>8.03 (2.89)<br>9.13 (2.52)<br>11.54 (2.14)<br>3.78 (0.32)<br>7.87 (2.09)<br>-0.34 (1.6)   | 7.74 (2.74)<br>4.29 (2.42)<br>-3.01 (1.47)<br>8.26 (2.84)<br>10.52 (2.5)<br>12.58 (1.96)<br>3.81 (0.47)<br>10.32 (2.3)<br>-0.04 (1.84)                                  |

Abbreviations: BHK, Concise Evaluation Scale for Children's Handwriting; DTVP-2, Developmental Test of Visual Perception, Second Edition; MABC-2, Movement Assessment Battery for Children, Second Edition; MRI, magnetic resonance imaging; NEPSY-II, Developmental Neuropsychological Assessment, Second Edition; ROCF, Rey-Osterreith Complex Figure Test for Children; WISC-V, Wechsler Intelligence Scale for Children, Fifth Edition. <sup>a</sup>The six largest Cohen's *d* values. Bold type, quantitative variables with positive contribution and qualitative variables with positive correlation; italic type, negative contribution. <sup>b</sup>Mean (SD) in age-standardized scores (BHK and DTVP-2 or ROCF). NEPSY-II VP 1/VP 2, visuomotor precision speed/accuracy; NEPSY-II DC 2/DC 3, design copying motor/local/global; NEPSY-II FT 1/FT 2, fingertip tapping, preferred hand/other hand; NEPSY-II RF, route finding; NEPSY-II BC, block construction; MABC-2 MD 1/2/3, MABC-2 manual dexterity preferred hand and other hand/bi-manual coordination/trail.



**FIGURE 3** Disentangling elementary reasoning and visuospatial skills along first principal component axis segregating the four clusters of patients. Comparison between (a) M1 (mean of elementary reasoning subtests) and (b) M2 (visuospatial subtests) distributions among clusters. Cluster 1 was mainly characterized by severe visuospatial treatment deficit (50th centile = 5.4) with medium reasoning level (50th centile = 9.75). Cluster 2 was the most homogeneous cognitive profile, with relative weakness in visuospatial skills (50th centile = 8.2) compared with a medium level of reasoning (50th centile = 11). All children with M2 less than 7 belonged to clusters 1 (23 children) and 2 (six children). The only child with an M1 less than 7 belonged to cluster 1. Clusters 3 and 4 were characterized by a level of elementary reasoning above the norm (50th centile = 13.0 in cluster 3, 15 in cluster 4) without substantial visuospatial impairment (50th centile = 9.4 in cluster 3, 50th centile = 11 in cluster 4). Verbal reasoning was in the high range of the norm or above the norm in all children (similarities mean 11, standard deviation [SD] 2.4, in cluster 1; 11.8, SD 2.0, in cluster 2; 13.9, SD 2.6, in cluster 3; 16.2, SD 2.3, in cluster 4).

The typical cognitive profile was characterized by a large discrepancy between the Verbal Comprehension Index (strong point) and Processing Speed Index (weak point). As previously reported,<sup>26,27</sup> we believe that the significant difference between Verbal Comprehension Index and Processing Speed Index associated with poor performance at the coding subtest of the Processing Speed Index is highly suggestive of a diagnosis of DCD. Likewise, 90% of our patients had handwriting impairment, of whom 52% had reduced speed, and 37% both reduced quality and speed, in line with recent findings.<sup>28</sup>

On the basis of a robust methodological approach without any supervision, we provide experimental data strongly supporting the distinction between mixed visuospatial and gestural difficulties and pure gestural disorders. In our study, 41% of the children, all belonging to clusters 1 and 2, had visuospatial impairment or weakness. This proportion is close to that found in the literature (37–58%).<sup>5,8,10–12,14</sup> The concept of children with a pure motor form of DCD is relatively new and their characteristics have been poorly studied.<sup>11,14</sup> Our finding that DCD characterized by pure gestural impairment with alteration of precision is distinct from DCD characterized by pure gestural impairment with reduced speed, is novel, and could help refine diagnosis. Such a distinction is meaningful as it may account for the clinical observation that children with DCD often have to favour precision over speed or the reverse, because they are unable to combine both<sup>29</sup> (Table 3). A recent study

identified a DCD subgroup with difficulties restricted to motor problems,<sup>11</sup> such as the children belonging to clusters 3 and 4. Although the focus of investigation and the tools used for motor evaluation were different from ours, we suspect an overlap between the subgroups identified by the two studies.

Patients from cluster 1 with a main deficit in visuospatial skills showed the poorest achievements in most evaluated domains compared with other clusters. This particular profile of children with DCD has been previously reported.<sup>5,8,12</sup> It may indicate that marked visuospatial alteration is a marker for a more severe and diffuse NDD. This would be consistent with the strong association between visuospatial treatment and elementary reasoning abilities in our population, but always with a large differential in favour of reasoning abilities. Hardly any of the children with DCD in our study had visuospatial skills above the norm. Thus, a relative weakness of visuospatial skills is probably a marker of DCD,<sup>26,30</sup> but having normal visuospatial abilities does not rule out a DCD diagnosis.

The overall structure of the DCD subgroups remained roughly unchanged when the analysis was based purely on the WISC-V. However, dedicated tests targeting visuospatial and motor skills are mandatory to avoid misclassification. We found a 51.2% overlap between DCD and ADHD, a result similar to the estimation of 50% found in the literature.<sup>31</sup> Although frequent, neither ADHD nor other NDDs influenced the classification.

DCD has a strong impact on school performance: 35.7% of the children had human assistance in class, and 13.4% of the children aged 11 years or older had already repeated a class in primary school, a high frequency reported in previous studies.<sup>11</sup> The significant impact of DCD on educational achievement persists after adjustment for sex, socioeconomic status, and IQ.<sup>32</sup>

Proper classification of DCD is highly relevant in clinical practice both as an indicator of prognosis and to guide the management of children with DCD taking into account their neuropsychological profile. Although not directly derived from our data, we can speculate about the usefulness and meaningfulness of our findings in clinical practice. Children with a combination of marked visuospatial and gestural impairment are most likely to experience difficulties at school and are thus expected to require more social support and pedagogical measures. Among the children with a pure gestural disorder, rehabilitation strategy and academic accommodation may differ between those with predominant slowness and those with predominant gesture imprecision.

#### CONFLICT OF INTEREST STATEMENT

The authors have stated that they had no interests that might be perceived as posing a conflict or bias.

#### DATA AVAILABILITY STATEMENT

Data available on request from the authors

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## SUPPORTING INFORMATION

The following additional material may be found online:

**Appendix S1:** Statistical analysis.

**Table S1:** Reported DCD subgroups in previous studies.

**Table S2:** Children not included in the study because of an alternative diagnosis.

**Table S3:** Perinatal history, MRI abnormalities and ophthalmologic history.

**Table S4:** The most characteristic variables according to each component.

**Table S5:** Descriptive statistics of the categorical variables (supplementary or illustrative variables) for each separate cluster.

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